

1 **Patterns of change and stability in caregiver burden and life satisfaction from 1 to 2**
2 **years after severe traumatic brain injury: A Norwegian longitudinal study**

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46 **Abstract**

47 **Objectives:** To assess burden and life satisfaction in caregivers of patients with severe
48 traumatic brain injury (sTBI) at 1 and 2 years post-injury, to examine if change in burden can
49 be predicted by caregiver and patient demographics, patient's functional status, caregiver's
50 social network or caregiver's level of burden at 1 year.

51 **Design:** Prospective national multicenter study. Self-report from caregivers, patient data
52 collected from a national cohort study on patients with sTBI.

53 **Participants:** 80 caregivers.

54 **Main outcome measure:** The Caregiver Burden Scale (CBS), life satisfaction.

55 **Results:** The total caregiver burden increased significantly between years 1 and 2 post-injury
56 ($p=0.04$). Life Satisfaction was significantly lower at 2 years follow-up ($p=0.03$) than at 1
57 year. Thirty percent of the caregivers reported an increased burden, 55% were stable, and 15%
58 had a decrease in burden between the two follow-up times. Logistic regression analyses
59 revealed that experiencing loneliness was an independent predictor of increased burden from
60 1 to 2 years post-injury ($OR=4.35$, $p<0.05$).

61 **Conclusions:** The results demonstrate a need for long-term follow-up of patients and
62 caregivers that particularly focuses on professional support to relieve caregiver burden and
63 risk of loneliness or social isolation. This group may benefit from additional follow-up
64 interventions tailored to their needs.

65

66 **Keywords:** caregiver burden, life satisfaction, traumatic brain injury, social network

67

68 INTRODUCTION

69 Severe traumatic brain injury (TBI) is a major public health challenge, as it is one of the most
70 common causes of death and disability in young adults (Sundstrom, Sollid, & Wester, 2005).
71 TBI is considered a “silent epidemic” because society seems to be unaware of the magnitude
72 and socioeconomic consequences of the injury (Roozenbeek, Maas, & Menon, 2013). Family
73 members serve a critical function as an extension of the health care system, but they lack
74 formal training and support to care for persons with severe TBI (Ramkumar & Elliott, 2010).
75 Caregiver burden has been defined as the social, psychological, physical, economic or
76 emotional strain that caregivers may experience (George & Gwyther, 1986). This burden may
77 also accumulate when providing care to an individual over a long period of time (Kasuya,
78 Polgar-Bailey, & Takeuchi, 2000). The concept of life satisfaction has been described as a
79 conscious cognitive judgment that compares one’s personal life circumstances with a set of
80 self-imposed standards of needs or wants (Diener, Oishi, & Lucas, 2003; Fugl-Meyer, Melin,
81 & Fugl-Meyer, 2002). In the present study, life satisfaction was considered the caregiver’s
82 view of satisfaction with his or her life.

83 In the past decades, a substantial amount of literature on the impact of severe TBI on
84 family members has pointed to a high level of caregiver burden, diminished life satisfaction,
85 unmet family needs, adaptation to new roles, anxiety, depression, social isolation, emotional
86 difficulties and challenges with family functioning (Bayen et al., 2012; Braine, 2011; Doser &
87 Norup, 2016; Douglas & Spellacy, 1996; Gillen, Tennen, Affleck, & Steinpreis, 1998;
88 Godwin & Kreutzer, 2013; Kolakowsky-Hayner, Miner, & Kreutzer, 2001; L. A. Livingston
89 et al., 2010; Manskow et al., 2014; McKinlay, Brooks, Bond, Martinage, & Marshall, 1981;
90 Norup, Siert, & Lykke Mortensen, 2010; Norup, Welling, Qvist, Siert, & Mortensen, 2012;
91 Oddy, Humphrey, & Uttley, 1978). When a close relative experience lower burden, less
92 anxiety and a good health, this is shown to have a positive impact on the person with severe

93 TBI (O'Neill & Carter, 1998). The close relation between the health and wellbeing of the
94 injured and the relatives makes it important to identify interventions to improve the situation
95 for both the patient and the relative/caregiver (Verhaeghe, Defloor, & Grypdonck, 2005).

96 Within the concept of caregiver burden or strain, several longitudinal studies have
97 been reported, with inconsistent findings: A study from New Zealand on caregiver burden in
98 52 caregivers of patients with severe TBI 6 and 12 months post-injury found no change in
99 burden over time (Marsh, Kersel, Havill, & Sleight, 2002), whereas Brooks et al. found greater
100 strain in caregivers 5 years post injury than at 1 year (Brooks, Campsie, Symington, Beattie,
101 & McKinlay, 1986). The French Paris-TBI study measured caregiver burden 1 and 4 years
102 post injury and reported a decrease in burden from 1 to 4 years after the injury (Bayen et al.,
103 2014; Bayen et al., 2012). Livingston and colleagues investigated 57 caregivers of patients
104 with severe TBI 3, 6 and 12 months post injury and found a high level of perceived burden at
105 all follow-up times, with a slight increase in burden at 6 and 12 months. Sander et al. found a
106 decrease in perceived burden during the first year post injury, investigating 69 caregivers at 3
107 different follow up times during the first year post injury (M. G. Livingston, Brooks, & Bond,
108 1985; Sander, High, Hannay, & Sherer, 1997). Nearly all prospective studies following the
109 same sample have been conducted during the first year post-injury.

110 Regarding predictors of burden, there is strong evidence that burden is correlated with
111 patient's neurobehavioral problems (Bayen et al., 2014; Bayen et al., 2012; Kreutzer,
112 Gervasio, & Camplair, 1994). Other studies have found that a patient's level of functioning is
113 connected to the caregiver's reported burden or strain (Boycott, Yeoman, & Vesey, 2013;
114 Brooks et al., 1986; Kreutzer et al., 2009; Manskow et al., 2014). Social network is a broad
115 concept that may include all social resources available to an individual; interpersonal ties,
116 health resources and professional support. The concept can also be defined more narrow as
117 the individuals' social network (A. G. Anke et al., 1997; Gottlieb & Bergen, 2010). A poor

118 social network have previously been shown to predict caregiver burden by Manskow et al.,
119 reporting that caregivers with a high burden 1 year post-injury were more lonely and met with
120 friends and family less frequently (Manskow et al., 2014). Knight and colleagues found no
121 relationship between caregiver burden and lack of a social network (Knight, Devereux, &
122 Godfrey, 1998), while other studies indicated that a lack of social network in terms of
123 perceived social support were related to a higher caregiver burden (Hanks, Rapport, &
124 Vangel, 2007; Kreutzer et al., 2009).

125 Few studies have reported on life satisfaction measured at several time points in
126 family members of patients with TBI. Kolakowsky-Hayner found diminished life satisfaction
127 in family members to patients with TBI several years post injury compared to the reported
128 level pre injury (Kolakowsky-Hayner et al., 2001). A large longitudinal study from the USA
129 reported on life satisfaction in family caregivers of patients with TBI 1 and 2 years post-injury
130 and found no changes in life satisfaction scores between years 1 and 2 (L. A. Livingston et al.,
131 2010). These findings are supported by a cross-sectional study by Kreutzer et al. that showed
132 no difference in life satisfaction compared to normative data 1, 2 or 5 years post injury
133 (Kreutzer et al., 2009). A simultaneous study of caregiver burden and life satisfaction in
134 caregivers of patients with stroke, found that the caregivers reporting lower life satisfaction
135 also reported significantly more caregiver burden.(Bergstrom, Eriksson, von Koch, & Tham,
136 2011)

137 To our knowledge, the present study is the first to analyze simultaneous changes in
138 caregiver burden and life satisfaction over time in a population of family members of patients
139 with severe TBI. In addition, a unique aspect of our study is the regression analysis of the
140 predictors of change in burden from 1 to 2 years post-injury. Based on the above literature
141 review of caregiver burden and life satisfaction over time in family members of patients with
142 severe TBI, we hypothesized that (1) the level of burden would be stable from 1 to 2 years

143 post-injury, (2) the level of burden would be associated with caregivers' satisfaction with life,
144 and (3) that an eventual change in burden would be predicted by the functional impact of
145 severe TBI and caregivers' social networks.

146

147 **METHODS**

148 **Design**

149 The study was a prospective, population-based, multicenter cohort study covering all regions
150 of Norway. We included adult family members or acquaintance of patients (≥ 16 years old)
151 with severe TBI injured between January 2009 and December 2011. All patients were
152 admitted to one of the four trauma referral centers in Norway. The inclusion criteria for the
153 participants in the present study were as follows:

- 154 • Closest family member or acquaintance of a patient included in the national
155 multicenter study on severe TBI (i.e., unседated Glasgow Coma Scale (GCS) score of
156 8 or less during the first 24 hours post-injury) (Andelic et al., 2012).
- 157 • Participant was listed as the patient's closest family member or acquaintance either by
158 the patient and/or in the patient's medical journal.
- 159 • Age ≥ 18 years.

160

161 **Data collection**

162 The regional project coordinator at each trauma center identified a close family member or
163 acquaintance. Written informed consent was required from both the person with severe TBI
164 and all participating family members. The family member consented on behalf of the patient
165 if he/she was unable to give consent due to cognitive impairment. Family members were then

166 contacted by telephone and informed about the study by the coordinator at the University
167 Hospital of North Norway (responsible for the database). The questionnaires and informed
168 consent form were sent by mail, and data were collected at 12 and 24 months post-injury. All
169 participants had the opportunity to withdraw from the study at any time. The study was
170 approved by the Committee for Medical Research Ethics for Southeast Norway number
171 2009/702. Data on patients were collected through the national multicenter patient study
172 (Andelic et al., 2012).

173

174 **Participants**

175 In the present study we choose to name the closest family member or acquaintance as “family
176 member”. As seen in Figure 1, a total of 171 family members were identified and contacted
177 for possible participation in the present study. At 1 year, 119 family members completed the
178 questionnaire, and 80 participated at both 1 and 2 years post-injury, giving a response rate of
179 70% and 47%, respectively (Figure 1).

180

181  Insert Figure 1 here

182

183 The characteristics of the family members not participating were not available. Patient
184 characteristics did not differ between those participating and non-participating, except the
185 proportion of male patients was higher among the participating group. Descriptive data for the
186 family members and the patient-related variables are presented in Table 1. There were no
187 statistically significant differences between participants at 1 year and participants at both 1
188 and 2 years post-injury. Around 75 % of family members were female, and nearly half were

189 married/cohabitant to the patient. We included only family members who responded at both 1
190 and 2 years (n=80) for the further analysis.

191

192 Insert Table 1 here

193

194 **Family member measures at 1 and 2 year follow-up**

195 A structured self-report questionnaire was used to obtain all information from the family
196 members. The Caregiver Burden Scale (CBS) was used to assess the family members burden.
197 The questionnaire is a generic 22-item scale developed to measure different dimensions of a
198 caregiver's subjective burden (Elmståhl, Malmberg, & Annerstedt, 1996). It has previously
199 been used to assess burden in caregivers of patients with stroke and dementia as well as in
200 caregivers of patients with severe TBI (Andren & Elmstahl, 2005; Dahlrup, Nordell, Andren,
201 & Elmståhl, 2011; Manskow et al., 2014). The CBS contains 5 subscales, representing each
202 dimension of the scale: general strain (8 items), isolation (3 items), disappointment (5 items),
203 emotional involvement (3 items) and environment (3 items). The construct validity and
204 internal consistency of the scale have been described previously, yielding Cronbach's alpha
205 coefficients of 0.70 to 0.87 for all subscales except for environment (0.53) (Elmståhl et al.,
206 1996). Elmståhl et al. (1996) also reported good test-retest reliability, with kappa values
207 ranging from 0.89-1.00 for all subscales except environment (0.69) (Elmståhl et al., 1996).
208 The items are scored from 1 to 4 (1; not at all; 2, seldom; 3; sometimes, 4; often), and the total
209 score for the whole scale ranges from 22-88. A high score indicates a high burden. Because
210 the subscales consist of a different number of questions, an index score was calculated by
211 using the sum score of each subscale divided by the total number of items. An index score of

212 1.0-1.99 indicated a low burden, 2.0-2.99 a moderate burden, and 3.0-3.99 a high burden
213 (Bergstrom et al., 2011; Elmståhl et al., 1996).

214 Life satisfaction was measured with one global item: “Overall, how satisfied are you
215 with your life right now?” The item was rated on a 5-point ordinal scale: 1 (very dissatisfied),
216 2 (dissatisfied), 3 (neither satisfied nor dissatisfied), 4 (satisfied) or 5 (very satisfied). This
217 item has been used and has shown validity previously for caregivers after traumatic brain
218 injury (Kolakowsky-Hayner et al., 2001). Based on previous results, two questions related to
219 the family member’s personal social network were included in the analysis (Manskow et al.,
220 2014). One questions tapped the social network quantity: “How often do you meet friends and
221 relatives with whom you do not live, for example, visits each other`s home, go out together,
222 talk on the phone?” (once a week, less than once a week but at least once a month, and less
223 than once a month). The other question assessed an aspect of the quality of their social
224 network that taps social support: “Do you ever feel lonely?” (never or rarely, sometimes,
225 often) (A. G. Anke et al., 1997).

226

227 **Patient outcome measures at baseline and 1 year follow-up**

228 Acute injury severity was assessed by the most commonly used and previously validated
229 scale: the abbreviated injury severity scale (AIS) (AAAM.org;, 2008). The AIS head score
230 was used in the present study to indicate the severity of the brain injury; a higher severity
231 score indicated a progressively more severe injury. The Glasgow Coma Scale (GCS) was used
232 to assess the patient’s level of consciousness in the acute phase of the TBI, and the lowest
233 GCS score within the first 24 hours was registered (Teasdale & Jennett, 1974). The Glasgow
234 Outcome Scale Extended (GOSE) was scored with the structured interview and used to
235 evaluate the patient’s functional level at the 1 year follow-up (Wilson, Pettigrew, & Teasdale,

236 1998). The GOSE categorizes patient status into one of eight categories: dead (1), vegetative
237 state (2), lower severe disability (3), upper severe disability (4), low to moderate disability
238 (5), upper to moderate disability (6), low to good recovery (7), and upper good recovery (8).

239

240 **Statistical analysis**

241 The Statistical Package for Social Sciences (SPSS) for Windows version 23.0 was used for
242 statistical analyses. The descriptive data are presented as the mean and standard deviation or
243 as proportions of subjects. Cross-tabulations with χ^2 -tests were performed for nominal data.
244 Assumptions of a normal distribution were visually inspected and tested with a skewness test
245 statistic. Non-parametric statistical analysis was applied when the data were not normally
246 distributed. The subscales Emotional Involvement and Environment of the CBS were skewed
247 in a positive direction. Independent sample t-tests were used to compare the means between
248 two or more groups. Paired sample t-tests or Wilcoxon Signed rank tests were used as the
249 parametric and non-parametric tests, respectively, to compare the CBS score at the 1 and 2
250 year follow-up. If there were 1 (or 2) missing data point(s) on the CBS, the data were replaced
251 with the caregiver's mean value on each subscale. Participants who had more than 2 missing
252 data points on the CBS scale were excluded.

253 We investigated how many participants in each group had a stable, improved or
254 worsened CBS sum score between 1 and 2 years. At first, a distribution-based method was
255 used to calculate the standard error of measurement (SEM, based on Cronbach's alpha and the
256 observed variance statistic), which indicated the smallest raw score change that reflected a
257 true change and not measurement error. A difference of at least one SEM has been used to
258 designate the minimal clinically important difference (Copay, Subach, Glassman, Polly, &
259 Schuler, 2007). However, we chose a stricter definition and required 2 SEM, which in
260 practice represented one-half the SD of the CBS total score, also used to indicate the minimal

261 clinically important difference (Norman, Sloan, & Wyrwich, 2003). The total CBS score at 1
262 and 2 years were inspected for each participant and differences of at least 2 SEM noted.

263 A chi-square test was performed to investigate the longitudinal change in caregiver
264 burden between 1 and 2 years, dichotomizing the participants into two groups at 1 year post-
265 injury: (1) low burden and (2) moderate to high burden. A binary logistic regression analysis
266 with backward stepwise regression was then conducted to assess predictors of worsened
267 caregiver burden from 1 to 2 years post-injury. Co-linearity was checked and only variables
268 with correlation coefficients <0.7 were entered together in the regression analyses. The CBS
269 dependent variable was coded as 0-stable/improved or 1-worsened.

270 Independent variables entered were for patients: age, gender, education, and GOSE;
271 for caregivers: gender, relation to patient, education, time spent with patient, and frequency of
272 meeting friends/family and loneliness measured at 1 year post-injury. First univariate analyses
273 were conducted to analyse the association between each independent and the dependent
274 variable. Then all variables were entered into the multivariate logistic regression analysis with
275 the Enter method and backward removal of insignificant variables. The results are presented
276 as adjusted odds ratios (ORs). Model fit was investigated by applying the Hosmer and
277 Lemeshow test, and the amount of explained variance in the outcome was investigated using
278 Nagelkerke's R^2 . The level of significance was set at $p < 0.05$.

279

280 **RESULTS**

281 At 1 year post-injury 12.5 % of caregiver experienced a high caregiver burden, 37.5 % a
282 moderate and 50 % a low burden. The mean total CBS sum score (Table 2) showed a
283 statistically significant increase in burden at 2 years compared with 1 year post-injury (paired
284 t-test, $p < 0.05$), and thereby rejecting our hypotheses of no change in CBS score between these

285 time-points. Supplementary analyses to identify subscales with significant changes showed
286 increases in the CBS subscales disappointment, emotional involvement and environment
287 increased significantly (Table 2). A proportion of 30% of the caregivers reported an increased
288 burden, 55% a stable burden and 15% a decrease in burden from 1 to 2 years post-injury.

289

290 Insert Table 2 here

291

292 Life satisfaction scores decreased significantly with a mean score of 3.7 (SD 0.85) at 1 year
293 to a mean score of 3.5 (SD 0.96) at 2 years post-injury ($p < 0.05$, Wilcoxon signed rank test).
294 As indicated in Figure 2, the proportion of caregivers scoring “very satisfied” decreased from
295 20% to 13% with a comparable increase in individuals dissatisfied with life as a whole.

296

297

298 Insert Figure 2 here

299

300 Testing our hypotheses nr. 2, the correlations between the total CBS scores and the Life
301 satisfaction scores were strongly negative at both 1 and 2 years post-injury (Spearman's
302 correlation = -.79 and -.79, respectively, $p < 0.001$ for both).

303

304 **Longitudinal individual changes in Caregiver Burden scores**

305 The CBS sum score at 1 year post-injury had a Cronbach's alpha of 0.96. The formula for the
306 standard error of measurement (SEM) led to 1 SEM equaling a change of 0.15 points. Hence a
307 change of 0.30 points or more (2 SEM) defined a minimal clinically important difference
308 between 1 and 2 years post-injury. Using 2 SEM individual changes in total CBS scores from
309 1 to 2 years post-injury are shown in Table 3 trichotomised into better, same and worse. The
310 longitudinal changes in CBS score were statistically significant for both those reporting a low
311 degree of burden at the 1-year follow-up, and those reporting an initially moderate/high
312 degree of burden ($p < 0.05$). A larger degree of change was observed in the moderate/high
313 burden group according to the reported p-values. By inspecting the cell numbers describing
314 the change, 53% (10 of 19) and 82% (14 of 17) of those changing did so in a negative
315 direction in the low and moderate/high burden groups, respectively (Table 3).

316

317 Insert Table 3 here

318

319 **Predictors of an increased caregiver burden from 1 to 2 years post-injury**

320 The analyses of predictors of worsened CBS from 1 to 2 years post-injury is shown in Table
321 4. In the univariate analyses p-values < 0.1 were observed for a low GOSE score (2-5),
322 caregiver male gender and caregiver feeling lonely often. Since the association between
323 worsened total CBS score and low GOSE score was marginally significant ($p = 0.067$), it was
324 not reported further. The final model retained gender ($p = 0.11$) and loneliness ($p = 0.04$),
325 explaining 12% (Nagelkerke R^2) of the variance in increase in burden and correctly
326 classifying 72.5% of the cases. The Hosmer and Lemeshow test indicated a good model fit
327 (chi-squared: .54, $df = 1$ and $p = .46$). As seen in Table 4, feeling of loneliness at 1 year post-

328 injury was the only statistically significant predictor of an increase in caregiver burden from 1
329 to 2 years post-injury in this model.

330 Insert Table 4 here

331

332 **DISCUSSION**

333 The present study found a significant increase in caregiver burden and a decrease in life
334 satisfaction at 2 years compared to 1 year post-injury, rejecting our hypotheses of no change
335 in CBS score between these time-points. The level of caregiver burden remained stable in
336 approximately half of the caregivers., We found as predicted a significant strong correlation
337 between burden and life satisfaction. A significant predictor of an increase in caregiver
338 burden was the feeling of loneliness at 1 year post-injury, which partly confirmed our third
339 hypothesis regarding social network as a predictor of an increase in burden.

340

341 **Caregiver burden**

342 Although in general the changes in burden in the present study were rather small and around
343 half of the participants reported stable values from 1 to 2 years post-injury, the finding that
344 30% of the participants had clinically significant increases in burden should be relevant for
345 health care services. In addition, caregiver burden increased more frequently among
346 caregivers with a moderate-high burden at 1 year post-injury than in caregivers who had a low
347 burden at 1 year post-injury. In a study by Marsh et al., no significant change in caregiver
348 burden was found between 6 and 12 months post-injury (Marsh et al., 2002). The Paris-TBI
349 study found a decrease in burden at 4 years compared with 1 year (Bayen et al., 2014; Bayen
350 et al., 2012). However, Bayen and colleagues did not compare the same population at these

351 two time points; they reported the degree of burden in two different studies at 1 and 4 years
352 post-injury. Both the different populations and the longer time span post-injury could explain
353 the discrepancy in results compared with the present study. The difference in tools used to
354 assess burden may have also influenced the results. In our study, the results were robust, as
355 the burden increased significantly both in mean CBS scores and when measured as numbers
356 with minimal clinically important different changes. A recent study from Denmark (2016)
357 assessed caregiver burden with the CBS 3-6 years after a severe TBI and found almost similar
358 burden scores within each subscale of the CBS as the present study, although the Danish
359 study only assessed the burden at one time point (Doser & Norup, 2016).

360 There may be several possible explanations for the findings of increased burden over
361 time in the present study. At 1 year post-injury, the caregivers may still hope for an
362 improvement in the patient's conditions and may not be fully aware of the long-term
363 consequences of the injury. At 2 years post-injury, the caregiver may also be receiving less
364 help from the community healthcare system than after 1 year. According to Lezak (Lezak,
365 1986), family members of a person with TBI go through 6 stages of reactions; from pleasure
366 of the patient returning home and optimism for the future, to anxiety when energy and
367 optimism decreases after 9-24 months post-injury and further. This last stage is often followed
368 by anxiety, depression, mourning and emotional disengagement. Psychological interventions
369 in addition to professional support in care may help families negotiate these stages and helps
370 them to understand the nature and long-term effects of the injury (Lezak, 1986).

371

372 **Correlation between caregiver burden and life satisfaction**

373 We found a strong correlation between caregiver burden and life satisfaction, indicating a
374 relationship in which a low burden was associated with a high satisfaction with life and vice

375 versa. The strong association between burden and life satisfaction was not unexpected. The
376 findings are in accordance with a Swedish cross-sectional study that investigated caregiver
377 burden and life satisfaction in caregivers of patients with stroke using the CBS and Li-Sat 11
378 and found that the caregivers who were not satisfied with life had a higher burden (Bergstrom
379 et al., 2011). The present study is the first to investigate caregiver burden and life satisfaction
380 simultaneously after severe TBI. To our knowledge, only two Swedish studies have
381 previously investigated the relationship between caregiver burden and life satisfaction;
382 Andren and Elmståhl reported that caregivers of patients with dementia could experience a
383 moderate burden and great satisfaction at the same time (Andren & Elmstahl, 2005), while the
384 Bergstrøm and colleagues found that caregivers of patients with stroke reporting lower life
385 satisfaction also reported significantly higher caregiver burden (Bergstrom et al., 2011). Both
386 studies used the same CBS as in the present study, although they used the Caregiver
387 Assessment of Satisfaction Index and the Life Sat11 respectively to measure life satisfaction.

388 Attention to the increasing burden and decrease in life satisfaction of caregivers needs
389 to be addressed in community health care (family doctors, district nurses, teachers, etc.) as
390 well as in specialized health care services. For patients, multidisciplinary rehabilitation
391 programs that have integrated or separate programs for family members should always be
392 considered (Becker, Kirmess, Tornas, & Lovstad, 2014). The use of programs for family
393 members focusing on education and coping strategies and providing information about
394 available services and social rights are indicated (Ramkumar & Elliott, 2010), and have in
395 previous studies shown to reduce the burden in family members of patients with TBI (Holland
396 & Shigaki, 1998; Perlesz & O'Loughlan, 1998).

397

398 **Life satisfaction**

399 The results in the present study are in reasonable accordance with a study by Livingston and
400 colleagues who reported small but non-significant decreases in life satisfaction in caregivers
401 of patients with TBI between 1 and 2 years post-injury, although they used the more extensive
402 Satisfaction With Life Scale (L. A. Livingston et al., 2010). A limitation in the present study
403 was the single item, but a previous study has confirmed validity (Kolakowsky-Hayner et al.,
404 2001), and in addition the high negative correlation to the CBS supports the validity of the
405 measure. Further, our findings indicate that the one-item question showed a good sensitivity
406 to change. Kolakowsky-Hayner et al. used the same life satisfaction item as in the present
407 study to compare an assessment after at least 4 years post injury with reported pre-injury
408 satisfaction in 57 caregivers of patients with TBI (Kolakowsky-Hayner et al., 2001). They
409 found a long term decrease in life satisfaction; 87 % of the caregivers reported to be satisfied
410 and very satisfied with life pre-injury, diminishing to 59 % satisfied/very satisfied at time of
411 assessment several years post injury, a somewhat higher proportion satisfied family members
412 than in our study at 2 years post-injury.

413

414 **Predictors of an increase in caregiver burden**

415 Feeling lonely was a significant independent predictor of an increase in burden from 1 to 2
416 years post-injury. Loneliness has been regarded as a psychological aspect of social isolation
417 and has been referred to as perceived subjective isolation (Cacioppo, Cacioppo, & Boomsma,
418 2014). Loneliness has previously been assessed by Anke and colleagues, who described
419 loneliness as an aspect of low perceived social support and an important indicator of social
420 network quality (A. G. Anke et al., 1997). Manskow et al. recently found that loneliness was
421 independent associated with a higher caregiver burden 1 year after severe TBI (Manskow et
422 al., 2014). However, loneliness and social isolation must be viewed as two separate concepts,

423 as feeling lonely does not necessarily mean that the person is socially isolated (Nicholson,
424 2012). Few studies have aimed to study caregivers' loneliness in the TBI population. Oddy et
425 al. reported loneliness as being the most difficult aspect of social adjustment in caregivers of
426 TBI survivors (Oddy, Coughlan, Tyerman, & Jenkins, 1985). In a qualitative study on
427 caregivers of patients with acquired brain injury, Braine found loneliness to be a key theme
428 contributing to their overwhelming sense of loss and burden (Braine, 2011).

429 In the present study, we revealed that men experienced an increase in burden over time
430 more often than women, although this was not statistically significant. In a clinical review on
431 caregiver burden gender was not mentioned as a risk factor for increase in burden over time,
432 and this issue should be explored in future studies (Adelman, Tmanova, Delgado, Dion, &
433 Lachs, 2014). The patient's level of functioning at 1 year post-injury was not a significant
434 independent predictor of an increased burden from 1 to 2 years in the present study, indicating
435 that an increase in burden over time was not influenced by the patients' functional status.
436 However, there was a tendency to a positive association between low GOSE score at 1 year
437 and increase in burden in the univariate analyze ($p=0.067$), indicating a risk for a statistical
438 Type 2-error. The results may also suggest that other factors such as no or little support from
439 health and/or social services, family and friends are more important factors contributing to
440 increases in burden (Hanks et al., 2007; Manskow et al., 2014).

441

442 **Consideration of methodology and design**

443 No established values to indicate a minimal clinically important change in CBS score at the
444 individual level have previously been reported. To investigate change and stability in CBS,
445 we decided to use a difference of 0.30 points between the two time points as the minimum
446 clinically important change. This corresponds to 10% of the range of the scale (1.0-4.0), a
447 percentage previously reported to indicate a minimal clinically important change in other

448 measures (Fayers, Langston, Robertson, & group, 2007). In general, the definition of the
449 minimal clinically important difference is debated in the literature, and no consensus has yet
450 been reached (Copay et al., 2007). Although one standard error of measurement (SEM) has
451 been reported to indicate a minimal clinically important difference (Norman et al., 2003), the
452 chosen value of 0.3 was twice the SEM and close to the alternative distribution-based method
453 using half the SD as the measurement of a minimal clinically important change (Copay et al.,
454 2007).

455 The Life satisfaction item used in the present study was uni-dimensional and only
456 assessed overall satisfaction with life, whereas there are other multidimensional scales
457 measuring satisfaction with life, for example the Life Satisfaction Index A (Neugarten,
458 Havighurst, & Tobin, 1961), and the Life satisfaction checklist (Fugl-Meyer et al., 2002). The
459 use of a one-item overall assessment of life satisfaction is transferrable to the use of a single
460 overall assessment of patients' health status and quality of life which has previously been
461 highlighted as an extremely useful indicator of a patients well-being (Fayers & Sprangers,
462 2002). The one-item questionnaire was chosen in accordance with previous studies, and
463 responsiveness and validity were supported in the present study (A. Anke et al., 2014; Fujita
464 & Diener, 2005; Kolakowsky-Hayner et al., 2001).

465

466 **Strengths and limitations of the study**

467 A clear strength is the study's design as a national prospective multicenter study. In addition,
468 the use of established and validated instruments and the assessment at two points in time are
469 strengths. The literature recommends prospective, longitudinal studies that use similar follow-
470 up times post-injury and the same level of TBI severity to assess burden over time
471 (Ramkumar & Elliott, 2010; Thompson, 2009). The participation rate for those who answered

472 at both follow-up times was 47%. Though lack of information regarding the non-consenting
473 family members, the patients in non-participating families did not differ significantly from
474 patients in participating families. However, we cannot exclude the possibility of selection
475 bias: Those who did not participate may have been family members with a higher or lower
476 burden than the participants. Although the number of participating family members were
477 higher than in most previous longitudinal studies, the relatively small number of participants
478 led to low statistical power.

479 A limitation could be the one-item question of loneliness tapping social support, but
480 the association between social support and burden is supported by several studies, also with
481 more extensive measures (Hanks et al., 2007; Kreutzer et al., 2009). Another limitation is the
482 lack of available neuropsychological data that would provide more information about the
483 patients cognitive functioning. We are also aware that other factors not measured in this study
484 are known to be important for family members burden and life satisfaction such as the
485 relatives coping style, resilience and mental health status (Ponsford & Schonberger, 2010;
486 Simpson & Jones, 2013). Whether these factors influence change in burden or life satisfaction
487 is not investigated in the present study. We acknowledge that our results may not be
488 generalizable to (all) other countries or cultures caused by national differences in systems for
489 support after discharge and traditions for family involvement. (Norup et al., 2015). Age of
490 family member was not measured in this study. However, as significant association between
491 age of family member and caregiver burden are not found in previous studies, we do not think
492 this weakens the results.

493

494 **CONCLUSION**

495 An increase in caregiver burden, a decrease in life satisfaction and a strong association
496 between caregiver burden and life satisfaction over time, has significant clinical implications.
497 Family members play a fundamental role in the long-term care and rehabilitation of patients
498 with severe TBI. The consequences of caregiving must be recognized and met with targeted
499 actions if caregivers are expected to effectively fulfill this role, participate in the society and
500 maintain their quality of life. The results demonstrate a need for future long-term follow-up of
501 patients and caregivers that particularly focuses on professional support to relieve caregiver
502 burden and risk of loneliness or social isolation. The ability to identify family members with a
503 moderate to high burden is crucial in order to develop relevant interventions.

504

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