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Being the next of kin of a person with a brain tumor: a metasynthesis focusing on coping factors and strategies

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ABSTRACT

Introduction: Being the next of kin of a person with a brain tumor is a stressful experience. For many, being a next of kin involves fear, insecurity, and overwhelming responsibility. The purpose of this study was to identify and synthesize qualitative primary studies to explore coping factors and strategies that next of kin use in their role.

Methods: A qualitative metasynthesis guided by Sandelowski and Barroso's guidelines was used. The databases Medline, CHINAL, and PsycINFO were searched for studies from January 2000 to March 3, 2021. The inclusion criteria were: qualitative primary studies focusing on factors or strategies used by the next of kin aged 18 years of age or older of persons with brain tumors.

Results: Of 1371 screened records, data from 19 studies, including 332 participants (200 female, 78 male, and 54 unclassified) were analyzed into metasummaries and a metasynthesis. The next of kin rely on coping factors such as their personal characteristics, finding meaning in their situation, external support, hope and religion, and having someone to talk to. Strategies to manage the situation involve regaining control, being proactive, and acceptance.

Conclusion: Coping factors and strategies within themselves, in their surroundings and assistance from a higher power are used by those who are next of kin for people with brain tumors. It is important that health-care professionals suggest and facilitate these coping factors and strategies because this could reduce stress and make the role of next of kin more manageable.

Keywords: brain tumor; coping factors; coping strategies; metasynthesis; next of kin; review

Strengths and limitations of the study

- The qualitative approach makes an important contribution to the research field by providing a deeper understanding of coping factors and strategies used by the next of kin of a person with a brain tumor.
- Most of the included studies in this metasynthesis were high-quality studies.

- Our sample is highly multicultural with different geographical origins represented and includes different welfare and health-care systems, and different cultures and religions.
- A limitation is that some of the subthemes, or parts of their content, could have been categorized in the other main theme.
- The majority of the sample comprised women. A more heterogeneous sample might have revealed more nuanced findings of the role of next of kin.

INTRODUCTION

In 2018, 885 people with central nervous system cancer were registered in Norway.[1] Worldwide there were 296,851 people.[2] The diagnosis is very confronting, with 56% of patients experiencing one or more symptoms. Hemiparesis and cognitive challenges are most frequently reported but also headache, nausea and vomiting, vision challenges, epileptic seizures, and personality changes are considered common symptoms.[3] Changes in behavior and personality are considered particularly challenging, both for the patient and for the next of kin, as this may include apathy, loss of initiative and empathy, indifference, selfishness, physical and mental aggression, impaired emotional control and social abilities, and tendencies toward childish behavior, among others.[4] Studies show that the disease can be more challenging and stressful for the next of kin than for the patients. The next of kin have high rates of depression, anxiety, diverse physical pain, difficulty adapting, loneliness, and absence from work, as well as a reduced quality of life.[5-9] Studies also show that both patients and next of kin miss out on additional follow-up, support, and information from health-care providers, family, friends, and the community in their struggle to cope with everyday life.[10 11]

Despite the severe challenges this disease imposes on next of kin, there are only a few primary qualitative studies that have investigated the coping factors that make everyday life more manageable or which strategies next of kin use to cope with their new role and tasks. To our knowledge, this research has not been synthesized. Such information is of great importance, especially for health-care providers working with this group of caregivers. With improved understanding, they could expect to be better able to facilitate more manageable everyday life among the next of kin.[12] There is some quantitative research directed at these aspects, but we wanted studies that were more complementary and personal, hence the choice of qualitative studies. Therefore, the purpose of this metasynthesis was to identify and synthesize evidence from primary qualitative studies regarding the experience of next of kin with coping factors and strategies in their role as next of kin for a person with a brain tumor. The findings are discussed in the context of Lazarus and Folkman's stress theory[13] and the approach to coping with stress to interpret our findings in a theoretical context.

METHODS

Design

The study was a metasynthesis within the interpretative paradigm. It was inspired by a phenomenological–hermeneutic design because the aim was to identify and synthesize qualitative primary studies that explored next of kin attitudes and experiences.[14] The metasynthesis process consisted of five steps: (1) formulating the purpose and rationale of the study; (2) searching for and retrieving relevant qualitative research studies; (3)

critically appraising the included studies; (4) classifying the findings, and finally; (5) synthesizing the findings.

Search strategy

In collaboration with an experienced librarian, we conducted a systematic search within the PsycINFO, OVID, CHINAL, and Medline databases via EBSCO host up from January 2000 until March 3, 2021.

To search the PsycINFO database, we used the following terms: *((qualitative adj2 (research* or design* or stud* or method*)) or hermeneutic* or "grounded theory" or "meta synthes*" or metasynthesis* or metaethnograph* or interview* or phenomenolog* or thematic or themes or experience*).ti,ab,hw,id. or exp qualitative methods or phenomenology AND (caregiver* or famil* or next of kin* or relatives or spous* or wife or husband* or sibling* or sister* or brother* or dependent* or loved one* or parent* or mother* or father* or carer* or care giver*).ti,ab,hw,id. AND glioma*.ti,ab,hw,id. OR (brain adj2 (cancer or neoplasm* or tumor*)).ti,ab,hw,id.*

In Medline and CHINAL, we used the following terms: *caregiver* OR famil* OR "next of kin*" OR relatives OR spous* OR wife OR husband* OR sibling* OR brother* OR sister* OR dependent* OR "loved one*" OR parent* OR mother* OR father* OR carer* OR "care giver*" AND (MH "Qualitative Studies+") OR (MH "Qualitative Research+") OR (MH "Grounded Theory") OR Interview* OR experienc* OR phenomenolog* OR (qualitative W1 (research* OR method* OR design* OR stud*)) OR themes OR thematic OR "audio recording" OR audiorecording OR metasynthes* OR "meta synthes*" OR metaetnograph* AND (MH "Glioma+") OR glioma OR gliomas OR glioblastom* OR brain W1 (cancer OR tumor* or neoplasm*).*

The inclusion criteria were qualitative primary studies in English or a Nordic language that aimed to explore the factors or strategies used by the next of kin of persons with brain tumors, regardless of tumor type and stage that enhanced their role as next of kin. The next of kin had to be 18 years of age or older. The exclusion criteria were studies that did not clearly identify coping factors or strategies, factors or strategies that included the participants' experiences in the role of bereaved and not next of kin, and studies including diagnoses other than a brain tumor.

Search outcome

The search strategy generated 1 371 unique citations. Titles and abstracts were screened by the authors using Rayyan.[15] Sixty-six papers were read in full and evaluated against the inclusion criteria by both authors; 19 of these were included in the metasynthesis. Figure 1 shows the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flowchart with a full overview of the screening process. The search output is presented in the PRISMA flowchart. A final consensus regarding the eligible articles was obtained through a group discussion between the authors. The authors read the full text of the eligible articles and independently extracted data from the included studies; this process is illustrated in Figure 1. Consensus for data extraction was obtained as part of a group discussion between the authors. Table 1 lists the title, author(s), study country, year of publication, aim, analysis, and study participants of all included studies. Most studies were from Europe: Sweden (3), Great Britain (3), Denmark (1), Belgium (1), and Turkey (1); seven were from Canada (3) and the USA (4), two were from Australia, and one was from Taiwan. The tumor type and stage varied. For details, see Table 1.

Figure 1 about here

Table 1 about here

Quality appraisal

The quality of the 19 papers was evaluated using the Critical Appraisal Skills Program (CASP) for qualitative studies. The first evaluation was conducted blinded and independently by AW and GR, whose CASP evaluations were then compared. Using the criteria in CASP for independent assessment, the authors mutually agreed on a final quality evaluation. For details, see Table 2.

The included studies appraised according to CASP are listed in Table 2. All studies had clearly stated the study aim and the qualitative methodologies were considered appropriate. Furthermore, several of the studies had been published in highly ranked journals. The most poorly addressed issue was the influence of the researcher on the research and vice versa.

Table 2 about here

Data abstraction and analyses

As suggested by Sandelowski and Barroso,[14] two approaches to qualitative synthesis were used. The first of these involved qualitative metasummaries of qualitative findings from the primary studies. This method is defined as qualitative, but the findings are presented quantitatively. The second involved a metasynthesis that developed new interpretations of the target findings from the primary studies.[14] The narrative analysis was inspired by Lindseth and Nordberg's phenomenological–hermeneutic methods.[16] Three steps were followed. First, the empirical materials were read several times. Second, after extraction, the target findings were imported into NVivo 11 data management software for further analysis.[17] The text was read line-by-line to identify meaning units, subthemes, and themes. Third, the researchers aimed to achieve a comprehensive understanding of the empirical materials, meaning units, and themes, and to relate these to the aim and research question of the metasynthesis.[16] The analytic themes were identified by AW and discussed with GR. The process of deriving the themes was inductive. The contribution of targeted findings from each of the included papers is outlined, and quotations are used to illustrate and support the findings, which increases the trustworthiness of the study. To validate the findings, both authors participated in discussions of the empirical analysis and in writing up the findings. Ethical approval was not required for the study.

Table 3 about here

Synthesis

Qualitative metasynthesis provides novel interpretations of the target findings from primary studies.[14] The two main themes of this metasynthesis were coping factors and coping strategies. The theme “coping factors” consisted of the subthemes, personal characteristics, meaningful external support and having someone to talk to, and hope and religion. The theme “coping strategies” consisted of the subthemes regain control, fight against, and acceptance. For a list of the studies that generated findings for the main themes and subthemes, see Table 3.

PATIENT AND PUBLIC INVOLVEMENT

This systematic review is based on published primary studies and do not involve public involvement.

RESULTS

The findings are presented as metasummaries supported by tables and figures, and as a metasynthesis presented under two themes.

Metasummaries

The 19 included studies consisted of 332 participants (200 women, 78 men, and 54 not classified). The focus was on the following themes: the needs of the next of kin;[3 18-22] their overall experiences as next of kin;[8 23 24] coping and coping mechanisms;[25-27] postoperative caregiving;[28 29] being a next of kin in the palliative phase;[30 31] support factors experienced;[32] how the caregiving changed over time;[33] and factors influencing treatment choice in the palliative phase.[34] Three of the studies were undertaken six months after diagnosis,[27 28 33] and three in the patients' palliative phase or postmortem.[30 31 34] In five studies, all the patients were the children of the informants.[8 26 27 30 31]

Metasynthesis

Main theme 1: Coping factors within the next of kin and as external support

Personal characteristics such as a strong and positive personality were important coping factors for next of kin in new challenging situations.[23 26 34] Showing empathy for the patient and also having health professionals available were important because the next of kin situation could easily engender feelings such as discouragement and reproach.[23] A positive mood and humor were also emphasized.[26]

The role as next of kin was considered to be complete and important.[21 23 25 28]

> *"But caring for him is something I will do—it is not a burden."*[28] (p. 81)

Engagement and commitment in the care of their relatives were highlighted by many next of kin, especially when the patients appreciated the help.[21] The engagement was even stronger when the emotional bond between patient and next of kin was strong.[18 19 26 32] However, other studies revealed less engagement and commitment, and underlined anger and reluctance with the new role and the heavy responsibilities and sacrifices of the next of kin that impacted their own needs and wishes.[19 20 23 28 30]

External support made the role of next of kin easier to cope with. The support was given by family, friends, neighbors, colleagues and workplaces, health personnel, schools, the religious community, people in the local community, and even strangers.[3 8 18-32] The support from health-care professionals was especially important. This support included emotional support and assistance during patient care and treatment.[3 8 18-24 26-32] The importance of assistance such as medical supervision and nursing care was emphasized,[8 20 26] with next of kin noting that this made it possible to feel like a partner again,[21] while concurrently allowing anticipated time alone.[22] A well-known health-care professional was crucial in making this possible, because it implied that the patient received the best care as they were known to the health-care professional, and also because the assistance was considered to be less intrusive.[21 22] To experience the assistance with care as a coping factor, it was crucial that care be compassionate and of the best quality. These qualities emphasized the health professional's genuine care and gave the next of kin hope and desire to fight the disease.[8 19 21 24 26]

> *"She (neurosurgeon) had to give us some bad news some of the time… and you couldn't ask for a better manner in her delivery of that bad news, or her support in what we were going through."*[32] (p. 8)

When next of kin experienced that their loved ones received a low quality of care or suffered malpractice, it implied mistrust of the health-care system and weakened the experience of health-care professionals as a support factor.[8 18 21 22] Emotional support from health-care professionals implied an acknowledgment that the disease affected not only the patients, but also their next of kin. It also implied that the health-care professionals recognized and met the wishes of the next of kin for active participation in monitoring the patient's disease course.[21 23 24 31] Next of kin who did not have such involvement felt ignored, useless, and helpless.[23 26]

Support from family and friends was invaluable in the care tasks and in coping with the role of the next of kin.

> *"Just support from family and friends, that was important to me, and just knowing that I could call on them…"*[20] (p. 1098)

Social, practical, and emotional support was emphasized, and included such things as economic help, childcare, transport, and housekeeping.[8 20 22 23 26-29 31 32] Some next of kin would have appreciated even more support and help from family and friends, preferably given on their own initiative.[18 20 22 23 32 33]

Having someone to confide in and talk to were also important in coping with the role as next of kin. Supportive conversations with health-care professionals were highly appreciated by many next of kin. However, this required the health-care professional's understanding and empathy for the situation of the patient as well as of their next of kin, and preferably that they should be available at all times.[19 21 24 27 28 34] Discussions with family and friends were also important,[19 22 23] and could even produce a stronger bond.[23] Such a bond required families and friends to understand and recognize the challenges faced by the next of kin.[22] Support groups and conversations with other next of kin were also highlighted. These conversations could be face-to-face or via the Internet.[3 20 22 27 31-33]

> *"From time to time, I need to be able to talk to someone. Because when I lay down in the evening, then it starts to work in the inside."*[21] (p. 411)

On the other hand, support groups were also considered demanding because it was difficult to listen to other families' stories. Furthermore, for some it was considered a waste of time to spend valuable hours with people other than their closest family members.[8 20 28]

Hope and religion were emphasized as important coping factors. The next of kin hoped that a miraculous treatment would be developed so that their loved ones could survive the disease or just have a better quality of life.[3 8 18-21 24 30 31]

> *You see a positive evolution, and everything that goes better is good for her. (...) Nobody can forbid us to have hope. And miracles happen. Whether we believe it or not, that's not the point, it is the only thing to focus on."*[21] (p. 409)

Hope gave a reason to fight, although it weakened in the palliative phase.[19 24 31] Faith strengthened the hope of healing during the treatment period and gave some form of peace in the final palliative phase. In most cases, hope was related to faith.[23 24 27 31 34]

Main theme 2: Coping strategies – control and proactivity

Regaining control of the situation was a frequent coping strategy and for most, this included being provided with enough information to allow an overview of what to expect, which implied some form of security.[8 18-21 27 32 34]

> *"So it's a, it's a roller coaster of emotion but for the most part I've been, 'What do we need to do? Where do we need to be?' And then just read, read, read whatever I can find out, whatever information because I feel like whatever I know, I can ask for."*[27] (p. 34)

The information provided should be adapted to the situation and the disease trajectory, and preferably given by health-care professionals.[18 20 21 23 26 34] The next of kin often hid this information from the patients to protect them and not diminish their hope.[8 24 27 28 31]

To regain control meant not only control of the diagnosis, but also personal control and control over their own reactions. In some cases, the next of kin denied their feelings. Some even denied the entire diagnosis,[18 23 26 27] and instead focused on being strong for the patient and the entire family.[21 23 27 29-31 33] One next of kin in Edvardsson and Ahlstroms' (2008) study[23] reported:

> *"I've sort of stowed it all away, I suppose. It is as if I'd experienced it from the outside or seen it on TV. It's often that way with sorrowful things."* (p. 588)

Being proactive and fighting the disease were also important coping strategies because they were better than not doing anything.[8 19 23 24 31]

> *"People ask you how you cope. But what if you were to give up? You've got to cope—and we do have each other! (...)."[23] (p. 588)*

This implied adopting a healthier lifestyle, including changing diet and exercise habits, hoping that this would improve the effects of medical treatment,[19 24] or trying alternative treatments.[8 31] However, an increasing feeling of powerlessness was emphasized if the fight, in the form of these actions and treatments, did not meet the hope of a cure.[19 21 24 31]

As the disease progressed and life went on, most next of kin accepted the diagnosis, prognosis, and a new pattern to everyday life.[8 24 25 27 31] There was a striving for normality, starting with recommencing hobbies, work, and school for children.[8 24 25 27 28 30 31 33] This was particularly important within families with children. At the same time, accepting disease progression or a bad diagnosis was most challenging when the patient was a child.[31]

DISCUSSION

This metasynthesis aimed to explore those factors and strategies that enhanced the ability of next of kin to cope with their experience as the next of kin of a person with a brain tumor. Valuable coping factors included personal characteristics, finding meaning in the situation, external support, hope and religion, and having someone to talk to. Strategies to manage the situation involved regaining control, being proactive, and acceptance.

We used Lazarus and Folkman's transactional stress theory published in 1984 in the discussion of our findings. Lazarus and Folkman define coping as a cognitive and behavioral endeavor under constant change, dealing with external and/or internal demands that a cognitive assessment indicates are stressful or that exceed personal resources.[13 35] Being the next of kin to a person with a brain tumor is considered to be a negative stressor because of the challenging life situation and care tasks. Nevertheless, several next of kin included in the metasynthesis expressed a desire to fight the disease and to gain control over the situation. This is described by Lazarus and Folkman[13] as a secondary assessment of the situation, in which the next of kin decide which measures to implement. One such measure could be to gain personal control—one of the most important and stress-reducing personal strategies available.[13]

A possible explanation for the proactive attitude of next of kin toward the disease may be their obligation and commitment to the patient. Commitment is an expression of something of great importance and can cause one to be willing to meet threats and challenges that he or she would otherwise avoid.[13] However, our findings revealed that the experience of contributing to something meaningful, not the obligation to do so, promoted coping in the situation. We consider that this is caused by the fact that obligation does not automatically make an action meaningful, but rather that it can be experienced as a compulsion. This assumption is strengthened by the findings that the tasks as next of kin could arouse emotions such as anger and aversion to the patient and to the diagnosis, rather than coping. Several studies refer to the same ambivalent experience regarding commitment and attitudes toward being a next of kin.[36 37]

External support was the factor that most relatives emphasized as promoting coping. It was described as invaluable, which was also confirmed in other studies,[38 39] and in Lazarus and Folkman's transactional stress theory.[13] At the same time, in both this metasynthesis and in other studies, next of kin voiced a strong desire and longing for even greater external support.[38 39] The findings of the metasynthesis also showed that the configuration and arrangement of the support, especially that given by health-care providers is of great importance. This may indicate that health-care providers do not offer support appropriate to individual needs of the next of kin and for the care situations, possibly as a consequence of a lack of knowledge among health-care providers about how this affects the experience of being next of kin.

The findings of this metasynthesis show that several next of kin considered hope to be an important coping factor, especially during the disease trajectory. Hope has also been shown to be an extensional coping factor in several studies,[40 41] and transactional stress theory states that faith and hope are two of the most important personal factors in the cognitive assessment of stressors.[13 35] Furthermore, according to Lazarus and Folkman,[13] the two factors are strongly related, which is consistent with the findings of our metasynthesis. For several next of kin, hope was strongly grounded in religion. This was especially prominent in the studies conducted in the palliative phase, which indicated that faith is strengthened when there is no hope of curative treatment. The same pattern has also been reported in other studies describing cancer patients' experiences of palliative care.[42 43]

As the disease progressed, several next of kin accepted the diagnosis and its burden. Their fight against the disease diminished to some extent, and the relatives instead tried to "normalize" everyday life as much as possible. Similar acceptance is also reported by next of kin of other cancer patients, especially in the palliative phase.[44 45] Lazarus and Folkman describe this as a reassessment, referring to a changed cognitive assessment of the stressor based on new information from the environment and/or the person.[13]

Strengths and limitations

A strength of this metasynthesis is that the primary search in the databases was conducted with the assistance of an experienced librarian, in an attempt to ensure that as many as possible of the relevant studies were included.[46] Furthermore, most of the included studies were of high methodological quality (see Table 2). Our sample was also highly multicultural (see Table 1). This attribute strengthens the validity of the metasynthesis since geographical origin could have affected the study sample because of different participant backgrounds related to different welfare and health-care systems, cultures, and/or religions.

A limitation of our metasynthesis is that one of the 66 articles intended to be read in full text could not be obtained.[47] The formation of the subthemes is also a possible limitation. Some of the subthemes, or parts of their content, could have been categorized in the other main theme. Both main themes and subthemes overlap in several cases, and we have read similar studies[24 27] where the findings are categorized differently than in metasynthesis. We chose to be true to the informants' statements and designated the location based on the informants' way of speaking and description of the experience. Another possible limitation is that our sample consisted mainly of women (see Table 1). A more heterogeneous sample might have revealed more nuanced findings and different experiences of the role of the next of kin.

CONCLUSION

The findings of this metasynthesis show that next of kin experience and use a range of coping factors and strategies in their role. Their experience is marked by individual differences. It is of great importance that health-care providers offer assistance that is individually adapted for these coping factors and strategies because this can reduce stress among the next of kin. The coping experience seems to go through phases, and further information is needed to understand fully how and when the various factors and strategies are used as the disease progresses. Longitudinal studies would therefore be of particular interest in this field.

Supplementary information

The manuscript has been edited by OnLine English (https://www.oleng.com.au) to comply with international publishing guidelines.

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Authors' contributions

AWL and GR designed the research project and developed the research plan. Librarian Ellen Sejersted at the University of Agder and AWL were responsible for the literature search, while AWL and GR were responsible for the analysis. Both authors were involved in the screening and inclusion of the studies, reviewed the manuscript, and contributed to the revision of the paper. Both authors read and approved the final version of the paper.

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Competing interests

The authors declare that they have no competing interests.

Patient consent for publication

Not required

Ethics approval

Ethical approval was not required, as no primary data were collected as part of this study.

Data availability

Data are available on reasonable request.

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Table 1: Characteristics of the included studies.

Author/year/country	Focus	Type of brain tumor and stage of treatment at interview	Recruitment	Participants, sex, and relationship	Method/design	Data collection/analysis
Arber et al. (2010).[3] United Kingdom (UK).	Caregivers' need for information.	Malignant. Stage of treatment not described.	Specialist hospital in England.	N = 22 M: 7 and F: 15 17 spouses 3 children 2 parents	Grounded theory.	Semistructured interview/comparative method for generating categories and topics.
Arber et al. (2013).[22] United Kingdom (UK).	Caregivers' need for support.	Malignant. Stage of treatment not described.	Recruited by a nurse at a cancer center in England.	N = 22 M: 7 and F: 15 17 spouses 3 children 2 parents	Grounded theory.	Semistructured interview/comparative method for generating categories and topics.
Coolbrandt et al. (2015).[21] Belgium.	Caregivers' experience and need for support.	High-grade. Radiation or chemotherapy, or in the follow-up phase after such treatment.	University Hospital in Leuven.	N = 16 M: 6 and F: 10 13 partners 2 parents 1 friend	Grounded theory.	Semistructured interview/thematic analysis inspired by the Qualitative Analysis Guide of Leuven.
Cutillo et al. (2018).[27] USA.	Which strategies caregivers of children with a brain tumor use in the postoperative phase.	15 benign. 25 malignant. Newly diagnosed and newly operated.	Pediatric hospital in the USA.	N = 22 M: 3 and F: 19 All parents	Triangulating mixed-method.	Semistructured interview/thematic analysis.

Edvardson & Ahlström (2008)[23]. Sweden.	Caregivers' experience.	25 low-grade. 2 high-grade. Stage of treatment not described.	The patients had participated in an earlier study.	N = 28 M: 8 and F: 20 15 partners, living together 3 partners, living apart 8 parents 1 sibling 1 child	Not described.	Semistructured interview/qualitative content analysis and quantitative analysis of how the topics were distributed among the participants.
Janda et al. (2006)[20]. Australia.	The need of support for brain tumor patients and their caregivers.	Different types. Treatment phase not described, but time since diagnosis stated: 1–2 years: 22 5 years: 5 More than 5 years: 11	Members of Queensland Cancer Fund's Brain Tumor Support Service.	N = 10 in focus group, n = 8 in semistructured interview M: 4 and F: 18 13 partners 5 children	Qualitative.	Focus group interview and semistructured interview/framework analysis.
Lipsman et al. (2007)[34]. Canada.	The experience of brain tumor patients and their caregivers, and how it affects the choice of treatment.	Malignant. Palliative phase.	Recruited by a neurosurgeon.	N = 22 Further participant information not described	Qualitative.	Semistructured interview/thematic analysis.

Lou et al. (2015).[31] Taiwan.	The experience and suffering of mothers waiting for their child to die from brain tumor.	Malignant. All patients deceased.	Not described.	N =10 F: 10 All mothers	Phenomenological.	In-depth interview/Colaizzi's analysis method.
Ownsworth et al. (2015).[32] Australia.	Caregivers' experience of support.	6 low-grade. 5 high-grade. All underwent surgery and radiation or chemotherapy. 9 months – 22 years since diagnosis.	Had participated in a different study.	N = 11 M: 6 and F: 5 8 spouses 3 parents	Phenomenological.	Semistructured interview/thematic analysis.
Piil et al. (2015).[19] Denmark.	Brain tumor patients' and their caregivers' experience, and their need for rehabilitation and support.	High-grade. The interviews conducted after: 1. Surgical diagnosis 2. Oncological treatment 3,4. Oncological treatment and scan showing treatment effect 5. After treatment	The University Hospital in Copenhagen.	N = 33 M: 10 and F: 23 23 spouses 2 girl/boyfriends 7 children 1 sister	Longitudinal and exploratory.	Semistructured interview/thematic analysis.

Russell et al. (2016)[8]. Canada.	The experience of children with a brain tumor and their caregivers.	Malignant. Diagnosed at least 3 months previously, stage of treatment not described.	Hospital in Toronto.	N = 12 Based on names: F: 11 stk., 1 stk. unknown All parents	Grounded theory.	Semistructured interview/comparative analysis.
Schmer et al. (2008)[28]. USA.	Caregivers' experience concerning care tasks after chemotherapy.	Malignant. During first 6 months of treatment.	The patients' treatment center.	N = 10 Sex unknown 7 spouses 2 daughters 1 son-in-law	Phenomenological.	Semistructured interview/Colaizzi's analysis method.
Schubart et al. (2008).[18] USA.	Caregivers' challenges and unmet needs.	Different types of brain cancer. 6 deceased 2 exacerbations 2 unstable 10 stable 1 terminal 3 recurrent 1 unclear	NeuroOncology Center.	N = 25 M: 7 and F: 18 18 spouses 4 parents 2 children 1 sibling	Grounded theory.	Semistructured interview/open coding and cross-case analysis.
Sherwood et al. (2011).[33] USA.	How caregivers adapt to their new role, and how this role changes during time.	Malignant. Interviewed 1 and 4 months after diagnosis.	A regional hospital.	N = 10 M: 2 and F: 8 5 spouses 2 parents	Longitudinal descriptive design.	Semistructured interview/thematic content analysis.

				1 child 1 nephew 1 friend		
Shortman et al. (2013). United Kingdom (UK).	Mothers of children with brain tumor—their experience and their coping mechanisms.	Different types and degrees. All underwent surgery, five radiation, and four chemotherapy. 17–35 months since diagnosis.	Also participated in another study.	N = 6 F: 6 All mothers.	Not described.	Semistructured interview/thematic content analysis.
Strang & Strang (2001).[25] Sweden.	The degree to which patients with a brain tumor and their caregivers cope, understand, and create meaning in the situation.	Malignant tumors, grade 2–4. Treatment stage not described.	Not described.	N = 16 Further participant information not described.	Hermeneutic phenomenological.	Semistructured interview/structural analysis based on hermeneutic circle described by Richoeur.
Tastan et al. (2011).[29] Turkey.	Caregivers' experience of postoperative phase and homecare.	Different types and degrees. All patients had undergone surgery and postoperative treatment and were being treated at home.	A research and military training hospital in Turkey.	N = 19 M: 4 and F: 6 4 spouses 4 children 1 parent 1 sibling	Descriptive qualitative study.	Semistructured interview/Colaizzi's analysis method.

Wideheim et al. (2002).[24] Sweden.	The experience of a brain tumor from a family perspective.	High-grade glioma. The interviews were conducted 2–3 weeks, 3 months, and 6 months postoperatively.	Not described.	N = 5 Sex unknown 2 spouses 2 parents 1 adult child	Descriptive qualitative study.	Qualitative interviews/inductive content analysis.
Zelcer et al. (2010).[30] Canada.	The experience of brain tumor patients and caregivers in the palliative phase.	Malignant. All patients deceased.	Children's Hospital, London Health Sciences Centre.	N = 25 M: 9 and F: 16 All parents	Qualitative	Semistructured interview/thematic content analysis.

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[1] M = Male, F = Female

Table 2: Critical appraisal of the included studies.

Criterion Y = yes N = no C = can't tell V = valuable NV = not valuable	1. Was there a clear statement of the aims?	2. Is a qualitative methodology appropriate?	3. Was the research design appropriate?	4. Was the recruitment strategy appropriate?	5. Were the data collected in a way that addressed the research issue?	6. Has the relationship between researcher and participants been adequately considered?	7. Have ethical issues been taken into consideration?	8. Was the data analysis sufficiently rigorous?	9. Is there a clear statement of findings?	10. How valuable is the research?	Impact factor
Arber et al. (2010)[3]	Y	Y	C	C	Y	N	Y	C	Y	V	Not found
Arber et al. (2013)[22]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	1.697
Coolbrant et al. (2015)[21]	Y	Y	Y	Y	Y	C	Y	Y	Y	V	2.022
Cutillo et al. (2018)[27]	Y	Y	Y	Y	Y	Y	N	Y	C	V	2.170
Edvardsson & Ahlström (2008)[23]	Y	Y	Y	Y	Y	N	N	Y	Y	V	3.470
Janda et al. (2006)[20]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	2.754
Lipsman et al. (2007)[34]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	2.922
Lou et al. (2015)[31]	C	Y	Y	C	Y	N	N	C	Y	V	2.022
Ownsworth et al. (2015)[32]	Y	Y	Y	Y	Y	C	C	Y	Y	V	4.137
Piil et al. (2015)[19]	Y	Y	Y	Y	Y	C	Y	Y	Y	V	1.096
Russel et al. (2016)[8]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	1.197

Schmer et al. (2008)[28]	Y	Y	N	Y	N	N	Y	N	Y	V	1.096
Schubart et al. (2008)[18]	C	Y	Y	Y	Y	N	N	Y	Y	V	3.470
Sherwood et al. (2011)[33]	Y	Y	Y	Y	C	Y	N	Y	N	V	1.438
Shortman et al. (2013)	Y	Y	Y	C	C	N	Y	N	Y	V	1.918
Strang & Strang (2001)[25]	C	C	Y	N	Y	N	Y	Y	Y	V	4.956
Tastan et al. (2011)[29]	Y	Y	N	Y	Y	N	Y	N	Y	V	1.096
Wideheim et al. (2002)[24]	Y	Y	Y	Y	Y	N	C	Y	Y	V	2.022
Zelcer et al. (2010)[30]	Y	Y	Y	Y	Y	N	C	Y	Y	V	5.731

Table 3: Thematic overview showing the studies' contribution to the different themes and subthemes.

	Coping factors					Coping mechanisms		
Author	Personal characteristics	Meaningful	External support	Hope and religion	Interlocutor	Gain control	Fight	Accept
Arber et al. (2010)[3]			V	V	V			
Arber et al. (2013)[22]			V		V			
Coolbrandt et al. (2015)[21]		V	V	V	V	V		
Cutillo et al. (2018)[27]			V	V	V	V		V
Edvardson & Ahlström (2008)[23]	V	V	V	V	V	V	V	
Janda et al. (2006)[20]			V	V	V	V		
Lipsman et al. (2007)[34]	V			V	V	V		
Lou et al. (2015)[31]			V	V	V	V	V	V
Ownsworth et al. (2015)[32]		V	V		V	V		
Piil et al. (2015)[19]		V	V	V	V	V	V	
Russell et al. (2016)[8]			V	V		V	V	V
Schmer et al. (2008)[28]		V	V		V			V
Schubart et al. (2008)[18]		V	V	V		V		
Sherwood et al. (2011)[33]					V	V		V
Shortman et al. (2013)[33]	V	V	V			V		
Strang & Strang (2001)[25]		V	V					V

Tastan et al. (2011)[29]			V			V		
Wideheim et al. (2002)[24]			V	V	V		V	V
Zelcer et al. (2010)[30]			V	V		V		V

PRISMA 2009 Flow Diagram

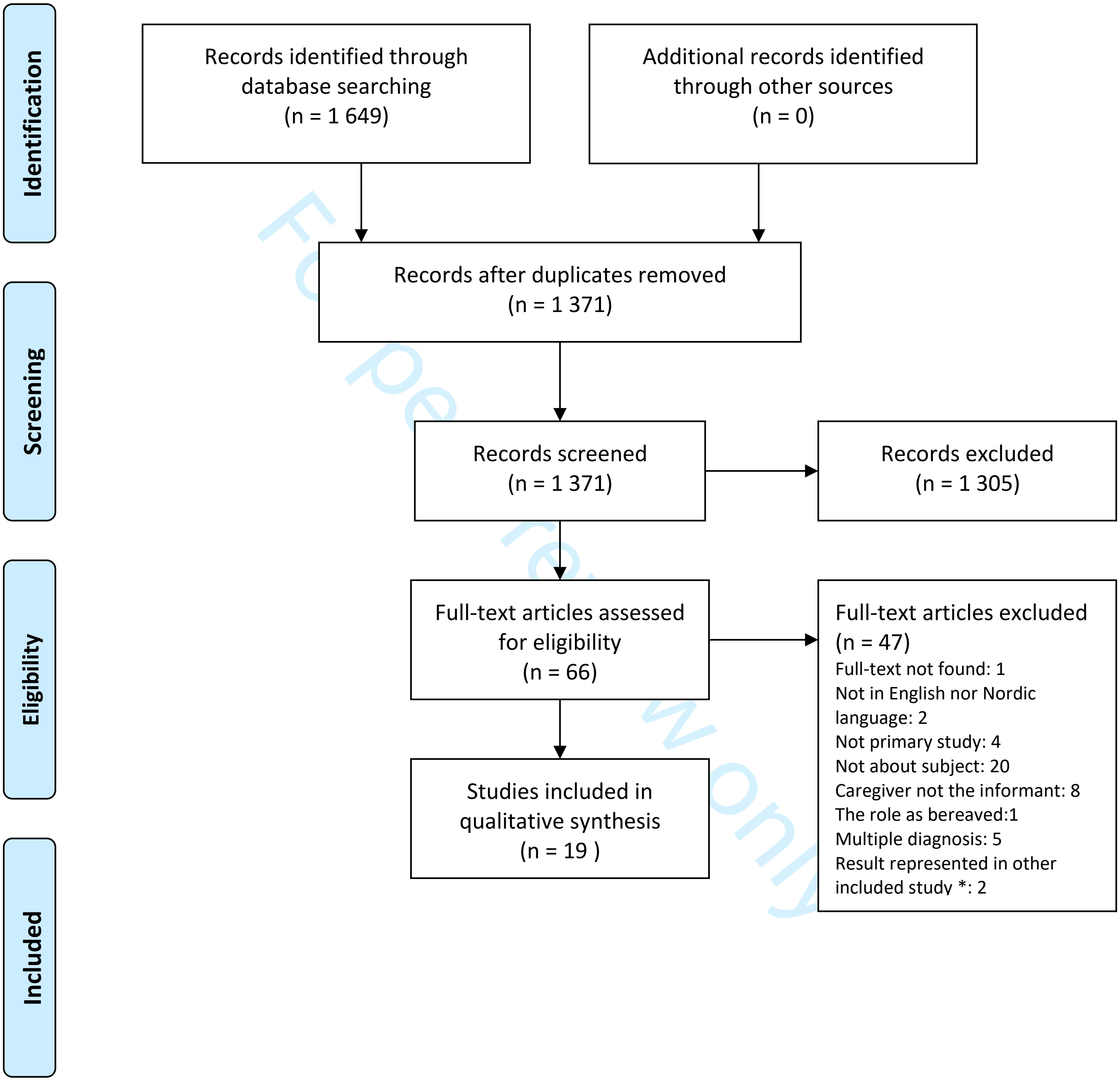

Figure 1. Flow chart of the inclusion process

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). *P*referred *R*eporting *I*tems for *S*ystematic Reviews and *M*eta-*A*nalyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097

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PRISMA NMA Checklist of Items to Include When Reporting A Systematic Review Involving a Network Meta-analysis

Section/Topic	Item #	Checklist Item	Reported on Page #
TITLE			
Title	1	Identify the report as a systematic review *incorporating a network meta-analysis (or related form of meta-analysis).*	***1***
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: **Background:** main objectives **Methods:** data sources; study eligibility criteria, participants, and interventions; study appraisal; and *synthesis methods, such as network meta-analysis.* **Results:** number of studies and participants identified; summary estimates with corresponding confidence/credible intervals; *treatment rankings may also be discussed. Authors may choose to summarize pairwise comparisons against a chosen treatment included in their analyses for brevity.* **Discussion/Conclusions:** limitations; conclusions and implications of findings. **Other:** primary source of funding; systematic review registration number with registry name.	1-2
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known*, including mention of why a network meta-analysis has been conducted.*	***3-4***
Objectives	4	Provide an explicit statement of questions being addressed, with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	4
METHODS			
Protocol and registration	5	Indicate whether a review protocol exists and if and where it can be accessed (e.g., Web address); and, if available, provide registration information, including registration number.	n.a
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale. *Clearly describe eligible treatments included in the treatment network, and note whether any have been clustered or merged into the same node (with justification).*	***6***
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	5-6
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	5-6
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable,	6-7

		included in the meta-analysis).	
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	7-8
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	5-6
Geometry of the network	**S1**	Describe methods used to explore the geometry of the treatment network under study and potential biases related to it. This should include how the evidence base has been graphically summarized for presentation, and what characteristics were compiled and used to describe the evidence base to readers.	***6***
Risk of bias within individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	7
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means). *Also describe the use of additional summary measures assessed, such as treatment rankings and surface under the cumulative ranking curve (SUCRA) values, as well as modified approaches used to present summary findings from meta-analyses.*	n.a
Planned methods of analysis	14	Describe the methods of handling data and combining results of studies for each network meta-analysis. This should include, but not be limited to: • *Handling of multi-arm trials;* • *Selection of variance structure;* • *Selection of prior distributions in Bayesian analyses; and* • *Assessment of model fit.*	7-8
Assessment of Inconsistency	**S2**	Describe the statistical methods used to evaluate the agreement of direct and indirect evidence in the treatment network(s) studied. Describe efforts taken to address its presence when found.	n.a
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	**n.a**
Additional analyses	16	Describe methods of additional analyses if done, indicating which were pre-specified. This may include, but not be limited to, the following: • Sensitivity or subgroup analyses; • Meta-regression analyses; • *Alternative formulations of the treatment network; and* • *Use of alternative prior distributions for Bayesian analyses (if applicable).*	***n.a***

RESULTS†			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	6-7, 9-10
Presentation of network structure	**S3**	Provide a network graph of the included studies to enable visualization of the geometry of the treatment network.	***6-7 and Figure 1***
Summary of network geometry	**S4**	Provide a brief overview of characteristics of the treatment network. This may include commentary on the abundance of trials and randomized patients for the different interventions and pairwise comparisons in the network, gaps of evidence in the treatment network, and potential biases reflected by the network structure.	***n.a***
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	9
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment.	n.a
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: 1) simple summary data for each intervention group, and 2) effect estimates and confidence intervals. *Modified approaches may be needed to deal with information from larger networks.*	***n.a***
Synthesis of results	21	Present results of each meta-analysis done, including confidence/credible intervals. *In larger networks, authors may focus on comparisons versus a particular comparator (e.g. placebo or standard care), with full findings presented in an appendix. League tables and forest plots may be considered to summarize pairwise comparisons.* If additional summary measures were explored (such as treatment rankings), these should also be presented.	
Exploration for inconsistency	**S5**	Describe results from investigations of inconsistency. This may include such information as measures of model fit to compare consistency and inconsistency models, *P* values from statistical tests, or summary of inconsistency estimates from different parts of the treatment network.	***n.a***
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies for the evidence base being studied.	n.a
Results of additional analyses	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression analyses, *alternative network geometries studied, alternative choice of prior distributions for Bayesian analyses,* and so forth).	***n.a***
DISCUSSION			
Summary of evidence	24	Summarize the main findings, including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy-makers).	17
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review level (e.g., incomplete retrieval of identified research, reporting bias). *Comment on the validity of the assumptions, such as transitivity and consistency. Comment*	16-17

		on any concerns regarding network geometry (e.g., avoidance of certain comparisons).	
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	17
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review. This should also include information regarding whether funding has been received from manufacturers of treatments in the network and/or whether some of the authors are content experts with professional conflicts of interest that could affect use of treatments in the network.	***n.a***

PICOS = population, intervention, comparators, outcomes, study design.
* Text in italics indicateS wording specific to reporting of network meta-analyses that has been added to guidance from the PRISMA statement.
† Authors may wish to plan for use of appendices to present all relevant information in full detail for items in this section.

Box. Terminology: Reviews With Networks of Multiple Treatments

Different terms have been used to identify systematic reviews that incorporate a network of multiple treatment comparisons. A brief overview of common terms follows.

Indirect treatment comparison: Comparison of 2 interventions for which studies against a common comparator, such as placebo or a standard treatment, are available (i.e., indirect information). The direct treatment effects of each intervention against the common comparator (i.e., treatment effects from a comparison of interventions made within a study) may be used to estimate an indirect treatment comparison between the 2 interventions (**Appendix Figure 1, A**). An indirect treatment comparison (ITC) may also involve multiple links. For example, in **Appendix Figure 1, B**, treatments B and D may be compared indirectly on the basis of studies encompassing comparisons of B versus C, A versus C, and A versus D.

Network meta-analysis or *mixed treatment comparison*: These terms, which are often used interchangeably, refer to situations involving the simultaneous comparison of 3 or more interventions. Any network of treatments consisting of strictly unclosed loops can be thought of as a series of ITCs (**Appendix Figure 1, A and B**). In mixed treatment comparisons, both direct and indirect information is available to inform the effect size estimates for at least some of the comparisons; visually, this is shown by closed loops in a network graph (**Appendix Figure 1, C**). Closed loops are not required to be present for every comparison under study. "Network meta-analysis" is an inclusive term that incorporates the scenarios of both indirect and mixed treatment comparisons.

Network geometry evaluation: The description of characteristics of the network of interventions, which may include use of numerical summary statistics. This does not involve quantitative synthesis to compare treatments. This evaluation describes the current evidence available for the competing interventions to identify gaps and potential bias. Network geometry is described further in **Appendix Box 4**.

Appendix Box 1. The Assumption of Transitivity for Network Meta-Analysis

Methods for indirect treatment comparisons and network meta-analysis enable learning about the relative treatment effects of, for example, treatments A and B through use of studies where these interventions are compared against a common therapy, C.

When planning a network meta-analysis, it is important to assess patient and study characteristics across the studies that compare pairs of treatments. These characteristics are commonly referred to as *effect modifiers* and include traits such as average patient age, gender distribution, disease severity, and a wide range of other plausible features.

For network meta-analysis to produce valid results, it is important that the distribution of effect modifiers is similar, for example, across studies of A versus B and A versus C. This balance increases the plausibility of reliable findings from an indirect comparison of B versus C through the common comparator A. When this balance is present, the assumption of transitivity can be judged to hold.

Authors of network meta-analyses should present systematic (and even tabulated) information regarding patient and study characteristics whenever available. This information helps readers to empirically evaluate the validity of the assumption of transitivity by reviewing the distribution of potential effect modifiers across trials.

Appendix Box 2. Differences in Approach to Fitting Network Meta-Analyses

Network meta-analysis can be performed within either a frequentist or a Bayesian framework. Frequentist and Bayesian approaches to statistics differ in their definitions of probability. Thus far, the majority of published network meta-analyses have used a Bayesian approach.

Bayesian analyses return the posterior probability distribution of all the model parameters given the data and prior beliefs (e.g., from external information) about the values of the parameters. They fully encapsulate the uncertainty in the parameter of interest and thus can make direct probability statements about these parameters (e.g., the probability that one intervention is superior to another).

Frequentist analyses calculate the probability that the observed data would have occurred under their sampling distribution for hypothesized values of the parameters. This approach to parameter estimation is more indirect than the Bayesian approach.

Bayesian methods have been criticized for their perceived complexity and the potential for subjectivity to be introduced by choice of a prior distribution that may affect study findings. Others argue that explicit use of a prior distribution makes transparent how individuals can interpret the same data differently. Despite these challenges, Bayesian methods offer considerable flexibility for statistical modeling.
In-depth introductions to Bayesian methods and discussion of these and other issues can be found elsewhere.

Appendix Box 3. Network Meta-Analysis and Assessment of Consistency

Network meta-analysis often involves the combination of direct and indirect evidence. In the simplest case, we wish to compare treatments A and B and have 2 sources of information: direct evidence via studies comparing A versus B, and indirect evidence via groups of studies comparing A and B with a common intervention, C. Together, this evidence forms a closed loop, ABC.

Direct and indirect evidence for a comparison of interventions should be combined only when their findings are similar in magnitude and interpretation. For example, for a comparison of mortality rates between A and B, an odds ratio determined from studies of A versus B should be similar to the odds ratio comparing A versus B estimated indirectly based on studies of A versus C and B versus C. This assumption of comparability of direct and indirect evidence is referred to as *consistency* of treatment effects.

When a treatment network contains a closed loop of interventions, it is possible to examine statistically whether there is agreement between the direct and indirect estimates of intervention effect.

Different methods to evaluate potential differences in relative treatment effects estimated by direct and indirect comparisons are grouped as *local approaches* and *global approaches.* Local approaches (e.g., the Bucher method or the node-splitting method) assess the presence of inconsistency for a particular pairwise comparison in the network, whereas global approaches (e.g., inconsistency models, I^2 measure for inconsistency) consider the potential for inconsistency in the network as a whole.

Tests for inconsistency can have limited power to detect a true difference between direct and indirect evidence. When multiple loops are being tested for inconsistency, one or a few may show inconsistency simply by chance. Further discussions of consistency and related concepts are available elsewhere.

Inconsistency in a treatment network can indicate lack of transitivity (see **Appendix Box 1**).

Appendix Box 4. Network Geometry and Considerations for Bias

The term *network geometry* is used to refer to the architecture of the treatment comparisons that have been made for the condition under study. This includes what treatments are involved in the comparisons in a network, in what abundance they are present, the respective numbers of patients randomly assigned to each treatment, and whether particular treatments and comparisons may have been preferred or avoided.

Networks may take on different shapes. Poorly connected networks depend extensively on indirect comparisons. Meta-analyses of such networks may be less reliable than those from networks where most treatments have been compared against each other.

Qualitative description of network geometry should be provided and accompanied by a network graph. Quantitative metrics assessing features of network geometry, such as *diversity* (related to the number of treatments assessed and the balance of evidence among them), *co-occurrence* (related to whether comparisons between certain treatments are more or less common), and *homophily* (related to the extent of comparisons between treatments in the same class versus competing classes), can also be mentioned.

Although common, established steps for reviewing network geometry do not yet exist, however examples of in-depth evaluations have been described related to treatments for tropical diseases and basal cell carcinoma and may be of interest to readers. An example based on 75 trials of treatments for pulmonary arterial hypertension (**Appendix Figure 3**) suggests that head-to-head studies of active therapies may prove useful to further strengthen confidence in interpretation of summary estimates of treatment comparisons.

Appendix Box 5. Probabilities and Rankings in Network Meta-Analysis

Systematic reviews incorporating network meta-analyses can provide information about the hierarchy of competing interventions in terms of treatment rankings.

The term *treatment ranking probabilities* refers to the probabilities estimated for each treatment in a network of achieving a particular placement in an ordering of treatment effects from best to worst. A network of 10 treatments provides a total of 100 ranking probabilities—that is, for each intervention, the chance of being ranked first, second, third, fourth, fifth, and so forth).

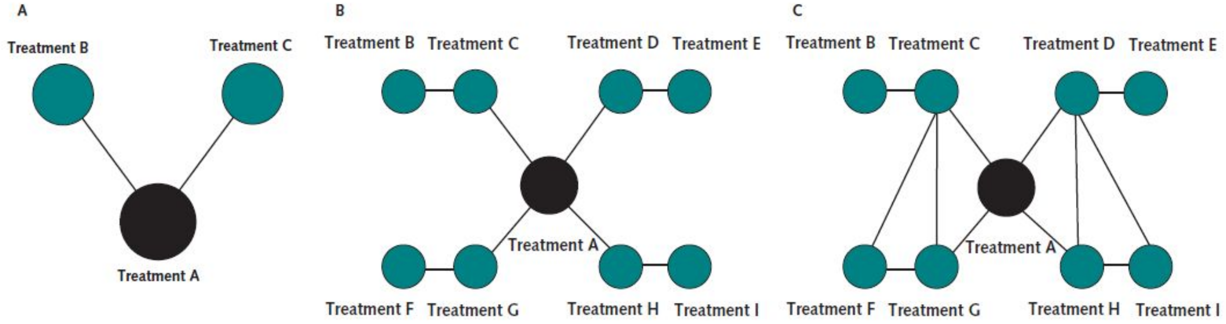
Several techniques are feasible to summarize relative rankings, and include graphical tools as well as different approaches for estimating ranking probabilities. **Appendix Figure 6** shows 2 approaches to presenting such information, on the basis of a comparison of adjuvant interventions for resected pancreatic adenocarcinoma.

Robust reporting of rankings also includes specifying median ranks with uncertainty intervals, cumulative probability curves, and the surface under the cumulative ranking (SUCRA) curve.

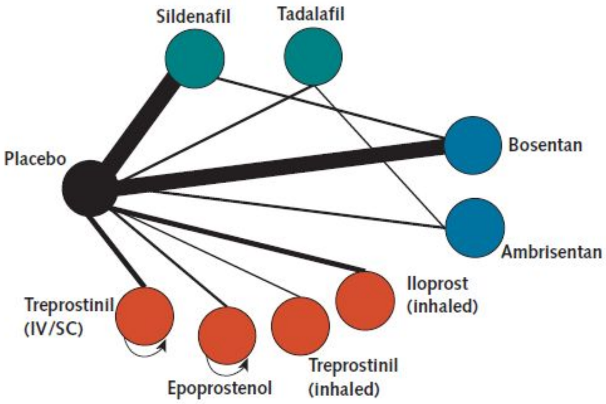
Rankings can be reported along with corresponding estimates of pairwise comparisons between interventions. Rankings should be reported with probability estimates to minimize misinterpretation from focusing too much on the most likely rank.

Rankings may exaggerate small differences in relative effects, especially if they are based on limited information. An objective assessment of the strength of information in the network and the magnitude of absolute benefits should accompany rankings to minimize potential biases.

Appendix Figure 1A-1C

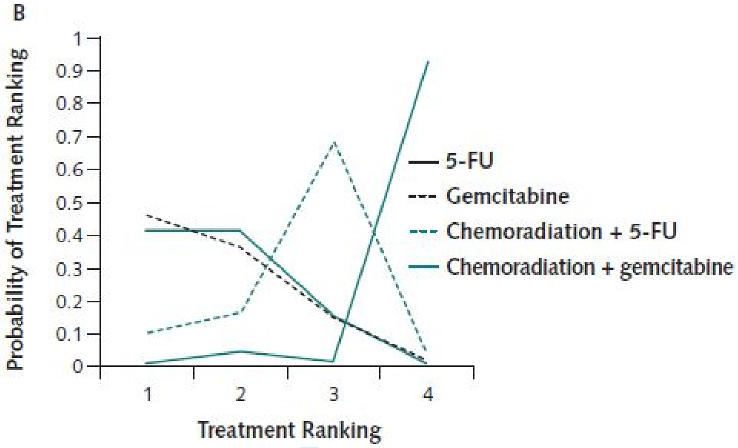


Appendix Figure 3



Appendix Figure 6

Ranking	Treatment and Cooresponding Ranking Probabilities Grade 3 or 4 Hematologic Toxicity			
	5-FU	Gemcitabine	Chemoradiation + 5-FU	Chemoradiation + gemcitabine
1	0.42	0.42	0.15	0.01
2	0.46	0.36	0.15	0.02
3	0.10	0.17	0.68	0.04
4	0.02	0.05	0.02	0.93



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Being the next of kin of a person with a brain tumor: a qualitative metasynthesis focusing on coping factors and strategies

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Being the next of kin of a person with a brain tumor: a qualitative metasynthesis focusing on coping factors and strategies

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ABSTRACT

Purpose: Being the next of kin of a person with a brain tumor is a stressful experience. For many, being a next of kin involves fear, insecurity, and overwhelming responsibility. The purpose of this study was to identify and synthesize qualitative original studies to explore coping factors and strategies that next of kin use in their role.

Method: A qualitative metasynthesis guided by Sandelowski and Barroso's guidelines was used. The databases Medline, CHINAL, and PsycINFO were searched for studies from January 2000 to January 18 2022. . Inclusion criteria were qualitative original studies focusing on coping factors or strategies used by the next of kin of persons with brain tumors. The study participants had to be aged 18 years of age or older

Results: Of 1 476 screened records, data from 20 studies, including 342 participants (207 female, 81 male, and 54 unclassified) were analyzed into metasummaries and a metasynthesis. The next of kin used coping factors such as their personal characteristics, finding meaning in their situation, external support, hope and religion, and findingg interlocutors. Coping strategies to manage the situation involved regaining control, being proactive and acceptance.

Conclusion: Next of kin of patients with brain tumor used coping factors and coping strategies gathered within themselves, in their surroundings and with assistance from a higher power to handle the situation and their role. It is important that health-care professionals suggest and facilitate these coping factors and strategies because this could reduce stress and make the role of next of kin more manageable.

Keywords: brain tumor; coping factors; coping strategies; metasynthesis; next of kin; review; qualitative studies

Strengths and limitations of the study

- The qualitative approach makes an important contribution to the research field by providing a deeper understanding of coping factors and strategies used by the next of kin of a person with a brain tumor.
- Most of the included studies in this metasynthesis were high-quality studies.

- Our sample is highly multicultural with different geographical origins represented and includes different welfare and health-care systems, and different cultures and religions.
- A limitation is that some of the subthemes, or parts of their content, could have been categorized in the other main theme.
- The majority of the sample comprised women. A more heterogeneous sample might have revealed more nuanced findings of the role of next of kin.

INTRODUCTION

In 2020 308,102 people worldwide with cancer in the central nervous system were registered.[1] The diagnosis brain tumor is very confronting, with 56% of patients experiencing one or more symptoms. Hemiparesis and cognitive challenges are most frequently reported but also headache, nausea and vomiting, vision challenges, epileptic seizures, and personality changes are considered common symptoms.[2-5] Changes in behavior and personality are considered particularly challenging, both for the patient and for the next of kin, as this may include apathy, loss of initiative and empathy, indifference, selfishness, physical and mental aggression, impaired emotional control and social abilities, and tendencies toward childish behavior, among others.[3 5 6] Studies show that the disease can be more challenging and stressful for the next of kin than for the patients. The next of kin have high rates of depression, anxiety, diverse physical pain, difficulty adapting, loneliness, and high absence from work, as well as a reduced quality of life.[7-11] Studies also show that both patients and next of kin miss additional follow-up, support, and information from health-care providers, family, friends, and the community in their struggle to cope with everyday life.[12 13]

All these strains can lead to next of kin experiencing stress and lack of coping. Lazarus and Folkman define coping as a cognitive and behavioral endeavor under constant change, dealing with external and/or internal demands that a cognitive assessment indicates are stressful or that exceed personal resources. When dealing with these demands, the next of kin has to review available coping factors; personal, external and characteristics of the situation itself.[14] This review will determine whether the situation is perceived as manageable or stressful, and secondly influence which coping strategy next of kin use. [9 13 14]

Despite this, there are only a few original qualitative studies that have investigated the coping factors that make everyday life more manageable or which strategies next of kin use to cope with their new role and tasks. To our knowledge, this research has not been synthesized. Such information is of great importance, especially for health-care providers working with this group of caregivers. With improved understanding, they could expect to be better able to facilitate more manageable everyday life among the next of kin. There is some quantitative research directed at these aspects,[8-11 15] but we wanted studies that were personal and focused on the lived experience of next of kin, hence the choice of qualitative studies. Therefore, the purpose of this metasynthesis was to identify and synthesize evidence from original qualitative studies regarding the experience of next of kin with coping factors and strategies in their role as next of kin for a person with a brain tumor. The findings are discussed in the context of Lazarus and Folkman's stress theory[14] and the approach to coping with stress to interpret our findings in a theoretical context.

METHODS

Design

The study was a metasynthesis within the interpretative paradigm. It was inspired by a phenomenological–hermeneutic design because the aim was to identify and synthesize qualitative original studies that explored next of kin attitudes and experiences.[16] The metasynthesis process consisted of five steps: (1) formulating the purpose and rationale of the study; (2) searching for and retrieving relevant qualitative research studies; (3) critically appraising the included studies; (4) classifying the findings, and finally; (5) synthesizing the findings.

Search strategy

In collaboration with an experienced librarian, we conducted a systematic search within the PsycINFO, OVID, CHINAL, and Medline databases via EBSCO host up from January 2000 until 18 January 2022. For search strategy see supplementary materials 1.

The inclusion criteria were qualitative original studies published in English, Norwegian, Swedish or Danish language that aimed to explore the factors or strategies used by the next of kin of persons with brain tumors, regardless of tumor type and stage, that enhanced their role as next of kin. The next of kin had to be 18 years of age or older. The exclusion criteria were studies that did not clearly identify coping factors or strategies, factors or strategies that included the participants' experiences in the role of bereaved and not next of kin, and studies including diagnoses other than a brain tumor.

Search outcome

The search strategy generated 1 476 unique citations. Titles and abstracts were screened by the authors using Rayyan, a systematic review management software.[17] A final consensus regarding the eligible articles was obtained through a group discussion between the authors. Seventy-two papers were read in full and evaluated against the inclusion criteria by both authors; 20 of these were included in the metasynthesis. Figure 1 shows the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flowchart with a full overview of the screening process. The search output is presented in the PRISMA flowchart. The authors read the full text of the eligible articles and independently extracted data from the included studies; this process is illustrated in Figure 1. Consensus for data extraction was obtained as part of a group discussion between the authors. Table 1 lists the title, author(s), study country, year of publication, aim, analysis, and study participants of all included studies. Most studies were from Europe: Sweden (3), Great Britain (3), Denmark (1), Belgium (1), and Turkey (1); seven were from Canada (3) and the USA (4), two from Australia and two from Taiwan. The tumor type and stage varied. For details, see supplementary materials 2.

Figure 1 about here

Quality appraisal

The quality of the 20 papers was evaluated using the Critical Appraisal Skills Program (CASP) for qualitative studies. The first evaluation was conducted blinded and independently by AWL and GR, whose CASP evaluations were then compared. Using the criteria in CASP for independent assessment, the authors mutually agreed on a final quality evaluation. For details, see Table 1.

The included studies appraised according to CASP are listed in Table 2. All studies had clearly stated the study aim and the qualitative methodologies were considered appropriate. Furthermore, several of the studies had been published in highly ranked journals. The most poorly addressed issue (criteria number 6 in the CASP list) was the influence of the researcher on the research and vice versa.

Table 1 about here

Data abstraction and analyses

As suggested by Sandelowski and Barroso,[16] two approaches to qualitative synthesis were used. The first of these involved qualitative metasummaries of qualitative findings from the original studies. This method is defined as qualitative, but the findings are presented quantitatively. The second involved a metasynthesis that developed new interpretations of the target findings from the original studies.[16] The narrative analysis was inspired by Lindseth and Nordberg's phenomenological–hermeneutic methods.[18] Three steps were followed. First, the empirical materials were read several times. Second, after extraction, the target findings were imported into NVivo 11 data management software for further analysis.[19] The text was read line-by-line to identify meaning units, subthemes, and themes. Third, the researchers aimed to achieve a comprehensive understanding of the empirical materials, meaning units, and themes, and to relate these to the aim and research question of the metasynthesis.[18] The analytic themes were identified by AWL and discussed with GR. The process of deriving the themes was inductive. The contribution of targeted findings from each of the included papers is outlined, and quotations are used to illustrate and support the findings, which increases the trustworthiness of the study. To validate the findings, both authors participated in discussions of the empirical analysis and in writing up the findings. Ethical approval was not required for the study.

Table 2 about here

Synthesis

Qualitative metasynthesis provides novel interpretations of the target findings from original studies.[16] The two main themes of this metasynthesis were coping factors within the next of kin and as external support and coping strategies – control and proactivity. The first main theme, coping factors, consisted of the subthemes, personal characteristics, meaningful, external support, having interlocutors, and hope and religion. The second main theme, coping strategies, consisted of the subthemes regain control, fight against, and acceptance. For a list of the studies that generated findings for the main themes and subthemes, see Table 2.

PATIENT AND PUBLIC INVOLVEMENT

This systematic review is based on published original studies and does not involve public involvement.

RESULTS

The findings are presented as metasummaries supported by tables and figures, and as a metasynthesis containing seven subthemes presented under two main themes. Each subtheme is supported by illustrative quotes from the original studies included.

Metasummaries

The 20 included studies consisted of 342 participants (207 women, 81 men, and 54 not classified). The focus was on the following themes: the needs of the next of kin;[2 20-24] their overall experiences as next of kin;[10 25-27] coping and coping mechanisms;[28-30] postoperative caregiving;[31 32] being a next of kin in the palliative phase;[33 34] support factors experienced;[35] how the caregiving changed over time;[36] and factors influencing treatment choice in the palliative phase.[37] Three of the studies were undertaken six months after diagnosis,[27 30 31 36] and three in the patients' palliative phase or postmortem.[33 34 37] In six studies, all the patients were children of the informants.[10 29 30 33 34]

Metasynthesis

Main theme 1: Coping factors within the next of kin and as external support

Personal characteristics such as a strong and positive personality were important coping factors for next of kin in new challenging situations.[25 29 37] Being able to show empathy for the patient and the health professionals were important, as if not the situation otherwise easily could engender feelings such as discouragement and reproach.[25] A positive mood and humor were also emphasized as for the same reasons.[29]

The role as next of kin was considered to be *meaningful* and important, as it made them feel needed and productive in the situation.[23 25 28 31] Engagement and commitment in the care of their relatives were highlighted by many next of kin, especially when the patients appreciated the help.[23] The engagement was even stronger when the emotional bond between patient and next of kin was strong.[20 21 29 35]

> *"But caring for him is something I will do—it is not a burden."*[31] (p. 81)

However, other studies revealed less engagement and commitment, and underlined anger and reluctance with the new role as the heavy responsibility and sacrifice impacted the next of kins own needs and wishes.[21 22 25 31 33]

External support made the role of next of kin easier to cope with. The support was given by family, friends, neighbors, colleagues and workplaces, health personnel, schools, the religious community, people in the local community, and even strangers.[2 10 20-35] The support from health-care professionals was especially important. This support included emotional support and assistance during patient care and treatment .[2 10 20-27 29-35] The importance of assistance such as medical supervision and nursing care was emphasized,[10 22 29] with next of kin noting that this made it possible to feel like a partner again,[23] while concurrently allowing anticipated time alone.[24] A well-known health-care professional was crucial in making this possible, because it implied that the patient received the best care as they were known to the health-care professional, and also because the assistance was considered to be less intrusive.[23 24] To experience the assistance with care as a coping factor, it was crucial that care be compassionate and of the best quality. These qualities emphasized the health professional's genuine care and gave the patients and the next of kin hope and desire to fight the disease.[10 21 23 26 27 29]

> *"She (neurosurgeon) had to give us some bad news some of the time… and you couldn't ask for a better manner in her delivery of that bad news, or her support in what we were going through."*[35] (p. 8)

When next of kin experienced that their loved ones received a low quality of care or suffered malpractice, it implied mistrust of the health-care system and weakened the experience of health-care professionals as a support factor.[10 20 23 24] Emotional support from health-care professionals implied an acknowledgment that the disease affected not only the patients, but also their next of kin. It also implied that the health-care professionals recognized and met the wishes of the next of kin for active participation in monitoring the patient's disease course.[23 25 26 34] Next of kin who did not have such involvement felt ignored, useless, and helpless.[25 29]

Support from family and friends was invaluable in the care tasks and in coping with the role of the next of kin.

> *"Just support from family and friends, that was important to me, and just knowing that I could call on them…"*[22] (p. 1098)

Social, practical, and emotional support was emphasized, and included such things as economic help, childcare, transport, and housekeeping.[10 22 24 25 29-32 34 35] Some next of kin would have appreciated even more support and help from family and friends, preferably given on their own initiative.[20 22 24 25 35 36]

Having *interlocutors,* meaning having someone to confide in and talk to, were also important in coping with the role as next of kin, as the situation, the responsibility and the impressions were though. Supportive conversations with health-care professionals were highly appreciated by many next of kin. However, this required the health-care professional's understanding and empathy for the situation of the patient as well as of their next of kin, and preferably that they should be available at all times.[21 23 26 30 31 37] Discussions with family and friends were also important,[21 24 25 27] and could even produce a stronger bond.[25] Such a bond required families and friends to understand and recognize the challenges faced by the next of kin.[24] Support groups and conversations with other next of kin were also highlighted,[2 22 24 30 34 35 37] as it could broaden the next of kins understanding of the tumor and what might to expect in the future.[27] These conversations could be face-to-face or via the Internet.[2 22 24 30 34 35 37]

> *"From time to time, I need to be able to talk to someone. Because when I lay down in the evening, then it starts to work in the inside."*[23] (p. 411)

On the other hand, support groups were also considered demanding because it was difficult to listen to other families' stories. Furthermore, for some it was considered a waste of time to spend valuable hours with people other than their closest family members.[10 22 31]

Hope and religion were emphasized as important coping factors. The next of kin hoped that a miraculous treatment would be developed so that their loved ones could survive the disease or just have a better quality of life.[2 10 20-23 26 33 34]

> *You see a positive evolution, and everything that goes better is good for her. (…) Nobody can forbid us to have hope. And miracles happen. Whether we believe it or not, that's not the point, it is the only thing to focus on."*[23] (p. 409)

Hope gave a reason to fight, although it weakened in the palliative phase.[21 26 34] Faith strengthened the hope of healing during the treatment period and gave some form of peace in the final palliative phase. In most cases, hope was related to faith.[25-27 30 34 37]

Main theme 2: Coping strategies – control and proactivity

Regaining control of the situation was a frequent coping strategy, and for most this included gathering enough information to allow an overview of what to expect, which implied some form of security.[10 20-23 27 30 35 37]

> *"So it's a, it's a roller coaster of emotion but for the most part I've been, 'What do we need to do? Where do we need to be?' And then just read,*

> *read, read whatever I can find out, whatever information because I feel like whatever I know, I can ask for.*"[30] (p. 34)

The information gathered and provided should preferably be adapted to the situation and the disease trajectory, and been given by health-care professionals .[20 22 23 25 27 29 37] The next of kin often hid this information from the patients to protect them and not diminish their hope.[10 26 30 31 34]

To regain control meant not only control of the diagnosis, but also personal control and control over their own reactions. In some cases, the next of kin denied their feelings. Some even denied the entire diagnosis,[20 25 29 30] and instead focused on being strong for the patient and the entire family.[23 25 30 32-34 36] One next of kin in Edvardsson and Ahlstroms' (2008) study[25] reported:

> *"I've sort of stowed it all away, I suppose. It is as if I'd experienced it from the outside or seen it on TV. It's often that way with sorrowful things." (p. 588)*

Being proactive, facilitate and encouraging the patient to fight the disease were also important coping strategies, as it felt better than accepting the morbid situation and not do anything.[10 21 25 26 34]

> *"People ask you how you cope. But what if you were to give up? You've got to cope—and we do have each other! (...)."[25] (p. 588)*

This implied adopting a healthier lifestyle, including changing diet and exercise habits, hoping that this would improve the effects of medical treatment,[21 26] or trying alternative treatments.[10 34] However, an increasing feeling of powerlessness was emphasized if the fight, in the form of these actions and treatments, did not meet the hope of a cure.[21 23 26 34]

As the disease progressed and life went on, most next of kin accepted the diagnosis, prognosis, and a new pattern to everyday life.[10 26 28 30 34] There was a striving for normality, starting with recommencing hobbies, work, and school for children.[10 26 28 30 31 33 34 36] This was particularly important within families with children. At the same time, accepting disease progression or a bad diagnosis was most challenging when the patient was a child.[34]

DISCUSSION

This metasynthesis aimed to explore those factors and strategies that enhanced the ability of next of kin to cope with their experience as the next of kin of a person with a brain tumor. Valuable coping factors included personal characteristics, finding meaning in the situation, external support, hope and religion, and interlocutors. Strategies to manage the situation involved regaining control, being proactive, and acceptance.[14 38]

Being the next of kin to a person with a brain tumor is considered to be a negative stressor because of the challenging life situation and care tasks. Nevertheless, several next of kin included in the metasynthesis expressed a desire to fight the disease and to gain control over the situation. This is described by Lazarus and Folkman[14] as a secondary assessment of the situation, in which the next of kin decide which measures to implement. One such measure could be to gain personal control—one of the most important and stress-reducing personal strategies available.[14]

A possible explanation for the proactive attitude of next of kin toward the disease may be their obligation and commitment to the patient. Commitment is an expression of something of great importance and can cause one to be willing to meet threats and challenges that he or she would otherwise avoid.[14] However, our findings revealed that the experience of contributing to something meaningful, not the obligation to do so, promoted coping in the situation. We consider that this is caused by the fact that obligation does not automatically make an action meaningful, but rather that it can be experienced as a compulsion. This assumption is strengthened by the findings that the tasks as next of kin could arouse emotions such as anger and aversion to the patient and to the diagnosis, rather than coping. Several studies refer to the same ambivalent experience regarding commitment and attitudes toward being a next of kin.[39 40]

External support was the factor that most relatives emphasized as promoting coping. It was described as invaluable, which was also confirmed in other studies,[41 42] and in Lazarus and Folkman's transactional stress theory.[14] At the same time, in both this metasynthesis and in other studies, next of kin voiced a strong desire and longing for even greater external support.[41 42] The findings of the metasynthesis also showed that the configuration and arrangement of the support, especially that given by health-care providers is of great importance. An explanation for the next of kins experience of unmet needs might be lack of knowledge among health-care providers about how to assist in due course. This may indicate that in some cases health-care providers should pay more attention to offer support in line with individual needs of the next of kin and for the care situations.

The findings of this metasynthesis show that several next of kin considered hope to be an important coping factor, especially during the disease trajectory. Hope has also been shown to be an extensional coping factor in several studies,[43 44] and transactional stress theory states that faith and hope are two of the most important personal factors in the cognitive assessment of stressors.[14 38] Furthermore, according to Lazarus and Folkman,[14] the two factors are strongly related, which is consistent with the findings of our metasynthesis. For several next of kin, hope was strongly grounded in religion. This was especially prominent in the studies conducted in the palliative phase, which indicated that faith is strengthened when there is no hope of curative treatment. The same pattern has also been reported in other studies describing cancer patients' experiences of palliative care.[45 46]

As the disease progressed, several next of kin accepted the diagnosis and its burden. Their fight against the disease diminished to some extent, and the relatives instead tried to "normalize" everyday life as much as possible. Similar acceptance is also reported by next of kin of other cancer patients, especially in the palliative phase.[47 48] Lazarus and Folkman describe this as a reassessment, referring to a changed cognitive assessment of the stressor based on new information from the environment and/or the person.[14]

Strengths and limitations

A strength of this metasynthesis is that the primary search in the databases was conducted with the assistance of an experienced librarian, in an attempt to ensure that as many as possible of the relevant studies were included.[49] Furthermore, most of the included studies were of high methodological quality (see Table 2). Our sample was also highly multicultural (see Table 1). This attribute strengthens the validity of the metasynthesis since geographical origin could have affected the study sample because of different participant backgrounds related to different welfare and health-care systems, cultures, and/or religions.

A limitation of our metasynthesis is that one of the 72 articles intended to be read in full text could not be obtained.[50] The formation of the subthemes is also a possible limitation. Some of the subthemes, or parts of their content, could have been categorized in the other main theme. Both main themes and subthemes overlap in several cases, and we have read similar studies[26 30] where the findings are categorized differently than in our metasynthesis. We chose to be true to the informants' statements and designated the location based on the informants' way of speaking and description of the experience. Another possible limitation is that our sample consisted mainly of women (see Table 1). A more heterogeneous sample might have revealed more nuanced findings and different experiences of the role of the next of kin.

CONCLUSION

The findings of this metasynthesis show that next of kin experience and use a range of coping factors and strategies in their role. Their experience is marked by individual differences. It is of great importance that health-care providers offer assistance that is individually adapted for these coping factors and strategies because this can reduce stress among the next of kin. The coping experience seems to go through phases, and further information is needed to understand fully how and when the various factors and strategies are used as the disease progresses. Longitudinal studies would therefore be of particular interest in this field.

Supplementary information

The manuscript has been edited by OnLine English (https://www.oleng.com.au) to comply with international publishing guidelines.

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Authors' contributions

AWL and GR designed the research project and developed the research plan. Librarian Ellen Sejersted at the University of Agder and AWL were responsible for the literature search, while AWL and GR were responsible for the analysis. Both authors were involved in the screening and inclusion of the studies, reviewed the manuscript, and contributed to the revision of the paper. Both authors read and approved the final version of the paper.

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Competing interests

The authors declare that they have no competing interests.

Patient consent for publication

Not required

Ethics approval

Ethical approval was not required, as no primary data were collected as part of this study.

Data availability

Data are available on reasonable request.

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Table 1: Critical appraisal of the included studies.

Criterion Y = yes N = no C = can't tell V = valuable NV = not valuable	1. Was there a clear statement of the aims?	2. Is a qualitative methodology appropriate?	3. Was the research design appropriate?	4. Was the recruitment strategy appropriate?	5. Were the data collected in a way that addressed the research issue?	6. Has the relationship between researcher and participants been adequately considered?	7. Have ethical issues been taken into consideration?	8. Was the data analysis sufficiently rigorous?	9. Is there a clear statement of findings?	10. How valuable is the research?	Impact factor
Arber et al. (2010)[2]	Y	Y	C	C	Y	N	Y	C	Y	V	Not found
Arber et al. (2013)[24]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	1.697
Coolbrant et al. (2015)[23]	Y	Y	Y	Y	Y	C	Y	Y	Y	V	2.022
Cutillo et al. (2018)[30]	Y	Y	Y	Y	Y	Y	N	Y	C	V	2.170
Edvardsson & Ahlström (2008)[25]	Y	Y	Y	Y	Y	N	N	Y	Y	V	3.470
Janda et al. (2006)[22]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	2.754
Huang et al. (2021)[27]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	2.592
Lipsman et al. (2007)[37]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	2.922
Lou et al. (2015)[34]	C	Y	Y	C	Y	N	N	C	Y	V	2.022
Ownsworth et al. (2015)[35]	Y	Y	Y	Y	Y	C	C	Y	Y	V	4.137
Piil et al. (2015)[21]	Y	Y	Y	Y	Y	C	Y	Y	Y	V	1.096
Russel et al. (2016)[10]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	1.197
Schmer et al. (2008)[31]	Y	Y	N	Y	N	N	Y	N	Y	V	1.096

Schubart et al. (2008)[20]	C	Y	Y	Y	Y	N	N	Y	Y	V	3.470
Sherwood et al. (2011)[36]	Y	Y	Y	Y	C	Y	N	Y	N	V	1.438
Shortman et al. (2013)	Y	Y	Y	C	C	N	Y	N	Y	V	1.918
Strang & Strang (2001)[28]	C	C	Y	N	Y	N	Y	Y	Y	V	4.956
Tastan et al. (2011)[32]	Y	Y	N	Y	Y	N	Y	N	Y	V	1.096
Wideheim et al. (2002)[26]	Y	Y	Y	Y	Y	N	C	Y	Y	V	2.022
Zelcer et al. (2010)[33]	Y	Y	Y	Y	Y	N	C	Y	Y	V	5.731

Table 2: Thematic overview showing the studies' contribution to the different themes and subthemes.

	Coping factors					Coping mechanisms		
Author	Personal characteristics	Meaningful	External support	Hope and religion	Interlocutor	Gain control	Fight	Accept
Arber et al. (2010)[2]			V	V	V			
Arber et al. (2013)[24]			V		V			
Coolbrandt et al. (2015)[23]		V	V	V	V	V		
Cutillo et al. (2018)[30]			V	V	V	V		V
Edvardson & Ahlström (2008)[25]	V	V	V	V	V	V	V	
Janda et al. (2006)[22]			V	V	V	V		
Huang et al. (2021)[27]			V	V	V	V		
Lipsman et al. (2007)[37]	V			V	V	V		
Lou et al. (2015)[34]			V	V	V	V	V	V
Ownsworth et al. (2015)[35]		V	V		V	V		
Piil et al. (2015)[21]		V	V	V	V	V	V	
Russell et al. (2016)[10]			V	V		V	V	V
Schmer et al. (2008)[31]		V	V		V			V
Schubart et al. (2008)[20]		V	V	V		V		
Sherwood et al. (2011)[36]					V	V		V
Shortman et al. (2013)[36]	V	V	V			V		

Strang & Strang (2001)[28]		V	V					V
Tastan et al. (2011)[32]			V			V		
Wideheim et al. (2002)[26]			V	V	V		V	V
Zelcer et al. (2010)[33]			V	V		V		V

Figure legend: Figure 1. Flow chart of the inclusion process

PRISMA 2009 Flow Diagram

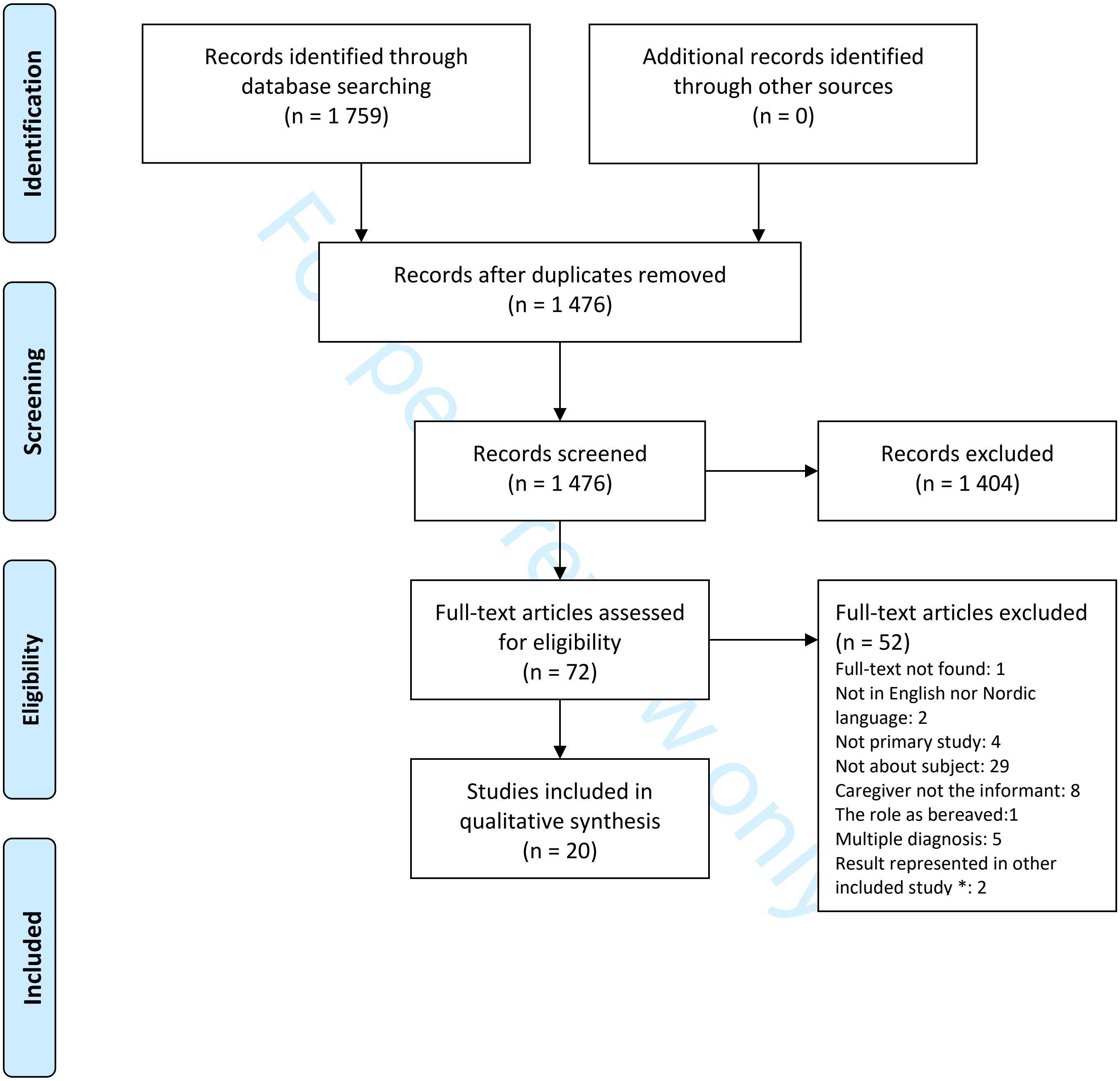


Figure 1. Flow chart of the inclusion process

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). *P*referred *R*eporting *I*tems for *S*ystematic Reviews and *M*eta-*A*nalyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097

For more information, visit www.prisma-statement.org.

The search strategy for the metasynthesis:

To search the PsycINFO database, we used the following terms: *((qualitative adj2 (research* or design* or stud* or method*)) or hermeneutic**

or "grounded theory" or "meta synthes" or metasynthesis* or metaethnograph* or interview* or phenomenolog* or thematic or themes or experience*).ti,ab,hw,id. or exp qualitative methods or phenomenology AND (caregiver* or famil* or next of kin* or relatives or spous* or wife or husband* or sibling* or sister* or brother* or dependent* or loved one* or parent* or mother* or father* or carer* or care giver*).ti,ab,hw,id. AND glioma*.ti,ab,hw,id. OR (brain adj2 (cancer or neoplasm* or tumor*)).ti,ab,hw,id.*

In Medline and CHINAL, we used the following terms: *caregiver* OR famil* OR "next of kin*" OR relatives OR spous* OR wife OR husband* OR sibling* OR brother* OR sister* OR dependent* OR "loved one*" OR parent* OR mother* OR father* OR carer* OR "care giver*" AND (MH "Qualitative Studies+") OR (MH "Qualitative Research+") OR (MH "Grounded Theory") OR Interview* OR experienc* OR phenomenolog* OR (qualitative W1 (research* OR method* OR design* OR stud*)) OR themes OR thematic OR "audio recording" OR audiorecording OR metasynthes* OR "meta synthes*" OR metaetnograph* AND (MH "Glioma+") OR glioma OR gliomas OR glioblastom* OR brain W1 (cancer OR tumor* or neoplasm*).*

Supplementary materials 2: Characteristics of the included studies.

Author/year/country	Focus	Type of brain tumor and stage of treatment at interview	Recruitment	Participants, sex, and relationship	Method/design	Data collection/analysis
Arber et al. (2010).[2] United Kingdom (UK).	Caregivers' need for information.	Malignant * Stage of treatment not described.	Specialist hospital in England.	N = 22 M: 7 and F: 15 17 spouses 3 children 2 parents	Grounded theory.	Semistructured interview/comparative method for generating categories and topics.
Arber et al. (2013).[24] United Kingdom (UK).	Caregivers' need for support.	Malignant * Stage of treatment not described.	Recruited by a nurse at a cancer center in England.	N = 22 M: 7 and F: 15 17 spouses 3 children 2 parents	Grounded theory.	Semistructured interview/comparative method for generating categories and topics.
Coolbrandt et al. (2015).[23] Belgium.	Caregivers' experience and need for support.	High-grade * Radiation or chemotherapy, or in the follow-up phase after such treatment.	University Hospital in Leuven.	N = 16 M: 6 and F: 10 13 partners 2 parents 1 friend	Grounded theory.	Semistructured interview/thematic analysis inspired by the Qualitative Analysis Guide of Leuven.
Cutillo et al. (2018).[30] USA.	Which strategies caregivers of children with a brain tumor use in the postoperative phase.	15 benign. 25 malignant. Newly diagnosed and newly operated.	Pediatric hospital in the USA.	N = 22 M: 3 and F: 19 All parents	Triangulating mixed-method.	Semistructured interview/thematic analysis.

Edvardson & Ahlström (2008)[25]. Sweden.	Caregivers' experience.	25 low-grade. 2 high-grade. Stage of treatment not described.	The patients had participated in an earlier study.	N = 28 M: 8 and F: 20 15 partners, living together 3 partners, living apart 8 parents 1 sibling 1 child	Not described.	Semistructured interview/qualitative content analysis and quantitative analysis of how the topics were distributed among the participants.
Janda et al. (2006)[22]. Australia.	The need of support for brain tumor patients and their caregivers.	Different types * Treatment phase not described, but time since diagnosis stated: 1–2 years: 22 5 years: 5 More than 5 years: 11	Members of Queensland Cancer Fund's Brain Tumor Support Service.	N = 10 in focus group, n = 8 in semistructured interview M: 4 and F: 18 13 partners 5 children	Qualitative.	Focus group interview and semistructured interview/framework analysis.
Lipsman et al. (2007)[37]. Canada.	The experience of brain tumor patients and their caregivers, and how it affects the choice of treatment.	Malignant * Palliative phase.	Recruited by a neurosurgeon.	N = 22 Further participant information not described	Qualitative.	Semistructured interview/thematic analysis.
Lou et al. (2015).[34] Taiwan.	The experience and suffering of mothers waiting for their child to die from brain tumor.	Malignant * All patients deceased.	Not described.	N =10 F: 10 All mothers	Phenomenological.	In-depth interview/Colaizzi's analysis method.

Ownsworth et al. (2015).[35] Australia.	Caregivers' experience of support.	6 low-grade. 5 high-grade. All underwent surgery and radiation or chemotherapy. 9 months – 22 years since diagnosis.	Had participated in a different study.	N = 11 M: 6 and F: 5 8 spouses 3 parents	Phenomenological.	Semistructured interview/thematic analysis.
Piil et al. (2015).[21] Denmark.	Brain tumor patients' and their caregivers' experience, and their need for rehabilitation and support.	High-grade * The interviews conducted after: 1. Surgical diagnosis 2. Oncological treatment 3,4. Oncological treatment and scan showing treatment effect 5. After treatment	The University Hospital in Copenhagen.	N = 33 M: 10 and F: 23 23 spouses 2 girl/boyfriends 7 children 1 sister	Longitudinal and exploratory.	Semistructured interview/thematic analysis.
Russell et al. (2016)[10]. Canada.	The experience of children with a brain tumor and their caregivers.	Malignant * Diagnosed at least 3 months previously, stage of treatment not described.	Hospital in Toronto.	N = 12 Based on names: F: 11 stk., 1 stk. unknown All parents	Grounded theory.	Semistructured interview/comparative analysis.
Schmer et al. (2008)[31]. USA.	Caregivers' experience concerning care tasks after chemotherapy.	Malignant * During first 6 months of treatment.	The patients' treatment center.	N = 10 Sex unknown 7 spouses 2 daughters 1 son-in-law	Phenomenological.	Semistructured interview/Colaizzi's analysis method.

Schubart et al. (2008).[20] USA.	Caregivers' challenges and unmet needs.	Different types of brain cancer * 6 deceased 2 exacerbations 2 unstable 10 stable 1 terminal 3 recurrent 1 unclear	NeuroOncology Center.	N = 25 M: 7 and F: 18 18 spouses 4 parents 2 children 1 sibling	Grounded theory.	Semistructured interview/open coding and cross-case analysis.
Sherwood et al. (2011).[36] USA.	How caregivers adapt to their new role, and how this role changes during time.	Malignant * Interviewed 1 and 4 months after diagnosis.	A regional hospital.	N = 10 M: 2 and F: 8 5 spouses 2 parents 1 child 1 nephew 1 friend	Longitudinal descriptive design.	Semistructured interview/thematic content analysis.
Shortman et al. (2013). United Kingdom (UK).	Mothers of children with brain tumor—their experience and their coping mechanisms.	Different types and degrees *. All underwent surgery, five radiation, and four chemotherapy. 17–35 months since diagnosis.	Also participated in another study.	N = 6 F: 6 All mothers.	Not described.	Semistructured interview/thematic content analysis.

Strang & Strang (2001).[28] Sweden.	The degree to which patients with a brain tumor and their caregivers cope, understand, and create meaning in the situation.	Malignant tumors, grade 2–4. Treatment stage not described.	Not described.	N = 16 Further participant information not described.	Hermeneutic phenomenological.	Semistructured interview/structural analysis based on hermeneutic circle described by Richoeur.
Tastan et al. (2011).[32] Turkey.	Caregivers' experience of postoperative phase and homecare.	Different types and degrees * All patients had undergone surgery and postoperative treatment and were being treated at home.	A research and military training hospital in Turkey.	N = 19 M: 4 and F: 6 4 spouses 4 children 1 parent 1 sibling	Descriptive qualitative study.	Semistructured interview/Colaizzi's analysis method.
Huang et al. (2021).[27] Taiwan	The lived experience of parents having a child with a brain tumor during the shared decision-making process of treatment	4 medulloblastoma 3 germ cell tumor 1 glioblastoma 1 astrocytoma 1 ependymoma The interviews were conducted between 1-6 months after the child received the diagnosis	A pediatric oncology ward at a medical center in Taiwan	N=10 M: 3 and F: 7	Descriptive phenomenological study	Semistructured interview/Colaizzi's analysis method.
Wideheim et al. (2002).[26] Sweden.	The experience of a brain tumor from a family perspective.	High-grade glioma. The interviews were conducted 2–3 weeks, 3 months, and 6 months postoperatively.	Not described.	N = 5 Sex unknown 2 spouses 2 parents	Descriptive qualitative study.	Qualitative interviews/inductive content analysis.

				1 adult child		
Zelcer et al. (2010).[33] Canada.	The experience of brain tumor patients and caregivers in the palliative phase.	Malignant * All patients deceased.	Children's Hospital, London Health Sciences Centre.	N = 25 M: 9 and F: 16 All parents	Qualitative	Semistructured interview/thematic content analysis.

1 M = Male, F = Female ∗=Tumor not further described

PRISMA NMA Checklist of Items to Include When Reporting A Systematic Review Involving a Network Meta-analysis

Section/Topic	Item #	Checklist Item	Reported on Page #
TITLE			
Title	1	Identify the report as a systematic review *incorporating a network meta-analysis (or related form of meta-analysis).*	***1***
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: **Background:** main objectives **Methods:** data sources; study eligibility criteria, participants, and interventions; study appraisal; and *synthesis methods, such as network meta-analysis.* **Results:** number of studies and participants identified; summary estimates with corresponding confidence/credible intervals; *treatment rankings may also be discussed. Authors may choose to summarize pairwise comparisons against a chosen treatment included in their analyses for brevity.* **Discussion/Conclusions:** limitations; conclusions and implications of findings. **Other:** primary source of funding; systematic review registration number with registry name.	1-2
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known*, including mention of why a network meta-analysis has been conducted.*	***3-4***
Objectives	4	Provide an explicit statement of questions being addressed, with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	4
METHODS			
Protocol and registration	5	Indicate whether a review protocol exists and if and where it can be accessed (e.g., Web address); and, if available, provide registration information, including registration number.	n.a
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale. *Clearly describe eligible treatments included in the treatment network, and note whether any have been clustered or merged into the same node (with justification).*	***6***
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	5-6
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	5-6
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable,	6-7

		included in the meta-analysis).	
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	7-8
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	5-6
Geometry of the network	**S1**	Describe methods used to explore the geometry of the treatment network under study and potential biases related to it. This should include how the evidence base has been graphically summarized for presentation, and what characteristics were compiled and used to describe the evidence base to readers.	***6***
Risk of bias within individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	7
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means). *Also describe the use of additional summary measures assessed, such as treatment rankings and surface under the cumulative ranking curve (SUCRA) values, as well as modified approaches used to present summary findings from meta-analyses.*	n.a
Planned methods of analysis	14	Describe the methods of handling data and combining results of studies for each network meta-analysis. This should include, but not be limited to: • *Handling of multi-arm trials;* • *Selection of variance structure;* • *Selection of prior distributions in Bayesian analyses; and* • *Assessment of model fit.*	7-8
Assessment of Inconsistency	**S2**	Describe the statistical methods used to evaluate the agreement of direct and indirect evidence in the treatment network(s) studied. Describe efforts taken to address its presence when found.	n.a
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	**n.a**
Additional analyses	16	Describe methods of additional analyses if done, indicating which were pre-specified. This may include, but not be limited to, the following: • Sensitivity or subgroup analyses; • Meta-regression analyses; • *Alternative formulations of the treatment network; and* • *Use of alternative prior distributions for Bayesian analyses (if applicable).*	***n.a***

RESULTS†			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	6-7, 9-10
Presentation of network structure	**S3**	Provide a network graph of the included studies to enable visualization of the geometry of the treatment network.	***6-7 and Figure 1***
Summary of network geometry	**S4**	Provide a brief overview of characteristics of the treatment network. This may include commentary on the abundance of trials and randomized patients for the different interventions and pairwise comparisons in the network, gaps of evidence in the treatment network, and potential biases reflected by the network structure.	***n.a***
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	9
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment.	n.a
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: 1) simple summary data for each intervention group, and 2) effect estimates and confidence intervals. *Modified approaches may be needed to deal with information from larger networks.*	***n.a***
Synthesis of results	21	Present results of each meta-analysis done, including confidence/credible intervals. *In larger networks, authors may focus on comparisons versus a particular comparator (e.g. placebo or standard care), with full findings presented in an appendix. League tables and forest plots may be considered to summarize pairwise comparisons.* If additional summary measures were explored (such as treatment rankings), these should also be presented.	
Exploration for inconsistency	**S5**	Describe results from investigations of inconsistency. This may include such information as measures of model fit to compare consistency and inconsistency models, *P* values from statistical tests, or summary of inconsistency estimates from different parts of the treatment network.	***n.a***
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies for the evidence base being studied.	n.a
Results of additional analyses	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression analyses, *alternative network geometries studied, alternative choice of prior distributions for Bayesian analyses,* and so forth).	***n.a***
DISCUSSION			
Summary of evidence	24	Summarize the main findings, including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy-makers).	17
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review level (e.g., incomplete retrieval of identified research, reporting bias). *Comment on the validity of the assumptions, such as transitivity and consistency. Comment*	16-17

		on any concerns regarding network geometry (e.g., avoidance of certain comparisons).	
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	17
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review. This should also include information regarding whether funding has been received from manufacturers of treatments in the network and/or whether some of the authors are content experts with professional conflicts of interest that could affect use of treatments in the network.	***n.a***

PICOS = population, intervention, comparators, outcomes, study design.

* Text in italics indicateS wording specific to reporting of network meta-analyses that has been added to guidance from the PRISMA statement.

† Authors may wish to plan for use of appendices to present all relevant information in full detail for items in this section.

Box. Terminology: Reviews With Networks of Multiple Treatments

Different terms have been used to identify systematic reviews that incorporate a network of multiple treatment comparisons. A brief overview of common terms follows.

Indirect treatment comparison: Comparison of 2 interventions for which studies against a common comparator, such as placebo or a standard treatment, are available (i.e., indirect information). The direct treatment effects of each intervention against the common comparator (i.e., treatment effects from a comparison of interventions made within a study) may be used to estimate an indirect treatment comparison between the 2 interventions (**Appendix Figure 1, A**). An indirect treatment comparison (ITC) may also involve multiple links. For example, in **Appendix Figure 1, B**, treatments B and D may be compared indirectly on the basis of studies encompassing comparisons of B versus C, A versus C, and A versus D.

Network meta-analysis or *mixed treatment comparison*: These terms, which are often used interchangeably, refer to situations involving the simultaneous comparison of 3 or more interventions. Any network of treatments consisting of strictly unclosed loops can be thought of as a series of ITCs (**Appendix Figure 1, A and B**). In mixed treatment comparisons, both direct and indirect information is available to inform the effect size estimates for at least some of the comparisons; visually, this is shown by closed loops in a network graph (**Appendix Figure 1, C**). Closed loops are not required to be present for every comparison under study. "Network meta-analysis" is an inclusive term that incorporates the scenarios of both indirect and mixed treatment comparisons.

Network geometry evaluation: The description of characteristics of the network of interventions, which may include use of numerical summary statistics. This does not involve quantitative synthesis to compare treatments. This evaluation describes the current evidence available for the competing interventions to identify gaps and potential bias. Network geometry is described further in **Appendix Box 4**.

Appendix Box 1. The Assumption of Transitivity for Network Meta-Analysis

Methods for indirect treatment comparisons and network meta-analysis enable learning about the relative treatment effects of, for example, treatments A and B through use of studies where these interventions are compared against a common therapy, C.

When planning a network meta-analysis, it is important to assess patient and study characteristics across the studies that compare pairs of treatments. These characteristics are commonly referred to as *effect modifiers* and include traits such as average patient age, gender distribution, disease severity, and a wide range of other plausible features.

For network meta-analysis to produce valid results, it is important that the distribution of effect modifiers is similar, for example, across studies of A versus B and A versus C. This balance increases the plausibility of reliable findings from an indirect comparison of B versus C through the common comparator A. When this balance is present, the assumption of transitivity can be judged to hold.

Authors of network meta-analyses should present systematic (and even tabulated) information regarding patient and study characteristics whenever available. This information helps readers to empirically evaluate the validity of the assumption of transitivity by reviewing the distribution of potential effect modifiers across trials.

Appendix Box 2. Differences in Approach to Fitting Network Meta-Analyses

Network meta-analysis can be performed within either a frequentist or a Bayesian framework. Frequentist and Bayesian approaches to statistics differ in their definitions of probability. Thus far, the majority of published network meta-analyses have used a Bayesian approach.

Bayesian analyses return the posterior probability distribution of all the model parameters given the data and prior beliefs (e.g., from external information) about the values of the parameters. They fully encapsulate the uncertainty in the parameter of interest and thus can make direct probability statements about these parameters (e.g., the probability that one intervention is superior to another).

Frequentist analyses calculate the probability that the observed data would have occurred under their sampling distribution for hypothesized values of the parameters. This approach to parameter estimation is more indirect than the Bayesian approach.

Bayesian methods have been criticized for their perceived complexity and the potential for subjectivity to be introduced by choice of a prior distribution that may affect study findings. Others argue that explicit use of a prior distribution makes transparent how individuals can interpret the same data differently. Despite these challenges, Bayesian methods offer considerable flexibility for statistical modeling.
In-depth introductions to Bayesian methods and discussion of these and other issues can be found elsewhere.

Appendix Box 3. Network Meta-Analysis and Assessment of Consistency

Network meta-analysis often involves the combination of direct and indirect evidence. In the simplest case, we wish to compare treatments A and B and have 2 sources of information: direct evidence via studies comparing A versus B, and indirect evidence via groups of studies comparing A and B with a common intervention, C. Together, this evidence forms a closed loop, ABC.

Direct and indirect evidence for a comparison of interventions should be combined only when their findings are similar in magnitude and interpretation. For example, for a comparison of mortality rates between A and B, an odds ratio determined from studies of A versus B should be similar to the odds ratio comparing A versus B estimated indirectly based on studies of A versus C and B versus C. This assumption of comparability of direct and indirect evidence is referred to as *consistency* of treatment effects.

When a treatment network contains a closed loop of interventions, it is possible to examine statistically whether there is agreement between the direct and indirect estimates of intervention effect.

Different methods to evaluate potential differences in relative treatment effects estimated by direct and indirect comparisons are grouped as *local approaches* and *global approaches.* Local approaches (e.g., the Bucher method or the node-splitting method) assess the presence of inconsistency for a particular pairwise comparison in the network, whereas global approaches (e.g., inconsistency models, I^2 measure for inconsistency) consider the potential for inconsistency in the network as a whole.

Tests for inconsistency can have limited power to detect a true difference between direct and indirect evidence. When multiple loops are being tested for inconsistency, one or a few may show inconsistency simply by chance. Further discussions of consistency and related concepts are available elsewhere.
Inconsistency in a treatment network can indicate lack of transitivity (see **Appendix Box 1**).

Appendix Box 4. Network Geometry and Considerations for Bias

The term *network geometry* is used to refer to the architecture of the treatment comparisons that have been made for the condition under study. This includes what treatments are involved in the comparisons in a network, in what abundance they are present, the respective numbers of patients randomly assigned to each treatment, and whether particular treatments and comparisons may have been preferred or avoided.

Networks may take on different shapes. Poorly connected networks depend extensively on indirect comparisons. Meta-analyses of such networks may be less reliable than those from networks where most treatments have been compared against each other.

Qualitative description of network geometry should be provided and accompanied by a network graph. Quantitative metrics assessing features of network geometry, such as *diversity* (related to the number of treatments assessed and the balance of evidence among them), *co-occurrence* (related to whether comparisons between certain treatments are more or less common), and *homophily* (related to the extent of comparisons between treatments in the same class versus competing classes), can also be mentioned.

Although common, established steps for reviewing network geometry do not yet exist, however examples of in-depth evaluations have been described related to treatments for tropical diseases and basal cell carcinoma and may be of interest to readers. An example based on 75 trials of treatments for pulmonary arterial hypertension (**Appendix Figure 3**) suggests that head-to-head studies of active therapies may prove useful to further strengthen confidence in interpretation of summary estimates of treatment comparisons.

Appendix Box 5. Probabilities and Rankings in Network Meta-Analysis

Systematic reviews incorporating network meta-analyses can provide information about the hierarchy of competing interventions in terms of treatment rankings.

The term *treatment ranking probabilities* refers to the probabilities estimated for each treatment in a network of achieving a particular placement in an ordering of treatment effects from best to worst. A network of 10 treatments provides a total of 100 ranking probabilities—that is, for each intervention, the chance of being ranked first, second, third, fourth, fifth, and so forth).

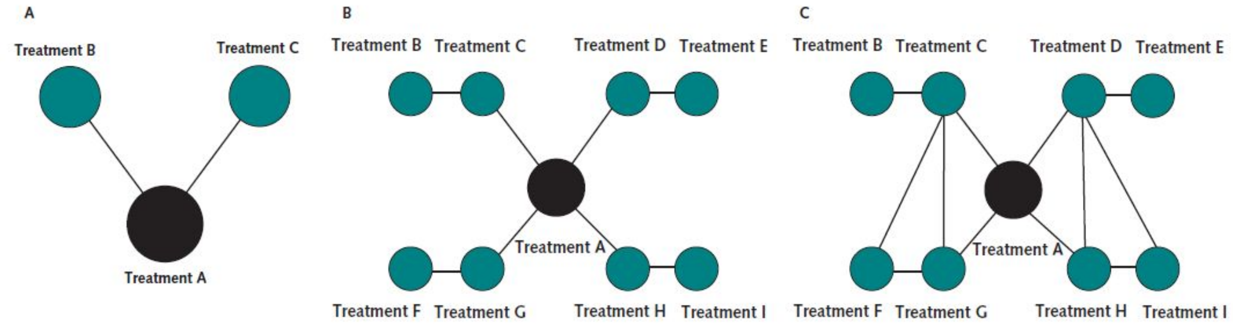
Several techniques are feasible to summarize relative rankings, and include graphical tools as well as different approaches for estimating ranking probabilities. **Appendix Figure 6** shows 2 approaches to presenting such information, on the basis of a comparison of adjuvant interventions for resected pancreatic adenocarcinoma.

Robust reporting of rankings also includes specifying median ranks with uncertainty intervals, cumulative probability curves, and the surface under the cumulative ranking (SUCRA) curve.

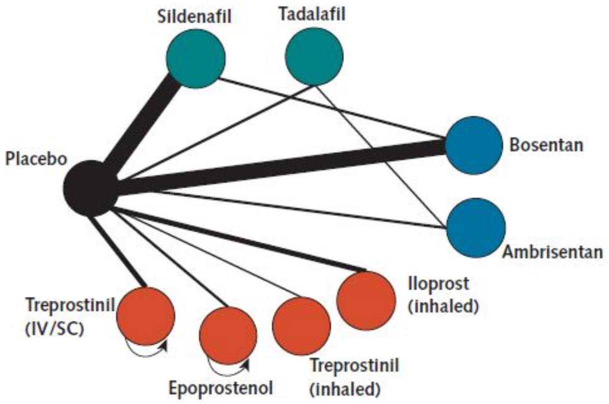
Rankings can be reported along with corresponding estimates of pairwise comparisons between interventions. Rankings should be reported with probability estimates to minimize misinterpretation from focusing too much on the most likely rank.

Rankings may exaggerate small differences in relative effects, especially if they are based on limited information. An objective assessment of the strength of information in the network and the magnitude of absolute benefits should accompany rankings to minimize potential biases.

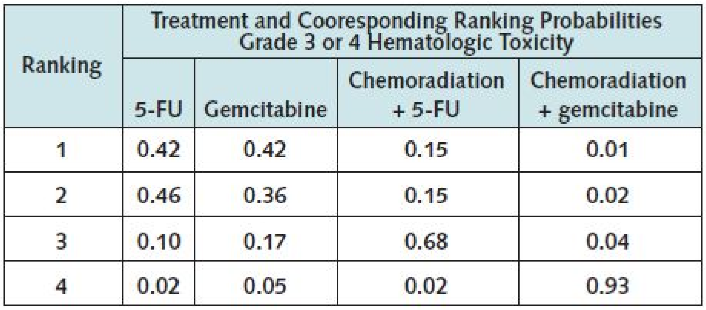
Appendix Figure 1A-1C



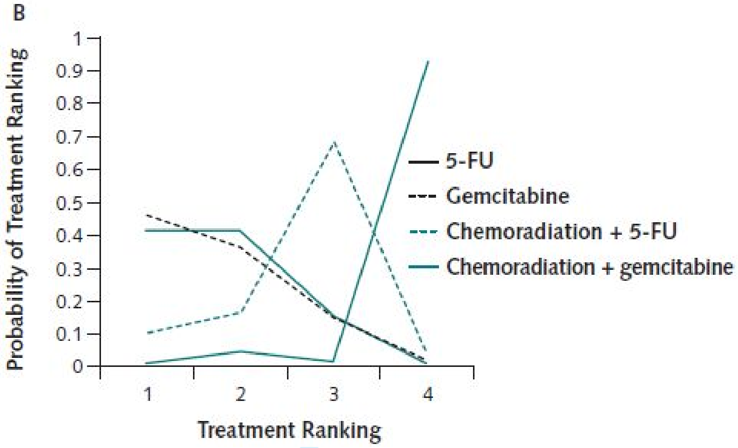
Appendix Figure 3



Appendix Figure 6



Ranking	Treatment and Cooresponding Ranking Probabilities Grade 3 or 4 Hematologic Toxicity			
	5-FU	Gemcitabine	Chemoradiation + 5-FU	Chemoradiation + gemcitabine
1	0.42	0.42	0.15	0.01
2	0.46	0.36	0.15	0.02
3	0.10	0.17	0.68	0.04
4	0.02	0.05	0.02	0.93



Coping in the role as next of kin of a person with a brain tumor: a qualitative metasynthesis

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Coping in the role as next of kin of a person with a brain tumor: a qualitative metasynthesis

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Word Count: 3981

ABSTRACT

Objective: Being the next of kin of a person with a brain tumor is a stressful experience. For many, being a next of kin involves fear, insecurity, and overwhelming responsibility. The purpose of this study was to identify and synthesize qualitative original studies that explore coping in the role as next of kin of a person with a brain tumor.

Methods: A qualitative metasynthesis guided by Sandelowski and Barroso's guidelines was used. The databases Medline, CHINAL, and PsycINFO were searched for studies from January 2000 to January 18, 2022. Inclusion criteria were qualitative original studies that aimed to explore experienced coping by the next of kin of a person with brain tumor. The next of kin had to be 18 years of age or older.

Results: Of 1 476 screened records, data from 20 studies, including 342 participants (207 female, 81 male, and 54 unclassified) were analyzed into metasummaries and a metasynthesis. The metasynthesis revealed that the next of kin experiences of coping were characterized by two main themes; 1) Coping factors within the next of kin and as external support, such as their personal characteristics, finding meaning in their situation, external support, hope and religion, and finding interlocutors. 2) Coping strategies – control and proactivit*y*, including regaining control, fight against, and acceptance

Conclusion: Next of kin of patients with brain tumor used coping factors and coping strategies gathered within themselves and in their surroundings to handle the situation and their role. It is important that health-care professionals suggest and facilitate these coping factors and strategies because this could reduce stress and make the role of next of kin more manageable.

Keywords: brain tumor; coping factors; coping strategies; metasynthesis; next of kin; review; qualitative studies

Strengths and limitations of the study

- The qualitative approach makes an important contribution to the research field by providing a deeper understanding of coping factors and strategies used by the next of kin of a person with a brain tumor.
- Most of the included studies in this metasynthesis were high-quality studies.

- Our sample is highly multicultural with different geographical origins represented and includes different welfare and health-care systems, and different cultures and religions.
- The majority of the sample comprised women., and a more heterogeneous sample might have revealed more nuanced findings of the role of next of kin.

INTRODUCTION

In 2020 308,102 people worldwide with cancer in the central nervous system were registered.[1] The diagnosis brain tumor is very confronting, with 56% of patients experiencing one or more symptoms. Hemiparesis and cognitive challenges are most frequently reported but also headache, nausea and vomiting, vision challenges, epileptic seizures, and personality changes are considered common symptoms.[2-5] Changes in behavior and personality are considered particularly challenging, both for the patient and for the next of kin, as this may include apathy, loss of initiative and empathy, indifference, selfishness, physical and mental aggression, impaired emotional control and social abilities, and tendencies toward childish behavior, among others.[3 5 6] Studies show that the disease can be more challenging and stressful for the next of kin than for the patients. The next of kin have high rates of depression, anxiety, diverse physical pain, difficulty adapting, loneliness, and high absence from work, as well as a reduced quality of life.[7-11] Studies also show that both patients and next of kin miss additional follow-up, support, and information from health-care providers, family, friends, and the community in their struggle to cope with everyday life.[12 13]

All these strains can lead to next of kin experiencing stress and lack of coping. Lazarus and Folkman define coping as a cognitive and behavioral endeavor under constant change, dealing with external and/or internal demands that a cognitive assessment indicates are stressful or that exceed personal resources. When dealing with these demands, the next of kin has to review available coping factors that could be able to making the situation more manageable; personal, external and characteristics of the situation itself.[14] This secondly influence which coping strategy, meaning active actions, next of kin use for further coping in the situation. [9 13 14]

There are some original qualitative studies that have explored coping in the role as next of kin of a person with a brain tumor. To our knowledge, this research has not been synthesized. Such information is of great importance, especially for health-care providers working with this group of caregivers. With improved understanding, they could expect to be better able to facilitate more manageable everyday life among the next of kin. There is also some quantitative research directed at these aspects,[8-11 15] but we wanted studies that were personal and focused on the lived experience of next of kin, hence the choice of qualitative studies. Therefore, the purpose of this metasynthesis was to identify and synthesize evidence from original qualitative studies regarding the experience of coping in the role as next of kin of a person with brain tumor. The findings are discussed in the context of Lazarus and Folkman's stress theory[14] and the approach to coping with stress to interpret our findings in a theoretical context.

METHODS

Design

The study was a metasynthesis within the interpretative paradigm. It was inspired by a phenomenological–hermeneutic design because the aim was to identify and synthesize qualitative original studies that explored next of kin attitudes and experiences.[16] The metasynthesis process consisted of five steps: (1) formulating the purpose and rationale of the study; (2) searching for and retrieving relevant qualitative research studies; (3) critically appraising the included studies; (4) classifying the findings, and finally; (5) synthesizing the findings.

Search strategy

In collaboration with an experienced librarian, we conducted a systematic search within the PsycINFO, OVID, CHINAL, and Medline databases via EBSCO host up from January 2000 until 18 January 2022. For search strategy see supplementary materials 1.

The inclusion criteria were qualitative original studies published in English, Norwegian, Swedish or Danish language that aimed to explore experienced coping by the next of kin of a person with a brain tumor, regardless of tumor type and stage, that enhanced their role as next of kin. The next of kin had to be 18 years of age or older. The exclusion criteria were studies that did not clearly identify coping, coping that included the participants' experiences in the role of bereaved and not next of kin, and studies including diagnoses other than a brain tumor.

Search outcome

The search strategy generated 1 476 unique citations. Titles and abstracts were screened by the authors using Rayyan, a systematic review management software.[17] A final consensus regarding the eligible articles was obtained through a group discussion between the authors. Seventy-two papers were read in full and evaluated against the inclusion criteria by both authors; 20 of these were included in the metasynthesis. Figure 1 shows the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flowchart with a full overview of the screening process. The search output is presented in the PRISMA flowchart. The authors read the full text of the eligible articles and independently extracted data from the included studies; this process is also illustrated in Figure 1. Consensus for data extraction was obtained as part of a group discussion between the authors. Supplementary materials 2 lists the title, author(s), study country, year of publication, aim, analysis, and study participants of all included studies. Most studies were from Europe: Sweden (3), Great Britain (3), Denmark (1), Belgium (1), and Turkey (1); seven were from Canada (3) and the USA (4), two from Australia and two from Taiwan. The tumor type and stage varied. For details, see supplementary materials 2.

Figure 1 about here

Quality appraisal

The quality of the 20 papers was evaluated using the Critical Appraisal Skills Program (CASP) for qualitative studies. The first evaluation was conducted blinded and independently by AWL and GR, whose CASP evaluations were then compared. Using the criteria in CASP for independent assessment, the authors mutually agreed on a final quality evaluation. For details, see Table 1.

The included studies appraised according to CASP are listed in Table 2. All studies had clearly stated the study aim and the qualitative methodologies were considered appropriate. Furthermore, several of the studies had been published in highly ranked journals. The most poorly addressed issue (criteria number 6 in the CASP list) was the influence of the researcher on the research and vice versa.

Table 1 about here

Data abstraction and analyses

As suggested by Sandelowski and Barroso,[16] two approaches to qualitative synthesis were used. The first of these involved qualitative metasummaries of qualitative findings from the original studies. This method is defined as qualitative, but the findings are presented quantitatively. The second involved a metasynthesis that developed new interpretations of the target findings from the original studies.[16] The narrative analysis was inspired by Lindseth and Nordberg's phenomenological–hermeneutic methods.[18] Three steps were followed. First, the empirical materials were read several times. Second, after extraction, the target findings were imported into NVivo 11 data management software for further analysis.[19] The text was read line-by-line to identify meaning units, subthemes, and themes. Third, the researchers aimed to achieve a comprehensive understanding of the empirical materials, meaning units, and themes, and to relate these to the aim and research question of the metasynthesis.[18] The analytic themes were identified by AWL and discussed with GR. The process of deriving the themes was inductive. The contribution of targeted findings from each of the included papers is outlined, and quotations are used to illustrate and support the findings, which increases the trustworthiness of the study. To validate the findings, both authors participated in discussions of the empirical analysis and in writing up the findings. Ethical approval was not required for the study.

Table 2 about here

Qualitative metasynthesis enables researchers to identify specific research questions, search for, apprise, summarize and combine qualitative evidence to address the research question. Metasynthesis provides novel interpretations of the target findings from original studies. [16] In our methasynthesis we identified two main themes: 1) coping factors within the next of kin and as external support and 2) coping strategies – tcontrol and proactivity, each comprising 3-5 sub themes. For a list of the studies that generated findings for the main themes and subthemes, see Table 2. When analyzing and organizing the results into themes and subthemes we chose to be in line with the content and meaning of coping in the original included studies, although some of the results could have been considered to also contributed and organized differently. The results will be elaborated below.

PATIENT AND PUBLIC INVOLVEMENT

Patient or patient organization were not involved in the planning of the study, the analyses and writing of this metasynthesis which are based on published original studies and of whom, some included patient involvement.

RESULTS

The results are presented as metasummaries supported by tables and figures, and as a metasynthesis containing two main themes. The themes are supported by illustrative quotes from the original studies included.

Metasummaries

The 20 included studies consisted of 342 participants (207 women, 81 men, and 54 not classified). The focus was on the following themes: the needs of the next of kin;[2 20-24] their overall experiences as next of kin;[10 25-27] coping and coping strategies;[28-30] postoperative caregiving;[31 32] being a next of kin in the palliative phase;[33 34] support factors experienced;[35] how the caregiving changed over time;[36] and factors influencing treatment choice in the palliative phase.[37] Three of the studies were undertaken six months after diagnosis,[27 30 31 36] and three in the patients' palliative phase or postmortem.[33 34 37] In six studies, all the patients were children of the informants.[10 29 30 33 34]

Metasynthesis

Main theme 1: Coping factors within the next of kin and as external support

Nineteen of the included studies provided data for the first main theme; *Coping factors within the next of kin and as external support* (see table 2). This main theme comprised the five sub themes: *personal characteristics, meaningful, external support, having interlocutors, and hope and religion.*

Personal characteristics such as a strong and positive personality were important coping factors for next of kin in new challenging situations.[25 29 37] Being able to show empathy for the patient and the health professionals were important, as if not the situation otherwise easily could engender feelings such as discouragement and reproach.[25] A positive mood and humor were also emphasized as for the same reasons.[29]

The role as next of kin was considered to be *meaningful* and important, as it made them feel needed and productive in the situation.[23 25 28 31] Engagement and commitment in the care of their relatives were highlighted by many next of kin, especially when the patients appreciated the help.[23] The engagement was even stronger when the emotional bond between patient and next of kin was strong.[20 21 29 35]

> *"But caring for him is something I will do—it is not a burden."*[31] (p. 81)

However, other studies revealed less engagement and commitment, and underlined anger and reluctance with the new role as the heavy responsibility and sacrifice impacted the next of kins own needs and wishes.[21 22 25 31 33]

External support made the role of next of kin easier to cope with. The support was given by family, friends, neighbors, colleagues and workplaces, health personnel, schools, the religious community, people in the local community, and even strangers.[2 10 20-35] The support from health-care professionals was especially important. This support included emotional support and assistance during patient care and treatment.[2 10 20-27 29-35] The importance of assistance such as medical supervision and nursing care was emphasized,[10 22 29] with next of kin noting that this made it possible to feel like a partner again,[23] while concurrently allowing anticipated time alone.[24] A well-known health-care professional was crucial in making this possible, because it implied that the patient received the best care as they were known to the health-care professional, and also because the assistance was considered to be less intrusive.[23 24] To experience the assistance with care as a coping factor, it was crucial that care be compassionate and of the best quality. These qualities emphasized the health professional's genuine care and gave the patients and the next of kin hope and desire to fight the disease.[10 21 23 26 27 29]

> *"She (neurosurgeon) had to give us some bad news some of the time… and you couldn't ask for a better manner in her delivery of that bad news, or her support in what we were going through."*[35] (p. 8)

When next of kin experienced that their loved ones received a low quality of care or suffered malpractice, it implied mistrust of the health-care system and weakened the experience of health-care professionals as a support factor.[10 20 23 24] Emotional support from health-care professionals implied an acknowledgment that the disease affected not only the patients, but also their next of kin. It also implied that the health-care professionals recognized and met the wishes of the next of kin for active participation in monitoring the patient's disease course.[23 25 26 34] Next of kin who did not have such involvement felt ignored, useless, and helpless.[25 29]

Support from family and friends was invaluable in the care tasks and in coping with the role of the next of kin.

> *"Just support from family and friends, that was important to me, and just knowing that I could call on them…"*[22] (p. 1098)

Social, practical, and emotional support was emphasized, and included such things as economic help, childcare, transport, and housekeeping.[10 22 24 25 29-32 34 35] Some next of kin would have appreciated even more support and help from family and friends, preferably given on their own initiative.[20 22 24 25 35 36]

Having *interlocutors,* meaning having someone to confide in and talk to, were also important in coping with the role as next of kin, as the situation, the responsibility and the impressions were though. Supportive conversations with health-care professionals were highly appreciated by many next of kin. However, this required the health-care professional's understanding and empathy for the situation of the patient as well as of their next of kin, and preferably that they should be available at all times.[21 23 26 30 31 37] Discussions with family and friends were also important,[21 24 25 27] and could even produce a stronger bond.[25] Such a bond required families and friends to understand and recognize the challenges faced by the next of kin.[24] Support groups and conversations with other next of kin were also highlighted,[2 22 24 30 34 35 37] as it could broaden the next of kins understanding of the tumor and what might to expect in the future.[27] These conversations could be face-to-face or via the Internet.[2 22 24 30 34 35 37]

> *"From time to time, I need to be able to talk to someone. Because when I lay down in the evening, then it starts to work in the inside."*[23] (p. 411)

On the other hand, support groups were also considered demanding because it was difficult to listen to other families' stories. Furthermore, for some it was considered a waste of time to spend valuable hours with people other than their closest family members.[10 22 31]

Hope and religion were emphasized as important coping factors. The next of kin hoped that a miraculous treatment would be developed so that their loved ones could survive the disease or just have a better quality of life.[2 10 20-23 26 33 34]

> *You see a positive evolution, and everything that goes better is good for her. (...) Nobody can forbid us to have hope. And miracles happen.*

> *Whether we believe it or not, that's not the point, it is the only thing to focus on."*[23] (p. 409)

Hope gave a reason to fight, although it weakened in the palliative phase.[21 26 34] Faith strengthened the hope of healing during the treatment period and gave some form of peace in the final palliative phase. In most cases, hope was related to faith.[25-27 30 34 37]

Main theme 2: Coping strategies – control and proactivity

Eighteen of the included studies provided data for the second main theme; *Coping strategies – control and proactivity* (see table 2). This main theme comprised the three sub themes: *regain control, fight against, and acceptance.*

Regaining control of the situation was a frequent coping strategy, and for most this included gathering enough information to allow an overview of what to expect, which implied some form of security.[10 20-23 27 30 35 37]

> *"So it's a, it's a roller coaster of emotion but for the most part I've been, 'What do we need to do? Where do we need to be?' And then just read, read, read whatever I can find out, whatever information because I feel like whatever I know, I can ask for.*"[30] (p. 34)

The information gathered and provided should preferably be adapted to the situation and the disease trajectory, and been given by health-care professionals.[20 22 23 25 27 29 37] The next of kin often hid this information from the patients to protect them and not diminish their hope.[10 26 30 31 34]

To regain control meant not only control of the diagnosis, but also personal control and control over their own reactions. In some cases, the next of kin denied their feelings. Some even denied the entire diagnosis,[20 25 29 30] and instead focused on being strong for the patient and the entire family.[23 25 30 32-34 36] One next of kin in Edvardsson and Ahlstroms' (2008) study[25] reported:

> *"I've sort of stowed it all away, I suppose. It is as if I'd experienced it from the outside or seen it on TV. It's often that way with sorrowful things." (p. 588)*

Being proactive, facilitate and encouraging the patient to fight the disease were also important coping strategies, as it felt better than accepting the morbid situation and not do anything.[10 21 25 26 34]

> *"People ask you how you cope. But what if you were to give up? You've got to cope—and we do have each other! (...)."*[25] *(p. 588)*

This implied adopting a healthier lifestyle, including changing diet and exercise habits, hoping that this would improve the effects of medical treatment,[21 26] or trying alternative treatments.[10 34] However, an increasing feeling of powerlessness was emphasized if the fight, in the form of these actions and treatments, did not meet the hope of a cure.[21 23 26 34]

As the disease progressed and life went on, most next of kin accepted the diagnosis, prognosis, and a new pattern to everyday life.[10 26 28 30 34] There was a striving for normality, starting with recommencing hobbies, work, and school for children.[10 26 28 30 31 33 34 36] This was particularly important within families with children. At the same time, accepting disease progression or a bad diagnosis was most challenging when the patient was a child.[34]

DISCUSSION

This metasynthesis aimed to explore coping in the role as next of kin of a person with a brain tumor. This generated two main themes;1) coping factors within the next of kin and as external support, 2) and coping strategies – control and proactivity. Valuable coping factors included personal characteristics, finding meaning in the situation, external support, hope and religion, and interlocutors. Active strategies to manage the situation involved regaining control, being proactive, and acceptance.[14 38]

Being the next of kin to a person with a brain tumor is considered to be a negative stressor because of the challenging life situation and care tasks. Nevertheless, several next of kin included in the metasynthesis expressed a desire to fight the disease and to gain control over the situation. This is described by Lazarus and Folkman[14] as a secondary assessment of the situation, in which the next of kin decide which measures to implement. One such measure could be to gain personal control—one of the most important and stress-reducing personal strategies available.[14]

A possible explanation for the proactive attitude of next of kin toward the disease may be their obligation and commitment to the patient. Commitment is an expression of something of great importance and can cause one to be willing to meet threats and challenges that he or she would otherwise avoid.[14] However, our findings revealed that the experience of contributing to something meaningful, not the obligation to do so, promoted coping in the situation. We consider that this is caused by the fact that obligation does not automatically make an action meaningful, but rather that it can be experienced as a compulsion. This assumption is strengthened by the findings that the tasks as next of kin could arouse emotions such as anger and aversion to the patient and to the diagnosis, rather than coping.

Several studies refer to the same ambivalent experience regarding commitment and attitudes toward being a next of kin.[39 40]

External support was the factor that most relatives emphasized as promoting coping. It was described as invaluable, which was also confirmed in other studies,[41 42] and in Lazarus and Folkman's transactional stress theory.[14] At the same time, in both this metasynthesis and in other studies, next of kin voiced a strong desire and longing for even greater external support.[41 42] The findings of the metasynthesis also showed that the configuration and arrangement of the support, especially that given by health-care providers is of great importance. An explanation for the next of kins experience of unmet needs might be lack of knowledge among health-care providers about how to assist in due course. This may indicate that in some cases health-care providers should pay more attention to offer support in line with individual needs of the next of kin and for the care situations.

The findings of this metasynthesis show that several next of kin considered hope to be an important coping factor, especially during the disease trajectory. Hope has also been shown to be an extensional coping factor in several studies,[43 44] and transactional stress theory states that faith and hope are two of the most important personal factors in the cognitive assessment of stressors.[14 38] Furthermore, according to Lazarus and Folkman,[14] the two factors are strongly related, which is consistent with the findings of our metasynthesis. For several next of kin, hope was strongly grounded in religion. This was especially prominent in the studies conducted in the palliative phase, which indicated that faith is strengthened when there is no hope of curative treatment. The same pattern has also been reported in other studies describing cancer patients' experiences of palliative care.[45 46]

As the disease progressed, several next of kin accepted the diagnosis and its burden. Their fight against the disease diminished to some extent, and the relatives instead tried to "normalize" everyday life as much as possible. Similar acceptance is also reported by next of kin of other cancer patients, especially in the palliative phase.[47 48] Lazarus and Folkman describe this as a reassessment, referring to a changed cognitive assessment of the stressor based on new information from the environment and/or the person.[14]

Strengths and limitations

A strength of this metasynthesis is that the primary search in the databases was conducted with the assistance of an experienced librarian, in an attempt to ensure that as many as possible of the relevant studies were included.[49] Furthermore, most of the included studies were of high methodological quality (see Table 2). Our sample was also highly multicultural (see Table 1). This attribute strengthens the validity of the metasynthesis since geographical origin could have affected the study sample because of different participant backgrounds related to different welfare and health-care systems, cultures, and/or religions.

A limitation of our metasynthesis is that one of the 72 articles intended to be read in full text could not be obtained.[50] The formation of the subthemes is also a possible limitation. Some of the subthemes, or parts of their content, could have been categorized in the other main theme. Both main themes and subthemes overlap in several cases, and we have read similar studies[26 30] where the findings are categorized differently than in our metasynthesis. We chose to be true to the informants' statements, the organization and meaning of the original studies included, and designated the location based on the informants' way of speaking and description of the experience. Another possible limitation is that our sample consisted mainly of women (see supplementary materials 2). A more heterogeneous sample might have revealed more nuanced findings and different experiences of the role of the next of kin.

CONCLUSION

The findings of this metasynthesis show that next of kin experience and use a range of coping factors and strategies in their role. Their experience is marked by individual differences. It is of great importance that health-care providers offer assistance that is individually adapted for these coping factors and strategies because this can reduce stress among the next of kin. The coping experience seems to go through phases, and further information is needed to understand fully how and when the various factors and strategies are used as the disease progresses. Longitudinal studies would therefore be of particular interest in this field.

Supplementary information

The manuscript has been edited by OnLine English (https://www.oleng.com.au) to comply with international publishing guidelines.

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Authors' contributions

AWL and GR designed the research project and developed the research plan. Librarian Ellen Sejersted at the University of Agder and AWL were responsible for the literature search, while AWL and GR were responsible for the analysis. Both authors were involved in the screening and inclusion of the studies, reviewed the manuscript, and contributed to the revision of the paper. Both authors read and approved the final version of the paper.

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Competing interests

The authors declare that they have no competing interests.

Patient consent for publication

Not required

Ethics approval

Ethical approval was not required, as no primary data were collected as part of this study.

Data availability

Data are available on reasonable request.

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Table 1: Critical appraisal of the included studies.

Criterion Y = yes N = no C = can't tell V = valuable NV = not valuable	1. Was there a clear statement of the aims?	2. Is a qualitative methodology appropriate?	3. Was the research design appropriate?	4. Was the recruitment strategy appropriate?	5. Were the data collected in a way that addressed the research issue?	6. Has the relationship between researcher and participants been adequately considered?	7. Have ethical issues been taken into consideration?	8. Was the data analysis sufficiently rigorous?	9. Is there a clear statement of findings?	10. How valuable is the research?	Impact factor
Arber et al. (2010)[2]	Y	Y	C	C	Y	N	Y	C	Y	V	Not found
Arber et al. (2013)[24]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	1.697
Coolbrant et al. (2015)[23]	Y	Y	Y	Y	Y	C	Y	Y	Y	V	2.022
Cutillo et al. (2018)[30]	Y	Y	Y	Y	Y	Y	N	Y	C	V	2.170
Edvardsson & Ahlström (2008)[25]	Y	Y	Y	Y	Y	N	N	Y	Y	V	3.470
Janda et al. (2006)[22]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	2.754
Huang et al. (2021)[27]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	2.592
Lipsman et al. (2007)[37]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	2.922
Lou et al. (2015)[34]	C	Y	Y	C	Y	N	N	C	Y	V	2.022
Ownsworth et al. (2015)[35]	Y	Y	Y	Y	Y	C	C	Y	Y	V	4.137
Piil et al. (2015)[21]	Y	Y	Y	Y	Y	C	Y	Y	Y	V	1.096
Russel et al. (2016)[10]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	1.197
Schmer et al. (2008)[31]	Y	Y	N	Y	N	N	Y	N	Y	V	1.096

Schubart et al. (2008)[20]	C	Y	Y	Y	Y	N	N	Y	Y	V	3.470
Sherwood et al. (2011)[36]	Y	Y	Y	Y	C	Y	N	Y	N	V	1.438
Shortman et al. (2013)	Y	Y	Y	C	C	N	Y	N	Y	V	1.918
Strang & Strang (2001)[28]	C	C	Y	N	Y	N	Y	Y	Y	V	4.956
Tastan et al. (2011)[32]	Y	Y	N	Y	Y	N	Y	N	Y	V	1.096
Wideheim et al. (2002)[26]	Y	Y	Y	Y	Y	N	C	Y	Y	V	2.022
Zelcer et al. (2010)[33]	Y	Y	Y	Y	Y	N	C	Y	Y	V	5.731

Table 2: Thematic overview showing the studies' contribution to the different themes and subthemes.sjekke

	Coping factors within the next of kin and as external support					*Coping strategies – control and proactivity*		
Author	Personal characteristics	Meaningful	External support	Hope and religion	Having interlocutor	Regain control	Fight against	Acceptance
Arber et al. (2010)[2]			V	V	V			
Arber et al. (2013)[24]			V		V			
Coolbrandt et al. (2015)[23]		V	V	V	V	V		
Cutillo et al. (2018)[30]			V	V	V	V		V
Edvardson & Ahlström (2008)[25]	V	V	V	V	V	V	V	
Janda et al. (2006)[22]			V	V	V	V		
Huang et al. (2021)[27]			V	V	V	V		
Lipsman et al. (2007)[37]	V			V	V	V		
Lou et al. (2015)[34]			V	V	V	V	V	V
Ownsworth et al. (2015)[35]		V	V		V	V		
Piil et al. (2015)[21]		V	V	V	V	V	V	
Russell et al. (2016)[10]			V	V		V	V	V
Schmer et al. (2008)[31]		V	V		V			V
Schubart et al. (2008)[20]		V	V	V		V		
Sherwood et al. (2011)[36]					V	V		V
Shortman et al. (2013)[36]	V	V	V			V		

Strang & Strang (2001)[28]		V	V					V
Tastan et al. (2011)[32]			V			V		
Wideheim et al. (2002)[26]			V	V	V		V	V
Zelcer et al. (2010)[33]			V	V		V		V

Figure legend: Figure 1. Flow chart of the inclusion process

PRISMA 2009 Flow Diagram

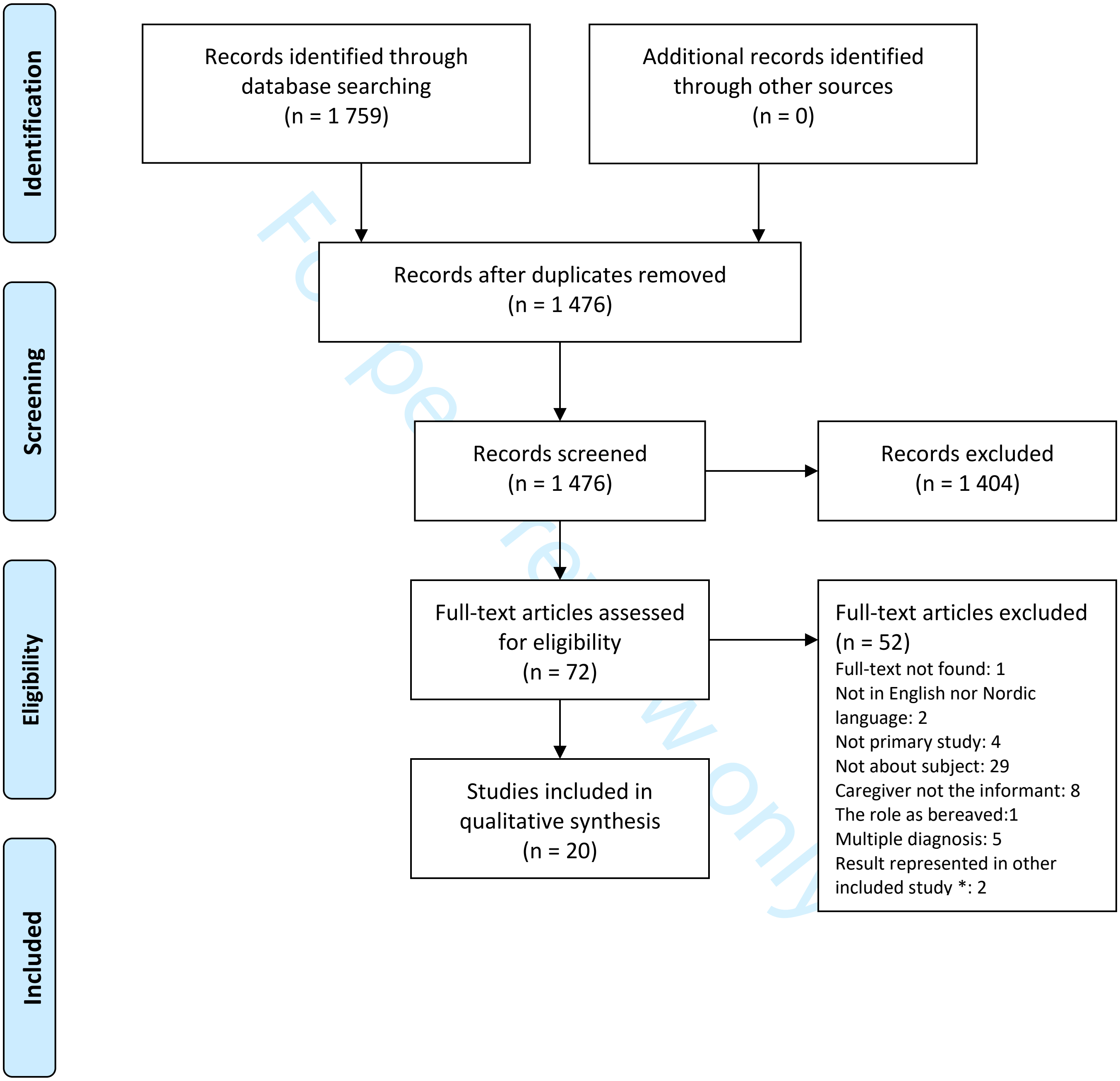

Figure 1. Flow chart of the inclusion process

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). *P*referred *R*eporting *I*tems for *S*ystematic Reviews and *M*eta-*A*nalyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097

For more information, visit www.prisma-statement.org.

The search strategy for the metasynthesis:

To search the PsycINFO database, we used the following terms: *((qualitative adj2 (research* or design* or stud* or method*)) or hermeneutic**

or "grounded theory" or "meta synthes" or metasynthesis* or metaethnograph* or interview* or phenomenolog* or thematic or themes or experience*).ti,ab,hw,id. or exp qualitative methods or phenomenology AND (caregiver* or famil* or next of kin* or relatives or spous* or wife or husband* or sibling* or sister* or brother* or dependent* or loved one* or parent* or mother* or father* or carer* or care giver*).ti,ab,hw,id. AND glioma*.ti,ab,hw,id. OR (brain adj2 (cancer or neoplasm* or tumor*)).ti,ab,hw,id.*

In Medline and CHINAL, we used the following terms: *caregiver* OR famil* OR "next of kin*" OR relatives OR spous* OR wife OR husband* OR sibling* OR brother* OR sister* OR dependent* OR "loved one*" OR parent* OR mother* OR father* OR carer* OR "care giver*" AND (MH "Qualitative Studies+") OR (MH "Qualitative Research+") OR (MH "Grounded Theory") OR Interview* OR experienc* OR phenomenolog* OR (qualitative W1 (research* OR method* OR design* OR stud*)) OR themes OR thematic OR "audio recording" OR audiorecording OR metasynthes* OR "meta synthes*" OR metaetnograph* AND (MH "Glioma+") OR glioma OR gliomas OR glioblastom* OR brain W1 (cancer OR tumor* or neoplasm*).*

Supplementary materials 2: Characteristics of the included studies.

Author/year/country	**Focus**	**Type of brain tumor and stage of treatment at interview**	**Recruitment**	**Participants, sex, and relationship**	**Method/design**	**Data collection/analysis**
Arber et al. (2010).[2] United Kingdom (UK).	Caregivers' need for information.	Malignant * Stage of treatment not described.	Specialist hospital in England.	N = 22 M: 7 and F: 15 17 spouses 3 children 2 parents	Grounded theory.	Semistructured interview/comparative method for generating categories and topics.
Arber et al. (2013).[24] United Kingdom (UK).	Caregivers' need for support.	Malignant * Stage of treatment not described.	Recruited by a nurse at a cancer center in England.	N = 22 M: 7 and F: 15 17 spouses 3 children 2 parents	Grounded theory.	Semistructured interview/comparative method for generating categories and topics.
Coolbrandt et al. (2015).[23] Belgium.	Caregivers' experience and need for support.	High-grade * Radiation or chemotherapy, or in the follow-up phase after such treatment.	University Hospital in Leuven.	N = 16 M: 6 and F: 10 13 partners 2 parents 1 friend	Grounded theory.	Semistructured interview/thematic analysis inspired by the Qualitative Analysis Guide of Leuven.
Cutillo et al. (2018).[30] USA.	Which strategies caregivers of children with a brain tumor use in the postoperative phase.	15 benign. 25 malignant. Newly diagnosed and newly operated.	Pediatric hospital in the USA.	N = 22 M: 3 and F: 19 All parents	Triangulating mixed-method.	Semistructured interview/thematic analysis.

Edvardson & Ahlström (2008)[25]. Sweden.	Caregivers' experience.	25 low-grade. 2 high-grade. Stage of treatment not described.	The patients had participated in an earlier study.	N = 28 M: 8 and F: 20 15 partners, living together 3 partners, living apart 8 parents 1 sibling 1 child	Not described.	Semistructured interview/qualitative content analysis and quantitative analysis of how the topics were distributed among the participants.
Janda et al. (2006)[22]. Australia.	The need of support for brain tumor patients and their caregivers.	Different types * Treatment phase not described, but time since diagnosis stated: 1–2 years: 22 5 years: 5 More than 5 years: 11	Members of Queensland Cancer Fund's Brain Tumor Support Service.	N = 10 in focus group, n = 8 in semistructured interview M: 4 and F: 18 13 partners 5 children	Qualitative.	Focus group interview and semistructured interview/framework analysis.
Lipsman et al. (2007)[37]. Canada.	The experience of brain tumor patients and their caregivers, and how it affects the choice of treatment.	Malignant * Palliative phase.	Recruited by a neurosurgeon.	N = 22 Further participant information not described	Qualitative.	Semistructured interview/thematic analysis.
Lou et al. (2015).[34] Taiwan.	The experience and suffering of mothers waiting for their child to die from brain tumor.	Malignant * All patients deceased.	Not described.	N =10 F: 10 All mothers	Phenomenological.	In-depth interview/Colaizzi's analysis method.

Ownsworth et al. (2015).[35] Australia.	Caregivers' experience of support.	6 low-grade. 5 high-grade. All underwent surgery and radiation or chemotherapy. 9 months – 22 years since diagnosis.	Had participated in a different study.	N = 11 M: 6 and F: 5 8 spouses 3 parents	Phenomenological.	Semistructured interview/thematic analysis.
Piil et al. (2015).[21] Denmark.	Brain tumor patients' and their caregivers' experience, and their need for rehabilitation and support.	High-grade * The interviews conducted after: 1. Surgical diagnosis 2. Oncological treatment 3,4. Oncological treatment and scan showing treatment effect 5. After treatment	The University Hospital in Copenhagen.	N = 33 M: 10 and F: 23 23 spouses 2 girl/boyfriends 7 children 1 sister	Longitudinal and exploratory.	Semistructured interview/thematic analysis.
Russell et al. (2016)[10]. Canada.	The experience of children with a brain tumor and their caregivers.	Malignant * Diagnosed at least 3 months previously, stage of treatment not described.	Hospital in Toronto.	N = 12 Based on names: F: 11 stk., 1 stk. unknown All parents	Grounded theory.	Semistructured interview/comparative analysis.
Schmer et al. (2008)[31]. USA.	Caregivers' experience concerning care tasks after chemotherapy.	Malignant * During first 6 months of treatment.	The patients' treatment center.	N = 10 Sex unknown 7 spouses 2 daughters 1 son-in-law	Phenomenological.	Semistructured interview/Colaizzi's analysis method.

Schubart et al. (2008).[20] USA.	Caregivers' challenges and unmet needs.	Different types of brain cancer * 6 deceased 2 exacerbations 2 unstable 10 stable 1 terminal 3 recurrent 1 unclear	NeuroOncology Center.	N = 25 M: 7 and F: 18 18 spouses 4 parents 2 children 1 sibling	Grounded theory.	Semistructured interview/open coding and cross-case analysis.
Sherwood et al. (2011).[36] USA.	How caregivers adapt to their new role, and how this role changes during time.	Malignant * Interviewed 1 and 4 months after diagnosis.	A regional hospital.	N = 10 M: 2 and F: 8 5 spouses 2 parents 1 child 1 nephew 1 friend	Longitudinal descriptive design.	Semistructured interview/thematic content analysis.
Shortman et al. (2013). United Kingdom (UK).	Mothers of children with brain tumor—their experience and their coping mechanisms.	Different types and degrees *. All underwent surgery, five radiation, and four chemotherapy. 17–35 months since diagnosis.	Also participated in another study.	N = 6 F: 6 All mothers.	Not described.	Semistructured interview/thematic content analysis.

Strang & Strang (2001).[28] Sweden.	The degree to which patients with a brain tumor and their caregivers cope, understand, and create meaning in the situation.	Malignant tumors, grade 2–4. Treatment stage not described.	Not described.	N = 16 Further participant information not described.	Hermeneutic phenomenological.	Semistructured interview/structural analysis based on hermeneutic circle described by Richoeur.
Tastan et al. (2011).[32] Turkey.	Caregivers' experience of postoperative phase and homecare.	Different types and degrees * All patients had undergone surgery and postoperative treatment and were being treated at home.	A research and military training hospital in Turkey.	N = 19 M: 4 and F: 6 4 spouses 4 children 1 parent 1 sibling	Descriptive qualitative study.	Semistructured interview/Colaizzi's analysis method.
Huang et al. (2021).[27] Taiwan	The lived experience of parents having a child with a brain tumor during the shared decision-making process of treatment	4 medulloblastoma 3 germ cell tumor 1 glioblastoma 1 astrocytoma 1 ependymoma The interviews were conducted between 1-6 months after the child received the diagnosis	A pediatric oncology ward at a medical center in Taiwan	N=10 M: 3 and F: 7	Descriptive phenomenological study	Semistructured interview/Colaizzi's analysis method.
Wideheim et al. (2002).[26] Sweden.	The experience of a brain tumor from a family perspective.	High-grade glioma. The interviews were conducted 2–3 weeks, 3 months, and 6 months postoperatively.	Not described.	N = 5 Sex unknown 2 spouses 2 parents	Descriptive qualitative study.	Qualitative interviews/inductive content analysis.

				1 adult child		
Zelcer et al. (2010).[33] Canada.	The experience of brain tumor patients and caregivers in the palliative phase.	Malignant * All patients deceased.	Children's Hospital, London Health Sciences Centre.	N = 25 M: 9 and F: 16 All parents	Qualitative	Semistructured interview/thematic content analysis.

1 M = Male, F = Female ∗=Tumor not further described

PRISMA 2020 Checklist

Section and Topic	Item #	Checklist item	Location where item is reported
TITLE			
Title	1	Coping in the role as next of kin of a person with a brain tumor: a qualitative metasynthesis *The page number refers to the copy without track changes*	Page 1, line 1-2
ABSTRACT			
Abstract	2	See the PRISMA 2020 for Abstracts checklist.	Page 1-2, line 18-39
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of existing knowledge.	Page 4, line 78-84
Objectives	4	Provide an explicit statement of the objective(s) or question(s) the review addresses.	Page 4, line 85-87
METHODS			
Eligibility criteria	5	Specify the inclusion and exclusion criteria for the review and how studies were grouped for the syntheses.	Page 5, line 100-109
Information sources	6	Specify all databases, registers, websites, organisations, reference lists and other sources searched or consulted to identify studies. Specify the date when each source was last searched or consulted.	Page 5, line 100-102
Search strategy	7	Present the full search strategies for all databases, registers and websites, including any filters and limits used.	Supplementary material 1
Selection process	8	Specify the methods used to decide whether a study met the inclusion criteria of the review, including how many reviewers screened each record and each report retrieved, whether they worked independently, and if applicable, details of automation tools used in the process.	Page 6, line 111-115
Data collection process	9	Specify the methods used to collect data from reports, including how many reviewers collected data from each report, whether they worked independently, any processes for obtaining or confirming data from study investigators, and if applicable, details of automation tools used in the process.	Page 6, line 120-125
Data items	10a	List and define all outcomes for which data were sought. Specify whether all results that were compatible with each outcome domain in each study were sought (e.g. for all measures, time points, analyses), and if not, the methods used to decide which results to collect.	na
	10b	List and define all other variables for which data were sought (e.g. participant and intervention characteristics, funding sources). Describe any assumptions made about any missing or unclear information.	na
Study risk of bias assessment	11	Specify the methods used to assess risk of bias in the included studies, including details of the tool(s) used, how many reviewers assessed each study and whether they worked independently, and if applicable, details of automation tools used in the process.	Page 6, line 128-132
Effect measures	12	Specify for each outcome the effect measure(s) (e.g. risk ratio, mean difference) used in the synthesis or presentation of results.	na
Synthesis methods	13a	Describe the processes used to decide which studies were eligible for each synthesis (e.g. tabulating the study intervention characteristics and comparing against the planned groups for each synthesis (item #5)).	Table 2
	13b	Describe any methods required to prepare the data for presentation or synthesis, such as handling of missing summary statistics, or data conversions.	Table 1 and 2
	13c	Describe any methods used to tabulate or visually display results of individual studies and syntheses.	Table 1 and 2

PRISMA 2020 Checklist

Section and Topic	Item #	Checklist item	Location where item is reported
	13d	Describe any methods used to synthesize results and provide a rationale for the choice(s). If meta-analysis was performed, describe the model(s), method(s) to identify the presence and extent of statistical heterogeneity, and software package(s) used.	Page 7-8
	13e	Describe any methods used to explore possible causes of heterogeneity among study results (e.g. subgroup analysis, meta-regression).	na
	13f	Describe any sensitivity analyses conducted to assess robustness of the synthesized results.	na
Reporting bias assessment	14	Describe any methods used to assess risk of bias due to missing results in a synthesis (arising from reporting biases).	na
Certainty assessment	15	Describe any methods used to assess certainty (or confidence) in the body of evidence for an outcome.	na
RESULTS			
Study selection	16a	Describe the results of the search and selection process, from the number of records identified in the search to the number of studies included in the review, ideally using a flow diagram.	Fig 1
	16b	Cite studies that might appear to meet the inclusion criteria, but which were excluded, and explain why they were excluded.	na
Study characteristics	17	Cite each included study and present its characteristics.	Supplementary materials 2 and page 8, line 177-184
Risk of bias in studies	18	Present assessments of risk of bias for each included study.	na
Results of individual studies	19	For all outcomes, present, for each study: (a) summary statistics for each group (where appropriate) and (b) an effect estimate and its precision (e.g. confidence/credible interval), ideally using structured tables or plots.	na
Results of syntheses	20a	For each synthesis, briefly summarise the characteristics and risk of bias among contributing studies.	na
	20b	Present results of all statistical syntheses conducted. If meta-analysis was done, present for each the summary estimate and its precision (e.g. confidence/credible interval) and measures of statistical heterogeneity. If comparing groups, describe the direction of the effect.	na
	20c	Present results of all investigations of possible causes of heterogeneity among study results.	na
	20d	Present results of all sensitivity analyses conducted to assess the robustness of the synthesized results.	na
Reporting biases	21	Present assessments of risk of bias due to missing results (arising from reporting biases) for each synthesis assessed.	na
Certainty of evidence	22	Present assessments of certainty (or confidence) in the body of evidence for each outcome assessed.	na
DISCUSSION			
Discussion	23a	Provide a general interpretation of the results in the context of other evidence.	Page 15-17, line 302-353
	23b	Discuss any limitations of the evidence included in the review.	Page 17-18, line 353-373
	23c	Discuss any limitations of the review processes used.	Page 17-18, line 353-373
	23d	Discuss implications of the results for practice, policy, and future research.	Page 18, line 376-383

PRISMA 2020 Checklist

Section and Topic	Item #	Checklist item	Location where item is reported
OTHER INFORMATION			
Registration and protocol	24a	Provide registration information for the review, including register name and registration number, or state that the review was not registered.	na
	24b	Indicate where the review protocol can be accessed, or state that a protocol was not prepared.	na
	24c	Describe and explain any amendments to information provided at registration or in the protocol.	na
Support	25	Describe sources of financial or non-financial support for the review, and the role of the funders or sponsors in the review.	na
Competing interests	26	Declare any competing interests of review authors.	na
Availability of data, code and other materials	27	Report which of the following are publicly available and where they can be found: template data collection forms; data extracted from included studies; data used for all analyses; analytic code; any other materials used in the review.	na

From: Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. BMJ 2021;372:n71. doi: 10.1136/bmj.n71

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Coping in the role as next of kin of a person with a brain tumor: a qualitative metasynthesis

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Coping in the role as next of kin of a person with a brain tumor: a qualitative metasynthesis

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Word Count: 3981

ABSTRACT

Objective: Being the next of kin of a person with a brain tumor is a stressful experience. For many, being a next of kin involves fear, insecurity, and overwhelming responsibility. The purpose of this study was to identify and synthesize qualitative original studies that explore coping in the role as next of kin of a person with a brain tumor.

Methods: A qualitative metasynthesis guided by Sandelowski and Barroso's guidelines was used. The databases Medline, CHINAL, and PsycINFO were searched for studies from January 2000 to January 18, 2022. Inclusion criteria were qualitative original studies that aimed to explore coping experience by the next of kin of a person with brain tumor. The next of kin had to be 18 years of age or older.

Results: Of a total of 1 476 screened records data from 20 studies, including 342 participants (207 female, 81 male, and 54 unclassified) were analyzed into metasummaries and a metasynthesis. The metasynthesis revealed that the next of kin coping experiences were characterized by two main themes: 1) Coping factors within the next of kin and as a support system, such as their personal characteristics, perceiving the role as meaningful, having a support system, and hope and religion. 2) Coping strategies – control and proactivit*y*, including regaining control, being proactive, and acceptance.

Conclusion: Next of kin of patients with brain tumors used coping factors and coping strategies gathered within themselves and in their surroundings to handle the situation and their role. It is important that health-care professionals suggest and facilitate these coping factors and strategies because this may reduce stress and make the role of next of kin more manageable.

Keywords: brain tumor; coping factors; coping strategies; metasynthesis; next of kin; review; qualitative studies

Strengths and limitations of the study

- The qualitative approach makes an important contribution to the research field by providing a deeper understanding of coping factors and strategies used by the next of kin of a person with a brain tumor.
- Most of the included studies in this metasynthesis were high-quality studies.

- Our sample is highly multicultural with different geographical origins represented and includes different welfare and health-care systems, and different cultures and religions.
- The majority of the sample were women, and a more heterogeneous sample might have revealed more nuanced findings regarding the role of next of kin.

INTRODUCTION

In 2020 308,102 people with cancer in the central nervous system were registered worldwide.[1] The diagnosis brain tumor is very confronting, with 56% of patients experiencing one or more symptoms. Hemiparesis and cognitive challenges are most frequently reported but also headaches, nausea and vomiting, vision challenges, epileptic seizures, and personality changes are considered common symptoms.[2-5] Changes in behavior and personality are considered particularly challenging, both for the patient and for the next of kin, as these may include apathy, loss of initiative and empathy, indifference, selfishness, physical and mental aggression, impaired emotional control and social skills, and tendencies toward childish behavior, among others.[3 5 6] Studies show that the disease can be more challenging and stressful for the next of kin than for the patients. The next of kin have high rates of depression, anxiety, various physical pain, adjustment difficulties, loneliness, and high work absence, as well as a reduced quality of life.[7-11] Studies also show that both patients and next of kin miss additional follow-up, support, and information from health-care providers, family, friends and the community in their struggle to cope with everyday life.[12 13]

All these strains can lead to next of kin experiencing stress and lack of coping. Lazarus and Folkman define coping as a cognitive and behavioral endeavor under constant change, dealing with external and/or internal demands that a cognitive assessment indicates as stressful or exceeding personal resources. When dealing with these demands, the next of kin has to review available coping strategies to be able to make the situation more manageable, meaning active actions the next of kin use to cope in the situation. [9 13 14]

There are some original qualitative studies that have explored coping in the role as next of kin of a person with a brain tumor. To our knowledge, this research has not been synthesized. Such information is of great importance, especially for health-care providers working with this group of caregivers. With improved understanding, they may be better equipped to facilitate a more manageable everyday life among the next of kin. Previous quantitative research directed at these aspects exist,[8-11 15] but we were interested in studies that were personal and focused on the lived experience of next of kin, hence the choice of qualitative studies. Therefore, the purpose of this metasynthesis was to identify and synthesize evidence from original qualitative studies regarding the experience of coping in the role as next of kin of a person with a brain tumor. The findings are discussed in the context of Lazarus' and Folkman's stress theory[14] and their approach to coping with stress in order to interpret our findings in a theoretical context.

METHODS

Design

The study is a metasynthesis within the interpretative paradigm. It was inspired by a phenomenological–hermeneutic design because the aim was to identify and synthesize qualitative original studies that explored next of kin attitudes and experiences.[16] The metasynthesis process consisted of five steps: (1) formulating the purpose and rationale of the study; (2) searching for and retrieving relevant qualitative research studies; (3) critically appraising the included studies; (4) classifying the findings, and finally; (5) synthesizing the findings.

Search strategy

In collaboration with an experienced librarian, we conducted a systematic search in the PsycINFO, OVID, CHINAL, and Medline databases via the EBSCO host from January 2000 until 18 January 2022. For search strategy see supplementary materials 1.

The inclusion criteria were qualitative original studies published in English, Norwegian, Swedish or Danish that aimed to explore coping experience by the next of kin of a person with a brain tumor, regardless of tumor type and stage which enhanced their role as next of kin. The next of kin had to be 18 years of age or older. The exclusion criteria were studies that did not clearly identify coping, coping that included the participants' experiences in the role of bereaved and not next of kin, and studies including diagnoses other than a brain tumor.

Search outcome

The search strategy generated 1 476 unique citations. Titles and abstracts were screened by the authors using Rayyan, a systematic review management software.[17] A final consensus regarding the eligible articles was obtained through a group discussion between the authors. Seventy-two papers were read in full and evaluated against the inclusion criteria by both authors; 20 of these were included in the metasynthesis. Figure 1 shows the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flowchart with a full overview of the screening process. The search output is presented in the PRISMA flowchart. The authors read the full text of the eligible articles and independently extracted data from the included studies; this process is also illustrated in Figure 1. Consensus for data extraction was obtained as part of a group discussion between the authors. Supplementary materials 2 lists the title, author(s), study country, year of publication, aim, analysis, and study participants of all included studies. Most studies were from Europe: Sweden (3), Great Britain (3), Denmark (1), Belgium (1), and Turkey (1); seven were from Canada (3) and the USA (4), two from Australia and two from Taiwan. The tumor type and stage varied. For details, see supplementary materials 2.

Figure 1 about here

Quality appraisal

The quality of the 20 papers was evaluated using the Critical Appraisal Skills Program (CASP) for qualitative studies. The first evaluation was conducted blinded and independently by AWL and GR whose CASP evaluations were then compared. Using the criteria in CASP for independent assessment, the authors mutually agreed on a final quality evaluation. For details, see Table 1.

The included studies that were appraised according to CASP are listed in Table 2. All studies had clearly stated the study aim and the qualitative methodologies were considered appropriate. Furthermore, several of the studies had been published in highly ranked journals. The most poorly addressed issue (criteria number 6 in the CASP list) was the influence of the researcher on the research and vice versa.

Table 1 about here

Data abstraction and analyses

As suggested by Sandelowski and Barroso,[16] two approaches to qualitative synthesis were used. The first of these involved qualitative metasummaries of qualitative findings from the original studies. This method is defined as qualitative, but the findings are presented quantitatively. The second involved a metasynthesis that developed new interpretations of the target findings from the original studies.[16] The narrative analysis was inspired by Lindseth and Nordberg's phenomenological–hermeneutic methods.[18] Three steps were followed. First, the empirical materials were read several times. Second, after extraction, the target findings were imported into NVivo 11 data management software for further analysis.[19] The text was read line-by-line to identify meaning units, subthemes, and themes. Third, the researchers aimed to achieve a comprehensive understanding of the empirical materials, meaning units, and themes, and to relate these to the aim and research question of the metasynthesis.[18] The analytic themes were identified by AWL and discussed with GR. The process of deriving the themes was inductive. The contribution of targeted findings from each of the included papers is outlined, and quotations are used to illustrate and support the findings, something which increases the trustworthiness of the study. To validate the findings, both authors participated in discussions of the empirical analysis and in writing up the findings. Ethical approval was not required for the study.

Table 2 about here

Qualitative metasynthesis enables researchers to identify specific research questions, search for, appraise, summarize, and combine qualitative evidence to address the research question. Metasynthesis provides novel interpretations of the target findings from the original studies. [16] In our methasynthesis we identified two main themes: 1) coping factors within the next of kin themselves and as a support system and 2) coping strategies –

control and proactivity, each comprising 3-4 subthemes. For a list of the studies that generated findings regarding the main themes and subthemes, see Table 2. When analyzing and organizing the results into themes and subthemes we chose to be in line with the content and meaning of coping in the original included studies, although some of the results could have been considered to also contributed and organized differently. The results will be elaborated below.

PATIENT AND PUBLIC INVOLVEMENT

No patients or patient organizations were involved in the planning of the study, the analyses or the writing of the metasynthesis. These were based on published original studies some of which included patient involvement.

RESULTS

The results are presented as metasummaries supported by tables and figures, and as a metasynthesis containing two main themes. The themes are supported by illustrative quotes from the included original studies.

Metasummaries

The 20 studies that were included comprised 342 participants (207 women, 81 men, and 54 not classified). The focus was on the following themes: the needs of the next of kin;[2 20-24] their overall experiences as next of kin;[10 25-27] coping and coping strategies;[28-30] postoperative caregiving;[31 32] being a next of kin in the palliative phase;[33 34] experienced support factors;[35] how the caregiving changed over time;[36] and factors influencing treatment choice in the palliative phase.[37] Three of the studies were undertaken six months after diagnosis,[27 30 31 36] and three in the patients' palliative phase or postmortem.[33 34 37] In six studies the patients were children of the informants.[10 29 30 33 34]

Metasynthesis

Main theme 1: Coping factors within the next of kin and as a support system

Nineteen of the included studies provided data regarding the first main theme; *Coping factors within the next of kin and as external support* (see table 2). This main theme comprised the following four sub themes: *personal characteristics, perceiving the role as meaningful, having a support system, and hope and religion.*

Personal characteristics such as a strong and positive personality were important coping factors for next of kin in new challenging situations.[25 29 37] Being able to show empathy for the patient and the health professionals was important, if not the situation could easily engender feelings such as discouragement and reproach.[25] A positive mood and a sense of humor were also emphasized for the same reasons.[29]

To perceive the role as next of kin as *meaningful* was important, as it made the next of kin feel needed and productive in the situation.[23 25 28 31] Engagement and commitment in the care of their relatives were highlighted as important by many next of kin, especially when the patients appreciated the help.[23] The engagement was even stronger when the emotional bond between patient and next of kin was strong.[20 21 29 35]

> *"But caring for him is something I will do—it is not a burden."*[31] (p. 81)

However, other studies revealed less engagement and commitment, and underlined anger and reluctance with the new role as the heavy responsibility and sacrifice impacted the next of kin's own needs and wishes.[21 22 25 31 33]

Having a support system made the role of next of kin easier to cope with. The support was given by family, friends, neighbors, colleagues and workplaces, health personnel, schools, the religious community, people in the local community, and even strangers.[2 10 20-35] The support from health-care professionals was especially important. This support included emotional support and assistance during patient care and treatment.[2 10 20-27 29-35] The importance of assistance such as medical supervision and nursing care was emphasized,[10 22 29] with next of kin noting that this made it possible to feel like a partner again,[23] while at the same time allowing for anticipated time alone.[24] A familiar health-care professional was crucial in making this possible, because it implied that the patient would receive the best care as they were known to the health-care professional, and also because the assistance was considered to be less intrusive.[23 24] To experience the assistance with care as a coping factor, it was crucial that the care was compassionate and of the best quality. These qualities emphasized the health professionals genuine care and gave the patients and the next of kin hope and a desire to fight the disease.[10 21 23 26 27 29]

> *"She (neurosurgeon) had to give us some bad news some of the time… and you couldn't ask for a better manner in her delivery of that bad news, or her support in what we were going through."*[35] (p. 8)

When next of kin experienced that their loved ones received a low quality of care or suffered malpractice it caused mistrust of the health-care system and weakened the experience of health-care professionals as a support factor.[10 20 23 24] Emotional support from health-care professionals implied an acknowledgment that the disease affected not only the patients, but also their next of kin. It also implied that the health-care professionals recognized and met the wishes of the next of kin for active participation in monitoring the patients disease course.[23 25 26 34] Next of kin who did not have such involvement felt ignored, useless and helpless.[25 29] Supportive conversations with health-care professionals were highly appreciated by many next of kin. However, this required the health-care professional's understanding and empathy for the situation of the patient as well as of their next of kin, and preferably that they should be always available.[21 23 26 30 31 37]

Support from family and friends was invaluable in the care tasks and in coping with the role of next of kin.

> *"Just support from family and friends, that was important to me, and just knowing that I could call on them…"*[22] (p. 1098)

Social, practical, and emotional support was emphasized, and included such things as economic help, childcare, transport and housekeeping.[10 22 24 25 29-32 34 35] Some next of kin would have appreciated even more support and help from family and friends, preferably given on the family and friends' own initiative.[20 22 24 25 35 36]

Discussions with family and friends were also important,[21 24 25 27] and could even create a stronger bond.[25] Such a bond required families and friends to understand and recognize the challenges faced by the next of kin.[24] Support groups and conversations with other next of kin were also highlighted as important,[2 22 24 30 34 35 37] as they might broaden the next of kin's understanding of the tumor and what they might expect in the future.[27] These conversations could be face-to-face or via the Internet.[2 22 24 30 34 35 37]

> *"From time to time, I need to be able to talk to someone. Because when I lay down in the evening, then it starts to work in the inside."*[23] (p. 411)

On the other hand, support groups were also considered demanding because it was difficult to listen to other families' stories. Furthermore, for some it was considered a waste of time to spend valuable hours with people other than their closest family members.[10 22 31]

Hope and religion were emphasized as important coping factors. The next of kin hoped that a miraculous treatment would be developed so that their loved ones could survive the disease or just have a better quality of life.[2 10 20-23 26 33 34]

> *You see a positive evolution, and everything that goes better is good for her. (...) Nobody can forbid us to have hope. And miracles happen. Whether we believe it or not, that's not the point, it is the only thing to focus on."*[23] (p. 409)

Hope gave a reason to fight, although it weakened in the palliative phase.[21 26 34] Faith strengthened the hope of healing during the treatment period and gave some form of peace in the final palliative phase. In most cases, hope was related to faith.[25-27 30 34 37]

Main theme 2: Coping strategies – control and proactivity

Eighteen of the included studies provided data regarding the second main theme; *Coping strategies – control and proactivity* (see table 2). This main theme comprised the three subthemes: *regaining control, being proactive and acceptance.*

Regaining control of the situation was a frequent coping strategy, and for most this included gathering enough information to allow an overview of what to expect, something which implied some form of security.[10 20-23 27 30 35 37]

> *"So it's a, it's a roller coaster of emotion but for the most part I've been, 'What do we need to do? Where do we need to be?' And then just read, read, read whatever I can find out, whatever information because I feel like whatever I know, I can ask for*."[30] (p. 34)

The information that was gathered and provided should preferably be adapted to the situation and the disease trajectory, and had been given by health-care professionals.[20 22 23 25 27 29 37] The next of kin often hid this information from the patients to protect them and not diminish their hope.[10 26 30 31 34]

To regain control meant not only control of the diagnosis, but also personal control and control over own reactions. In some cases, the next of kin denied their feelings. Some even denied the entire diagnosis,[20 25 29 30] and instead focused on being strong for the patient and the entire family.[23 25 30 32-34 36] One next of kin in Edvardsson and Ahlstroms' (2008) study[25] reported:

> *"I've sort of stowed it all away, I suppose. It is as if I'd experienced it from the outside or seen it on TV. It's often that way with sorrowful things." (p. 588)*

Being proactive, facilitating and encouraging the patient to fight the disease were also important coping strategies, as it felt better than accepting the morbid situation and not do anything.[10 21 25 26 34]

> *"People ask you how you cope. But what if you were to give up? You've got to cope—and we do have each other! (...)."[25] (p. 588)*

This implied adopting a healthier lifestyle, including a change in diet and exercise habits, hoping that this would improve the effects of medical treatment,[21 26] or trying alternative treatments.[10 34] However, an increasing feeling of powerlessness was emphasized if the fight, in the form of these actions and treatments, did not meet the hope of a cure.[21 23 26 34]

As the disease progressed and life went on there was a strive for normality, particularly in families with children. This lead most next of kin into a strategy of *acceptance,* as *e*veryday life continued. This involved work, school for children and hobbies. [10 26 28 30 34 10 26 28 30 31 33 34 36] Although this was an important and expected strategy, accepting disease progression or a bad diagnosis was challenging, especially when the patient was a child.[34]

DISCUSSION

This metasynthesis aimed to explore coping in the role as next of kin of a person with a brain tumor. This generated two main themes:1) coping factors within the next of kin and as a support system, 2) and coping strategies – control and proactivity. Valuable coping factors included personal characteristics, perceiving the role as next of kin as meaningful, having a support system, and hope and religion. Active strategies to manage the situation involved regaining control, being proactive, and acceptance.[14 38]

Being the next of kin to a person with a brain tumor is considered to be a negative stressor because of the challenging life situation and care tasks. Nevertheless, several next of kin who were included in the metasynthesis expressed a desire to be proactive, fight the disease and to gain control over the situation. This is described by Lazarus and Folkman[14] as a secondary assessment of the situation, in which the next of kin decide which measures to implement. One such measure could be to gain personal control—one of the most important and stress-reducing personal strategies available.[14]

A possible explanation for the proactive attitude of next of kin toward the disease may be their obligation and commitment to the patient. Commitment is an expression of something of great importance and can cause some to be willing to meet threats and challenges that he or she would otherwise avoid.[14] However, our findings revealed that the experience of contributing to something meaningful, not the obligation to do so, promoted coping in the situation. We consider that this is caused by the fact that obligation does not automatically make an action meaningful, but rather that it can be experienced as a compulsion. This assumption is strengthened by the findings that the tasks as next of kin may arouse emotions such as anger and aversion towards the patient and the diagnosis, rather than coping. Several studies refer to the same ambivalent experience regarding commitment and attitudes toward being a next of kin.[39 40]

Having a support system was the factor that most relatives emphasized as promoting coping. It was described as invaluable something which was also confirmed in other studies,[41 42] and in Lazarus and Folkman's transactional stress theory.[14] At the same time, in both this metasynthesis and in other studies, next of kin voiced a strong desire and longing for even greater external support.[41 42] The findings of the metasynthesis also showed that the configuration and arrangement of the support, especially that given by health-care providers are of great importance. An explanation for the next of kin's experience of unmet needs might be lack of knowledge among health-care providers about how to assist at the right time. This may indicate that in some cases health-care providers should pay more attention to offering support in line with the individual needs of the next of kin and the care situations.

The findings of this metasynthesis show that several next of kin considered hope to be an important coping factor, especially during the disease trajectory. Hope has also been shown to be an strengthening coping factor in several studies,[43 44] and transactional stress theory states that faith and hope are two of the most important personal factors in the cognitive assessment of stressors.[14 38] Furthermore, according to Lazarus and Folkman,[14] the two factors are strongly related, which is consistent with the findings in our metasynthesis. For several next of kin, hope was strongly grounded in religion. This was especially prominent in the studies conducted in the palliative phase, which indicated that faith is strengthened when there is no hope of curative treatment. The same pattern has also been reported in other studies describing cancer patients' experiences of palliative care.[45 46]

As the disease progressed, several next of kin chose an acceptance strategy toward the diagnosis and its burden. Their fight against the disease diminished to some extent, and instead the relatives tried to "normalize" everyday life as much as possible. A similar strategy is also reported by next of kin of other cancer patients, especially in the palliative phase.[47 48] Lazarus and Folkman describe this as a reassessment, referring to a changed cognitive assessment of the stressor based on new information from the environment and/or the person himself or herself.[14]

Strengths and limitations

A strength of this metasynthesis is that the primary search in the databases was conducted with the assistance of an experienced librarian in an attempt to ensure that as many as possible of the relevant studies were included.[49] Furthermore, most of the included studies were of high methodological quality (see Table 2). Our sample was also highly multicultural (see Table 1). This attribute strengthens the validity of the metasynthesis since geographical origin might have affected the study sample because of different participant backgrounds related to different welfare and health-care systems, cultures, and/or religions.

A limitation of our metasynthesis is that one of the 72 articles that was intended to be read in full text could not be obtained.[50] The formation of the subthemes is also a possible limitation. Some of the subthemes, or parts of their content, could also have been categorized in the other main theme. Both main themes and subthemes overlap in several cases, and we have read similar studies[26 30] where the findings are categorized differently than in our metasynthesis. We chose to be true to the informants' statements, the organization and meaning of the original studies that were included, and allocated the findings based on the informants' way of speaking and description of the experience. Another possible limitation is that our sample consisted mainly of women (see supplementary materials 2). A more heterogeneous sample might have revealed more nuanced findings and different experiences of the role of the next of kin.

CONCLUSION

The findings of this metasynthesis show that next of kin experience and use a range of coping factors and strategies in their role. Their experience is marked by individual differences. It is of great importance that health-care providers offer assistance which is individually adapted to these coping factors and strategies because this may reduce stress among the next of kin. The coping experience seems to go through phases, and further information is needed to fully understand how and when the various factors and strategies are used as the disease progresses. Longitudinal studies would therefore be of particular interest in this field.

Supplementary information

The manuscript has been edited by OnLine English (https://www.oleng.com.au) to comply with international publishing guidelines.

Authors' contributions

AWL and GR designed the research project and developed the research plan. AWL was responsible for the literature search, while AWL and GR were responsible for the analysis. Both authors were involved in the screening and inclusion of the studies, reviewed the manuscript, and contributed to the revision of the paper. Both authors read and approved the final version of the paper.

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Competing interests

The authors declare that they have no competing interests.

Patient consent for publication

Not required

Ethics approval

Ethical approval was not required, as no primary data were collected as part of this study.

Data availability

Data are available on reasonable request.

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Table 1: Critical appraisal of the included studies.

Criterion Y = yes N = no C = can't tell V = valuable NV = not valuable	1. Was there a clear statement of the aims?	2. Is a qualitative methodology appropriate?	3. Was the research design appropriate?	4. Was the recruitment strategy appropriate?	5. Were the data collected in a way that addressed the research issue?	6. Has the relationship between researcher and participants been adequately considered?	7. Have ethical issues been taken into consideration?	8. Was the data analysis sufficiently rigorous?	9. Is there a clear statement of findings?	10. How valuable is the research?	Impact factor
Arber et al. (2010)[2]	Y	Y	C	C	Y	N	Y	C	Y	V	Not found
Arber et al. (2013)[24]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	1.697
Coolbrant et al. (2015)[23]	Y	Y	Y	Y	Y	C	Y	Y	Y	V	2.022
Cutillo et al. (2018)[30]	Y	Y	Y	Y	Y	Y	N	Y	C	V	2.170
Edvardsson & Ahlström (2008)[25]	Y	Y	Y	Y	Y	N	N	Y	Y	V	3.470
Janda et al. (2006)[22]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	2.754
Huang et al. (2021)[27]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	2.592
Lipsman et al. (2007)[37]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	2.922
Lou et al. (2015)[34]	C	Y	Y	C	Y	N	N	C	Y	V	2.022
Ownsworth et al. (2015)[35]	Y	Y	Y	Y	Y	C	C	Y	Y	V	4.137
Piil et al. (2015)[21]	Y	Y	Y	Y	Y	C	Y	Y	Y	V	1.096
Russel et al. (2016)[10]	Y	Y	Y	Y	Y	N	Y	Y	Y	V	1.197
Schmer et al. (2008)[31]	Y	Y	N	Y	N	N	Y	N	Y	V	1.096

Schubart et al. (2008)[20]	C	Y	Y	Y	Y	N	N	Y	Y	V	3.470
Sherwood et al. (2011)[36]	Y	Y	Y	Y	C	Y	N	Y	N	V	1.438
Shortman et al. (2013)	Y	Y	Y	C	C	N	Y	N	Y	V	1.918
Strang & Strang (2001)[28]	C	C	Y	N	Y	N	Y	Y	Y	V	4.956
Tastan et al. (2011)[32]	Y	Y	N	Y	Y	N	Y	N	Y	V	1.096
Wideheim et al. (2002)[26]	Y	Y	Y	Y	Y	N	C	Y	Y	V	2.022
Zelcer et al. (2010)[33]	Y	Y	Y	Y	Y	N	C	Y	Y	V	5.731

Table 2: Thematic overview showing the studies' contribution to the different themes and subthemes.

	Coping factors within the next of kin and as a support system				*Coping strategies – control and proactivity*		
Author	Personal characteristics	Perceiving the role as meaningful	Having a support system	Hope and religion	Regain control	Proactivity	Acceptance
Arber et al. (2010)[2]			V	V			
Arber et al. (2013)[24]			V				
Coolbrandt et al. (2015)[23]		V	V	V	V		
Cutillo et al. (2018)[30]			V	V	V		V
Edvardson & Ahlström (2008)[25]	V	V	V	V	V	V	
Janda et al. (2006)[22]			V	V	V		
Huang et al. (2021)[27]			V	V	V		
Lipsman et al. (2007)[37]	V			V	V		
Lou et al. (2015)[34]			V	V	V	V	V
Ownsworth et al. (2015)[35]		V	V		V		
Piil et al. (2015)[21]		V	V	V	V	V	
Russell et al. (2016)[10]			V	V	V	V	V
Schmer et al. (2008)[31]		V	V				V
Schubart et al. (2008)[20]		V	V	V	V		
Sherwood et al. (2011)[36]			V		V		V

Shortman et al. (2013)[36]	V	V	V		V		
Strang & Strang (2001)[28]		V	V				V
Tastan et al. (2011)[32]			V		V		
Wideheim et al. (2002)[26]			V	V		V	V
Zelcer et al. (2010)[33]			V	V	V		V

Figure legend: Figure 1. Flow chart of the inclusion process

PRISMA 2009 Flow Diagram

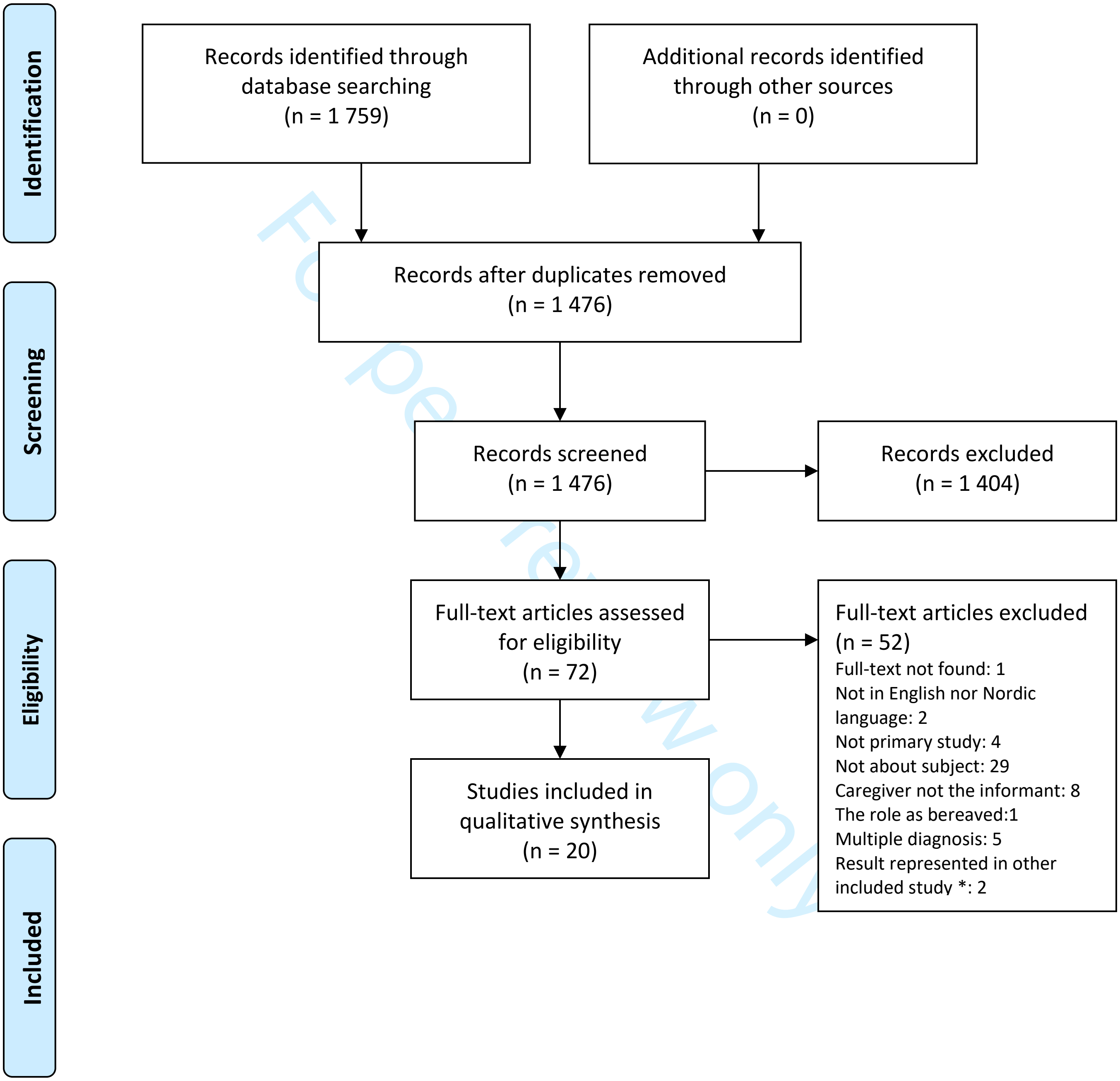

Figure 1. Flow chart of the inclusion process

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). *P*referred *R*eporting *I*tems for *S*ystematic Reviews and *M*eta-*A*nalyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097

For more information, visit www.prisma-statement.org.

The search strategy for the metasynthesis:

To search the PsycINFO database, we used the following terms: *((qualitative adj2 (research* or design* or stud* or method*)) or hermeneutic* or "grounded theory" or "meta synthes*" or metasynthesis* or metaethnograph* or interview* or phenomenolog* or thematic or themes or experience*).ti,ab,hw,id. or exp qualitative methods or phenomenology AND (caregiver* or famil* or next of kin* or relatives or spous* or wife or husband* or sibling* or sister* or brother* or dependent* or loved one* or parent* or mother* or father* or carer* or care giver*).ti,ab,hw,id. AND glioma*.ti,ab,hw,id. OR (brain adj2 (cancer or neoplasm* or tumor*)).ti,ab,hw,id.*

In Medline and CHINAL, we used the following terms: *caregiver* OR famil* OR "next of kin*" OR relatives OR spous* OR wife OR husband* OR sibling* OR brother* OR sister* OR dependent* OR "loved one*" OR parent* OR mother* OR father* OR carer* OR "care giver*" AND (MH "Qualitative Studies+") OR (MH "Qualitative Research+") OR (MH "Grounded Theory") OR Interview* OR experienc* OR phenomenolog* OR (qualitative W1 (research* OR method* OR design* OR stud*)) OR themes OR thematic OR "audio recording" OR audiorecording OR metasynthes* OR "meta synthes*" OR metaetnograph* AND (MH "Glioma+") OR glioma OR gliomas OR glioblastom* OR brain W1 (cancer OR tumor* or neoplasm*).*

Supplementary materials 2: Characteristics of the included studies.

Author/year/country	Focus	Type of brain tumor and stage of treatment at interview	Recruitment	Participants, sex, and relationship	Method/design	Data collection/analysis
Arber et al. (2010).[2] United Kingdom (UK).	Caregivers' need for information.	Malignant * Stage of treatment not described.	Specialist hospital in England.	N = 22 M: 7 and F: 15 17 spouses 3 children 2 parents	Grounded theory.	Semistructured interview/comparative method for generating categories and topics.
Arber et al. (2013).[24] United Kingdom (UK).	Caregivers' need for support.	Malignant * Stage of treatment not described.	Recruited by a nurse at a cancer center in England.	N = 22 M: 7 and F: 15 17 spouses 3 children 2 parents	Grounded theory.	Semistructured interview/comparative method for generating categories and topics.
Coolbrandt et al. (2015).[23] Belgium.	Caregivers' experience and need for support.	High-grade * Radiation or chemotherapy, or in the follow-up phase after such treatment.	University Hospital in Leuven.	N = 16 M: 6 and F: 10 13 partners 2 parents 1 friend	Grounded theory.	Semistructured interview/thematic analysis inspired by the Qualitative Analysis Guide of Leuven.
Cutillo et al. (2018).[30] USA.	Which strategies caregivers of children with a brain tumor use in the postoperative phase.	15 benign. 25 malignant. Newly diagnosed and newly operated.	Pediatric hospital in the USA.	N = 22 M: 3 and F: 19 All parents	Triangulating mixed-method.	Semistructured interview/thematic analysis.

Edvardson & Ahlström (2008)[25]. Sweden.	Caregivers' experience.	25 low-grade. 2 high-grade. Stage of treatment not described.	The patients had participated in an earlier study.	N = 28 M: 8 and F: 20 15 partners, living together 3 partners, living apart 8 parents 1 sibling 1 child	Not described.	Semistructured interview/qualitative content analysis and quantitative analysis of how the topics were distributed among the participants.
Janda et al. (2006)[22]. Australia.	The need of support for brain tumor patients and their caregivers.	Different types * Treatment phase not described, but time since diagnosis stated: 1–2 years: 22 5 years: 5 More than 5 years: 11	Members of Queensland Cancer Fund's Brain Tumor Support Service.	N = 10 in focus group, n = 8 in semistructured interview M: 4 and F: 18 13 partners 5 children	Qualitative.	Focus group interview and semistructured interview/framework analysis.
Lipsman et al. (2007)[37]. Canada.	The experience of brain tumor patients and their caregivers, and how it affects the choice of treatment.	Malignant * Palliative phase.	Recruited by a neurosurgeon.	N = 22 Further participant information not described	Qualitative.	Semistructured interview/thematic analysis.
Lou et al. (2015).[34] Taiwan.	The experience and suffering of mothers waiting for their child to die from brain tumor.	Malignant * All patients deceased.	Not described.	N =10 F: 10 All mothers	Phenomenological.	In-depth interview/Colaizzi's analysis method.

Ownsworth et al. (2015).[35] Australia.	Caregivers' experience of support.	6 low-grade. 5 high-grade. All underwent surgery and radiation or chemotherapy. 9 months – 22 years since diagnosis.	Had participated in a different study.	N = 11 M: 6 and F: 5 8 spouses 3 parents	Phenomenological.	Semistructured interview/thematic analysis.
Piil et al. (2015).[21] Denmark.	Brain tumor patients' and their caregivers' experience, and their need for rehabilitation and support.	High-grade * The interviews conducted after: 1. Surgical diagnosis 2. Oncological treatment 3,4. Oncological treatment and scan showing treatment effect 5. After treatment	The University Hospital in Copenhagen.	N = 33 M: 10 and F: 23 23 spouses 2 girl/boyfriends 7 children 1 sister	Longitudinal and exploratory.	Semistructured interview/thematic analysis.
Russell et al. (2016)[10]. Canada.	The experience of children with a brain tumor and their caregivers.	Malignant * Diagnosed at least 3 months previously, stage of treatment not described.	Hospital in Toronto.	N = 12 Based on names: F: 11 stk., 1 stk. unknown All parents	Grounded theory.	Semistructured interview/comparative analysis.
Schmer et al. (2008)[31]. USA.	Caregivers' experience concerning care tasks after chemotherapy.	Malignant * During first 6 months of treatment.	The patients' treatment center.	N = 10 Sex unknown 7 spouses 2 daughters 1 son-in-law	Phenomenological.	Semistructured interview/Colaizzi's analysis method.

Schubart et al. (2008).[20] USA.	Caregivers' challenges and unmet needs.	Different types of brain cancer * 6 deceased 2 exacerbations 2 unstable 10 stable 1 terminal 3 recurrent 1 unclear	NeuroOncology Center.	N = 25 M: 7 and F: 18 18 spouses 4 parents 2 children 1 sibling	Grounded theory.	Semistructured interview/open coding and cross-case analysis.
Sherwood et al. (2011).[36] USA.	How caregivers adapt to their new role, and how this role changes during time.	Malignant * Interviewed 1 and 4 months after diagnosis.	A regional hospital.	N = 10 M: 2 and F: 8 5 spouses 2 parents 1 child 1 nephew 1 friend	Longitudinal descriptive design.	Semistructured interview/thematic content analysis.
Shortman et al. (2013). United Kingdom (UK).	Mothers of children with brain tumor—their experience and their coping mechanisms.	Different types and degrees *. All underwent surgery, five radiation, and four chemotherapy. 17–35 months since diagnosis.	Also participated in another study.	N = 6 F: 6 All mothers.	Not described.	Semistructured interview/thematic content analysis.

Strang & Strang (2001).[28] Sweden.	The degree to which patients with a brain tumor and their caregivers cope, understand, and create meaning in the situation.	Malignant tumors, grade 2–4. Treatment stage not described.	Not described.	N = 16 Further participant information not described.	Hermeneutic phenomenological.	Semistructured interview/structural analysis based on hermeneutic circle described by Richoeur.
Tastan et al. (2011).[32] Turkey.	Caregivers' experience of postoperative phase and homecare.	Different types and degrees * All patients had undergone surgery and postoperative treatment and were being treated at home.	A research and military training hospital in Turkey.	N = 19 M: 4 and F: 6 4 spouses 4 children 1 parent 1 sibling	Descriptive qualitative study.	Semistructured interview/Colaizzi's analysis method.

Huang et al. (2021).[27] Taiwan	The lived experience of parents having a child with a brain tumor during the shared decision-making process of treatment	4 medulloblastoma 3 germ cell tumor 1 glioblastoma 1 astrocytoma 1 ependymoma The interviews were conducted between 1-6 months after the child received the diagnosis	A pediatric oncology ward at a medical center in Taiwan	N=10 M: 3 and F: 7	Descriptive phenomenological study	Semistructured interview/Colaizzi's analysis method.
Wideheim et al. (2002).[26] Sweden.	The experience of a brain tumor from a family perspective.	High-grade glioma. The interviews were conducted 2–3 weeks, 3 months, and 6 months postoperatively.	Not described.	N = 5 Sex unknown 2 spouses 2 parents	Descriptive qualitative study.	Qualitative interviews/inductive content analysis.

				1 adult child		
Zelcer et al. (2010).[33] Canada.	The experience of brain tumor patients and caregivers in the palliative phase.	Malignant * All patients deceased.	Children's Hospital, London Health Sciences Centre.	N = 25 M: 9 and F: 16 All parents	Qualitative	Semistructured interview/thematic content analysis.

1 M = Male, F = Female ∗=Tumor not further described

PRISMA 2020 Checklist

Section and Topic	Item #	Checklist item	Location where item is reported
TITLE			
Title	1	Coping in the role as next of kin of a person with a brain tumor: a qualitative metasynthesis *The page number refers to the copy without track changes*	Page 1, line 1-2
ABSTRACT			
Abstract	2	See the PRISMA 2020 for Abstracts checklist.	Page 1-2, line 18-39
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of existing knowledge.	Page 4, line 78-84
Objectives	4	Provide an explicit statement of the objective(s) or question(s) the review addresses.	Page 4, line 85-87
METHODS			
Eligibility criteria	5	Specify the inclusion and exclusion criteria for the review and how studies were grouped for the syntheses.	Page 5, line 100-109
Information sources	6	Specify all databases, registers, websites, organisations, reference lists and other sources searched or consulted to identify studies. Specify the date when each source was last searched or consulted.	Page 5, line 100-102
Search strategy	7	Present the full search strategies for all databases, registers and websites, including any filters and limits used.	Supplementary material 1
Selection process	8	Specify the methods used to decide whether a study met the inclusion criteria of the review, including how many reviewers screened each record and each report retrieved, whether they worked independently, and if applicable, details of automation tools used in the process.	Page 6, line 111-115
Data collection process	9	Specify the methods used to collect data from reports, including how many reviewers collected data from each report, whether they worked independently, any processes for obtaining or confirming data from study investigators, and if applicable, details of automation tools used in the process.	Page 6, line 120-125
Data items	10a	List and define all outcomes for which data were sought. Specify whether all results that were compatible with each outcome domain in each study were sought (e.g. for all measures, time points, analyses), and if not, the methods used to decide which results to collect.	na
	10b	List and define all other variables for which data were sought (e.g. participant and intervention characteristics, funding sources). Describe any assumptions made about any missing or unclear information.	na
Study risk of bias assessment	11	Specify the methods used to assess risk of bias in the included studies, including details of the tool(s) used, how many reviewers assessed each study and whether they worked independently, and if applicable, details of automation tools used in the process.	Page 6, line 128-132
Effect measures	12	Specify for each outcome the effect measure(s) (e.g. risk ratio, mean difference) used in the synthesis or presentation of results.	na
Synthesis methods	13a	Describe the processes used to decide which studies were eligible for each synthesis (e.g. tabulating the study intervention characteristics and comparing against the planned groups for each synthesis (item #5)).	Table 2
	13b	Describe any methods required to prepare the data for presentation or synthesis, such as handling of missing summary statistics, or data conversions.	Table 1 and 2
	13c	Describe any methods used to tabulate or visually display results of individual studies and syntheses.	Table 1 and 2

PRISMA 2020 Checklist

Section and Topic	Item #	Checklist item	Location where item is reported
	13d	Describe any methods used to synthesize results and provide a rationale for the choice(s). If meta-analysis was performed, describe the model(s), method(s) to identify the presence and extent of statistical heterogeneity, and software package(s) used.	Page 7-8
	13e	Describe any methods used to explore possible causes of heterogeneity among study results (e.g. subgroup analysis, meta-regression).	na
	13f	Describe any sensitivity analyses conducted to assess robustness of the synthesized results.	na
Reporting bias assessment	14	Describe any methods used to assess risk of bias due to missing results in a synthesis (arising from reporting biases).	na
Certainty assessment	15	Describe any methods used to assess certainty (or confidence) in the body of evidence for an outcome.	na
RESULTS			
Study selection	16a	Describe the results of the search and selection process, from the number of records identified in the search to the number of studies included in the review, ideally using a flow diagram.	Fig 1
	16b	Cite studies that might appear to meet the inclusion criteria, but which were excluded, and explain why they were excluded.	na
Study characteristics	17	Cite each included study and present its characteristics.	Supplementary materials 2 and page 8, line 177-184
Risk of bias in studies	18	Present assessments of risk of bias for each included study.	na
Results of individual studies	19	For all outcomes, present, for each study: (a) summary statistics for each group (where appropriate) and (b) an effect estimate and its precision (e.g. confidence/credible interval), ideally using structured tables or plots.	na
Results of syntheses	20a	For each synthesis, briefly summarise the characteristics and risk of bias among contributing studies.	na
	20b	Present results of all statistical syntheses conducted. If meta-analysis was done, present for each the summary estimate and its precision (e.g. confidence/credible interval) and measures of statistical heterogeneity. If comparing groups, describe the direction of the effect.	na
	20c	Present results of all investigations of possible causes of heterogeneity among study results.	na
	20d	Present results of all sensitivity analyses conducted to assess the robustness of the synthesized results.	na
Reporting biases	21	Present assessments of risk of bias due to missing results (arising from reporting biases) for each synthesis assessed.	na
Certainty of evidence	22	Present assessments of certainty (or confidence) in the body of evidence for each outcome assessed.	na
DISCUSSION			
Discussion	23a	Provide a general interpretation of the results in the context of other evidence.	Page 15-17, line 302-353
	23b	Discuss any limitations of the evidence included in the review.	Page 17-18, line 353-373
	23c	Discuss any limitations of the review processes used.	Page 17-18, line 353-373
	23d	Discuss implications of the results for practice, policy, and future research.	Page 18, line 376-383

PRISMA 2020 Checklist

Section and Topic	Item #	Checklist item	Location where item is reported
OTHER INFORMATION			
Registration and protocol	24a	Provide registration information for the review, including register name and registration number, or state that the review was not registered.	na
	24b	Indicate where the review protocol can be accessed, or state that a protocol was not prepared.	na
	24c	Describe and explain any amendments to information provided at registration or in the protocol.	na
Support	25	Describe sources of financial or non-financial support for the review, and the role of the funders or sponsors in the review.	na
Competing interests	26	Declare any competing interests of review authors.	na
Availability of data, code and other materials	27	Report which of the following are publicly available and where they can be found: template data collection forms; data extracted from included studies; data used for all analyses; analytic code; any other materials used in the review.	na

From: Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. BMJ 2021;372:n71. doi: 10.1136/bmj.n71

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