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Research Article

Coping and Managing ALS Disease in the Family during COVID-19: Caregivers' Perspective

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Abstract

Amyotrophic Lateral Sclerosis (ALS) is a progressive neurodegenerative disease that affects motor neurons, the nerve cells in the brain and the spinal cord that enable voluntary muscle movement. Managing ALS is complex and increasingly requires informal care, most often by the patient's companions/spouses or children. The COVID-19 pandemic posed additional critical issues, particularly the disruption of home care and the increased time caregivers spent on patient care. One aim of this research was to assess caregivers' health conditions and to understand how the lockdown has affected their lives and the management of their relatives' illnesses. Another was to observe whether a psychological support intervention for caregivers' minor children could indirectly impact caregivers. The study involved Italian participants: 26 caregivers (31% males and 69% females) aged between 20 and 69 years (M = 43.85 years, SD

= 10.17). The following variables were measured at t0 and t1: reflective functioning with the Reflective Functioning Questionnaire (RFQ), depression with the Beck Depression Inventory-I (BDI-I), hopelessness with the Beck Hopelessness Scale (BHS) and burden of care with Family Strain Questionnaire (FSQ). At t1 only 18 participants also participated in a semi-structured interview to explore the impact of the COVID-19 pandemic, specifically on caregivers of the psychological support participants' children received. The results at t0 showed that caregivers exhibit significant distress symptoms, high stress levels and burden of care. At t1 it was found that the pandemic hurt caregivers' emotions; however, they do not show a worsening but a decrease in hopelessness. The interviews showed that the intervention on their children had a positive effect by allowing containment of the negative effects on well-being. This suggests that it is essential that ALS management also includes a whole-family intervention.

Keywords

Amyotrophic lateral sclerosis; informal caregivers; COVID-19; mixed methods research

1. Introduction

Amyotrophic Lateral Sclerosis (ALS) is a neurodegenerative disease that affects motor neurons. It is characterized by progressive muscle paralysis, coinciding with degeneration of motor neurons in the primary motor cortex, corticospinal tract, brainstem and anterior horn cells in the spinal cord [1]. Its progression is rapid and involves symptoms such as weakness and spasticity of the limbs, fasciculations and cramps, dysarthria, dyspnoea, chronic hypoventilation, sialorrhea, dysphagia and emotional lability [2]; moreover, about 30%-50% of ALS patients experience cognitive degeneration and behavioral disturbances, as the brain areas responsible for controlling the cognitive and emotional components of empathy are affected (e.g., the amygdala), in particular the ability to process emotions, especially with regards to adverse emotional information [3, 4]. Motor degeneration involves voluntary muscles, thus compromising some vital functions, such as breathing, swallowing, speech, walking, gripping, etc. [1]. In particular, symptoms impact communication, which becomes increasingly complex: understanding the mental states and emotional experiences of the patient becomes extremely difficult for family members, and even more so for children [5, 6]. For a person with this diagnosis, every day can present new cognitive or physical losses, and the disease is characterized by the progressive necessity to depend on others [7]. It is mainly the family who takes care of the ALS patient: the patient needs daily assistance which is the main caregivers often the patient's partner, and it is common for children to participate in home care management [8].

This leads to a consistent burden, defined as the impact on the caregiver's emotional health, physical health, social life and financial situation due to adopting the caregiving role [9]. The well-being of caregivers is essential in the care of ALS since their ability to adequately care for their loved one represents an important factor that allows patients to stay at home until the end of their life rather than having to spend their last moments in a care facility. Furthermore, studies have shown a high correlation between patient and caregiver well-being, indicating that reduced caregiver well-being can also hurt the patient [10]. In the presence of a serious degenerative pathology such as

ALS, there is often the transition from being a close relative to almost exclusively being a caregiver; for this reason, caregivers can often experience severe problems of depression, anxiety, frustration and dissatisfaction [11]. To these elements, other aspects should be added as well such as the difficult conditions of the patient, the compromised communication and the difficult emotional reading caused by facial paralysis, which creates a peculiar aspect of ambiguity: the person is physically present but not psychologically and, consequently, a condition of anticipatory mourning can be experienced, even in the absence of a definitive loss [12, 13].

1.1 Caregivers' Management of ALS during the COVID-19 Pandemic

The outbreak of the global COVID-19 pandemic in 2020 represented a further difficulty for the caregivers of people with ALS: patients with diseases affecting the respiratory system and those with pathologies that present a rapid progression, such as ALS patients, found themselves having a greater concern about contracting the virus [14]. On average, death for ALS patients occurs between 3-5 years after their diagnosis, due to respiratory problems [15]. Moreover, disease management is often shared between different caregivers, both informal and formal ones, but during the COVID-19 pandemic there was a need to cancel face-to-face appointments and switch to telemedicine, thus reducing the risk of infection during hospital visits [16]; therefore, all the burden of care had to be managed by family caregivers [17]. Patients with ALS are also particularly vulnerable to the medical complications of infection or emotional distress in terms of anxiety and depression, as well as maladaptive coping strategies, which make them even more susceptible to the harmful neuropsychiatric effects of the pandemic [18]. The social isolation and loneliness increased by the pandemic seem to have increased anxiety levels resulting from a feeling of abandonment or rejection by health professionals [14]. Some studies have even reported that in this period, regardless of the severity of the disease, family visits, even the shortest ones, were abruptly interrupted, probably due to the fear of spreading the disease to patients [19]. Therefore, studies conducted during COVID-19 have reported high levels of anger, frustration, uncertainty about the future, anxiety, depression, anguish, loneliness, and a sense of oppression between caregivers and patients [18-21].

During this pandemic period, the concerns that have mostly affected the caregivers have been contracting the virus and the possibility of having to go through the quarantine, as well as the risk, as a consequence, of not being able to be present for the care of their loved one affected by ALS, thus causing a lack of important support [22]. Furthermore, caregiver, often had to struggle with the loss of income, job losses and lack of childcare, which have become even more widespread during the pandemic [23]. In the most serious cases, these situations could also lead to burnout, which can be defined as a syndrome that is developed in response to the stress that the context of caregiving can represent, with possible experiences of emotional exhaustion, that is, a feeling of overload, of no longer being able to continue, and being emotionally drained when it comes to dealing with caregiving [24]. In the most serious cases, the depersonalization of the loved one affected by ALS could also occur, with a detached response in the relationship with the person, who loses his/her basic characteristics of a human being [25].

Differently from other studies involving caregivers or patients with ALS during the COVID-19 pandemic, the novelty of the present study is that it observed the impact of this situation on caregivers by also taking into account the presence of minors who were meanwhile receiving psychological support, and investigated its indirect effects [26-28]. It is already known in the literature how much the well-being or malaise of caregivers affects that of their children and this was all the more accentuated during the Covid-19 pandemic, where the worsening of parents' mental health occurred together with the worsening of children's behavioral health: parents already exposed to a higher risk of distress also due to situations prior to lockdown had compromised being supportive caregivers by compromising children's mental health [29-31].

2. Materials and Methods

2.1 The Present Study

This study is part of a larger project initiated two months before the outbreak of the COVID-19 pandemic in northern Italy.

The project was designed to understand how ALS emotionally and effectively affected children and adolescents with a sick parent or grandparent and the patient's primary caregivers, in addition to observing the effectiveness of a psychological support intervention with minors, the direct beneficiaries of the project. Immediately after the t0 assessment, the COVID-19 pandemic broke out and necessitated redefining some intervention modalities alternative to those originally thought [32, 33]. The intervention had the objective of helping the participants process the experience of ambiguous loss and the representation of the ill parent’s mental condition through psychodrama and art therapy activities [34, 35].

The intervention was immediately repurposed and implemented to support families struggling with the difficulties of ALS. It was maintained at a distance and resumed in person at the end of the first lockdown in the spring of 2020.

In parallel with the intervention, in order to explore its possible effects, another study has been implemented, involving the parents of children and adolescents involved in the psychological intervention. While the data concerning minors have already been published [32, 33], the present article focuses on the parents’ experience. More specifically, these children’s parents did not personally take part in the support intervention. However, it was hypothesized that they could also indirectly benefit from it. The researchers were also interested in exploring the points of view and experiences of adults who are the primary caregivers of loved ones affected by ALS more broadly, particularly related to the current pandemic period and the associated lockdown measures.

2.2 Aims

This research aimed to understand whether and how isolation affected the lives of caregivers of patients with ALS, and how it changed the management of their relatives' illness. Additionally, we intended to observe whether a psychological support intervention aimed at the minor dependents of ALS caregivers could indirectly impact the caregivers, and the burden of care due to the difficulties of caring for ALS patients, which have already been widely highlighted in the literature.

2.3 Participants and Methodology

The research team adopted a mixed-method approach; therefore the study followed both quantitative and qualitative methodologies. This was deemed necessary to obtain faceted and combined information concerning the participants' condition, needs and difficulties, also considering the eventual changes that occurred in their families as a consequence of the intervention of psychological support their children had received. Therefore, the study required both objective and more subjective, in-depth measures.

Regarding the quantitative measures, these have been administered both before the psychological support intervention offered to minors and seven months after it, to detect any changes in the caregivers, since the study hypothesis was that they could indirectly benefit as well from such intervention. The qualitative part of the research was instead conducted after the intervention, to explore participants' direct points of view.

In the following sections, the quantitative and the qualitative data collection and analysis procedures adopted will be described in detail.

The quantitative section of the study was conducted in two phases, before the intervention of psychological support with minors and seven months later. For the first phase, pre-intervention, 26 caregivers from 25 families were recruited from different offices of non-profit organizations throughout Italy. To be eligible to participate, having a close relative with ALS and a minor family member who was indirectly involved in managing the disease was necessary. The participants were predominantly female (8 males, 31% of the sample and 16 females, or 69% of the sample), aged between 20 and 69 years (M = 43.85 years, SD = 10.17). Almost all (25 people, 96% of the sample) declared themselves Catholic. Finally, in most cases (16 people, or 62% of the sample) the participants were caregivers of their spouse, 6 (23%) were caregivers of a parent. In contrast, the remaining 4 (each corresponding to 4% of the sample) presented a different relationship with the person they cared for: father-in-law, brother-in-law, son/daughter, grandson/granddaughter.

Participants were recruited from the parents of some of the minors who took part in the parallel study conducted by the same researchers [32, 33]; therefore their recruitment followed purposive sampling, in particular homogeneous sampling. Psychologists of different offices of the non-profit association involved in the project proposed participation to some parents of children/teens who had taken part in the psychological support intervention, who were also caregivers of a close relative with ALS. The detailed objectives of the study were carefully explained to all potential participants. Those who declared interest received a detailed explanation of the research methodology and procedures, and the confidentiality of their answers was guaranteed. Written informed consent was obtained from all participants before proceeding with data collection. The study followed the American Psychological Association’s Ethical Principles of Psychologists and Code of Conduct and the principles of the Declaration of Helsinki. It received the approval of the Health Sciences and Science Research Ethics Committee of the University of Padua (reference: C3DD8C5FCE1C26C7E80954B4EC34DC16).

Only 18 participants took part in the second phase of research, post-test, and the qualitative part of the study, which was carried out after the support intervention with minors too because between the two assessments their relative with ALS either died or they simply dropped out of the study giving no further reasons. The characteristics of the 18 caregivers who participated in the study are presented in Table 1.

Table 1 Participants (N = 18).

Caregivers' characteristics	**N (%)**	**Range**	**M (DS)**
Gender			
Male	6 (33%)		
Female	12 (66.7%)		
Age		27-69	46.83 (9.44)
Marital status			
Unmarried	1 (5.6%)		
Coniugato/a	16 (88.9%)		
Widowed	1 (5.6%)		
Nationality			
Italian	18 (100%)		
Degree			
8th grade diploma	4 (22.2%)		
High school diploma	9 (50%)		
Degree	3 (16.7%)		
PhD	2 (11.1%)		
Religion			
Christian Catholic	18 (100%)		
Family members		3-5	4.06 (0.64)
Relative with ALS			
Husband/Wife	11 (61.1%)		
Father/Mother	3 (16.7%)		
Father-in-law	1 (5.6%)		
Sister-in-law	1 (5.6%)		
Daughter	1 (5.6%)		
Granddaughter	1 (5.6%)		

2.3.1 Quantitative Data Collection and Analysis

The constructs that the quantitative section of the study intended to investigate were reflective functioning, that is the ability to mentalize, therefore to reflect upon and understand oneself and others in terms of behaviors guided by mental states, the eventual presence of depressive symptoms and hopelessness, as well as the presence of burden perceived because of their role. To achieve this, four standardized questionnaires have been used: Reflective Functioning Questionnaire (RFQ) [36], Beck Depression Inventory-I (BDI-I) [37], Beck Hopelessness Scale (BHS) [38], and Family Strain Questionnaire (FSQ) [39]. Caregivers completed the self-report questionnaires at t0 independently in the presence of the project psychologist to whom they could ask for information. At t1, when possible, questionnaires were completed as at t0 or using the online meeting platform Zoom. The compilation lasted about 30 minutes.

The RFQ is a self-report questionnaire used to evaluate the psychological processes underlying the ability to mentalize. For the present study, the questionnaire's short version was chosen, consisting of eight items divided into two subscales concerning one's own and others' mental states:

Certainty (RFQ_C) and Uncertainty (RFQ_U). Participants need to choose between 1 and 7 to indicate how much they agree or disagree with each of the statements, where 1 corresponds to 'Strongly disagree' and 7 to 'Strongly agree.' The average scores on both scales indicate good mentalization ability. At the same time, certainty scores that are too high indicate a lack of awareness in believing that the mental processes of others are not completely knowable and remain partially incomprehensible (hyper-mentalization). Finally, uncertainty scores that are too low indicate poor mentalization (hypo-mentalization).

The BDI-SF is a 13-item short-form of the BDI and is a self-report inventory measuring depression in adults. It has an internal consistency comparable to that of the long form. Each item is a list of four statements arranged in increasing severity about a particular area of functioning that could be affected in case of depression (sadness, pessimism, sense of failure, self-satisfaction, sense of guilt, self-criticism, suicide thoughts, loss of interest, decision-making, self-esteem, loss of energy, fatigue, appetite) and respondents need to choose their answers on a 4 points Likert scale, from 0 (which indicates the absence of that symptom) to 3 (which indicates obviousness of that symptom in the last two weeks). The higher the total score results, the more likely it is for the respondents to present a condition of clinical depression, with a cut-off generally considered to be 20 points signaling the passage between a condition of dysphoria and one of clinical depression.

The BHS is a 20-item self-report inventory that measures the condition of hopelessness declined into three aspects: feelings about the future, loss of motivation, and expectations. Each item is dichotomous (therefore it simply requires a 'true' or 'false' possible answer); the higher the total score, the higher the hopelessness the person reports.

Finally, the FSQ consists of a brief semi-structured interview and 44 dichotomous items, which investigate the level of burden perceived by informal caregivers by focusing on 5 macro-areas: emotional burden, problems in social involvement, need for knowledge about the disease, satisfaction with family relationships, and thoughts about death. The higher the total score, the more fragile the caregiver is at risk of a pathopsychological condition.

2.3.2 Qualitative Data Collection and Analysis

Only 18 participants took part in the qualitative part of the study, conducted in parallel with the quantitative post-test phase, the same ones who also participated in the post-test phase, as has already been stated. Interviews were conducted following the administration of the questionnaires and, as mentioned above, were done in person where possible, based on the Covid-related situation, and alternatively were done on Zoom. It took about 45 minutes to complete the interview.

The characteristics of the qualitative participant group were the following: 18 caregivers, 6 males and 12 females, aged between 27 and 69 years old (M = 46,83; SD = 9,44). All of them reported considering themselves Catholic. Moreover, the majority of them (11) were caring for their spouse, while the remaining were either caring for one of their parents (3 participants), for their daughter (1 participant) or for other relatives such as a sister-in-law (1 participant), a niece/nephew (1 participant), and a father-in-law (1 participant).

Data were collected through a semi-structured interview, which represents a flexible methodology that allows the researcher to accurately explore some central themes with the participants, following their point of view and being able to adapt the interview to each participant's specific answers, thus allowing the person to focus more on the aspects he/she believes to be of greater importance based on his/her own experience [40]. More specifically, the interview investigated the level of appreciation of the psychological support the participants' children received, the emotional experiences and the general difficulties of managing their loved one's ALS with a focus on the impact of the COVID-19-related lockdown period as well, the support received during this period, the presence or absence of family dialogue regarding the condition of the patient, his/her prognosis and the prospect of his/her death, and any suggestions they would have liked to provide to other families in a similar situation.

Each interview lasted about 40 minutes and was audio-recorded and subsequently transcribed verbatim. The researchers subsequently analysed the written text obtained from the transcriptions of the interviews following the thematic analysis approach, which allows the identification, analysis and reporting of the central themes that emerge from qualitative data [41]. The textual analysis followed six main phases: engaging in the preparatory organization, reading the texts carefully, coding data, interpreting themes, searching for alternative explanations and producing the final report [41]. The procedure was conducted by identifying and highlighting meaningful concepts in each text, and subsequently comparing these elements with those found in the other participants' narrations. Lastly, broader themes common to all the texts were identified and adequate thematic categories were created. Therefore, the process adopted was bottom-up, since all the obtained categories emerged directly from the texts without predefined ones available. The analysis was carried out in parallel by two researchers, with the support of the *Atlas. Ti* qualitative analysis software [42], compared and combined their results.

3. Results

3.1 Quantitative Results

Regarding the reflective functioning measured by the RFQ, the first survey conducted before the psychological support intervention was carried out on the children of some participants, revealed a mean score on the certainty scale of 0.85 (SD = 0.93) for the caregivers and a score of 0.65 (SD = 0.52) on the uncertainty scale, indicating a significant uncertainty of the caregivers' mental states. The sub-scale of certainty of one's mental states has good reliability (RFQ_C alpha = 0.88) while the uncertainty scale is somewhat low (RFQ_U alpha = 0.59).

Concerning the depressive condition, as measured by the BDI-SF, the total depression scale has good internal consistency (alpha = 0.88).

The mean score of caregivers' responses was 6.92 (SD = 5.63). The reconverted scores for scoring ranged from 0 to 3, where 0 represents No Depression and 3 Severe Depression. Caregivers in the present sample are distributed as follows: 20 (77%) experienced absence of depression; 5 (19%) experienced mild depression; 1 experienced moderate depression (4%); and none (0%) had severe depression.

The dimension of hopelessness, as measured by the BHS, the scale had good reliability (alpha = 0.92). The mean score of caregivers is 8.23 (SD = 5.81). Compared with the normative values, the sample ranks as follows: 9 people (35%) fall under Absence of hopelessness; 3 (12%) under Mild hopelessness; 9 (35%) under Moderate hopelessness; and 5 (19%) under Severe hopelessness.

Finally, regarding caregivers' perceived burden, as measured by the FSQ scale, which shows good reliability (alpha = 0.91), the mean score is 19.38 (SD = 7.01). The Italian validation [34] allows caregivers to be placed in degrees of stress and burden of care that are: 'OK,' which indicates a good reaction to the situation; 'R' which indicates a good reaction with some inability to adapt, and psychological consultation is recommended if symptoms worsen; 'SR' in which psychological support is strongly recommended due to evident stress and burden; and 'U,' which indicates the presence of relevant stress and burden at high psychological risk, and the caregiver should be seen by a psychologist urgently. Concerning these areas, our sample ranks as follows: none (0%) in OK; 6 (23%) in the R; 10 (39%) in the SR; 10 (39%) in the U, indicating the presence of relevant burden for the caregiver (see Table 2).

Table 2 Descriptive statistics of study variables for target group at the pre-test administration.

Caregivers (N = 26)					
Measures		alpha	Range	*M*	*DS*
RFQ	Certainty about mental states[1]	0.88	0-3	0.81	0.90
RFQ	Uncertainty about mental states[1]	0.60	0-2	0.66	0.51
BDI	Total Depression	0.88	0-20	6.92	5.63
BHS	Total Hopelessness	0.92	1-20	8.23	5.81
FSQ	Total Caregiver Strain	0.91	8-30	19.38	7.01

1 A transformation by the square root of the values was adopted for both RFQ factors to normalize the distribution.

Regarding the reflective function as measured by the RFQ in the post-intervention surveys, in which only 18 caregivers participated as indicated earlier, the sub-scale of certainty of one's mental states at the post-test has good reliability (RFQ_C alpha = 0.89) and also that of the uncertainty scale (RFQ_U alpha = 0.77) which is better than the pre-test (alpha = 0.59).

The caregivers' mean score on the certainty scale is 0.60 (SD = 0.58), while that of the uncertainty scale is 0.68 (SD = 0.41). The scores on both scales did not change from the pre-test. The scores between the two scales were insignificant (t(17) = -0.34; p = 0.737).Concerning the results obtained from the re-administration of the BDI-SF scale, the total depression scale shows good internal consistency (alpha = 0.75). The mean score of caregivers' responses is 7.23 (SD = 4.12). Thus, there was no significant change between the pre-test and post-test; therefore, with respect to depression, caregivers maintained stable scores, experiencing neither significant worsening nor improvement.

The caregivers in our sample were distributed as follows: 13 (72%) placed themselves in the no-depression group and 5 (28%) in the mild depression group. None placed themselves in the moderate depression or severe depression ranges.

The scale has good reliability in the domain of hopelessness, detected through the BHS (alpha = 0.93). The mean score of caregivers is 6.72 (SD = 5.41). Concerning hopelessness, therefore, a significant change over time was observed (t(17) = 2.22; p = 0.041), i.e., a decrease in values compared to the pre-test (M = 8.83, SD = 5.40).

Compared with the normative values, the sample ranks as follows: 7 people (39%) fall into the absence of hopelessness, 5 (28%) into mild hopelessness, 4 (22%) into moderate hopelessness, and 2 (11%) into severe hopelessness.

Finally, about the burden of care experienced by caregivers, as measured by the FSQ, the caregiver's burden and stress scale for caring for a sick relative shows good reliability (alpha = 0.94).

The caregivers' mean score was 19.33 (SD = 7.88). No significant change over time was observed in this scale, so caregivers remained stable over time experiencing neither a worsening nor an improvement between the pre-test and post-test.

More specifically, our sample ranks as follows: 1 (5%) in the 'OK' area, 3 (17%) in the 'R' area, 5 (28%) in the 'SR' area, and 9 (50%) in the 'U' area (see Table 3).

Table 3 Descriptive statistics of study variables for target group at the post-test.

Caregivers (N = 18)					
Measures		alpha	Range	*M*	*DS*
RFQ	Certainty about mental states[1]	0.89	0-1.58	0.60	0.58
RFQ	Uncertainty about mental states[1]	0.77	0-1.47	0.68	0.41
BDI	Total Depression	0.75	2-15	7.23	4.12
BHS	Total Hopelessness	0.93	0-17	6.72	5.41
FSQ	Total Caregiver Strain	0.94	4-30	19.33	7.88

1A transformation by the square root of the values was adopted for both RFQ factors to normalize the distribution.

3.2 Qualitative Results

From the qualitative data analysis, four fundamental thematic areas emerged: 'Feedback on the intervention and support received from the association,' 'Lockdown: difficulties and sources of support,' 'Representations of death and dialogue in the family' and 'Emotional states, burden of caregiving and suggestions caregivers would like to give to others in their situation.'

3.2.1 Feedback on the Intervention and Support Received from the Association

The participants' feedback was collectively very positive, concerning both the actual intervention offered to their children and the effects on a family level that the participants detected, and, more generally, the support network provided by the association. The intervention of the project allowed caregivers to better manage their relationships with their children and with the patient; it was a chance for their children to be listened to and understood by, trusted external people and ALS experts. Manuela, for example, the 48-year-old caregiver of her husband, referring to her own change since the beginning of her journey with the association, said:

"I was certainly able to rationalize the disease, try to find my identity and not focus only on it; my life goes on, luckily with the rest of my family. Unfortunately, these are things that can happen; I was able, how can I say, to understand pain more, to rationalize pain more".

Another aspect that the participants reported as of great use regarding to being part of an association that supports people in their condition was the value of sharing, that is, the possibility to talk to a professional, discuss, ask for advice and be listened to. Giulia, a 51-year-old woman and caregiver of her husband underlined how this possibility of speaking with someone and receiving support was, in her opinion, a fundamental help:

"The fact of being able to speak freely about our difficulties with a competent person, conversations that we would never have had with other people ... the fact of having been listened to and being understood in our difficulties".

Many participants, including Manuela, described the association as a safe space, a network they could count on:

“Surely, with their advice ... knowing that the association was there for me, it was almost a safety net, a mattress I had in case I fell. I knew that I could make a phone call and meet with qualified people who know me and know what we are going through [...] I can talk about it to the general practitioner. I can tell a friend, but it's not like talking about it with professionals who have unfortunately seen many situations like ours”.

With regard instead to the actual psychological support intervention implemented with minors, caregivers have noticed many positive changes in their children, including greater serenity and awareness of the disease and its outcomes. In turn, caregivers have also impacted their lives, feeling calmer and more aware, rationalizing the illness and pain more, understanding how their children are coping, and feeling more supported and defended where before they felt weak and helpless.

For example, Silvia, a 52-year-old woman who is her husband's caregiver, reported:

“The fact that my son is calmer, yes. It also makes us parents calmer as well and gives us peace of mind because at the beginning he was very agitated, and very nervous. And, even though he initially did not want to take part in the intervention, when he had finished the sessions, he did not complain or say "I don't want to do it anymore" ... he was calm and serene".

Manuela, agreeing with Silvia, reported that this experience has made her daughter much more mature, more responsible and more aware:

“In my opinion she has matured, she has matured. Every now and then I asked her if she needed a chat and she said no, but maybe she too saw it as a kind of ... a lifeline, if one needed advice".

3.2.2 Lockdown: Difficulties and Sources of Support

Regarding the COVID-19 pandemic and, in particular, the moments of national lockdown imposed by the Italian government, some participants have noticed how, for them, accustomed to a rather withdrawn way of life, dedicated predominantly to the family and to the care of their loved one suffering from ALS, in fact there have been no striking changes in daily life, which was already poor in social activities even before the pandemic. Giorgio, a 41-year-old man, caregiver for his wife, for example, said:

“Our social life is very limited; therefore, let's say that we did not feel the difference between the normal period and lockdown periods in terms of social life; the changes were absolutely minimal".

Agreeing with him, Maria, the 49-year-old caregiver of her husband reported that the period of restrictions was not particularly burdensome, as she was already in the habit of staying at home.

“No, but it hasn't changed for me, because I'm already at home ... nothing changes for me, indeed, if I go out every day, half an hour in the morning is already a lot ... but nothing changes; I like to stay at home. I didn’t experience anything unusual”.

However, for most participants, further challenges and complications emerged due to the pandemic. Many have reported negative emotions such as loneliness, terror, isolation, abandonment, feeling at the mercy of events, difficulty staying away from the sick person to protect him or her, confusion, lack of control, lack of interest, indifference, fear of contagion, grief, despair, psychological fatigue, bewilderment, feeling helpless, and sadness. In particular, the greatest difficulty generally encountered was the imposed necessity to take care of the patient entirely by themselves, in the absence of the support that was previously provided by healthcare professionals (nurses, physiotherapists and doctors), who in that period were instead involved in the fight against the pandemic and therefore very much less able to deal with other types of pathologies. Alice, a 48-year-old female, caregiver for her father, said:

“In short, this lockdown did not help us; even the physiotherapy was suspended, precisely because the operators advised us not to go to the hospital. There was not enough personnel. Therefore the physiotherapist told me "no Alice" she made me understand that it was better to avoid it."

This situation meant that many participants felt alone, with all the weight of their family's care on them, and therefore helpless, as expressed, for example, by Giulia:

“Terror, loss, helplessness, the feeling of not being able to do anything. Yes. The lockdown period was the worst ever. [...] We have experienced the lockdown with my husband's illness, loneliness, everything. Precisely, "terror" is the exact word."

However, although it was much more difficult to receive adequate support from healthcare professionals during the lockdown, many participants reported that the proximity of family and their network of friends greatly supported them. Alice, for example, reported how her daughter gave her great support:

“[The daughter] helps me in the daily activities already with the mere fact that many times she does the shopping for me, she does the shopping for her grandparents, she collaborates like this ... then during the day she prepares meals for her father and her brother because I can no longer burden my mother with the thought of having to prepare food as well…".

Moreover, some participants also found support online, for example by sharing their experiences with groups of people who were in the same condition, as expressed for example by Francesca:

“I also subscribed to a Facebook group for patients and family members ... if I hadn't had this reference, I honestly don't know how I would have gotten out of this situation".

Another considerably problematic aspect that emerged significantly was also a conflict in the management of caregivers’ proximity to the patient: on the one hand, indeed, a person with ALS needs constant care that also includes physical proximity; on the other, the caregiver could be the cause of an involuntary contagion from COVID-19 and therefore a painful dilemma arises between having to take care of the loved one while fearing that the very act of caring for him/her might cause his/her health deterioration.

Francesca, a 45-year-old woman, and caregiver of her father, said in this regard, for example:

“Being away from people ... the more you want to protect them the more you have to stay away ... and there are people who can't stay away from you, who need you to be close to them, and so this has been complicated”.

This aspect has also led many caregivers to experience intense fear of contracting the COVID-19 virus. They were afraid of both becoming seriously ill themselves and, more often, of passing it on to a relative with ALS. The awareness also aggravated this fear that it was impossible to have total control over something as complex as a potential contagion, as Francesca expressed:

“First of all the fear of the virus entering the house, and the realization that the control of this thing does not totally depend on us."

Related in part to the previous theme, participants also reported difficulty in trusting people who could come in contact with their loved ones, even when those people were healthcare professionals, with a consequent increase in their sense of precariousness and anxiety. In this regard, Claudio, a 41-year-old man and caregiver of his father-in-law, said:

"Yes, because it is something that you cannot control, you just have to hope to be able to implement a series of precautions that can limit the risk, but you are always in the position of not being able to manage it completely, because you cannot know what contact a nurse has had, even just the fact of coming by public transport and going to my mother-in-law's house ... anything can happen".

3.2.3 Representations of Death and Dialogue in the Family

Regarding the sensitive theme of the possible prospect of their family member's death and how caregivers deal with it, many of the participants said they find significant comfort in their spirituality, and more specifically, in their case, in the Catholic religious faith. Many participants found comfort in believing that the loved one would still be able to live in the afterlife, in heaven, after his/her death, as indicated, for example, by Giuseppe, a 42-year-old man and caregiver of his wife:

"Ah, what can happen? Unfortunately, I hope she will be in the hands of God because she is a good person ... she has always been a good woman, very sensitive for her children, just as much as for the family ... I hope this".

However, other participants reported a more secular vision, and sometimes experiences of uncertainty and spiritual crisis resulting from the pain of the loss of the person they loved, as described, for example, by Giorgio, a 41-years-old man and caregiver of his wife_

"Let's say that at this stage I have a love-and-hate relationship with God. I have enormous hatred. I once told Dr M that I did not use to, but now I curse God, but on the other hand I am hopeful ... I hope to be understood".

On the other hand, dealing with the issue of death in the family was described as very difficult for caregivers. Many of them explained that this topic, referring to the conditions of the sick relative, is almost taboo. Silvia, the 52-years-old female caregiver of her husband, for example, said she does not want to talk about it with her children, but that everyone in the family knows what will happen anyway:

"It is something that I think and believe we often think about, but we avoid discussing it. I don't talk about it with my children, but we know it will happen sooner or later. We have also seen other stories of other sick people".

However, other participants instead reported that they tried to address the issue of the possible death of their loved one, particularly with their children, as reported, for example, by Riccardo, a 52-year-old male caregiver to his wife. Riccardo said that even though he did not try to directly and explicitly talk about it, he still tried to give his children an adequate temporal perspective concerning their mother's life, who was affected by ALS:

"The word [death] was not used very much, I think it was not directly spoken, it came out, but without directly saying it. We often talk about the future, tomorrow, ... I don't know. To give an example, I say to them: "Mom will no longer be there, but I'm here, I want ..." With M (son) I joked, "I want to become a grandpa. Let me become a grandfather immediately because, of course, you have to give me time. But remember, when you grow up and have children, I will be the grandfather who will take them for a walk and make them play", always without taking out that word [death] specifically".

Finally, others, such as Claudio, have instead declared that they have decided to be as transparent as possible with their children, and speak to them in a totally open way:

“We made this choice, in all things, beautiful or not, to always be clear and transparent, in order not to sell them a reality that is actually different. With the right ways, even though you never really know which are the right ones, but, let's say, with the right awareness of making them participate in everything that happens. With them we faced the fact that this thing will happen anyway when, we don't even know, but it will happen. This is synonymous with suffering, synonymous with detachment, but in my opinion the boys become even more aware in this way; I'm not saying they are ready, because they never will be, but perhaps with a few more elements to be able to face that moment”.

3.2.4 Emotional States, Burden of Caregiving and Suggestions Caregivers Would Like to Give to Others in Their Situation

Many participants described a significant burden with regard to their personal emotions and experiences related to their loved one’s disease, with an impact on the person's entire family, as Elisabetta, for example expressed:

“The disease does not only concern the sick person but all the people who live with him/her. I have always said, and I will always say that M. [sister-in-law] got sick but in reality, the whole family got sick with her".

Furthermore, from some interviews, caregivers expressed how they felt that taking care of their loved ones was exclusively their responsibility and that asking for help was imporssible. Flavia, a 36-year-old woman and her husband's caregiver, said, for example, that, in some moments of despair, she thought that the caregiver was the only one to carry this burden:

“Sometimes I felt like crying, other times I isolated myself, I tended not to speak, but in any case, I had to go on because I am one with that difficulty at home."

Furthermore, often taking care of a family member affected by ALS could become an all-encompassing element, which ended up completely characterizing the life of the caregiver, who was almost dedicated to this, as indicated, for example, by Giorgio:

“Finding a solution means annihilating the life even of the person who does not suffer from this disease, life understood as existence, unless this person's life is completely and solely contained in the personal relationship with the sick person. A disease of this kind means that the social, professional, cultural life of the person who takes care of the sick person is annihilated or greatly sacrificed”.

Another element that assumed a considerable negative weight in caregivers' lives was that they often perceived that others only perceived the suffering of their family members, which they recognized as enormous, but this did not allow validation of their difficulties and pains, which automatically became less important. This was, for example, underlined by Giorgio, who stated:

“This is not well understood either by A. [his wife with ALS], which is normal, or by outsiders who tend to think only of the patient. This is a sign of misunderstanding; if I may say, it is incommunicability in short ... it is also normal that in the face of the greater evil, the lesser evil would disappear [...]. However, it does not mean that that one does not count, it is obscured but also important."

About the weight that the same symptomatology can have in the caregiver's relationship with the loved one with ALS, one of the aspects that caregivers found more difficult to manage emotionally was the loss of their ability to communicate. This difficulty exacerbated the feeling that the patient was already gone, or that he/she was no longer really himself/herself. For example, Francesca reported:

“Sometimes it seems to me that I live as if he is almost no longer there, because in any case this fact that we cannot communicate ... he has somewhat gone out of our lives”.

Feelings of anger, frustration and helplessness also emerged in this regard. Elizabeth, for example, recounted her experience concerning these feelings regarding her sister-in-law's illness:

“Well initially [I felt] a great helplessness because when one does not understand and does not know what to do, he is faced with this disease and says «And now what do I do, how do I do it, what can I do?». [...] Sometimes [I felt] so much anger, so much anger, because it is unclear why we, or just her, could have suffered such a fate, such a disease ... yes, this is the inability to understand ... that's what's left. And we still don't know”.

In contrast, however, positive feelings were also experienced by the participants. Francesca, for example, believed that the disease should be considered an opportunity for growth and self-discovery, addressing this situation actively and proactively:

“I think these diseases are not just punishment; that's it. They could also be, for those who want to deal with them in an active and not passive way, a way to ... well, life has given you this thing, I had my father with ALS ... and so what do we do with this stuff? Let's try to understand if there can be a way to face life differently, as if grandfather and his illness could have given us a new perspective”.

Lastly, regarding what advice the participants would have liked to be able to give other people who were living with the same condition, an aspect reported by many was the importance of asking for help whenever they needed to, as reported by Giulia:

“Ask for help, help, help. Alone you are helpless in the face of certain things. Ask for help from the outside, whit support of all kinds. You need the support of all kinds. Psychological, medical, physical, because it is such a great thing, especially when one is young like my husband”.

Another aspect that emerged significantly was also the importance of being able to live one day at a time, trying to find a form of normality in this situation, as expressed, for example by Claudio:

“I would tell them to live day by day; whatever comes, it is always something more. Because there will be an end anyway, and this end you never know when it will come, so live every day in an attempt to make it as normal as possible. For us today this has probably become normality, absurdly enough. [...] the idea of living things day by day with absolute normality, with the awareness that it will have an end, because there will be, but to take everything beautiful you can find in the meantime".

4. Discussion

The present study aimed to understand how the lockdown has influenced the life of caregivers, how it has changed the management of their loved one’s ALS, their representations of the illness and the perspective of the loved one’s death, as well as the effects that have been observed following the psychological intervention received by the caregivers’ children, through both a quantitative approach and a qualitative one. In the following sections, both the quantitative and qualitative results will be discussed in detail.

4.1 Quantitative Discussion

The quantitative component of the study had the objective of assessing the caregivers' condition related to their ability to reflect and understand themselves and others in terms of behaviors guided by mental states, the eventual presence of depressive symptoms and hopelessness, as well as the presence of burden perceived because of their role, comparing the results with the condition presented by the caregivers following the performance of a psychological support intervention aimed at their children.

From the analysis of the collected data, it emerged that, at the pre-test phase, the caregivers presented a significant uncertainty regarding their mental states, in line with what has been observed in current literature concerning the impact that stressful and potentially traumatic events can have on people's reflective functioning [36]. It has indeed been observed how these events can affect one's ability and desire to identify with other people to understand their mental states, which can make traumatic situations more difficult to understand, reducing resilience, increasing the vulnerability of the person and leading to a 'selective switching off' of the prefrontal cortex, which is responsible for the reflective capacity [43]. Reflective functioning, which is context- and affect-dependent, could also present distinct levels of functioning to keep separate some contexts that would normally tend to integrate. In these cases, the self is organized in such a way that some internal models of functioning include reflective and highly functioning components, while other relational models appear to be depleted, indicating minimal mentalization skills [44].

Another important aspect that emerged was the fact that 19 participants (79%) had an absence of depression, 4 (17%) experienced mild depression, and 1 experienced moderate depression (4%). In addition, 9 of them (37.5%) did not live in a state of despair and hopelessness, 3 (12.5%) experienced mild despair, 7 (29%) experienced moderate despair and 5 (21%) felt severe despair. These results, in particular the presence of mild and (in one case) moderate depression, are in line with the current literature, given that several studies have found that depression in caregivers worsens over time with the severity of the disease and the increase of physical disability in patients with ALS [45-47]. Even regarding the experiences of despair and lack of hope, the results are consistent with what has emerged in previous research, particularly the lack of hope as one of the central cognitive components of depression. According to Beck, the author of the BDI and the BHS, what has been observed is in line with the literature concerning the presence of depressive symptoms in caregivers of relatives affected by neurodegenerative diseases reported above [48, 49]. Finally, regarding the caregivers' burden, the majority of the participants, 10 people (42%) found themselves in a condition of significant perceived weight and at high risk of a consequent disruption of their psychological well-being. This data is also supported by evidence in the literature, with numerous studies that have underlined the stress perceived by caregivers who take care of their relatives, with descriptions of feelings of overload, increasing fatigue, frustration and anger towards the disease, as well as conditions of economic burden [50, 51]. Often, this can lead to a series of hardships for the caregiver who has little time to dedicate to himself/herself and other significant relationships. This has frequently also negatively impacted intimacy in the relationship [8, 52].

On the other hand, the post-test results did not show significant changes in the previously described dimensions, which remained almost constant, except for the dimension of hopelessness, which instead showed a significant change over time, decreasing in comparison to the pre-test phase. More specifically, 7 people (39%) resulted in no condition of hopelessness, 5 (28%) in mild despair, 4 (22%) in moderate despair, and 2 (11%) in grave despair.

In any case, the fact that the other measures remained unaltered could also be understandable in light of the situation experienced by the participants during the period in which the study took place, namely the COVID-19 pandemic and the related lockdown. Indeed, some studies have shown that the impact of the pandemic was significant for both patients and caregivers, among whom higher levels of anxiety and depression, feelings of loneliness, greater vulnerability to distress, and greater symptoms of fatigue were observed, such as headaches, muscle aches and abdominal discomfort [18, 53, 54]. The results and comparison between the pre-and post-test phase suggest that the intervention improved the negative effects of caregiving on participants regarding a lack of hope for the future, and containment of the other dimensions of distress, since there is no worsening of depression, stress and care burden.

4.2 Qualitative Discussion

The qualitative component of the study investigated in depth the direct point of view of the participants, and, some central themes emerged from the data analysis. In particular, a central element reported by the participants was the usefulness and importance of the psychological support intervention received by their children, which gave them greater tranquillity and serenity, as reported for example by Silvia, and gave them a more mature perspective on the situation, as indicated for example by Manuela. The participants also noted the fundamental value of an adequate social networksupporting the families of people with ALS. Specifically, some participants (in particular Manuela and Giulia) underlined how being able to express their suffering and difficulties freely was for them, and how the presence of an association dedicated to people with ALS and their families was an important safety net for them. These aspects are in line with other studies in the literature, which have highlighted the importance of an adequate support network, including psychological resources, for people with ALS and their loved ones. which can allow them to adequately express their suffering [54-56].

Another aspect that has emerged from the research results was linked with managing family duties during the lockdown and the pandemic. While some participants (specifically Giorgio and Maria) did not notice any particular change during the lockdown, most participants reported that this period had a particularly intense impact on their daily life, and represented a significant challenge. This happened mostly because of the fear of getting sick and, both through one's proximity to and through external help from other people, of causing the sickness of loved ones suffering from ALS, thus aggravating their health condition (as reported, for example, by Francesca and Claudio). Another intense cause for aggravated pressure on caregivers during the lockdown and the pandemic in general was related to other practical difficulties such as, having to take care of their loved one alone, without being able to count on the professional support of healthcare professionals (as indicated in particular by Alice and Giulia). These aspects are in line with recent literature as well, since other studies have also highlighted how the COVID-19 pandemic period and the consequent lockdown have had a further negative weight on caregivers, both because of their intense fear of transmitting the infection to their relatives with ALS [14, 57], and because of an increase in the experiences of isolation and less support available from healthcare professionals [23, 58].

However, some participants also reported that during this period they fortunately also benefited from the support of their family and friends. This element partially compensated for the reduced availability of healthcare professionals, as indicated for example by Alice. Online support groups via social networks also represented an important source of support for some participants, as Elisabetta reported. All these elements have also been confirmed by other studies in the field, which have observed how mutual support groups (in person and online) provide effective and highly supportive social relationships [59, 60].

Furthermore, the participants also reflected on the complex issue of the life expectancy of their loved one with ALS and in this regard some of them (in particular Patrizia) underlined how death is still an almost taboo subject, difficult and painful to deal with in the family, albeit present in an unspoken way [26, 27]. This aspect has already been highlighted by much other literature on the subject, with studies that have indicated how it is still difficult, especially within Western societies, to accept the death of a loved one, an element that often leads to slipping into situations of the so-called 'Conspiracy of silence' in which no one talks about the imminent end of the life of a loved one even if everyone is aware of it [61-63]. However, other participants also reported how they chose to be open concerning this topic and discuss it in the family, especially with their children, as told, for example, by Riccardo and Claudio. There is growing evidence in the literature that talking about death does not frighten or make children and adolescents sad but allows them to have answers to questions about death and build resources to prepare for it and to grieve [64, 65].

Furthermore, participants also presented their own vision of what they believe could happen after death, with some of them strongly convinced that there will be a peaceful afterlife (Giuseppe and Elisabetta) and who therefore found comfort from their faith. In contrast, others showed instead greater skepticism and sometimes anger and signs of spiritual crisis, consequent to losing their loved one (such as Giacomo, for example). These different possible approaches to the end-of-life theme are both supported by other studies in the literature, with research that has confirmed the protective value of spirituality for people suffering from severe terminal illnesses and for their families [66-69], who manage to maintain higher levels of hope and less despair. In contrast, other studies have reported that the illness of a loved one can contribute significantly to spiritual crises and a perceived loss of meaning in existence [70].

Lastly, a final theme that emerged from the interviews was linked to the complexity of their caregiving experiences in general. In this regard, many participants reported how a diagnosis of ALS has an impact not only on the person who is affected but also on the whole family, representing a load of really intense care and commitment that can become all-encompassing and make the caregiver feel alone and crushed by it (as reported for example by Elisabetta, Flavia and Giorgio).

The main emotions that emerged from the interviews were sometimes suffering, a sense of helplessness and even anger particularly in cases where the disease made communication complex on the part of the patient, who is therefore present but ceases to be really active in the emotional relationship, as reported, for example. by Francesca and Elisabetta. This makes the person physically present but not psychologically; consequently, an actual situation of ambiguous loss is experienced, even without a definitive loss, as also pointed out in the literature [12, 13].

Another aspect that has been reported as particularly painful was also the realization by the caregivers, of how, since the suffering of their loved one is really enormous, their difficulties, their problems tend to be no longer considered, both by their loved one and from the rest of society, an aspect that increases their sense of isolation, as told for example by Giorgio. All these aspects are confirmed by other studies in the literature as well, with some articles that have underlined the considerable burden that the care of a family member with ALS can assume for the caregiver, with a possible increased risk of developing, for example, a major depressive disorder [11] and a higher correlation to the risk of hypertension [58].

However, testimonies from different perspectives have also emerged; more specifically, some caregivers reported how, although their condition is complex, their loved one's illness also represented an opportunity for them to re-evaluate what really matters in life, and to learn to enjoy every moment they can dedicate to being with their loved one, as reported by Francesca for example. Similar results have also emerged in other studies in which positive emotions were associated with the ability to draw strength from the patient and search emotionally for the best way to cope with this situation [46].

Lastly, about this last aspect, some participants also reported some advice that they would like to give to other people in the same situation as themselves. In this regard, many of them agreed that an important aspect is not to be afraid to ask for help if a person feels he/she needs it, as suggested, for example by Giulia, while others, such as Claudio, have emphasized how it is really essential to be able to go on day by day, appreciating every moment one can have with the loved one affected by ALS. In the current literature, some studies have confirmed that being able to face one day at a time with positivity and hope can produce a sense of reciprocity in the caregivers and in the ill person, which, consequently can lead to feelings of well-being both in the patient and in the caregiver, a resilience factor that can mitigate the pain that this situation can generate [59, 71].

5. Conclusion

This research aimed to understand how COVID-19 lockdowns have affected the lives of caregivers, how caregivers' management of their relatives' illnesses has changed, representations of illness and death, and the effects of the psychological interventions received by the children and adolescents. From what emerged from the study, the interventions made on families positively affected children and their parents. Caregivers reported that they perceived a climate of greater serenity within families. This research has made it possible to understand the greatest sources of suffering experienced by families with a relative living with ALS, exacerbated by the COVID-19 pandemic. The intervention was carried out during the lockdown period. This allowed us to highlight social isolation's impact on caregivers and their families and how this has aggravated an already very complex situation. Despite this, however, being already accustomed to a life of renunciation, sacrifice and constant presence for the patient had meant that some caregivers did not see differences with everyday life before COVID-19 and that they had more time together. Family and friends have proved to be a great source of support, albeit with this period's limitations. The support received has also shown positive sides to their difficulties.This study also aimed to assess the caregivers' condition about their ability to reflect and understand themselves and others in terms of behaviors guided by mental states, the eventual presence of depressive symptoms and hopelessness, as well as the presence of burden perceived because of their role. As the results show and in line with previous studies, the impact of the disease on informal caregivers is significant. It is essential that the management of patients with ALS also includes an intervention aimed at the whole family.

6. Limitations and Future Directions

One limitation of this study is that it investigated the experiences and emotional status of caregivers indirectly investigated caregiver's experiences and emotional status, without providing a specific psychological intervention for them. We we believe that the main limitation of the research is the lack of a targeted psychological intervention for caregivers. Future projects should consider this, as it would be helpful to assist caregivers in coping with the difficulties they encounter in managing patient and family relationships, as well as in dealing with negative emotions such as anxiety, loss of hope, and loneliness. Another limitation of the project is that it did not explore varying experiences of caregivers based on the degree of kinship with the patient or the stage of each ALS patient, which could affect the psychological state and burden of the caregiver and other family members.

Author Contributions

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Competing Interests

The authors have declared that no competing interests exist.

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