

Assessment of needs, psychological impact and quality of life in families of patients with locked-in syndrome

Zulay Lugo, Frédéric Pellas, Veronique Blandin, Steven Laureys & Olivia Gosseries

To cite this article: Zulay Lugo, Frédéric Pellas, Veronique Blandin, Steven Laureys & Olivia Gosseries (2017) Assessment of needs, psychological impact and quality of life in families of patients with locked-in syndrome, *Brain Injury*, 31:12, 1590-1596, DOI: [10.1080/02699052.2017.1347277](https://doi.org/10.1080/02699052.2017.1347277)

To link to this article: <https://doi.org/10.1080/02699052.2017.1347277>



Published online: 24 Aug 2017.



Submit your article to this journal [↗](#)



Article views: 156



View related articles [↗](#)



View Crossmark data [↗](#)



Assessment of needs, psychological impact and quality of life in families of patients with locked-in syndrome

Zulay Lugo^{a,b}, Frédéric Pellas^{b,c}, Veronique Blandin^b, Steven Laureys^a, and Olivia Gosseries^{a,b}

^aGIGA-consciousness, GIGA-consciousness, Coma Science Group, University and University Hospital of Liège, Liège, Belgium; ^bFrench Association for Locked-In Syndrome (ALIS), Paris, France; ^cComa Arousal Unit, PMR Department, Nîmes University Hospital, Nîmes, France

ABSTRACT

Objective. Family needs of patients with acquired brain injuries have been studied for about three decades. In this study, we assessed the needs, the quality of life and the psychological state of relatives of patients with locked-in syndrome (LIS).

Design: A survey was carried out using the family needs' questionnaire (FNQ).

Subjects and methods: Thirty relatives of patients with LIS fully completed the FNQ.

Results: The most important need reported by families corresponded to the need for medical information. The highest percentage of satisfaction (66%) was also observed for this need. Among the needs for information, specifically, the most important for LIS' relatives was to know that the patient's needs and wishes were respected by the professional staff, which were fulfilled in 93% of the sample. The need for emotional support was the least important and had the lowest percentage of satisfaction (55%). The number of met/unmet needs correlated with the quality of life of the families. Most of the relatives reported anxiety feelings and depressive thoughts.

Conclusions: Receiving accurate medical information is the most important need for family of patients with LIS and their quality of life is correlated with the fulfilment of their needs.

ARTICLE HISTORY

Received 21 July 2016
Revised 2 March 2017
Accepted 5 May 2017
Published online 14 Sep 2017

KEYWORDS

Locked-in syndrome; family needs' questionnaire; quality of life; psychological state

Introduction

The study of the needs, psychological state and quality of life of families of patients with severe acquired brain injuries has gained considerable importance over the last 30 years [1,2]. Similarities in the needs of the relatives have been reported in various clinical settings. Honest and accurate information on the patient's medical condition, treatment and prognosis has been found as one of the most important needs in relatives of patients in the acute and post-acute states after brain injuries [3–10] as well as in patients with spinal cord injuries [11]. Other studies have reported a higher rate of anxiety and depressive symptoms as well as a decreased quality of life in relatives of patients with acquired brain injuries, as compared to normal population [7,8,12–18].

The psychological and social dysfunctions of the closest relatives of patients with brain injuries have been related to the changes in patients' behaviour, personality or mental state rather than to the severity of the injury or the physical disability [19–21]. No study has however addressed the needs, quality of life and psychological well-being of relatives of patients with a locked-in syndrome (LIS). LIS is a rare neurological condition, which is mainly due to a ventral lesion of the pons producing quadriplegia or quadriplegia, anarthria and severe dysphagia [22]. These patients cannot move but their cognitive abilities and sensory pathways are usually preserved, and they are fully conscious [23,24].

Classically, communication remains possible by means of spared vertical eye movements or blinking. Patients with LIS can be easily misdiagnosed with other types of severe cortical and thalamic injuries such as patients with disorders of consciousness (i.e., unresponsive wakefulness syndrome/vegetative state and the minimally conscious state).

Daily care of these patients is difficult as they are fully dependent on others and in the case of patients with LIS, communication should be a priority even if it requires high dedication and motivation of caregivers. The following three cases illustrate different characteristics of the LIS and how patients and their families cope with everyday life.

Case 1

Mr D., born in 1945, suffered from an ischaemic stroke in the brainstem in May 2000. After being hospitalized in a rehabilitation centre for several months, he returned home. He lives with his wife and life assistants help him daily. For instance, a nurse comes everyday for his treatment. He no longer has tracheotomy or gastrostomy because swallowing is possible again and he takes all his meals mixed. He uses a computer with a virtual keyboard with his left hand. He also drives his electric wheelchair with a joystick control using this hand. Communication through the eyes has been progressively replaced by the use of words and short sentences. His wife says that she is exhausted by this accompaniment for 17 years.

Case 2

Ms B., born in 1964, suffered in 2007 from a cerebral haemorrhage in the brainstem of unknown origin. Before the accident, she worked and lived independently. She is currently anarthric and quadriplegic, but she can communicate through eye movements. She also uses a computer with a contactor adapted to a slight movement that she has recovered in one of her fingers. She lives at home and her parents take care of her. Her father complains on the difficulty of having continuous rehabilitation therapy for his daughter and on the difficulty of being well-informed about the LIS.

Case 3

Mr. N., born in 1957, fell from high altitude and suffered a traumatic brain injury with intracerebral haemorrhage primarily affecting the brainstem in 2007. After several days in coma, he regained consciousness but remained anarthric and quadriplegic, with only preserved eye movements to communicate. Currently, he lives at home being cared by his relatives and assisted by physiotherapists and life assistants. His wife reports difficulties in finding nurses.

These three cases illustrate the differences that may exist in the conditions of these patients suffering from the same pathology, as well as the different needs expressed by their relatives. Thus, the aim of this study is to characterize the needs of family members of patients with LIS and to assess their quality of life and psychological well-being.

Methods

Two hundred and four relatives of patients who are members of the French Association of Locked-In Syndrome (ALIS) were asked to evaluate their own needs using the family needs' questionnaire (FNQ). This questionnaire encompasses 40 statements of needs which are divided into six subscales: 1) health information (HI), 2) emotional support (ES), 3) instrumental support (IS), 4) professional support (PS), 5) community support network (CSN) and 6) involvement with patient's care (IWC). Each statement can be rated from 1 (not important) to 4 (very important) and by the extent to which each need is filled (met, partially met or not met) [25].

Previous studies using the FNQ have made minor changes to adapt the questionnaire to the studied group, for example, by adding the category 'non-applicable' to the responses [8]. Considering the particular characteristics of patients with LIS, three questions of the questionnaire were removed and three questions specific to LIS were added. More specifically, the statements 'to be told why my family member acts in ways that are different, difficult or strange', 'to be told if I am making the best possible decisions about my family member' and 'to have different staff members agree on the best way to help my family member' were removed (note that these statements did not belong to any of the subscales). The statements 'the need of having a physician knowing the specific problems of the LIS', 'the need of having information on equipment for the house' and 'the need of having external help in case of health problems or absence of the usual caregiver' were added. A second modification was introduced in

the statement 'I need to have other family members understand how difficult it is for me' which was changed to 'I need to have the patient understand how difficult it is for me'. This change was made in view of the particular characteristics of patients with LIS who, despite their severe deficits, are fully aware [22,23], so their opinion regarding the behaviour of their relatives has a lot of weight on their care. Finally, the question about the need to learn about the treatment of alcohol or drug intake was considered as not applicable to these patients and was replaced by the need to have complete information on the LIS treatment.

In addition to the FNQ, the Anamnestic Comparative Self-Assessment (ACSA) scale was also included to evaluate the quality of life of the relatives [26]. This scale takes as end-points the worst and the best times of the subject's life, assigning a score of -5 and +5, respectively, to these moments. The participant is then asked to compare their current life situation (i.e., the last 2 weeks) with those moments, and to assign a score in between that range. This scale has been used in a previous study to evaluate the quality of life of patients with LIS [27].

To evaluate the emotional state of the relatives, questions about the presence of anxiety feelings (i.e., never, moderately or extremely anxious) and depressive thoughts (i.e., never, occasionally or frequently) were added. A question about end-of-life decisions was also included at the end of the questionnaire and concerned the past and current opinions about the possibility of euthanasia for the patient. Finally, open questions about care management problems of the patients and a final free commentary space completed the survey.

Questionnaires were sent by a post mail to family members of patients with LIS through the ALIS. The study was approved by the ethical committee of the University of Liège and informed consent was obtained from all the participants.

Data were analysed with Statistica version 10 (www.statsoft.fr). A normal distribution of quantitative variables was assessed with the Shapiro–Wilk test. For descriptive analyses, we used subject counts, percentages, mean, standard deviation (SD) and range. For the comparison of quantitative variables, we used the Mann–Whitney *U* test and for qualitative variables, we used the chi-squared test. The association between the number of needs rated as 'met' and the number of needs rated as 'unmet' and the quality of life scores were evaluated using Spearman's rank coefficient correlation test (partially fulfilled needs were not included for this analysis). The level of significance for the tests was set at $p < 0.05$.

Results

Thirty-four questionnaires were received from the ALIS (response rate 17%). Four questionnaires were excluded because of incomplete responses to the FNQ. Thus, answers from 30 questionnaires were evaluated.

Responders were mainly women (73%) and in most cases, the partner of the patient. The mean age of the responders was 56 ± 12 years. Regarding the patients, the mean age was 51 ± 14 years, and the mean time since the LIS onset was 8 ± 7 years (range: 6 months to 28 years) with 63% of the patients living at home. Table 1 shows the socio-demographic characteristics of the sample.

Table 1. Socio-demographic characteristics of the sample.

Socio-demographic variable	n=30
Gender	
Female	22
Male	8
Age (mean years \pm SD)	56.3 \pm 11.6
(range)	28-81
Relationship to the patient	
Wife	9
Husband	4
Partner (female)	4
Partner (male)	3
Father	1
Mother	6
Daughter	2
Not answered	1
Caregiver's occupation	
Actively working	13
Retired	8
Not answered	9
Data from patients:	
Age (mean years \pm SD)	51 \pm 14
(range)	(18-75)
Time since LIS onset in years (mean \pm SD)	8 \pm 7
Place of patient's living	
Home	19
Institution	11
Reeducation centre	8
Nursing and care centre	3
Presence of gastrostomy or tracheostomy	
Only gastrostomy	10
Only tracheostomy	1
Both	10
None	9

The importance of all the statement's needs was, on average, rated between 3 and 4 (i.e., quite important to very important). Among the five statements with the highest scores of importance, four corresponded to the need for HI. The lowest scores of importance corresponded to the need for ES.

Needs with the highest percentage of fulfilment also corresponded to the need for HI. Families were globally satisfied with their need of being informed about all changes in the patient's medical status (met in 66%), need for honest answers to the questions (met in 64%) and need of having complete information on the treatments (met in 62%). The lowest level of fulfilment was for the emotional need, including getting help to prepare for the worst (unmet in 55%) and to appease doubts and fears (unmet in 44%). The need of getting external/institutional aide in the case of caregiver's absence (which does not belong to any of the subscales) was the third least satisfied need (unmet in 42%). Table 2 displays the list of all the needs with mean scoring in the order of importance and percentage of fulfilment of each need.

Regarding the quality of life, 26 out of 30 participants rated the ACSA scale for their current quality of life. The mean score for the group was -0.62 (± 2.4 SD). Figure 1 displays the distribution of participants according to the ACSA scores. As previously performed in a study evaluating the quality of life in patients with LIS [27], we divided the respondents into two groups based on the ACSA scores: those with ACSA scores ≥ 0 (14 participants, 58%, mean 1 ± 1.4 SD) and those with scores below 0 (11 participants, 42%, mean -2.9 ± 2.3 SD). There was no significant difference between the two groups in terms of gender, age, time since LIS onset, caregiver's occupa-

tion (active vs. retired) and patient's place of living (home/institution). Spearman's rank correlation coefficient showed a significant positive correlation between the number of fulfilled needs and the ACSA scoring ($r = 0.43$, $p = 0.02$) and a negative correlation with the number of unfulfilled needs ($r = -0.39$, $p = 0.04$) (Figure 2).

The presence of anxiety feelings was expressed by 86% of the participants (56% 'moderately' and 32% 'extremely') and 64% reported the presence of depressive thoughts (43% 'sometimes' and 21% 'frequently'). The analysis of the socio-demographic variables did not show significant differences between the presence or absence of anxiety feelings or depressive thoughts and the participant's gender and age, patient's place of living, time since onset of the LIS and occupation of the relative.

When analysing the presence of those feelings regarding the quality of life, participants with the ACSA score above or equal to 0 reported depressive thoughts in 53% and anxiety feelings in 80% of the cases. Participants with the ACSA score below 0 reported more depressed feelings (73%) and more anxiety feelings (91%). Due to the small sample size, this difference between the group was, however, not significant ($p = 0.31$ for depressive thought and $p = 0.44$ for anxiety feelings).

Regarding the end-of-life decisions, 78% of the participants never considered euthanasia for their proxy, 15% had considered this decision in the past but not currently and only 7% expressed the desire of euthanasia for the patient at the moment of the interview.

Finally, 90% of the participants gave their opinion on the problems concerning the care of the patient and the possible solutions. Two main kinds of difficulties were identified. The first issue was related to the large number of administrative formalities to be carried out with a difficulty to find appropriate rehabilitation centres and the lack of adequate information on health services or support institutions. In this category, about half of the respondents (44%) had encountered difficulties. The second kind of problems corresponded to the difficulties with the medical and paramedical staff: difficulty of finding home care (e.g., speech therapist, physiotherapist, auxiliary caregivers, etc.) as well as disagreements with the professional team. Twenty-two percent of the relatives expressed dissatisfaction on these aspects. A third group of complaints was mainly related to the lack of ES for the caregivers (19%). Finally, one caregiver expressed the main problem to be the patient's needs for companionship, recreation and social expansion. Note that most of the relatives expressed opinions on more than one aspects. Table 3 shows examples of the main problems expressed in each of the four categories.

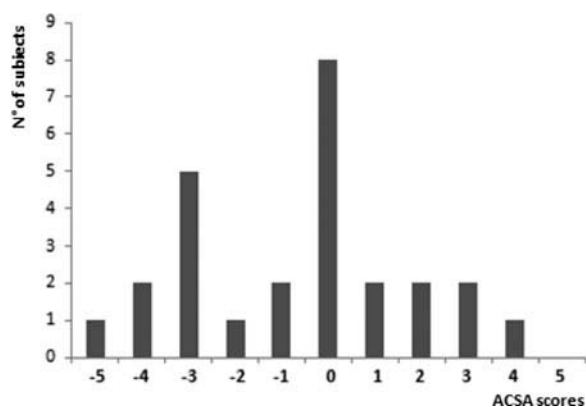
Discussion

The aim of this study was to evaluate the needs of relatives of patients with LIS using a structured questionnaire as well as assessing their quality of life and emotional status. Despite the particular condition of these patients who are paralysed and unable to speak but retaining full consciousness and mostly normal cognitive functioning, we found several similarities with other studies of relatives of patients with different cerebral

Table 2. Scoring and percentage of fulfilment for each item of the FNQ ordered from highest to lowest importance.

Item	I need	Score (mean)	Met (%)	Partially met (%)	Not met (%)	Sub-scale
1	To be shown that medical educational or rehabilitation staff respect the patient's needs or wishes	4	44.82	48.27	6.89	HI
5	To be assured that the best possible medical care is being given to the patient	3.93	51.72	41.37	6.89	HI
9	To have a professional to turn to for advice or services when the patient needs help.	3.93	41.37	44.82	13.79	CSN
16	To have information on the patient's rehabilitative or educational progress	3.90	55.17	34.48	10.34	HI
7	To have my questions answered honestly.	3.89	64.28	28.57	7.14	HI
11	To have complete information on the patient's physical problems (e.g., weakness, headaches, dizziness, problems with vision or walking)	3.89	46.42	39.28	14.28	HI
19	To have enough resources for myself or the family (e.g., financial or legal counselling, respite care, counselling, nursing or day care)	3.88	34.61	61.53	3.84	PS
40	To have the possibility of external/institutional aide in case of caregiver absence	3.86	35.71	21.42	42.85	(*)
12	To have complete information on the patient's problems with thinking (e.g., confusion, memory, or communication)	3.82	42.85	42.85	14.28	HI
22	To get enough rest or sleep	3.82	35.71	32.14	32.14	IS
38	To have a physician knowing all the problems related to the locked-in syndrome	3.80	36.66	30	33.33	(*)
20	To have help keeping the house (e.g., shopping, cleaning, cooking, etc.)	3.77	30.76	53.84	15.38	IS
13	To have complete information on the treatment	3.76	62.06	27.58	10.34	HI
4	To be told about all the changes in the patient's medical status	3.76	65.51	24.13	10.34	HI
18	To have enough resources for the patient (e.g., rehabilitation programs, physical therapy, counselling, job counselling)	3.76	44.82	37.93	17.24	PS
6	To have explanations from professionals given in terms I can understand	3.72	56.62	41.37	0	HI
8	To be shown that my opinions are used in planning the patient's treatment, rehabilitation, or education	3.71	55.17	41.37	3.44	IWC
10	To have complete information on the medical care of traumatic injuries	3.71	50	21.42	28.57	HI
15	To be shown what to do when the patient is upset or acting strange	3.71	32.14	42.85	25	PS
14	To be told how long each of the patient's problems is expected to last	3.68	35.71	25	39.28	PS
39	To receive information on the existing home equipment and other materials aid	3.65	36.66	30	33.33	(*)
34	Help getting over my doubts and fears about the future	3.63	33.3	20	46.66	ES
17	To have help in deciding how much to let the patient do by himself/herself	3.61	42.85	35.71	21.42	PS
28	To know that other family members understand the patient's problems	3.57	36.66	43.33	20	CSN
2	To be told daily what is being done with or for the patient	3.57	50	42.85	7.14	IWC
21	To have help from other members of the family in taking care of the patient	3.55	31.03	41.37	27.58	IS
35	Help in remaining hopeful about patient's future	3.55	31.03	41.37	27.58	ES
23	To get a break from my problems and responsibilities	3.50	28.57	32.14	39.28	IS
37	To be encouraged to ask others to help out	3.50	33.33	36.66	30	ES
25	To pay attention to my own needs, job or interests	3.50	35.71	53.57	10.71	IS
29	To have patient's friends understand her/his problems	3.48	31.03	34.48	34.48	CSN
27	To have my partner or friends understand how difficult it is for me	3.47	43.33	40	16.66	ES
31	To discuss my feelings about the patient with someone who has gone through the same experience	3.36	39.28	25	35.71	ES
30	To have the patient's employer, coworkers, or teachers understand his/her problems	3.35	34.78	34.78	30.43	CSN
24	To spend time with my friends	3.31	34.48	37.93	27.58	IS
32	To discuss my feelings about the patient with other friends or family	3.29	46.42	39.28	14.28	CSN
26	To have the patient understand how difficult it is for me	3.27	53.33	40	6.66	ES
3	To give my opinions daily to others involved in the patient's care, rehabilitation or education	3.22	59.25	25.92	14.81	IWC
36	Help to preparing for the worst	3.17	24.13	20.68	55.17	ES
33	To be reassured that it is usual to have strong negative feelings about the patient	3.07	35.71	25	39.28	ES

HI, health information; ES, emotional support; IS, instrumental support; PS, professional support; CSN, community support network; IWC, involvement with patient care, (*): not included in any of the six subscales.

**Figure 1.** Distribution of the ACSA scores in the group.

lesions or spinal cord injury [3–11]. Our results showed that the need for HI was rated as the most important by the relatives of patients with LIS, as found in the previous studies.

The importance of this need in our cohort (belonging to an association and giving continuous support and counselling to its members) could be interpreted not only as the need for medical information about the pathology but also as the need of being updated about the most recent technical advances allowing better and easier home care, communication and greater autonomy for the patient. Our results also seem to be in line with the findings of Stebbins and Leung [8] who found in their longitudinal study that the initial need for information does not diminish with time, and that other needs appear such as CSN, IS and ES.

The need for information is expressed with slight variations among the different studies. In our sample, the highest score was associated with the need for the staff to respect the wishes and needs of the patient. This may be related to the fact that the patients with LIS are fully conscious and have their cognitive abilities preserved, so they can make decisions and participate in their treatment decisions. This might also reflect that relatives need to be sure that the staff takes into account the patient's

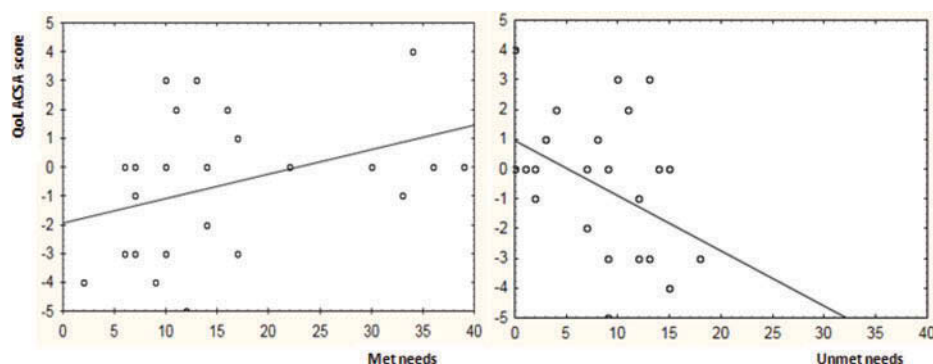


Figure 2. Correlation coefficients plots QoL/met and QoL/unmet needs.

Table 3. Examples of difficulties expressed by LIS patients' relatives.

- 1) *Problems with administrative/financials formalities*
 "Administrative problems, always paperwork..."
 "To find a host establishment, to be properly informed about the disease, to get closer to the place of living"
 "Misguided"
- 2) *Problems with medical/paramedical staff*
 "Insensitivity of doctors to the LIS..."
 "To find a physiotherapist, a speech therapist, to find the most appropriate cares..."
 "Staff problems: physiotherapist, nurse, doctors"
- 3) *Emotional and social relative's difficulties*
 "A great inexperience on the paralysis and the handicap, much clumsiness, difficulties, incomprehension..."
 "Being alone for all steps..."
- 4) *Patient social needs*
 "...to find people who enable her to evolve in her passions: art, music, literature. That she can integrate a social group despite her difficulties"

opinion despite difficult communication. In that sense, our result contrasts with those of Meade et al. [11] in patients with spinal cord injuries. In that study, eight of the 10 needs which were scored as the most important concerned the need for information, but the need to respect the patient's wishes was not included, possibly because patients with spinal cord injury can easily express themselves verbally (unless the injury is at the high cervical level and requires intubation or if there are other brain injuries). Patients with LIS, although mostly unable to communicate by words, have other non-verbal ways to communicate with the outside world, for example, with yes-no eye code, alphabet communication systems or even chin-controlled Morse system decoded by a computer [28]. Thus, the recognition of the LIS condition, time availability and learning how to use the codes are some requirements to effectively communicate with patients with LIS in order to know their own needs.

The need for honest answers (considered fifth most important in our group) is one of the most frequently encountered needs expressed by relatives of patients both in the acute and the chronic settings [3,4]. The study from Leonardi et al. [10] on the relatives of patients with disorders of consciousness, such as unresponsive wakefulness and minimally conscious state, showed that the most important information need was to know what had been done to their relative. In the second place, the authors found the need to be involved in the decisions that might affect the patient, and in the third position, the need was to maintain satisfactory communication with the entire therapeutic team. All these needs for information expressed by relatives of patients with different brain

injuries and in different settings (acute/chronic) confirm the importance of studying and individualizing as much as possible the relative's needs in order to provide care in each case or to a particular group of patients.

Similar to other studies [6,7,9], the needs for ES were perceived as most frequently not met and in our group, they were among the needs rated with the lowest levels of importance. However, this level of importance remained high (ranging from quite important to very important). Thus, it cannot be interpreted as a disregard towards their own needs. It rather seems that the relatives of patients with LIS simply put the patient's needs and well-being before their own. As described in the open commentaries of the participants, most of the difficulties and complaints were related to the large number of activities to be undertaken to provide adequate care to the patients. In this sense, despite a real need for ES, there is a family imbalance in which most of the care and attention are invested in the patient and not in the relatives themselves. The score in the FNQ suggests that relatives seem to be aware of their own emotional needs, but possibly they have not enough time to devote to fulfil them.

The quality of life of relatives of patients with acquired brain injury has been studied through semi-structured interviews and standardized measures [7,15,16,18,29] or more indirectly under other means which can be assimilated to a description or a component of quality of life such as stress, burden, strain or social functioning [10,13,17,19-21]. Despite the methodological differences, most studies agree on the findings of a decrease in the quality of life after the accident and a sense of overload [7,12-16,18]. In our group, most participants (58%) reported neutral to positive scores, which is in line with the findings of Koskinen [29] who found in a longitudinal study that most relatives reported a 'rather good' quality of life. They found that the main deterioration in the quality of life occurred during the first year after the injury and that there was stabilization over time but without reaching the levels prior to the injury. An important finding in our study was the significant correlation between the number of met/unmet needs and the quality of life in relatives, showing that the quality of life increased as the number of met needs increased, and that a lower quality of life was related to an increased number of unmet needs. In their study, Moules and Chandler also found a significant correlation between high levels of unmet needs and a perceived low quality of life in relatives of patients with traumatic brain injury [7].

The variability observed in the physical condition and communication limitations in patients with LIS [28] might have an impact on the caregiver's perception of burden, strain or stress. Nevertheless, the presence of stress or psycho-social dysfunction in relatives of patients with brain injuries has been associated in previous studies with the perception of changes in the patient's personality rather than with the severity of the injury or the inability to resume previous activities [19] and to the patient's complaints [13]. We know that all the patients in our study have at least a basic code for yes/no communication with relatives and caregivers, some of them also use technical means to communicate and others have recovered some limited but functional movements facilitating communication. On the other hand, Bruno et al. [27] reported that patients with LIS have a rather good quality of life. None of the open comments on the problems encountered by the relatives in our study were on the difficulties of communication or on the physical limitations of the patients but mainly, as already pointed out, they referred to the numerous administrative procedures to be carried out, the lack of adapted centres, the difficulties with medical and paramedical staff and the lack of ES that some feel in face of such heavy situation.

With respect to the psychological status, we found a high percentage of anxiety and depressed feelings in relatives of patients with LIS, in line with other studies evaluating the well-being of relatives of patients with acquired brain injury both in the acute and the chronic states [7,10,12,13,16–18]. Others findings have evaluated the psychological impact from the point of view of the 'perceived burden' or 'perceived strain' [10,13,17,19–21]. Results are widely consistent showing a great psychological and social impact on these relatives and different predictors of such psychological distress have been described. Nabors et al. [30] found that the principal predictor of burden was the importance of needs of the caregiver and that the percentage of met needs was also a predictor of psychological or behavioural burden. Moules and Chandler found that the relative's psychological distress was associated with the level of needs' satisfaction [7]. A recent study on the caregivers of patients with disorders of consciousness found that the combination of a preoccupied attachment style and a high degree of hopelessness predicted 49% of the total variability of burden [31]. In our study, the presence of anxiety and depressed feelings also seems to be related to the degree of needs' satisfaction, as the relatives with lower quality of life were also the ones with a higher percentage of unmet needs. Given that the highest percentage of unmet needs in our group corresponded to the need for ES, it seems that the main need to be filled in the relatives of patients with LIS refers to this specific aspect. This also suggests an urgent necessity to address the personal needs of families of patients with LIS to improve their quality of life and emotional well-being.

Finally, most of the families (almost 80%) never considered euthanasia as a possibility for their relatives. This could be explained by the fact that the patients with LIS are conscious and can take their own decisions. This result might also in principle partly reflect the mentality of people living in a country (France) where euthanasia or physician-assisted suicides are not legally permitted, so these decisions are not an option to consider at the first glance even when facing a devastating neurological condition like the LIS. To this, it

can be added to the wide dissemination that the LIS has had in this country since the foundation of ALIS in 1997. Considering that the mean time in the LIS of the patients in our group is less than 10 years, their relatives possibly may have had very early access to information on the possibilities for these patients to have meaningful lives and received practical help to achieve these goals.

Our study has some limitations. The small sample size of our studied group though similar to other studies with mailed questionnaires [30] restrains the generalization of the results to all the relatives of patients with LIS and can be an index of a selective bias. As they are the members of ALIS, an association giving counselling and support since almost 20 years to the patients with LIS and their families, it would be possible that those relatives who answered reflect a minority with the not-satisfied needs. In addition, many patients who used to live in urban cities prior to their brain injuries are now living in distant institutions where most of the care falls on the staff and not on the family members. These relatives thus might have been less interested on the survey. In that context, the participation of a greater number of families would be desirable in future studies and the inclusion of family partnerships of LIS organizations from other European countries (www.alis-asso.fr/lisef/) along with other methods of approach for the evaluation of relatives would be recommended. Standardized questionnaires of depression and anxiety should also be employed in future studies to fine-tune the assessment of these symptoms.

In conclusion, we described the principal needs of a group of relatives of patients with LIS and evaluated their quality of life and psychological status. To the best of our knowledge, this is the first study addressing these questions in the relatives of patients with this type of neurological handicap. We found that the need for the respect of patients' wishes was the main concern, but other needs such as having honest and accurate information were also rated as very important. It is worth to note the high rate of unmet needs for ES in our group of relatives, which in our opinion must be urgently addressed in order to improve their quality of life and psychological well-being.

Acknowledgments

The authors thank all participating families of patients with LIS for their cooperation.

Competing interests

The authors declare no conflict of interests.

Funding

This research was supported by the French Association of Locked-in Syndrome (ALIS), the Belgian National Funds for Scientific Research (FNRS), the European Commission (European ICT Programme Projects FP7-247919, DECODER), the Belgian American Education Foundation, the Wallonie-Bruxelles International, the Fondazione Europea di Ricerca Biomedica, FEDER structural fund RADIOMED-930549, Fonds Léon Fredericq, the James McDonnell Foundation, the Mind Science Foundation, the French Speaking Community Concerted Research Action (ARC-06/11-340), Human Brain Project (EU-H2020-FETFLAGSHIP-HBP-SGA1-GA720270), LUMINOUS project (EU-

H2020-FETOPEN-GA686764) and the University and University Hospital of Liège. OG is a post-doctoral fellow and SL is a research director at FRS-FNRS. The funding sources are not liable for any use that may be made of the information contained therein.

References

- Sinnakaruppan I, Williams DM. Family carers and the adult head-injured: a critical review of carers' needs. *Brain Inj.* 2001;15:653–672.
- Thompson HJ. A critical analysis of measures of caregiver and family functioning following traumatic brain injury. *J Neurosci Nurs.* 2009;41:148–158.
- Mathis M. Personal needs of family members of critically ill patients with and without acute brain injury. *J Neurosurg Nurs.* 1984;16:36–44.
- Engli M, Kirsivali-Farmer K. Needs of family members of critically ill patients with and without acute brain injury. *J Neurosci Nurs.* 1993;25:78–85.
- Kreutzer JS, Devany Serio C, Bergquist S. Family needs after brain injury: A quantitative analysis. *J Head Trauma Rehabil.* 1994;9:104–115.
- Witol AD, Sander AM, Kreutzer JS. A longitudinal analysis of family needs following traumatic brain injury. *NeuroRehabilitation.* 1996;7:175–187.
- Moules S, Chandler BJ. A study of the health and social needs of carers of traumatically brain injured individuals served by one community rehabilitation team. *Brain Inj.* 1999;13:983–993.
- Stebbins P, Leung P. Changing family needs after brain injury. *J Rehabil.* 1998;64:15–22.
- Murray HM, Maslany GW, Jeffery B. Assessment of family needs following acquired brain injury in Saskatchewan. *Brain Inj.* 2006;20:575–585.
- Leonardi M, Giovannetti AM, Pagani M, Raggi A, Sattin D. Burden and needs of 487 caregivers of patients in vegetative state and in minimally conscious state: results from a national study. *Brain Inj.* 2012;26:1201–1210.
- Meade MA, Taylor LA, Kreutzer JS, Marwitz JH, Thomas V. A preliminary study of acute family needs after spinal cord injury: analysis and implications. *Rehabil Psychol.* 2004;49:150–155.
- Livingston MG, Brooks DN, Bond MR. Three months after severe head injury: psychiatric and social impact on relatives. *J Neurol Neurosurg Psychiatry.* 1985;48:870–875.
- Livingston MG, Brooks DN, Bond MR. Patient outcome in the year following severe head injury and relatives' psychiatric and social functioning. *J Neurol Neurosurg Psychiatry.* 1985;48:876–881.
- Kreutzer JS, Gervasio AH, Camplair PS. Primary caregivers' psychological status and family functioning after traumatic brain injury. *Brain Inj.* 1994;8:197–210.
- Kolakowsky-Hayner SA, Miner KD, Kreutzer JS. Long-term life quality and family needs after traumatic brain injury. *J Head Trauma Rehabil.* 2001;16:374–385.
- Norup A, Siert L, Lykke Mortensen E. Emotional distress and quality of life in relatives of patients with severe brain injury: the first month after injury. *Brain Inj.* 2010;24:81–88.
- Moretta P, Estraneo A, De Lucia L, Cardinale V, Loreto V, Trojano L. A study of the psychological distress in family caregivers of patients with prolonged disorders of consciousness during in-hospital rehabilitation. *Clin Rehabil.* 2014;28:717–725.
- Giovannetti AM, Covelli V, Sattin D, Leonardi M. Caregivers of patients with disorder of consciousness: burden, quality of life and social support. *Acta Neurol Scand.* 2015;132:259–269.
- Oddy M, Humphrey M, Uttley D. Stresses upon the relatives of head-injured patients. *Br J Psychiatry.* 1978;133:507–513.
- Brooks N, Campsie L, Symington C, Beattie A, McKinlay W. The five year outcome of severe blunt head injury: a relative's view. *J Neurol Neurosurg Psychiatry.* 1986;49:764–770.
- Knight RG, Devereux R, Godfrey HP. Caring for a family member with a traumatic brain injury. *Brain Inj.* 1998;12:467–481.
- Giacino JT, Zasler ND, Whyte J, Katz DI, Glen M, Andary, M. Recommendations for use of uniform nomenclature pertinent to patients with severe alterations in consciousness. *Arch Phys Med Rehabil.* 1995;76:205–209.
- Laureys S, Pellas F, Van Eeckhout P, Ghorbel S, Schnakers C, Perrin F, Berré J, Faimonville ME, Pantke KH, Damas F, et al. The locked-in syndrome : what is it like to be conscious but paralyzed and voiceless? *Prog Brain Res.* 2005;150:495–511.
- Schnakers C, Majerus S, Goldman S, Boly M, Van Eeckhout P, Gay S, Pellas F, Bartsch V, Peigneux P, Moonen G, et al. Cognitive function in the locked-in syndrome. *J Neurol.* 2008;255:323–330.
- Serio CD, Kreutzer JS, Witol AD. Family needs after traumatic brain injury: a factor analytic study of the Family Needs Questionnaire. *Brain Inj.* 1997;11:1–9.
- Bernheim JL. How to get serious answers to the serious question: "How have you been?": subjective quality of life (QOL) as an individual experiential emergent construct. *Bioethics.* 1999;13:272–287.
- Bruno MA, Bernheim JL, Ledoux D, Pellas F, Demertzi A, Laureys S. A survey on self-assessed well-being in a cohort of chronic locked-in syndrome patients: happy majority, miserable minority. *BMJ Open.* 2011;1(1):e000039.
- Lugo ZR, Bruno MA, Gosseries O, Demertzi A, Heine L, Thonnard M, Blandin V, Pellas F, Laureys S. Beyond the gaze: communicating in chronic locked-in syndrome. *Brain Inj.* 2015;29:1056–1061.
- Koskinen S. Quality of life 10 years after a very severe traumatic brain injury (TBI): the perspective of the injured and the closest relative. *Brain Inj.* 1998;12:631–648.
- Nabors N, Seacat J, Rosenthal M. Predictors of caregiver burden following traumatic brain injury. *Brain Inj.* 2002;16:1039–1050.
- Romaniello C, Farinelli M, Matera N, Bertoletti E, Pedone V, Northoff G. Anxious attachment style and hopelessness as predictors of burden in caregivers of patients with disorders of consciousness : A pilot study. *Brain Inj.* 2015;9052:466–472.