



Patient and carer involvement in the formulation of research questions: findings from the Italian guideline on palliative care in adults with glioma

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Abstract

Background In 2017, the European Association for Neuro-Oncology (EANO) published the guideline for palliative care (PC) in adults with glioma. The Italian Society of Neurology (SIN), the Italian Association for Neuro-Oncology (AINO), and the Italian Society for Palliative Care (SICP) joined forces to update and adapt this guideline to the Italian context and aimed to involve patients and carers in the formulation of the clinical questions.

Methods During semi-structured interviews with glioma patients and focus group meetings (FGMs) with family carers of deceased patients, participants rated the importance of a set of pre-specified intervention topics, shared their experience, and suggested additional topics. Interviews and FGMs were audio-recorded, transcribed, coded, and analyzed (framework and content analysis).

Results We held 20 interviews and five FGMs (28 carers). Both parties considered the pre-specified topics as important, chiefly information/communication, psychological support, symptoms management, and rehabilitation. Patients aired the impact of focal neurological and cognitive deficits. Carers reported difficulties in dealing with patient's behavior and personality changes and appreciated the preservation of patient's functioning via rehabilitation. Both affirmed the importance of a dedicated healthcare path and patient's involvement in the decision-making process. Carers expressed the need to be educated and supported in their caregiving role.

Conclusions Interviews and FGMs were well informative and emotionally challenging. Both parties confirmed the importance of the pre-specified topics, and carers suggested one additional topic: education/support to caregivers. Our findings strengthen the importance of a comprehensive care approach and of addressing the needs of both patients and their family carers.

Keywords Brain tumors · Gliomas · Palliative care · Practice guideline · Consumer involvement · Qualitative study

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Introduction

In 2017, the European Association of Neuro-Oncology (EANO) published the guideline for palliative care (PC) in adults with glioma [1]. Based on the most recent definition, PC is the active holistic care of individuals across all ages with serious health-related suffering, because of severe illness, and especially of those near the end of life [2]. PC aims to improve the quality of life of the patients, their families, and their caregivers [3]. Glioma sufferers and their families have to deal with the many health issues of a rapidly progressive condition, making PC important throughout the whole disease trajectory. The EANO guideline has identified three main areas of PC for adult patients with glioma: symptom management, patient and caregivers needs, and PC and end-of-life phase [1]. The guideline is

currently being updated and adapted to the Italian context by an interdisciplinary panel appointed by the Italian Neurological Society (SIN), the Italian Society for Palliative Care (SICP), and the Italian Association of Neuro-Oncology (AINO). The panel decided to involve healthcare professionals (HPs), glioma patients, and caregivers in the formulation of the clinical questions to increase the relevance and trustworthiness of the SIN/SICP/AINO guideline.

Patient and public involvement in clinical guideline production and dissemination has long been recommended [4]. However, such involvement remains suboptimal, and the best strategies of engagement are a matter of discussion [5–8].

The methods of our consultation are outlined in Fig. 1.

Briefly, the guideline panel devised 14 intervention topics (Table 1). The importance of each topic and suggestions of additional topics were appraised by the stakeholders

Fig. 1 Outline of the mixed-method project. The blue frame identifies the qualitative study reported herein. EANO, European Association for Neuro-Oncology; HP, healthcare professional; FGM, focus group meeting

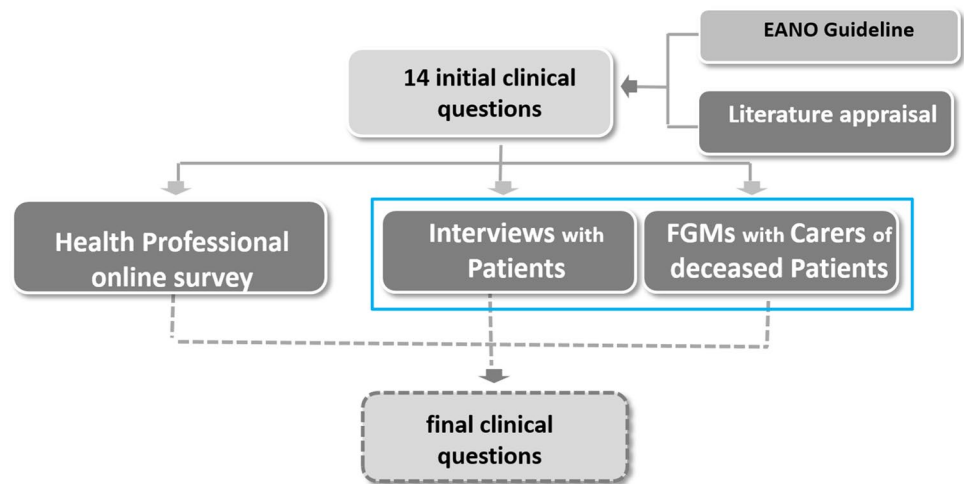


Table 1 The 14 intervention topics devised by the Italian guideline panel

No	Description	Source
1	Symptoms management	EANO guideline
2	Rehabilitation	EANO guideline
3	Information and communication	EANO guideline
4	Psychological support	EANO guideline
5	Spiritual and existential support	New
6	Social support	EANO guideline
7	Service organization	EANO guideline
8	Advance care planning	EANO guideline
9	End of life care	EANO guideline
10	Bereavement support	New
11	Psychological support to healthcare professionals	New
12	Training in palliative care of healthcare professionals	New
13	Early palliative care	EANO guideline
14	Care pathways	New

EANO, European Association for Neuro-Oncology

Topics 1/8 were presented during the interviews and topics 1/10 during the focus group meetings

using a mixed-model design: an HP online survey, whose results have been described elsewhere [9], and a qualitative study, consisting of personal interviews with patients with glioma, and focus group meetings (FGMs) with caregivers of bereaved patients.

Materials and methods

This qualitative, multicenter study consisted of two parts: personal, semi-structured interviews with glioma patients, and FGMs with family carers of recently deceased patients. Participants were selected using a purposeful sampling technique. The patient's referring physician at the center approached potentially eligible participants, informed them about the study aims and what their participation in the study consisted of, and gathered the signed informed consent form. The consolidated criteria for reporting qualitative study (COREQ) checklist [10] are reported in Supplementary file 1.

Personal semi-structured interviews

We recruited adult outpatients with grade III/IV gliomas, followed at one of the five participating centers. We excluded patients with severe cognitive compromise (clinical judgment) or impairment precluding communication. The intention was to recruit patients who varied in terms of age, gender, and disease trajectory. Interviews were face-to-face, they were arranged in a quiet room at the center, and lasted for a maximum of 60 min. The interviewers were trained researchers; they used a semi-structured interview guide produced by the guideline panel, consisting of four sections: (I) introduction and recap of the objectives of the interview. (II) Exploration of the main problems encountered by the patient along his/her disease. (III) Presentation of 8 pre-specified topics and discussion of the most important ones based on patient's own experience. (IV) Probing additional topics that the patient considered important (Supplementary file 2).

Focus group meetings

Participants in the FGMs were recruited from family carers of glioma patients deceased in the preceding 6 to 24 months. The intention was to recruit carers varied in terms of age, gender, and relation with the patient. The time from bereavement was chosen to exclude the early phase of grieving, at the same time, assuring that the whole disease trajectory could be remembered with sufficient detail. Each FGM was planned to include 6–10 participants, plus 2 moderators; it lasted for a maximum of 90 min and was held online (Teams or Lepida conference system). One moderator (facilitator) had no previous contact with participants; his/her task was to engage participants,

promote exchanges, modulate conflicts, and ensure that all the topics were adequately covered, while allowing time for exploration of any pertinent issues arising. The co-moderator took notes, noted relevant non-verbal communication, assisted with logistics, and oversaw the audio recording. The moderators used a semi-structured FGM guide that consisted of four parts: (I) outline of the objectives of the study, meeting procedure, and invitation of each participant to introduce him/herself. (II) Presentation of 10 pre-specified topics and identification by each carer of the three most important ones, based on carer's own experience. (III) Discussion of the topics, starting from those rated as the most important. (IV) Probing additional topics that the carers considered important (Supplementary file 2).

Analysis

Interviews and FGMs were audio-recorded and transcribed. We planned a minimum of 12 interviews and five FGMs (one at each center). FGM reports were sent to participants for approval.

Two researchers (EB, BL) analyzed the data using framework and content analysis, in a mainly inductive approach, working with Microsoft Word files within an online shared folder [11]. This analytic approach was considered appropriate for making themes emerge and allowing inter-coder agreement. EB and BL independently read the transcripts extensively, wrote comments and initial thoughts in a memo, and coded the text of the interviews and FGMs. They then jointly discussed the list of provisional themes, renamed the themes when needed, and defined subthemes. Finally, they extracted the most meaningful narratives from the interviews and FGMs to draft the final report, which was discussed with SV, LDP, AS, and AP.

Ethics statement

The research project was approved by the Ethics Committees the IRCCS-Regina Elena National Cancer Institute (ref. 1240/19); Fondazione IRCCS Istituto Neurologico Carlo Besta (ref. 65/18.09.2019); Azienda USL-IRCCS di Reggio Emilia (ref. 2020/0063420); ASST Papa Giovanni XXIII, Bergamo (ref. 219/19). All participants provided signed informed consent. They did not receive any financial compensation for their participation in the study.

Results

Between February and October 2020, 20 outpatients with glioma were approached (four from each participating center), all of whom participated. The interviewers were three neurologists, two bioethicists, and a research nurse experienced in conducting interviews and FGMs; they were

trained in the study procedures and use of the interview guide (Supplementary file 3). The patients' general and clinical characteristics are reported on Table 2: their median age was 56 years (range 38–78), 70% were men, and 60% were full-time employed. The Median Karnofsky Performance Scale score was 90 (range 60–100), and one-quarter had mild cognitive impairment. All the patients had received surgical therapy, and 20% re-surgery for tumor recurrence; 80% were on treatment, mainly chemotherapy.

The five FGMs were held between December 2020 and March 2021 and lasted 71 to 127 min (median 97 min). The moderators were two neurologists, two bioethicists, a palliative care physician, and a psychologist. All were experienced in conducting FGMs and trained in the study procedures and use of the interview guide (Supplementary file 3). The 28 family carers had a median age of 53 years (range 25–67), 82% were women, and 62% were the patient spouse or partner. Time from patient's death ranged between 6 and 27 months (Table 3).

We found the following protocol deviations: ten interviews (2 in Turin, 4 in Bergamo and Rome) and one FGM (Rome) were conducted by HPs involved in patient's care. At Reggio Emilia, the patient referring neurologist was present during the interview, with patient agreement. Finally, two FGMs (Bergamo and Reggio Emilia) had only four participants, and participants to these FGMs did not receive the report for approval (Supplementary file 1). Meeting reports were approved by all participants in the Milan, Turin, and Rome FGMs.

Both patients and family carers prioritized four of the pre-specified topics: information and communication, psychological support, symptoms management, and rehabilitation. The qualitative analysis identified 37 sub-themes, grouped into eight themes (Table 4). Quotations for each theme and subtheme are reported in Supplementary file 3. Six of the themes were common to patients and carers; of these, four corresponded to the prioritized topics reported above, plus decision making and service organization. Participation was a theme identified by patients only, and economic and social issues are themes identified by carers only. Most of the themes were related each other, in variable degree (e.g. information/communication and decision-making). Except for HP psychological support, all the pre-specified intervention topics (including those not presented during interviews and FGMs) pertained to one or more themes (Table 4).

Information and communication

Patients emphasized the importance of being listened to and of a personalized communication based on their current needs and awareness. They wanted to receive clear information about their condition, its course and treatments, but also to maintain hope:

Table 2 General and clinical features of the 20 interviewed patients

Characteristic	N (%)
Women	6 (30)
Age, years ¹	56 (38–78)
Education	
Middle school	3 (15)
High school	10 (50)
Bachelor's degree	1 (5)
Master's degree or higher	6 (30)
Employment	
Employed	12 (60)
Retired (age)	5 (25)
Retired (disability)	1 (5)
Homemaker	1 (5)
Not reported	1 (5)
Marital status	
Spouse/partner	18 (90)
Divorced	1 (5)
Widower	1 (5)
Living with:	
Partner and children	10 (50)
Partner	9 (45)
Children	1 (5)
Years since diagnosis	
≤ 1	5 (25)
2–5	10 (50)
> 5	5 (25)
Tumor grade	
IV	13 (65)
III	6 (30)
II	1 (5)
Histology	
Glioblastoma	13 (65)
Anaplastic astrocytoma	5 (25)
Anaplastic oligodendroglioma	2 (10)
Karnofsky performance status score ¹	90 (60–100)
Cognitive impairment	
Absent	15 (75)
Mild	5 (25)
Previous treatment (in addition to surgery)	
Chemotherapy and radiotherapy	9 (45)
Radiotherapy	4 (20)
Re-surgery, chemotherapy and radiotherapy	4 (20)
Chemotherapy	2 (10)
No treatment	1 (5)
Current treatment	
Chemotherapy	14 (70)
No treatment	5 (25)
Chemotherapy and radiotherapy	1 (5)

¹Median (min–max)

Table 3 Characteristics of the 28 family carers

Characteristic	N (%)
Women	23 (82)
Age, years ¹	53 (25–67)
Education	
Middle school	1 (4)
High school	17 (61)
Master's degree or higher	10 (35)
Relationship with the patient	
Spouse/partner	18 (64)
Son/daughter	8 (29)
Sister	2 (7)
Months since patient's death ¹	20 (6–27)
Patient's place of death	
Home	15 (54)
Hospice	6 (21)
Hospital	3 (11)
Not reported	4 (14)

¹Median (min–max)

“I believe that some information was not given so as not to add fears upon fears. However, communication is important. It is important to say: we'll do this first and then that” (47-year-old man, KPS 90, Reggio Emilia).

Family carers of bereaved patients agreed on their role as mediators of communication between patients and HPs. They took responsibility for deciding how much to inform the patient, a role that they perceived as an additional burden:

“I found myself always going an hour before the visit to explain the things she knew and didn't know. I was afraid that it would come out too much. Eventually my hands were tied: when a person is too lucid it becomes difficult to tell her false truths!” (40-year-old son, Turin).

Carers valued HPs who communicate with the patient in a measured, individualized approach. In addition to information provision, HPs should listen to the patient, be sensitive, tactful, and supportive:

“It's important that the patient knows, that he is aware of what is happening to him. But in my opinion you (the HP) have to be good in doing that. You must understand whom you are dealing with... There are people you can push harder and people you can't” (51-year-old wife, Rome).

Psychological support

Patients reported many psychological symptoms, including loneliness, rage, anger, depression, and fear of dying.

Feeling as a family burden was an issue for many of them. Nevertheless, they recognized the importance of psychological support mainly on a theoretical basis. Rather, patients relied to the support received within the family:

“My loved ones know perfectly what I am like. They told me everything that could be good and everything that could be bad” (68-year-old woman, KPS 100, Reggio Emilia).

Carers found that HPs scan sparsely the mood symptoms of the patients, preventing appropriate referral and treatment. Carers' own mood symptoms originate from many issues, including a sense of loneliness in dealing with patient's complex medical problems, patient's cognitive and behavioral changes, and role changes within the family. For many of them, these psychological needs become apparent after patient's death:

“I had incredible strength because I never collapsed, I was always there. Now instead, a few months after his disappearance, in a period in which there are no more commitments... I've collapsed” (67-year-old sister, Turin).

Decision-making

Patients expressed doubts about the degree of their involvement in the decision-making process; they did not feel competent in the medical issues and were concerned about making the doctor waste his/her time. Nevertheless, they considered as important to be informed about the treatment options and to express their wishes and preferences:

“I'm not a doctor and I can't judge the goodness of the treatments they offer me. I entrust myself to the doctor I trust and I accept what he proposes but I request complete information” (70-year-old man, KPS 80, Bergamo).

None of the patients had drawn up their advance care plan, and few knew about the Italian Law 219/2017 (on advance directives and advance care plan). A few of the patients reported they had informally shared their wishes with a family member:

“My family is close to me, we have talked about my thoughts and wishes [...] As long as there is something to do, I hope I don't give up and go on fighting” (69-year-old man, KPS 80, Bergamo).

Carers reported that they took responsibility of the patient's medical decisions, and they still did not know whether they had made the right decisions:

“Perhaps the thing was, after recurrence, choosing for him whether to continue or not (...). He knew

Table 4 Findings of the combined qualitative analysis

Patients		Carers	
Theme	Sub-theme	Theme	Sub-theme
Information and communication - <i>ACP</i> - <i>HP training in PC</i> - <i>Early PC</i>	<ul style="list-style-type: none"> • Listening to the patient • Telling the truth • Personalized information in a progressive approach • Informing while preserving hope 	Information and communication - <i>ACP</i> - <i>HP training in PC</i> - <i>Early PC</i>	<ul style="list-style-type: none"> • HP competences in communication • Filtering information • Tailoring information on patient's needs
Psychological support - <i>Symptoms management</i> - <i>Spiritual/existential support</i> - <i>ACP</i> - <i>HP training in PC</i>	<ul style="list-style-type: none"> • Mood and behavioral problems • Fear of dying • Feeling as a burden • Receiving support 	Psychological support - <i>Symptoms management</i> - <i>Spiritual/existential support</i> - <i>ACP</i> - <i>Bereavement support</i> - <i>HP training in PC</i>	<ul style="list-style-type: none"> • Role change within the family • Patient's psychological needs • Carer's psychological needs during disease and bereavement
Decision making - <i>Information/communication</i> - <i>ACP</i>	<ul style="list-style-type: none"> • Patient's engagement and involvement • Respect of patient's values and preferences 	Decision making - <i>Information/communication</i> - <i>ACP</i>	<ul style="list-style-type: none"> • Carer's understanding and respect of beloved's values and preferences • Patient's engagement and involvement • Carer's knowledge of disease prognosis
Symptoms management - <i>Rehabilitation</i> - <i>Service organization</i> - <i>HP training in PC</i> - <i>Early PC</i> - <i>Care pathways</i>	<ul style="list-style-type: none"> • Focal neurological symptoms • Cognitive impairment 	Symptoms management - <i>Rehabilitation</i> - <i>Psychological support</i> - <i>Service organization</i> - <i>HP training in PC</i> - <i>Early PC</i> - <i>Care pathways</i>	<ul style="list-style-type: none"> • Focal neurological symptoms • Cognitive and behavioral symptoms • Epileptic seizures
Rehabilitation - <i>Symptoms management</i> - <i>Information/communication</i> - <i>Service organization</i> - <i>Early PC</i> - <i>Care pathways</i>	<ul style="list-style-type: none"> • Physiotherapy for motor problems • Rehab of speech and cognitive problems 	Rehabilitation - <i>Symptoms management</i> - <i>Information/communication</i> - <i>Service organization</i> - <i>End of life care</i> - <i>HP training in PC</i> - <i>Early PC</i> - <i>Care pathways</i>	<ul style="list-style-type: none"> • Along the disease trajectory, goals change over time • Preserving wellbeing and dignity
Service organization - <i>Service organization</i> - <i>Early PC</i> - <i>Care pathways</i>	<ul style="list-style-type: none"> • Care team approach • Service co-operation 	Service organization - <i>Service organization</i> - <i>Early PC</i> - <i>Care pathways</i>	<ul style="list-style-type: none"> • Need for of palliative care and other services • Need for care continuity
Participation - <i>Symptoms management</i> - <i>Rehabilitation</i> - <i>Social support</i> - <i>Service organization</i>	<ul style="list-style-type: none"> • Preserving personal and social role • Car driving • Work ability 	Economic and social issues - <i>Social support</i> - <i>Service organization</i> - <i>Early PC</i> - <i>Care pathways</i>	<ul style="list-style-type: none"> • Professional caregiving • Economic and working problems for family members

Six of the eight themes were common across patients and carers. The (additional) pre-specified intervention topics related to each theme are reported in italics

ACP, advance care planning; *HP*, healthcare professional; *PC*, palliative care

very well that the tumor he had returned, and it was no longer possible to operate. But let's say that it was not easy to choose to do the second line. I wasn't in his head so I can say what I would have done for me, and I don't know yet what the best thing is" (25-year-old daughter, Milan).

Patient's clinical worsening was often unexpected to family carers, and quickly new decisions had to be taken:

"We find ourselves facing deterioration that we didn't know how to manage; this is the main thing we had difficulty with; we lacked information of what we should envisage; I expected the doctor to prepare us for this" (56-year-old wife, Milan).

Most of the carers considered essential to involve the patient in the decision-making process. Carers had little knowledge of the Law 219/2017.

Symptom management

Besides mood and behavioral changes reported above, patients reported symptoms due to focal deficits (e.g., mobility problems or speech problems) as a challenge. Cognitive compromises, such as memory loss, attention difficulties, and speech problems, were also taxing. Three subthemes emerged from the FGMs: focal symptoms; cognitive and behavioral symptoms; epileptic seizures; and other emergencies. Focal symptoms, such as aphasia, paralysis, bowel and bladder incontinence, dysphagia, and difficulty with walking, resulted in increased care needs and restriction of patient's autonomy and dignity. Cognitive and behavioral symptoms, such as mood symptoms (e.g., agitation, depression), personality changes (e.g., disinhibition, aggressiveness, and apathy), disorientation, memory loss, and reduced decisional capacity, were the most complex aspects for carers to manage. Carers felt alone in understanding and coping with these changes:

"It took him away from me first as a person, and then as a living being; he got lost, never got home, he was no longer attentive to his daughters, he was very selfish... My husband eventually became violent; he also raised his hands, with me and with my children" (58-year-old wife, Reggio Emilia).

The management of epileptic seizures and of patient's choking was an emergency for which carers did not feel prepared or educated:

"I also learned to intubate her with a piece of cannula because one evening she had an epileptic attack and she swallowed her tongue. I was in a panic because the ambulance didn't arrive... I broke two of her front teeth with the cannula" (49-year-old husband, Milan).

Carers pointed to the need for informing them on "what to expect", educating them on "what to look for", and particularly "what to do" in case of uncontrolled symptoms and emergencies.

Rehabilitation

Patients recognized the importance of physiotherapy, speech therapy, and cognitive retraining for returning to a "normal" life. However, physiotherapy was the only type of rehabilitation they had experienced. Patients expressed the desire to receive cognitive rehabilitation:

"I wish there was more attention to rehabilitation of memory, my current only problem. But I know the therapies for this problem are limited..." (49-year-old man, KPS 80, Turin).

Rehabilitation was especially relevant for carers, from first surgery to the end of life period. Goals differed along the disease trajectory and ranged from preservation of functional independence:

"He had great results in the first phase, they really got it back on its feet, it's been a phenomenal year" (65-year-old husband, Rome).

to the relief of pain, dyspnea, and other symptoms in the end-of-life phase:

"It gave her back and limbs relief" (67-year-old sister, Turin).

Carers commented that by working on functional independence, rehabilitation had a substantial impact on the patient's well-being and quality of life. Moreover, benefits of rehabilitation extend to the family carer, in terms of improved physical and psychological well-being and reduced caregiving burden:

"It was key, because my husband remained "standing up" to a month before... He was self-employed and I could go to work, rehab gave strength to both him and me, to manage with everyday life" (51-year-old wife, Rome).

Service organization

Both patients and carers argued that fragmentation of care and silo working are problems. Patients tended to report good experiences, while carers were more critical about the difficulties encountered during the disease trajectory. Patients recognized some qualities of the professionals who cared for them: expertise, humanity, availability, and punctuality. Especially, they felt protected by being in a path of care:

"The outpatient path is managed well; I always interface with the same people and this makes me very happy; not having to travel around so many clinics is convenient and having a well-organized itinerary is practical" (70-year-old man, KPS 80, Bergamo).

Carers too emphasized that continuity of care was vital. Many had trouble in having access to services, including PC (either at home or in hospice), social services, and rehabilitation services. They voiced the need of implementation of the patient's care plan in a responsive way, ensuring a smooth flow of information across services:

"For home assistance, there were a thousand and one problems, a thousand and one documents to present ... which means time, bureaucratic difficulties... Finally, at the hospice I felt relieved of many responsibilities" (57-year-old wife, Reggio Emilia).

Participation

This theme pertained to the patients only, who were concerned about preserving their role within their community. Car driving and seizure control were among patient

priorities. Retaining work was another issue, with important personal and economic implications:

“Memory is a bit of a problem at work, but I’ve organized myself, I write everything down; even at home, I’m full of notes ...” (49-year-old man, KPS 80, Turin).

Economic and social issues

Family caregivers felt overwhelmed by various social and financial problems, which added to the difficulty of coping with their loved one’s diagnosis. Accompanying the patient to the hospital for consultations, exams, therapies, obtaining mobility aids, and other equipment, extricating from bureaucracy led to a rapid restriction of social life both for the patient and for family carers:

“My sister and I took care of everything, with the help of few other people, because our family consisted of just the three of us. My sister had applied for the “104” (Italian Law 104 on paid leave from work), which was revoked when my father was well. If the bureaucracy were more streamlined!” (35-year-old daughter, Milan).

Most family carers described a laborious and frustrating search for a professional caregiver who could help with patient care, including personal hygiene, mobility in or out of the home, and other activities of daily living:

“For home assistance, there were a thousand and one problems, a thousand and one documents to present ... which means time, bureaucratic difficulties... Finally, at the hospice, I felt relieved of many responsibilities” (57-year-old wife, Reggio Emilia).

Financial and employment matters, including periods of protected leave from work, were another concern.

Discussion

Patient engagement in the development of clinical practice guidelines is recommended, with the aim to increase guideline legitimacy and transparency and the likelihood of their adoption [6, 7, 12]. Specifically, the AGREE II instrument emphasizes the importance of consultation with patients to determine the priority topics, in order to ensure that the guideline developers are addressing patient’s needs [6]. The present study focused on the appraisal and input on the guidelines’ clinical questions by patients with glioma and family carers of recently deceased patients. Involving both patients and family carers was vital for two main reasons: first, PC typically addresses the patient and his/her caregiver as a dyad, and some issues (e.g. bereavement support) are specific to the caregiver; second, carers function as proxy informants of critically ill patients and of patients who

have lost decisional capacity. The ultimate goal of our project was the meaningful and equitable engagement of stakeholder groups (patients, family carers, and HPs) living in Italy, in order to ground the clinical questions to the needs of those potentially affected by the SIN/SICP/AINO guideline recommendations.

Glioma patients and carers participated openly and wholeheartedly in the study, sharing their experiences, thoughts, and feelings. Some interviews and FGMs lasted longer than scheduled to accommodate to the information richness of the participants, and emotions were often tangible. This was the case particularly for the FGMs, where carers recounted and shared their recent experiences. Both parties considered all the pre-selected intervention topics as important and prioritized four of them: information and communication, psychological support, symptoms management, and rehabilitation. Most of the themes identified were related to each other, in variable degree (e.g. information/communication and decision-making). Furthermore, three of the four topics devised by the guideline panel that were not presented during interviews and FGMs pertained to most of the themes: HP training in PC, early PC, and care pathways (Table 4). From the FGMs, it emerged a need of the family carers to be educated and supported in their caregiving role. This additional intervention topic was also suggested in the online HP survey that was part of our mixed-method consultation project [9]. It is worth mentioning the recent publication of a randomized controlled study assessing the effectiveness of a supportive educational intervention for carers of glioma patients, showing a significant improvement in carer preparedness, while carer distress was not reduced [13].

The management of mood symptoms and behavioral changes was critical to both patients and carers. This is in line with qualitative studies where family carers reported changes in family relationships, loss of their old life and patient’s old self, while patients described their life as marked by loss [14–16].

Service organization was a theme connected with all the other ones. Many caregivers reported difficulties in having access to rehabilitation, psychological support, or PC services. They complained for care fragmentation and huge bureaucracy. This is consistent with findings from our online survey, where HPs considered key the early integration of neuro-oncology and PC [9]. While patients tended to appreciate the relational competences of the HPs, carers’ experiences were mixed, and they asked for improved competences of the HPs in communication and in shared decision-making.

Strengths of the study are the well-varied characteristics of the participants and the fundamental contribution provided by family carers of recently deceased patients. Carers helped understand the challenges of severely affected patients and patients lacking decisional capacity. They also shared their grieving experience and needs.

Limitations of the study include the fact that the FGMs were online due to the pandemic; not all of the pre-specified intervention topics could be discussed in detail during each interview/FGM; there were some protocol deviations, which could have

biased the results. These include the fact that half of the interviews and one FGM were held by HPs involved in patient's care, and in four interviews, the patient referring neurologist was also present. Finally, we did not include a formal evaluation of participant's satisfaction of the engagement experience and suggestions for improving engagement in the future [12].

Conclusions

Patient interviews and carer FGMs allowed to confirm the importance of the intervention topics identified by the guideline panel and to propose one additional topic. These data, together with those obtained from HPs, will inform the formulation of the final guideline PICO questions.

Supplementary Information The online version contains supplementary material available at <https://doi.org/10.1007/s10072-023-06687-y>.

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Data availability The data that support the findings of this study are available on request from the corresponding author, AS. The data are not publicly available as they contain information that could compromise the privacy of research participants.

Declarations

Ethics approval The research project was approved by the Ethics Committees the IRCCS-Regina Elena National Cancer Institute (ref. 1240/19); Fondazione IRCCS Istituto Neurologico Carlo Besta (ref. 65/18.09.2019); Azienda USL-IRCCS di Reggio Emilia (ref. 2020/0063420); ASST Papa Giovanni XXIII, Bergamo (ref. 219/19). All participants provided signed informed consent. They did not receive any financial compensation for their participation in the study.

Conflict of interest Dr. Solari reports grants from the Italian Multiple Sclerosis Foundation (FISM) and the European Academy of Neurology, during the conduct of the study; she serves as board member for Merck Serono and received personal fees from Almirall and Merck Serono, outside the submitted work. Dr. Rudà received honoraria for lectures or advisory boards from Bayer, Genenta, Novocure, and UCB. All the other authors have nothing to declare.

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