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

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14

15 **Word Count:** 4014

16

17 **ABSTRACT**

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

22 **Methods:** A qualitative metasynthesis guided by Sandelowski and Barroso's guidelines
23 was used. The databases Medline, CHINAL, and PsycINFO were searched for studies
24 from January 2000 to March 3, 2021. The inclusion criteria were: qualitative primary

1
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3 25 studies focusing on factors or strategies used by the next of kin aged 18 years of age or
4
5 26 older of persons with brain tumors.
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7

8 27 **Results:** Of 1371 screened records, data from 19 studies, including 332 participants (200
9
10 28 female, 78 male, and 54 unclassified) were analyzed into metasummaries and a
11
12 29 metasynthesis. The next of kin rely on coping factors such as their personal characteristics,
13
14 30 finding meaning in their situation, external support, hope and religion, and having someone
15
16 31 to talk to. Strategies to manage the situation involve regaining control, being proactive, and
17
18 32 acceptance.
19
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21

22 33 **Conclusion:** Coping factors and strategies within themselves, in their surroundings and
23
24 34 assistance from a higher power are used by those who are next of kin for people with brain
25
26 35 tumors. It is important that health-care professionals suggest and facilitate these coping
27
28 36 factors and strategies because this could reduce stress and make the role of next of kin
29
30 37 more manageable.
31
32
33

34 38 **Keywords:** brain tumor; coping factors; coping strategies; metasynthesis; next of kin;
35
36 39 review
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40

41 **Strengths and limitations of the study**

- 42
- 43 • The qualitative approach makes an important contribution to the research field by
44 providing a deeper understanding of coping factors and strategies used by the next
45 of kin of a person with a brain tumor.
 - 46 • Most of the included studies in this metasynthesis were high-quality studies.
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3 46 • Our sample is highly multicultural with different geographical origins represented
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5 47 and includes different welfare and health-care systems, and different cultures and
6
7 48 religions.
8
9
10 49 • A limitation is that some of the subthemes, or parts of their content, could have
11
12 50 been categorized in the other main theme.
13
14
15 51 • The majority of the sample comprised women. A more heterogeneous sample
16
17 52 might have revealed more nuanced findings of the role of next of kin.
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20 53

54 INTRODUCTION

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

1
2
3 68 information from health-care providers, family, friends, and the community in their
4
5 69 struggle to cope with everyday life.^{10 11}
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7
8 70 Despite the severe challenges this disease imposes on next of kin, there are only a few
9
10 71 primary qualitative studies that have investigated the coping factors that make everyday
11
12 72 life more manageable or which strategies next of kin use to cope with their new role and
13
14 73 tasks. To our knowledge, this research has not been synthesized. Such information is of
15
16 74 great importance, especially for health-care providers working with this group of
17
18 75 caregivers. With improved understanding, they could expect to be better able to facilitate
19
20 76 more manageable everyday life among the next of kin.¹² There is some quantitative
21
22 77 research directed at these aspects, but we wanted studies that were more complementary
23
24 78 and personal, hence the choice of qualitative studies. Therefore, the purpose of this
25
26 79 metasynthesis was to identify and synthesize evidence from primary qualitative studies
27
28 80 regarding the experience of next of kin with coping factors and strategies in their role as
29
30 81 next of kin for a person with a brain tumor. The findings are discussed in the context of
31
32 82 Lazarus and Folkman's stress theory¹³ and the approach to coping with stress to interpret
33
34 83 our findings in a theoretical context.
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41 **METHODS**

42 *Design*

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47 86 The study was a metasynthesis within the interpretative paradigm. It was inspired by a
48
49 87 phenomenological–hermeneutic design because the aim was to identify and synthesize
50
51 88 qualitative primary studies that explored next of kin attitudes and experiences.¹⁴ The
52
53 89 metasynthesis process consisted of five steps: (1) formulating the purpose and rationale of
54
55 90 the study; (2) searching for and retrieving relevant qualitative research studies; (3)
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3 91 critically appraising the included studies; (4) classifying the findings, and finally; (5)
4
5 92 synthesizing the findings.
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8 93 ***Search strategy***
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11 94 In collaboration with an experienced librarian, we conducted a systematic search within the
12
13 95 PsycINFO, OVID, CHINAL, and Medline databases via EBSCO host up from January
14
15 96 2000 until March 3, 2021.
16
17

18 97 To search the PsycINFO database, we used the following terms: *((qualitative adj2*
19
20 98 *(research* or design* or stud* or method*)) or hermeneutic* or “grounded theory” or*
21
22 99 *“meta synthes*” or metasynthesis* or metaethnograph* or interview* or phenomenolog**
23
24 100 *or thematic or themes or experience*).ti,ab,hw,id. or exp qualitative methods or*
25
26 101 *phenomenology AND (caregiver* or famil* or next of kin* or relatives or spous* or wife*
27
28 102 *or husband* or sibling* or sister* or brother* or dependent* or loved one* or parent* or*
29
30 103 *mother* or father* or carer* or care giver*).ti,ab,hw,id. AND glioma*.ti,ab,hw,id. OR*
31
32 104 *(brain adj2 (cancer or neoplasm* or tumor*).ti,ab,hw,id.*
33
34
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36

37 105 In Medline and CHINAL, we used the following terms: *caregiver* OR famil* OR “next of*
38
39 106 *kin*” OR relatives OR spous* OR wife OR husband* OR sibling* OR brother* OR sister**
40
41 107 *OR dependent* OR “loved one*” OR parent* OR mother* OR father* OR carer* OR*
42
43 108 *“care giver*” AND (MH “Qualitative Studies+”) OR (MH “Qualitative Research+”) OR*
44
45 109 *(MH “Grounded Theory”) OR Interview* OR experienc* OR phenomenolog* OR*
46
47 110 *(qualitative W1 (research* OR method* OR design* OR stud*)) OR themes OR thematic*
48
49 111 *OR “audio recording” OR audiorecording OR metasynthes* OR “meta synthes*” OR*
50
51 112 *metaetnograph* AND (MH “Glioma+”) OR glioma OR gliomas OR glioblastom* OR*
52
53 113 *brain W1 (cancer OR tumor* or neoplasm*).*
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3 114 The inclusion criteria were qualitative primary studies in English or a Nordic language that
4
5 115 aimed to explore the factors or strategies used by the next of kin of persons with brain
6
7 116 tumors, regardless of tumor type and stage that enhanced their role as next of kin. The next
8
9 117 of kin had to be 18 years of age or older. The exclusion criteria were studies that did not
10
11 118 clearly identify coping factors or strategies, factors or strategies that included the
12
13 119 participants' experiences in the role of bereaved and not next of kin, and studies including
14
15 120 diagnoses other than a brain tumor.

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19
20 121 ***Search outcome***

21
22 122 The search strategy generated 1 371 unique citations. Titles and abstracts were screened by
23
24 123 the authors using Rayyan.¹⁵ Sixty-six papers were read in full and evaluated against the
25
26 124 inclusion criteria by both authors; 19 of these were included in the metasynthesis. Figure 1
27
28 125 shows the Preferred Reporting Items for Systematic Reviews and Meta-Analyses
29
30 126 (PRISMA) flowchart with a full overview of the screening process. The search output is
31
32 127 presented in the PRISMA flowchart. A final consensus regarding the eligible articles was
33
34 128 obtained through a group discussion between the authors. The authors read the full text of
35
36 129 the eligible articles and independently extracted data from the included studies; this
37
38 130 process is illustrated in Figure 1. Consensus for data extraction was obtained as part of a
39
40 131 group discussion between the authors. Table 1 lists the title, author(s), study country, year
41
42 132 of publication, aim, analysis, and study participants of all included studies. Most studies
43
44 133 were from Europe: Sweden (3), Great Britain (3), Denmark (1), Belgium (1), and Turkey
45
46 134 (1); seven were from Canada (3) and the USA (4), two were from Australia, and one was
47
48 135 from Taiwan. The tumor type and stage varied. For details, see Table 1.

49
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54
55 136 Figure 1 about here
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2
3 137 Table 1 about here
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6 138 ***Quality appraisal***
7

8
9 139 The quality of the 19 papers was evaluated using the Critical Appraisal Skills Program
10
11 140 (CASP) for qualitative studies. The first evaluation was conducted blinded and
12
13 141 independently by AW and GR, whose CASP evaluations were then compared. Using the
14
15 142 criteria in CASP for independent assessment, the authors mutually agreed on a final quality
16
17 143 evaluation. For details, see Table 2.
18
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20
21 144 The included studies appraised according to CASP are listed in Table 2. All studies had
22
23 145 clearly stated the study aim and the qualitative methodologies were considered appropriate.
24
25 146 Furthermore, several of the studies had been published in highly ranked journals. The most
26
27 147 poorly addressed issue was the influence of the researcher on the research and vice versa.
28
29

30 148 Table 2 about here
31
32

33 149 ***Data abstraction and analyses***
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35
36 150 As suggested by Sandelowski and Barroso,¹⁴ two approaches to qualitative synthesis were
37
38 151 used. The first of these involved qualitative metasummaries of qualitative findings from
39
40 152 the primary studies. This method is defined as qualitative, but the findings are presented
41
42 153 quantitatively. The second involved a metasynthesis that developed new interpretations of
43
44 154 the target findings from the primary studies.¹⁴ The narrative analysis was inspired by
45
46 155 Lindseth and Nordberg's phenomenological–hermeneutic methods.¹⁶ Three steps were
47
48 156 followed. First, the empirical materials were read several times. Second, after extraction,
49
50 157 the target findings were imported into NVivo 11 data management software for further
51
52 158 analysis.¹⁷ The text was read line-by-line to identify meaning units, subthemes, and
53
54 159 themes. Third, the researchers aimed to achieve a comprehensive understanding of the
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3 160 empirical materials, meaning units, and themes, and to relate these to the aim and research
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5 161 question of the metasynthesis.¹⁶ The analytic themes were identified by AW and discussed
6
7 162 with GR. The process of deriving the themes was inductive. The contribution of targeted
8
9 163 findings from each of the included papers is outlined, and quotations are used to illustrate
10
11 164 and support the findings, which increases the trustworthiness of the study. To validate the
12
13 165 findings, both authors participated in discussions of the empirical analysis and in writing
14
15 166 up the findings. Ethical approval was not required for the study.
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19

20 167 Table 3 about here
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22

23 168 *Synthesis*

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25
26 169 Qualitative metasynthesis provides novel interpretations of the target findings from
27
28 170 primary studies.¹⁴ The two main themes of this metasynthesis were coping factors and
29
30 171 coping strategies. The theme “coping factors” consisted of the subthemes, personal
31
32 172 characteristics, meaningful external support and having someone to talk to, and hope and
33
34 173 religion. The theme “coping strategies” consisted of the subthemes regain control, fight
35
36 174 against, and acceptance. For a list of the studies that generated findings for the main
37
38 175 themes and subthemes, see Table 3.
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43 176 **PATIENT AND PUBLIC INVOLVEMENT**

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47 177 This systematic review is based on published primary studies and do not involve public
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49 178 involvement.
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179 RESULTS

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

182 *Metasummaries*

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

191 *Metasynthesis*

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

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

198 The role as next of kin was considered to be complete and important.^{21 23 25 28}

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

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2
3 200 Engagement and commitment in the care of their relatives were highlighted by many next
4
5 201 of kin, especially when the patients appreciated the help.²¹ The engagement was even
6
7 202 stronger when the emotional bond between patient and next of kin was strong.^{18 19 26 32}
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9
10 203 However, other studies revealed less engagement and commitment, and underlined anger
11
12 204 and reluctance with the new role and the heavy responsibilities and sacrifices of the next of
13
14 205 kin that impacted their own needs and wishes.^{19 20 23 28 30}
16
17 206 External support made the role of next of kin easier to cope with. The support was given
18
19 207 by family, friends, neighbors, colleagues and workplaces, health personnel, schools, the
20
21 208 religious community, people in the local community, and even strangers.^{3 8 18-32} The
22
23 209 support from health-care professionals was especially important. This support included
24
25 210 emotional support and assistance during patient care and treatment.^{3 8 18-24 26-32} The
26
27 211 importance of assistance such as medical supervision and nursing care was emphasized,^{8 20}
28
29 212 ²⁶ with next of kin noting that this made it possible to feel like a partner again,²¹ while
30
31 213 concurrently allowing anticipated time alone.²² A well-known health-care professional was
32
33 214 crucial in making this possible, because it implied that the patient received the best care as
34
35 215 they were known to the health-care professional, and also because the assistance was
36
37 216 considered to be less intrusive.^{21 22} To experience the assistance with care as a coping
38
39 217 factor, it was crucial that care be compassionate and of the best quality. These qualities
40
41 218 emphasized the health professional's genuine care and gave the next of kin hope and desire
42
43 219 to fight the disease.^{8 19 21 24 26}
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49

50 220 *“She (neurosurgeon) had to give us some bad news some of the time...*
51
52 221 *and you couldn't ask for a better manner in her delivery of that bad*
53
54 222 *news, or her support in what we were going through.”³² (p. 8)*
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3 223 When next of kin experienced that their loved ones received a low quality of care or
4
5 224 suffered malpractice, it implied mistrust of the health-care system and weakened the
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8 225 experience of health-care professionals as a support factor.^{8 18 21 22} Emotional support from
9
10 226 health-care professionals implied an acknowledgment that the disease affected not only the
11
12 227 patients, but also their next of kin. It also implied that the health-care professionals
13
14 228 recognized and met the wishes of the next of kin for active participation in monitoring the
15
16 229 patient's disease course.^{21 23 24 31} Next of kin who did not have such involvement felt
17
18 230 ignored, useless, and helpless.^{23 26}
19
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21
22 231 Support from family and friends was invaluable in the care tasks and in coping with the
23
24 232 role of the next of kin.
25

26
27 233 *“Just support from family and friends, that was important to me, and just*
28
29 234 *knowing that I could call on them...”²⁰ (p. 1098)*
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31

32
33 235 Social, practical, and emotional support was emphasized, and included such things as
34
35 236 economic help, childcare, transport, and housekeeping.^{8 20 22 23 26-29 31 32} Some next of kin
36
37 237 would have appreciated even more support and help from family and friends, preferably
38
39 238 given on their own initiative.^{18 20 22 23 32 33}
40
41
42
43 239 Having someone to confide in and talk to were also important in coping with the role as
44
45 240 next of kin. Supportive conversations with health-care professionals were highly
46
47 241 appreciated by many next of kin. However, this required the health-care professional's
48
49 242 understanding and empathy for the situation of the patient as well as of their next of kin,
50
51 243 and preferably that they should be available at all times.^{19 21 24 27 28 34} Discussions with
52
53 244 family and friends were also important,^{19 22 23} and could even produce a stronger bond.²³
54
55 245 Such a bond required families and friends to understand and recognize the challenges faced
56
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1
2
3 246 by the next of kin.²² Support groups and conversations with other next of kin were also
4
5 247 highlighted. These conversations could be face-to-face or via the Internet.^{3 20 22 27 31-33}
6
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8
9 248 *“From time to time, I need to be able to talk to someone. Because when I*
10
11 249 *lay down in the evening, then it starts to work in the inside.”²¹ (p. 411)*
12
13

14 250 On the other hand, support groups were also considered demanding because it was difficult
15
16 251 to listen to other families’ stories. Furthermore, for some it was considered a waste of time
17
18 252 to spend valuable hours with people other than their closest family members.^{8 20 28}
19
20 253 Hope and religion were emphasized as important coping factors. The next of kin hoped
21
22 254 that a miraculous treatment would be developed so that their loved ones could survive the
23
24 255 disease or just have a better quality of life.^{3 8 18-21 24 30 31}
25
26
27
28

29 256 *You see a positive evolution, and everything that goes better is good for*
30
31 257 *her. (...) Nobody can forbid us to have hope. And miracles happen.*
32
33 258 *Whether we believe it or not, that’s not the point, it is the only thing to*
34
35 259 *focus on.”²¹ (p. 409)*
36
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38
39

40 260 Hope gave a reason to fight, although it weakened in the palliative phase.^{19 24 31} Faith
41
42 261 strengthened the hope of healing during the treatment period and gave some form of peace
43
44 262 in the final palliative phase. In most cases, hope was related to faith.^{23 24 27 31 34}
45
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48 263 *Main theme 2: Coping strategies – control and proactivity*
49

50
51 264 Regaining control of the situation was a frequent coping strategy and for most, this
52
53 265 included being provided with enough information to allow an overview of what to expect,
54
55 266 which implied some form of security.^{8 18-21 27 32 34}
56
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1
2
3 267 *“So it’s a, it’s a roller coaster of emotion but for the most part I’ve been,*
4
5 268 *‘What do we need to do? Where do we need to be?’ And then just read,*
6
7 269 *read, read whatever I can find out, whatever information because I feel*
8
9
10 270 *like whatever I know, I can ask for.”*²⁷ (p. 34)
11
12

13 271 The information provided should be adapted to the situation and the disease trajectory, and
14
15 272 preferably given by health-care professionals.^{18 20 21 23 26 34} The next of kin often hid this
16
17 273 information from the patients to protect them and not diminish their hope.^{8 24 27 28 31}
18
19 274 To regain control meant not only control of the diagnosis, but also personal control and
20
21 275 control over their own reactions. In some cases, the next of kin denied their feelings. Some
22
23 276 even denied the entire diagnosis,^{18 23 26 27} and instead focused on being strong for the
24
25 277 patient and the entire family.^{21 23 27 29-31 33} One next of kin in Edvardsson and Ahlstroms’
26
27
28 278 (2008) study²³ reported:
29
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31

32
33 279 *“I’ve sort of stowed it all away, I suppose. It is as if I’d experienced it*
34
35 280 *from the outside or seen it on TV. It’s often that way with sorrowful*
36
37 281 *things.”* (p. 588)
38
39
40

41 282 Being proactive and fighting the disease were also important coping strategies because
42
43 283 they were better than not doing anything.^{8 19 23 24 31}
44
45

46 284 *“People ask you how you cope. But what if you were to give up? You’ve*
47
48 285 *got to cope—and we do have each other! (...).”*²³ (p. 588)
49
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51
52 286 This implied adopting a healthier lifestyle, including changing diet and exercise habits,
53
54 287 hoping that this would improve the effects of medical treatment,^{19 24} or trying alternative
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3 288 treatments.^{8 31} However, an increasing feeling of powerlessness was emphasized if the
4
5 289 fight, in the form of these actions and treatments, did not meet the hope of a cure.^{19 21 24 31}
6
7
8 290 As the disease progressed and life went on, most next of kin accepted the diagnosis,
9
10 291 prognosis, and a new pattern to everyday life.^{8 24 25 27 31} There was a striving for normality,
11
12 292 starting with recommencing hobbies, work, and school for children.^{8 24 25 27 28 30 31 33} This
13
14
15 293 was particularly important within families with children. At the same time, accepting
16
17 294 disease progression or a bad diagnosis was most challenging when the patient was a
18
19 295 child.³¹
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21
22

23 296 **DISCUSSION**

24
25
26 297 This metasynthesis aimed to explore those factors and strategies that enhanced the ability
27
28 298 of next of kin to cope with their experience as the next of kin of a person with a brain
29
30 299 tumor. Valuable coping factors included personal characteristics, finding meaning in the
31
32 300 situation, external support, hope and religion, and having someone to talk to. Strategies to
33
34 301 manage the situation involved regaining control, being proactive, and acceptance.
35
36
37 302 We used Lazarus and Folkman's transactional stress theory published in 1984 in the
38
39 303 discussion of our findings. Lazarus and Folkman define coping as a cognitive and
40
41 304 behavioral endeavor under constant change, dealing with external and/or internal demands
42
43 305 that a cognitive assessment indicates are stressful or that exceed personal resources.^{13 35}
44
45
46 306 Being the next of kin to a person with a brain tumor is considered to be a negative stressor
47
48 307 because of the challenging life situation and care tasks. Nevertheless, several next of kin
49
50 308 included in the metasynthesis expressed a desire to fight the disease and to gain control
51
52 309 over the situation. This is described by Lazarus and Folkman¹³ as a secondary assessment
53
54 310 of the situation, in which the next of kin decide which measures to implement. One such
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3 311 measure could be to gain personal control—one of the most important and stress-reducing
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5 312 personal strategies available.¹³
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8 313 A possible explanation for the proactive attitude of next of kin toward the disease may be
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10 314 their obligation and commitment to the patient. Commitment is an expression of something
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12 315 of great importance and can cause one to be willing to meet threats and challenges that he
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14 316 or she would otherwise avoid.¹³ However, our findings revealed that the experience of
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16 317 contributing to something meaningful, not the obligation to do so, promoted coping in the
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18 318 situation. We consider that this is caused by the fact that obligation does not automatically
19
20 319 make an action meaningful, but rather that it can be experienced as a compulsion. This
21
22 320 assumption is strengthened by the findings that the tasks as next of kin could arouse
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24 321 emotions such as anger and aversion to the patient and to the diagnosis, rather than coping.
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26 322 Several studies refer to the same ambivalent experience regarding commitment and
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28 323 attitudes toward being a next of kin.^{36 37}
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34 324 External support was the factor that most relatives emphasized as promoting coping. It was
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36 325 described as invaluable, which was also confirmed in other studies,^{38 39} and in Lazarus and
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38 326 Folkman's transactional stress theory.¹³ At the same time, in both this metasynthesis and in
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40 327 other studies, next of kin voiced a strong desire and longing for even greater external
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42 328 support.^{38 39} The findings of the metasynthesis also showed that the configuration and
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44 329 arrangement of the support, especially that given by health-care providers is of great
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46 330 importance. This may indicate that health-care providers do not offer support appropriate
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48 331 to individual needs of the next of kin and for the care situations, possibly as a consequence
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50 332 of a lack of knowledge among health-care providers about how this affects the experience
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52 333 of being next of kin.
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3 334 The findings of this metasynthesis show that several next of kin considered hope to be an
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5 335 important coping factor, especially during the disease trajectory. Hope has also been
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7 336 shown to be an extensional coping factor in several studies,^{40 41} and transactional stress
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10 337 theory states that faith and hope are two of the most important personal factors in the
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12 338 cognitive assessment of stressors.^{13 35} Furthermore, according to Lazarus and Folkman,¹³
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14 339 the two factors are strongly related, which is consistent with the findings of our
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16 340 metasynthesis. For several next of kin, hope was strongly grounded in religion. This was
17
18 341 especially prominent in the studies conducted in the palliative phase, which indicated that
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20 342 faith is strengthened when there is no hope of curative treatment. The same pattern has also
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22 343 been reported in other studies describing cancer patients' experiences of palliative care.^{42 43}
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25 344 As the disease progressed, several next of kin accepted the diagnosis and its burden. Their
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27 345 fight against the disease diminished to some extent, and the relatives instead tried to
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29 346 "normalize" everyday life as much as possible. Similar acceptance is also reported by next
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31 347 of kin of other cancer patients, especially in the palliative phase.^{44 45} Lazarus and Folkman
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33 348 describe this as a reassessment, referring to a changed cognitive assessment of the stressor
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35 349 based on new information from the environment and/or the person.¹³
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41 **Strengths and limitations**

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45 351 A strength of this metasynthesis is that the primary search in the databases was conducted
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47 352 with the assistance of an experienced librarian, in an attempt to ensure that as many as
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49 353 possible of the relevant studies were included.⁴⁶ Furthermore, most of the included studies
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51 354 were of high methodological quality (see Table 2). Our sample was also highly
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53 355 multicultural (see Table 1). This attribute strengthens the validity of the metasynthesis
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55 356 since geographical origin could have affected the study sample because of different
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3 357 participant backgrounds related to different welfare and health-care systems, cultures,
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5 358 and/or religions.
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8 359 A limitation of our metasynthesis is that one of the 66 articles intended to be read in full
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10 360 text could not be obtained.⁴⁷ The formation of the subthemes is also a possible limitation.
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12 361 Some of the subthemes, or parts of their content, could have been categorized in the other
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15 362 main theme. Both main themes and subthemes overlap in several cases, and we have read
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17 363 similar studies^{24 27} where the findings are categorized differently than in metasynthesis. We
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19 364 chose to be true to the informants' statements and designated the location based on the
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21 365 informants' way of speaking and description of the experience. Another possible limitation
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23 366 is that our sample consisted mainly of women (see Table 1). A more heterogeneous sample
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25 367 might have revealed more nuanced findings and different experiences of the role of the
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27 368 next of kin.
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31 32 369 **CONCLUSION** 33 34

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36 370 The findings of this metasynthesis show that next of kin experience and use a range of
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38 371 coping factors and strategies in their role. Their experience is marked by individual
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40 372 differences. It is of great importance that health-care providers offer assistance that is
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42 373 individually adapted for these coping factors and strategies because this can reduce stress
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44 374 among the next of kin. The coping experience seems to go through phases, and further
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46 375 information is needed to understand fully how and when the various factors and strategies
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48 376 are used as the disease progresses. Longitudinal studies would therefore be of particular
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50 377 interest in this field.
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3 379 **Supplementary information**
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6 380 The manuscript has been edited by OnLine English (<https://www.oleng.com.au>) to comply
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8 381 with international publishing guidelines.
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17 384 search strategy.
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20
21 385 **Authors' contributions**
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24 386 AWL and GR designed the research project and developed the research plan. Librarian
25
26 387 Ellen Sejersted at the University of Agder and AWL were responsible for the literature
27
28 388 search, while AWL and GR were responsible for the analysis. Both authors were involved
29
30 389 in the screening and inclusion of the studies, reviewed the manuscript, and contributed to
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32 390 the revision of the paper. Both authors read and approved the final version of the paper.
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45 394 **Competing interests**
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49 395 The authors declare that they have no competing interests.
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52 396 **Patient consent for publication**
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55 397 Not required
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58 398 **Ethics approval**
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3 399 Ethical approval was not required, as no primary data were collected as part of this study.
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6 400 **Data availability**
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8 401 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.

1 2 3 4 5 6 7 8 9 10 11 12 13 14 15 16	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.
17 18 19 20 21 22 23 24 25	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.
26 27 28 29 30 31 32 33 34 35 36 37 38 39 40 41 42 43 44 45 46	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.

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

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.

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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 Ricoeur.
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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557 1 M = Male, F = Female

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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
Owensworth 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.

Author	Coping factors					Coping mechanisms		
	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
Owensworth 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

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PRISMA 2009 Flow Diagram

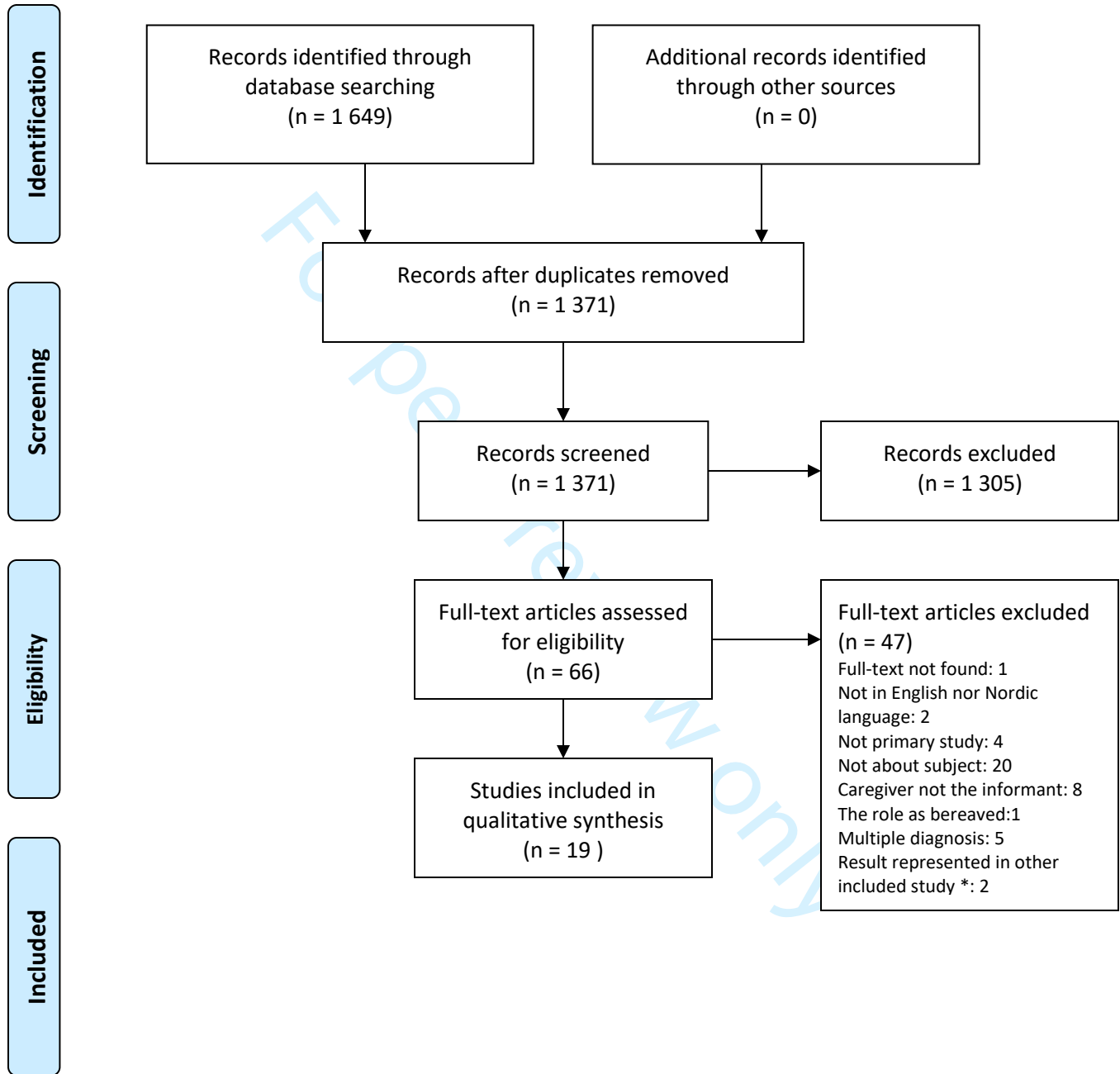


Figure 1. Flow chart of the inclusion process

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: 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 <i>incorporating a network meta-analysis (or related form of meta-analysis)</i> .	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 <i>synthesis methods, such as network meta-analysis</i> . Results: number of studies and participants identified; summary estimates with corresponding confidence/credible intervals; <i>treatment rankings may also be discussed. Authors may choose to summarize pairwise comparisons against a chosen treatment included in their analyses for brevity.</i> 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, <i>including mention of why a network meta-analysis has been conducted</i> .	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. <i>Clearly describe eligible treatments included in the treatment network, and note whether any have been clustered or merged into the same node (with justification)</i> .	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). <i>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.</i>	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: <ul style="list-style-type: none"> • <i>Handling of multi-arm trials;</i> • <i>Selection of variance structure;</i> • <i>Selection of prior distributions in Bayesian analyses; and</i> • <i>Assessment of model fit.</i> 	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: <ul style="list-style-type: none"> • Sensitivity or subgroup analyses; • Meta-regression analyses; • <i>Alternative formulations of the treatment network; and</i> • <i>Use of alternative prior distributions for Bayesian analyses (if applicable).</i> 	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. <i>Modified approaches may be needed to deal with information from larger networks.</i>	n.a
Synthesis of results	21	Present results of each meta-analysis done, including confidence/credible intervals. <i>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.</i> 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, <i>P</i> 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, <i>alternative network geometries studied, alternative choice of prior distributions for Bayesian analyses, and so forth</i>).	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). <i>Comment on the validity of the assumptions, such as transitivity and consistency.</i> Comment	16-17

1		<i>on any concerns regarding network geometry (e.g., avoidance</i>	
2		<i>of certain comparisons).</i>	
3			
4	Conclusions	26	Provide a general interpretation of the results in the context of
5			other evidence, and implications for future research.
6			
7	FUNDING		
8	Funding	27	Describe sources of funding for the systematic review and other
9			support (e.g., supply of data); role of funders for the systematic
10			review. This should also include information regarding whether
11			funding has been received from manufacturers of treatments in
12			the network and/or whether some of the authors are content
13			experts with professional conflicts of interest that could affect
14			use of treatments in the network.
15			
16			

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.

review only

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.

Review only

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.

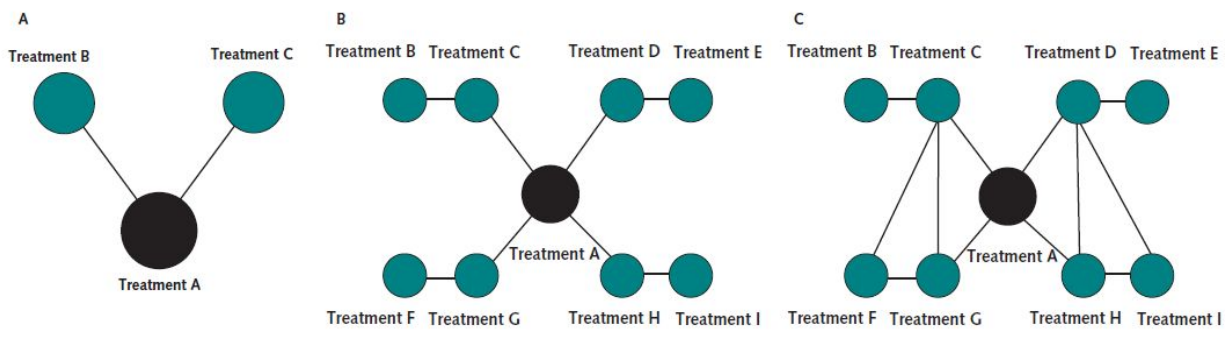
Robust reporting of rankings also includes specifying median ranks with uncertainty intervals, cumulative probability curves, and the surface under the cumulative ranking (SUCRA) curve.

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.

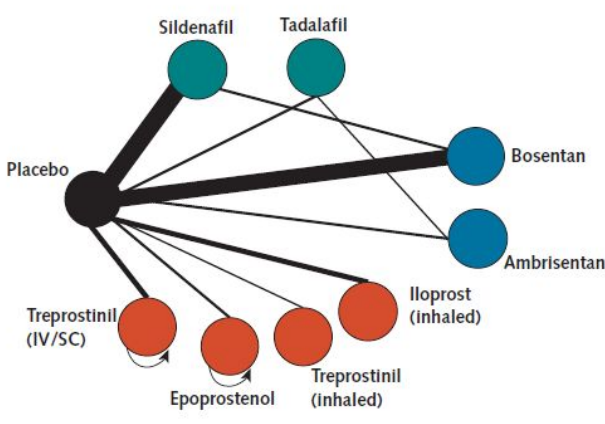
Rankings may exaggerate small differences in relative effects, especially if they are based on limited information. An objective assessment of the strength of information in the network and the magnitude of absolute benefits should accompany rankings to minimize potential biases.

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Appendix Figure 1A-1C

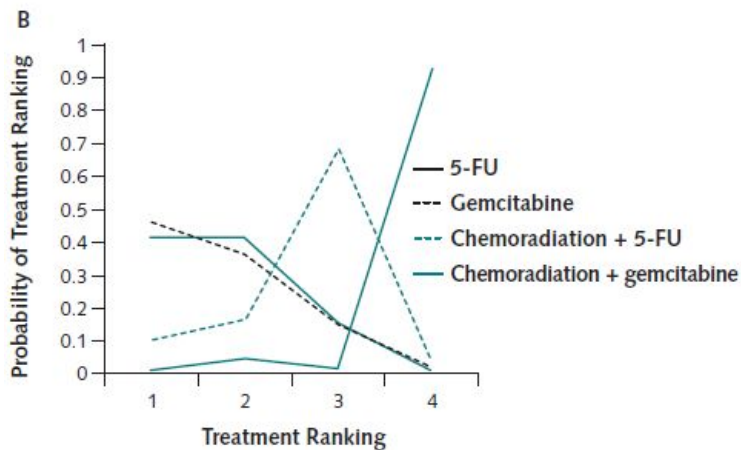


Appendix Figure 3



Appendix Figure 6

Ranking	Treatment and Coresponding 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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Primary Subject Heading:	Palliative care
Secondary Subject Heading:	Health policy
Keywords:	MEDICAL ETHICS, ONCOLOGY, Head & neck tumours < ONCOLOGY

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

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15 **Word Count:** 3981

16

17 **ABSTRACT**

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

22 **Method:** A qualitative metasynthesis guided by Sandelowski and Barroso's guidelines was
23 used. The databases Medline, CHINAL, and PsycINFO were searched for studies from
24 January 2000 to January 18 2022. . Inclusion criteria were qualitative original studies

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3 25 focusing on coping factors or strategies used by the next of kin of persons with brain
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5 26 tumors. The study participants had to be aged 18 years of age or older
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8 27 **Results:** Of 1 476 screened records, data from 20 studies, including 342 participants (207
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10 28 female, 81 male, and 54 unclassified) were analyzed into metasummaries and a
11
12 29 metasynthesis. The next of kin used coping factors such as their personal characteristics,
13
14 30 finding meaning in their situation, external support, hope and religion, and findingg
15
16 31 interlocutors. Coping strategies to manage the situation involved regaining control, being
17
18 32 proactive and acceptance.
19
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21
22 33 **Conclusion:** Next of kin of patients with brain tumor used coping factors and coping
23
24 34 strategies gathered within themselves, in their surroundings and with assistance from a
25
26 35 higher power to handle the situation and their role. It is important that health-care
27
28 36 professionals suggest and facilitate these coping factors and strategies because this could
29
30 37 reduce stress and make the role of next of kin more manageable.
31
32
33
34 38 **Keywords:** brain tumor; coping factors; coping strategies; metasynthesis; next of kin;
35
36 39 review; qualitative studies
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41 **Strengths and limitations of the study**

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

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

54 INTRODUCTION

55 In 2020 308,102 people worldwide with cancer in the central nervous system were
56 registered.¹ The diagnosis brain tumor is very confronting, with 56% of patients
57 experiencing one or more symptoms. Hemiparesis and cognitive challenges are most
58 frequently reported but also headache, nausea and vomiting, vision challenges, epileptic
59 seizures, and personality changes are considered common symptoms.²⁻⁵ Changes in
60 behavior and personality are considered particularly challenging, both for the patient and
61 for the next of kin, as this may include apathy, loss of initiative and empathy, indifference,
62 selfishness, physical and mental aggression, impaired emotional control and social
63 abilities, and tendencies toward childish behavior, among others.^{3 5 6} Studies show that the
64 disease can be more challenging and stressful for the next of kin than for the patients. The
65 next of kin have high rates of depression, anxiety, diverse physical pain, difficulty
66 adapting, loneliness, and high absence from work, as well as a reduced quality of life.⁷⁻¹¹
67 Studies also show that both patients and next of kin miss additional follow-up, support,

1
2
3 68 and information from health-care providers, family, friends, and the community in their
4
5 69 struggle to cope with everyday life.^{12 13}
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8 70 All these strains can lead to next of kin experiencing stress and lack of coping. Lazarus and
9
10 71 Folkman define coping as a cognitive and behavioral endeavor under constant change,
11
12 72 dealing with external and/or internal demands that a cognitive assessment indicates are
13
14 73 stressful or that exceed personal resources. When dealing with these demands, the next of
15
16 74 kin has to review available coping factors; personal, external and characteristics of the
17
18 75 situation itself.¹⁴ This review will determine whether the situation is perceived as
19
20 76 manageable or stressful, and secondly influence which coping strategy next of kin use.^{9 13}
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27 78 Despite this, there are only a few original qualitative studies that have investigated the
28
29 79 coping factors that make everyday life more manageable or which strategies next of kin
30
31 80 use to cope with their new role and tasks. To our knowledge, this research has not been
32
33 81 synthesized. Such information is of great importance, especially for health-care providers
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35 82 working with this group of caregivers. With improved understanding, they could expect to
36
37 83 be better able to facilitate more manageable everyday life among the next of kin. There is
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39 84 some quantitative research directed at these aspects,^{8-11 15} but we wanted studies that were
40
41 85 personal and focused on the lived experience of next of kin, hence the choice of qualitative
42
43 86 studies. Therefore, the purpose of this metasynthesis was to identify and synthesize
44
45 87 evidence from original qualitative studies regarding the experience of next of kin with
46
47 88 coping factors and strategies in their role as next of kin for a person with a brain tumor.
48
49 89 The findings are discussed in the context of Lazarus and Folkman's stress theory¹⁴ and the
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51 90 approach to coping with stress to interpret our findings in a theoretical context.
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91 **METHODS**

92 *Design*

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

100 *Search strategy*

101 In collaboration with an experienced librarian, we conducted a systematic search within the
102 PsycINFO, OVID, CHINAL, and Medline databases via EBSCO host up from January
103 2000 until 18 January 2022. For search strategy see supplementary materials 1.

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

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3 111 ***Search outcome***
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5
6 112 The search strategy generated 1 476 unique citations. Titles and abstracts were screened by
7
8 113 the authors using Rayyan, a systematic review management software.¹⁷ A final consensus
9
10 114 regarding the eligible articles was obtained through a group discussion between the
11
12 115 authors. Seventy-two papers were read in full and evaluated against the inclusion criteria
13
14 116 by both authors; 20 of these were included in the metasynthesis. Figure 1 shows the
15
16 117 Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA)
17
18 118 flowchart with a full overview of the screening process. The search output is presented in
19
20 119 the PRISMA flowchart. The authors read the full text of the eligible articles and
21
22 120 independently extracted data from the included studies; this process is illustrated in Figure
23
24 121 1. Consensus for data extraction was obtained as part of a group discussion between the
25
26 122 authors. Table 1 lists the title, author(s), study country, year of publication, aim, analysis,
27
28 123 and study participants of all included studies. Most studies were from Europe: Sweden (3),
29
30 124 Great Britain (3), Denmark (1), Belgium (1), and Turkey (1); seven were from Canada (3)
31
32 125 and the USA (4), two from Australia and two from Taiwan. The tumor type and stage
33
34 126 varied. For details, see supplementary materials 2.
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41 127 Figure 1 about here
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43
44 128 ***Quality appraisal***
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46 129 The quality of the 20 papers was evaluated using the Critical Appraisal Skills Program
47
48 130 (CASP) for qualitative studies. The first evaluation was conducted blinded and
49
50 131 independently by AWL and GR, whose CASP evaluations were then compared. Using the
51
52 132 criteria in CASP for independent assessment, the authors mutually agreed on a final quality
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54 133 evaluation. For details, see Table 1.
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3 134 The included studies appraised according to CASP are listed in Table 2. All studies had
4
5 135 clearly stated the study aim and the qualitative methodologies were considered appropriate.
6
7 136 Furthermore, several of the studies had been published in highly ranked journals. The most
8
9 137 poorly addressed issue (criteria number 6 in the CASP list) was the influence of the
10
11 138 researcher on the research and vice versa.
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15 139 Table 1 about here
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17 18 140 ***Data abstraction and analyses*** 19

20
21 141 As suggested by Sandelowski and Barroso,¹⁶ two approaches to qualitative synthesis were
22
23 142 used. The first of these involved qualitative metasummaries of qualitative findings from
24
25 143 the original studies. This method is defined as qualitative, but the findings are presented
26
27 144 quantitatively. The second involved a metasynthesis that developed new interpretations of
28
29 145 the target findings from the original studies.¹⁶ The narrative analysis was inspired by
30
31 146 Lindseth and Nordberg's phenomenological–hermeneutic methods.¹⁸ Three steps were
32
33 147 followed. First, the empirical materials were read several times. Second, after extraction,
34
35 148 the target findings were imported into NVivo 11 data management software for further
36
37 149 analysis.¹⁹ The text was read line-by-line to identify meaning units, subthemes, and
38
39 150 themes. Third, the researchers aimed to achieve a comprehensive understanding of the
40
41 151 empirical materials, meaning units, and themes, and to relate these to the aim and research
42
43 152 question of the metasynthesis.¹⁸ The analytic themes were identified by AWL and
44
45 153 discussed with GR. The process of deriving the themes was inductive. The contribution of
46
47 154 targeted findings from each of the included papers is outlined, and quotations are used to
48
49 155 illustrate and support the findings, which increases the trustworthiness of the study. To
50
51 156 validate the findings, both authors participated in discussions of the empirical analysis and
52
53 157 in writing up the findings. Ethical approval was not required for the study.
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3 158 Table 2 about here
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6 159 ***Synthesis***
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9 160 Qualitative metasynthesis provides novel interpretations of the target findings from
10
11 161 original studies.¹⁶ The two main themes of this metasynthesis were coping factors within
12
13 162 the next of kin and as external support and coping strategies – control and proactivity. The
14
15 163 first main theme, coping factors, consisted of the subthemes, personal characteristics,
16
17 164 meaningful, external support, having interlocutors, and hope and religion. The second main
18
19 165 theme, coping strategies, consisted of the subthemes regain control, fight against, and
20
21 166 acceptance. For a list of the studies that generated findings for the main themes and
22
23 167 subthemes, see Table 2.
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29 168 **PATIENT AND PUBLIC INVOLVEMENT**
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33 169 This systematic review is based on published original studies and does not involve public
34
35 170 involvement.
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39 171 **RESULTS**
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41

42 172 The findings are presented as metasummaries supported by tables and figures, and as a
43
44 173 metasynthesis containing seven subthemes presented under two main themes. Each
45
46 174 subtheme is supported by illustrative quotes from the original studies included.
47
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49

50 175 ***Metasummaries***
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52

53 176 The 20 included studies consisted of 342 participants (207 women, 81 men, and 54 not
54
55 177 classified). The focus was on the following themes: the needs of the next of kin;^{2 20-24} their
56
57 178 overall experiences as next of kin;^{10 25-27} coping and coping mechanisms;²⁸⁻³⁰ postoperative
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3 179 caregiving,^{31 32} being a next of kin in the palliative phase,^{33 34} support factors
4
5 180 experienced,³⁵ how the caregiving changed over time;³⁶ and factors influencing treatment
6
7 181 choice in the palliative phase.³⁷ Three of the studies were undertaken six months after
8
9 182 diagnosis,^{27 30 31 36} and three in the patients' palliative phase or postmortem.^{33 34 37} In six
10
11 183 studies, all the patients were children of the informants.^{10 29 30 33 34}
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14

15 184 *Metasynthesis*

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18 185 *Main theme 1: Coping factors within the next of kin and as external support*

19
20 186 *Personal characteristics* such as a strong and positive personality were important coping
21
22 187 factors for next of kin in new challenging situations.^{25 29 37} Being able to show empathy for
23
24 188 the patient and the health professionals were important, as if not the situation otherwise
25
26 189 easily could engender feelings such as discouragement and reproach.²⁵ A positive mood
27
28 190 and humor were also emphasized as for the same reasons.²⁹
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30
31

32 191 The role as next of kin was considered to be *meaningful* and important, as it made them
33
34 192 feel needed and productive in the situation.^{23 25 28 31} Engagement and commitment in the
35
36 193 care of their relatives were highlighted by many next of kin, especially when the patients
37
38 194 appreciated the help.²³ The engagement was even stronger when the emotional bond
39
40 195 between patient and next of kin was strong.^{20 21 29 35}
41
42
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44
45 196 *“But caring for him is something I will do—it is not a burden.”³¹ (p. 81)*

46
47
48 197 However, other studies revealed less engagement and commitment, and underlined anger
49
50 198 and reluctance with the new role as the heavy responsibility and sacrifice impacted the
51
52 199 next of kin's own needs and wishes.^{21 22 25 31 33}
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3 200 *External support* made the role of next of kin easier to cope with. The support was given
4
5 201 by family, friends, neighbors, colleagues and workplaces, health personnel, schools, the
6
7 202 religious community, people in the local community, and even strangers.^{2 10 20-35} The
8
9 203 support from health-care professionals was especially important. This support included
10
11 204 emotional support and assistance during patient care and treatment .^{2 10 20-27 29-35} The
12
13 205 importance of assistance such as medical supervision and nursing care was emphasized,¹⁰
14
15 206 ^{22 29} with next of kin noting that this made it possible to feel like a partner again,²³ while
16
17 207 concurrently allowing anticipated time alone.²⁴ A well-known health-care professional was
18
19 208 crucial in making this possible, because it implied that the patient received the best care as
20
21 209 they were known to the health-care professional, and also because the assistance was
22
23 210 considered to be less intrusive.^{23 24} To experience the assistance with care as a coping
24
25 211 factor, it was crucial that care be compassionate and of the best quality. These qualities
26
27 212 emphasized the health professional's genuine care and gave the patients and the next of kin
28
29 213 hope and desire to fight the disease.^{10 21 23 26 27 29}

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35
36 214 *“She (neurosurgeon) had to give us some bad news some of the time...*
37
38 215 *and you couldn't ask for a better manner in her delivery of that bad*
39
40 216 *news, or her support in what we were going through.”³⁵ (p. 8)*

41
42
43
44 217 When next of kin experienced that their loved ones received a low quality of care or
45
46 218 suffered malpractice, it implied mistrust of the health-care system and weakened the
47
48 219 experience of health-care professionals as a support factor.^{10 20 23 24} Emotional support from
49
50 220 health-care professionals implied an acknowledgment that the disease affected not only the
51
52 221 patients, but also their next of kin. It also implied that the health-care professionals
53
54 222 recognized and met the wishes of the next of kin for active participation in monitoring the
55
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3 223 patient's disease course.^{23 25 26 34} Next of kin who did not have such involvement felt
4
5 224 ignored, useless, and helpless.^{25 29}
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7

8 225 Support from family and friends was invaluable in the care tasks and in coping with the
9
10 226 role of the next of kin.
11

12
13
14 227 *“Just support from family and friends, that was important to me, and just*
15
16 228 *knowing that I could call on them... ”*²² (p. 1098)
17

18
19 229 Social, practical, and emotional support was emphasized, and included such things as
20
21 230 economic help, childcare, transport, and housekeeping.^{10 22 24 25 29-32 34 35} Some next of kin
22
23 231 would have appreciated even more support and help from family and friends, preferably
24
25 232 given on their own initiative.^{20 22 24 25 35 36}
26
27

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29 233 Having *interlocutors*, meaning having someone to confide in and talk to, were also
30
31 234 important in coping with the role as next of kin, as the situation, the responsibility and the
32
33 235 impressions were though. Supportive conversations with health-care professionals were
34
35 236 highly appreciated by many next of kin. However, this required the health-care
36
37 237 professional's understanding and empathy for the situation of the patient as well as of their
38
39 238 next of kin, and preferably that they should be available at all times.^{21 23 26 30 31 37}
40
41

42
43 239 Discussions with family and friends were also important,^{21 24 25 27} and could even produce a
44
45 240 stronger bond.²⁵ Such a bond required families and friends to understand and recognize the
46
47 241 challenges faced by the next of kin.²⁴ Support groups and conversations with other next of
48
49 242 kin were also highlighted,^{2 22 24 30 34 35 37} as it could broaden the next of kins understanding
50
51 243 of the tumor and what might to expect in the future.²⁷ These conversations could be face-
52
53 244 to-face or via the Internet.^{2 22 24 30 34 35 37}
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1
2
3 245 *“From time to time, I need to be able to talk to someone. Because when I*
4
5 246 *lay down in the evening, then it starts to work in the inside.”*²³ (p. 411)
6
7

8
9 247 On the other hand, support groups were also considered demanding because it was difficult
10
11 248 to listen to other families’ stories. Furthermore, for some it was considered a waste of time
12
13 249 to spend valuable hours with people other than their closest family members.^{10 22 31}
14
15

16 250 *Hope and religion* were emphasized as important coping factors. The next of kin hoped
17
18 251 that a miraculous treatment would be developed so that their loved ones could survive the
19
20 252 disease or just have a better quality of life.^{2 10 20-23 26 33 34}
21
22

23
24 253 *You see a positive evolution, and everything that goes better is good for*
25
26 254 *her. (...) Nobody can forbid us to have hope. And miracles happen.*
27
28 255 *Whether we believe it or not, that’s not the point, it is the only thing to*
29
30 256 *focus on.”*²³ (p. 409)
31
32

33
34 257 Hope gave a reason to fight, although it weakened in the palliative phase.^{21 26 34} Faith
35
36 258 strengthened the hope of healing during the treatment period and gave some form of peace
37
38 259 in the final palliative phase. In most cases, hope was related to faith.^{25-27 30 34 37}
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41
42 260 *Main theme 2: Coping strategies – control and proactivity*
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45
46 261 *Regaining control* of the situation was a frequent coping strategy, and for most this
47
48 262 included gathering enough information to allow an overview of what to expect, which
49
50 263 implied some form of security.^{10 20-23 27 30 35 37}
51
52

53
54 264 *“So it’s a, it’s a roller coaster of emotion but for the most part I’ve been,*
55
56 265 *‘What do we need to do? Where do we need to be?’ And then just read,*
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58

1
2
3 266 *read, read whatever I can find out, whatever information because I feel*
4
5 267 *like whatever I know, I can ask for.*³⁰ (p. 34)
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8
9 268 The information gathered and provided should preferably be adapted to the situation and
10
11 269 the disease trajectory, and been given by health-care professionals.^{20 22 23 25 27 29 37} The next
12
13 270 of kin often hid this information from the patients to protect them and not diminish their
14
15 271 hope.^{10 26 30 31 34}
16

17
18 272 To regain control meant not only control of the diagnosis, but also personal control and
19
20 273 control over their own reactions. In some cases, the next of kin denied their feelings. Some
21
22 274 even denied the entire diagnosis,^{20 25 29 30} and instead focused on being strong for the
23
24 275 patient and the entire family.^{23 25 30 32-34 36} One next of kin in Edvardsson and Ahlstroms'
25
26 276 (2008) study²⁵ reported:
27

28
29
30 277 *"I've sort of stowed it all away, I suppose. It is as if I'd experienced it from the*
31
32 278 *outside or seen it on TV. It's often that way with sorrowful things."* (p. 588)
33
34

35 279 *Being proactive, facilitate and encouraging the patient to fight the disease were also*
36
37 280 *important coping strategies, as it felt better than accepting the morbid situation and not do*
38
39 281 *anything.*^{10 21 25 26 34}
40
41

42
43 282 *"People ask you how you cope. But what if you were to give up? You've*
44
45 283 *got to cope—and we do have each other! (...)." ²⁵ (p. 588)*
46
47

48
49 284 This implied adopting a healthier lifestyle, including changing diet and exercise habits,
50
51 285 hoping that this would improve the effects of medical treatment,^{21 26} or trying alternative
52
53 286 treatments.^{10 34} However, an increasing feeling of powerlessness was emphasized if the
54
55 287 fight, in the form of these actions and treatments, did not meet the hope of a cure.^{21 23 26 34}
56
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3 288 As the disease progressed and life went on, most next of kin accepted the diagnosis,
4
5 289 prognosis, and a new pattern to everyday life.^{10 26 28 30 34} There was a striving for normality,
6
7 290 starting with recommencing hobbies, work, and school for children.^{10 26 28 30 31 33 34 36} This
8
9 291 was particularly important within families with children. At the same time, accepting
10
11 292 disease progression or a bad diagnosis was most challenging when the patient was a
12
13 293 child.³⁴
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18 294 **DISCUSSION**

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20
21 295 This metasynthesis aimed to explore those factors and strategies that enhanced the ability
22
23 296 of next of kin to cope with their experience as the next of kin of a person with a brain
24
25 297 tumor. Valuable coping factors included personal characteristics, finding meaning in the
26
27 298 situation, external support, hope and religion, and interlocutors. Strategies to manage the
28
29 299 situation involved regaining control, being proactive, and acceptance.^{14 38}
30
31
32
33 300 Being the next of kin to a person with a brain tumor is considered to be a negative stressor
34
35 301 because of the challenging life situation and care tasks. Nevertheless, several next of kin
36
37 302 included in the metasynthesis expressed a desire to fight the disease and to gain control
38
39 303 over the situation. This is described by Lazarus and Folkman¹⁴ as a secondary assessment
40
41 304 of the situation, in which the next of kin decide which measures to implement. One such
42
43 305 measure could be to gain personal control—one of the most important and stress-reducing
44
45 306 personal strategies available.¹⁴
46
47
48
49 307 A possible explanation for the proactive attitude of next of kin toward the disease may be
50
51 308 their obligation and commitment to the patient. Commitment is an expression of something
52
53 309 of great importance and can cause one to be willing to meet threats and challenges that he
54
55 310 or she would otherwise avoid.¹⁴ However, our findings revealed that the experience of
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1
2
3 311 contributing to something meaningful, not the obligation to do so, promoted coping in the
4
5 312 situation. We consider that this is caused by the fact that obligation does not automatically
6
7 313 make an action meaningful, but rather that it can be experienced as a compulsion. This
8
9 314 assumption is strengthened by the findings that the tasks as next of kin could arouse
10
11 315 emotions such as anger and aversion to the patient and to the diagnosis, rather than coping.
12
13 316 Several studies refer to the same ambivalent experience regarding commitment and
14
15 317 attitudes toward being a next of kin.^{39 40}
16
17 318 External support was the factor that most relatives emphasized as promoting coping. It was
18
19 319 described as invaluable, which was also confirmed in other studies,^{41 42} and in Lazarus and
20
21 320 Folkman's transactional stress theory.¹⁴ At the same time, in both this metasynthesis and in
22
23 321 other studies, next of kin voiced a strong desire and longing for even greater external
24
25 322 support.^{41 42} The findings of the metasynthesis also showed that the configuration and
26
27 323 arrangement of the support, especially that given by health-care providers is of great
28
29 324 importance. An explanation for the next of kin's experience of unmet needs might be lack
30
31 325 of knowledge among health-care providers about how to assist in due course. This may
32
33 326 indicate that in some cases health-care providers should pay more attention to offer support
34
35 327 in line with individual needs of the next of kin and for the care situations.
36
37 328 The findings of this metasynthesis show that several next of kin considered hope to be an
38
39 329 important coping factor, especially during the disease trajectory. Hope has also been
40
41 330 shown to be an extensional coping factor in several studies,^{43 44} and transactional stress
42
43 331 theory states that faith and hope are two of the most important personal factors in the
44
45 332 cognitive assessment of stressors.^{14 38} Furthermore, according to Lazarus and Folkman,¹⁴
46
47 333 the two factors are strongly related, which is consistent with the findings of our
48
49 334 metasynthesis. For several next of kin, hope was strongly grounded in religion. This was
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3 335 especially prominent in the studies conducted in the palliative phase, which indicated that
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5 336 faith is strengthened when there is no hope of curative treatment. The same pattern has also
6
7 337 been reported in other studies describing cancer patients' experiences of palliative care.^{45 46}
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10 338
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13 339 As the disease progressed, several next of kin accepted the diagnosis and its burden. Their
14
15 340 fight against the disease diminished to some extent, and the relatives instead tried to
16
17 341 "normalize" everyday life as much as possible. Similar acceptance is also reported by next
18
19 342 of kin of other cancer patients, especially in the palliative phase.^{47 48} Lazarus and Folkman
20
21 343 describe this as a reassessment, referring to a changed cognitive assessment of the stressor
22
23 344 based on new information from the environment and/or the person.¹⁴
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27

28 345 **Strengths and limitations**

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30
31 346 A strength of this metasynthesis is that the primary search in the databases was conducted
32
33 347 with the assistance of an experienced librarian, in an attempt to ensure that as many as
34
35 348 possible of the relevant studies were included.⁴⁹ Furthermore, most of the included studies
36
37 349 were of high methodological quality (see Table 2). Our sample was also highly
38
39 350 multicultural (see Table 1). This attribute strengthens the validity of the metasynthesis
40
41 351 since geographical origin could have affected the study sample because of different
42
43 352 participant backgrounds related to different welfare and health-care systems, cultures,
44
45 353 and/or religions.

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49
50 354 A limitation of our metasynthesis is that one of the 72 articles intended to be read in full
51
52 355 text could not be obtained.⁵⁰ The formation of the subthemes is also a possible limitation.
53
54 356 Some of the subthemes, or parts of their content, could have been categorized in the other
55
56 357 main theme. Both main themes and subthemes overlap in several cases, and we have read

1
2
3 358 similar studies^{26 30} where the findings are categorized differently than in our metasynthesis.
4
5 359 We chose to be true to the informants' statements and designated the location based on the
6
7 360 informants' way of speaking and description of the experience. Another possible limitation
8
9 361 is that our sample consisted mainly of women (see Table 1). A more heterogeneous sample
10
11 362 might have revealed more nuanced findings and different experiences of the role of the
12
13 363 next of kin.
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18 364 **CONCLUSION**

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21 365 The findings of this metasynthesis show that next of kin experience and use a range of
22
23 366 coping factors and strategies in their role. Their experience is marked by individual
24
25 367 differences. It is of great importance that health-care providers offer assistance that is
26
27 368 individually adapted for these coping factors and strategies because this can reduce stress
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29 369 among the next of kin. The coping experience seems to go through phases, and further
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31 370 information is needed to understand fully how and when the various factors and strategies
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33 371 are used as the disease progresses. Longitudinal studies would therefore be of particular
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35 372 interest in this field.
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44 374 **Supplementary information**

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47 375 The manuscript has been edited by OnLine English (<https://www.oleng.com.au>) to comply
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49 376 with international publishing guidelines.
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8 379 search strategy.
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12 380 **Authors' contributions**
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15 381 AWL and GR designed the research project and developed the research plan. Librarian
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17 382 Ellen Sejersted at the University of Agder and AWL were responsible for the literature
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37 389 **Competing interests**
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40 390 The authors declare that they have no competing interests.
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43 391 **Patient consent for publication**
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46 392 Not required
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49 393 **Ethics approval**
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52 394 Ethical approval was not required, as no primary data were collected as part of this study.
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55 395 **Data availability**
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58 396 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
Cuttilo 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
Owensworth 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

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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

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

Author	Coping factors					Coping mechanisms		
	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
Owensworth 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		

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3								
4	Strang & Strang (2001) ²⁸	V	V					V
5								
6	Tastan et al. (2011) ³²		V			V		
7								
8								
9	Wideheim et al. (2002) ²⁶		V	V	V		V	V
10								
11	Zelcer et al. (2010) ³³		V	V		V		V
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3 Figure legend: Figure 1. Flow chart of the inclusion process
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PRISMA 2009 Flow Diagram

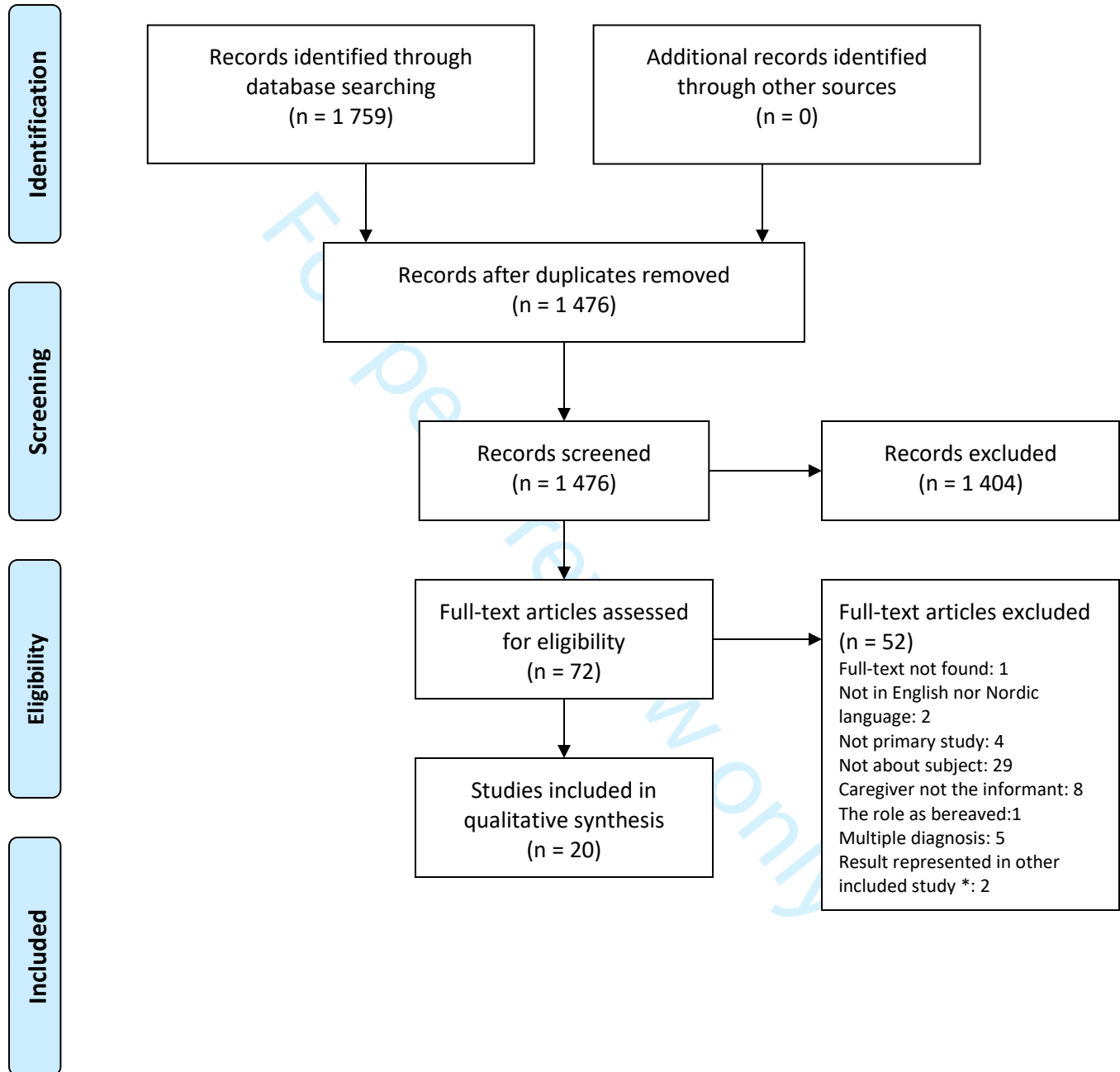


Figure 1. Flow chart of the inclusion process

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097

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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 WI (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 WI (cancer OR tumor* or neoplasm*).*

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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.
Cuttillo 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.

1 2 3 4 5 6 7 8 9 10 11 12 13 14	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.
15 16 17 18 19 20 21 22 23 24	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.
25 26 27 28 29 30 31	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.
32 33 34 35 36 37 38 39 40 41 42 43 44 45 46	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.

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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.

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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 Ricoeur.
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 <i>incorporating a network meta-analysis (or related form of meta-analysis)</i> .	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 <i>synthesis methods, such as network meta-analysis</i> . Results: number of studies and participants identified; summary estimates with corresponding confidence/credible intervals; <i>treatment rankings may also be discussed. Authors may choose to summarize pairwise comparisons against a chosen treatment included in their analyses for brevity.</i> 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, <i>including mention of why a network meta-analysis has been conducted</i> .	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. <i>Clearly describe eligible treatments included in the treatment network, and note whether any have been clustered or merged into the same node (with justification)</i> .	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). <i>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.</i>	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: <ul style="list-style-type: none"> • <i>Handling of multi-arm trials;</i> • <i>Selection of variance structure;</i> • <i>Selection of prior distributions in Bayesian analyses; and</i> • <i>Assessment of model fit.</i> 	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: <ul style="list-style-type: none"> • Sensitivity or subgroup analyses; • Meta-regression analyses; • <i>Alternative formulations of the treatment network; and</i> • <i>Use of alternative prior distributions for Bayesian analyses (if applicable).</i> 	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. <i>Modified approaches may be needed to deal with information from larger networks.</i>	n.a
Synthesis of results	21	Present results of each meta-analysis done, including confidence/credible intervals. <i>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.</i> 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, <i>P</i> 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, <i>alternative network geometries studied, alternative choice of prior distributions for Bayesian analyses, and so forth</i>).	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). <i>Comment on the validity of the assumptions, such as transitivity and consistency.</i> Comment	16-17

1		<i>on any concerns regarding network geometry (e.g., avoidance</i>	
2		<i>of certain comparisons).</i>	
3			
4	Conclusions	26	Provide a general interpretation of the results in the context of
5			other evidence, and implications for future research.
6			17
7	FUNDING		
8	Funding	27	Describe sources of funding for the systematic review and other
9			support (e.g., supply of data); role of funders for the systematic
10			review. This should also include information regarding whether
11			funding has been received from manufacturers of treatments in
12			the network and/or whether some of the authors are content
13			experts with professional conflicts of interest that could affect
14			use of treatments in the network.
15			<i>n.a</i>

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.

review only

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.

Review only

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.

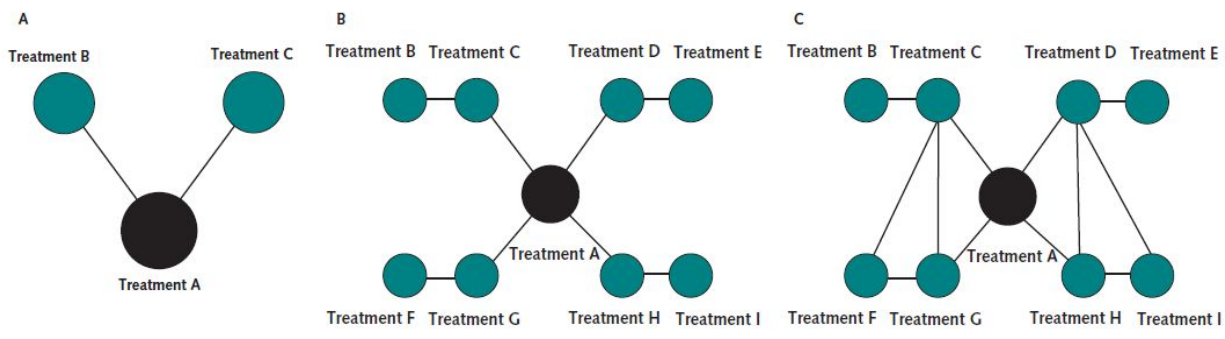
Robust reporting of rankings also includes specifying median ranks with uncertainty intervals, cumulative probability curves, and the surface under the cumulative ranking (SUCRA) curve.

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.

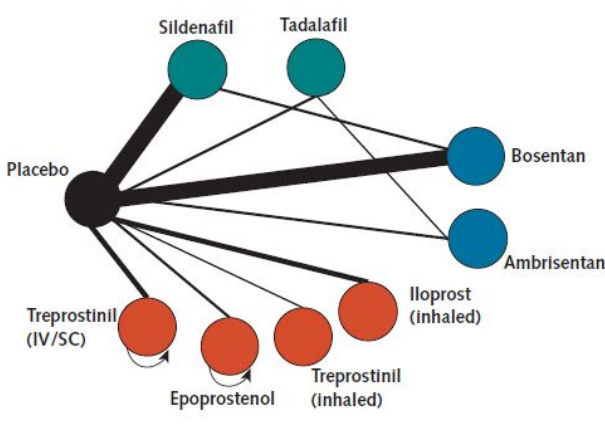
Rankings may exaggerate small differences in relative effects, especially if they are based on limited information. An objective assessment of the strength of information in the network and the magnitude of absolute benefits should accompany rankings to minimize potential biases.

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Appendix Figure 1A-1C

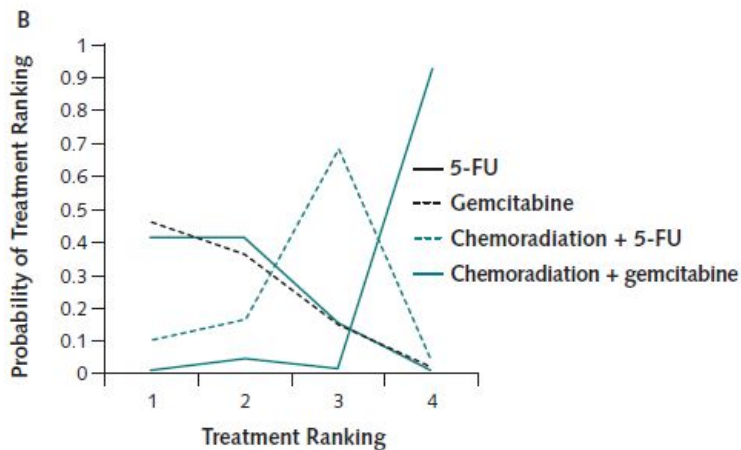


Appendix Figure 3



Appendix Figure 6

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

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Secondary Subject Heading:	Health policy
Keywords:	MEDICAL ETHICS, ONCOLOGY, Head & neck tumours < ONCOLOGY

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

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15
16 **Word Count:** 3981

17
18 **ABSTRACT**

19 **Objective:** Being the next of kin of a person with a brain tumor is a stressful experience.

20 For many, being a next of kin involves fear, insecurity, and overwhelming responsibility.

21 The purpose of this study was to identify and synthesize qualitative original studies that

22 explore coping in the role as next of kin of a person with a brain tumor.

23 **Methods:** A qualitative metasynthesis guided by Sandelowski and Barroso's guidelines

24 was used. The databases Medline, CHINAL, and PsycINFO were searched for studies

25 from January 2000 to January 18, 2022. Inclusion criteria were qualitative original studies

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3 26 that aimed to explore experienced coping by the next of kin of a person with brain tumor.
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5 27 The next of kin had to be 18 years of age or older.
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8 28 **Results:** Of 1 476 screened records, data from 20 studies, including 342 participants (207
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10 29 female, 81 male, and 54 unclassified) were analyzed into metasummaries and a
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12 30 metasynthesis. The metasynthesis revealed that the next of kin experiences of coping were
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14 31 characterized by two main themes; 1) Coping factors within the next of kin and as external
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16 32 support, such as their personal characteristics, finding meaning in their situation, external
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18 33 support, hope and religion, and finding interlocutors. 2) Coping strategies – control and
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20 34 proactivity, including regaining control, fight against, and acceptance
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25 35 **Conclusion:** Next of kin of patients with brain tumor used coping factors and coping
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27 36 strategies gathered within themselves and in their surroundings to handle the situation and
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29 37 their role. It is important that health-care professionals suggest and facilitate these coping
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31 38 factors and strategies because this could reduce stress and make the role of next of kin
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33 39 more manageable.
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37 40 **Keywords:** brain tumor; coping factors; coping strategies; metasynthesis; next of kin;
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39 41 review; qualitative studies
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43 **Strengths and limitations of the study**

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

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2
3 70 All these strains can lead to next of kin experiencing stress and lack of coping. Lazarus and
4
5 71 Folkman define coping as a cognitive and behavioral endeavor under constant change,
6
7 72 dealing with external and/or internal demands that a cognitive assessment indicates are
8
9 73 stressful or that exceed personal resources. When dealing with these demands, the next of
10
11 74 kin has to review available coping factors that could be able to making the situation more
12
13 75 manageable; personal, external and characteristics of the situation itself.¹⁴ This secondly
14
15 76 influence which coping strategy, meaning active actions, next of kin use for further coping
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17 77 in the situation.^{9 13 14}
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21
22 78 There are some original qualitative studies that have explored coping in the role as next of
23
24 79 kin of a person with a brain tumor. To our knowledge, this research has not been
25
26 80 synthesized. Such information is of great importance, especially for health-care providers
27
28 81 working with this group of caregivers. With improved understanding, they could expect to
29
30 82 be better able to facilitate more manageable everyday life among the next of kin. There is
31
32 83 also some quantitative research directed at these aspects,^{8-11 15} but we wanted studies that
33
34 84 were personal and focused on the lived experience of next of kin, hence the choice of
35
36 85 qualitative studies. Therefore, the purpose of this metasynthesis was to identify and
37
38 86 synthesize evidence from original qualitative studies regarding the experience of coping in
39
40 87 the role as next of kin of a person with brain tumor. The findings are discussed in the
41
42 88 context of Lazarus and Folkman's stress theory¹⁴ and the approach to coping with stress to
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44 89 interpret our findings in a theoretical context.
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90 **METHODS**

91 *Design*

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

99 *Search strategy*

100 In collaboration with an experienced librarian, we conducted a systematic search within the
101 PsycINFO, OVID, CHINAL, and Medline databases via EBSCO host up from January
102 2000 until 18 January 2022. For search strategy see supplementary materials 1.

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

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3 110 ***Search outcome***
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6 111 The search strategy generated 1 476 unique citations. Titles and abstracts were screened by
7
8 112 the authors using Rayyan, a systematic review management software.¹⁷ A final consensus
9
10 113 regarding the eligible articles was obtained through a group discussion between the
11
12 114 authors. Seventy-two papers were read in full and evaluated against the inclusion criteria
13
14 115 by both authors; 20 of these were included in the metasynthesis. Figure 1 shows the
15
16 116 Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA)
17
18 117 flowchart with a full overview of the screening process. The search output is presented in
19
20 118 the PRISMA flowchart. The authors read the full text of the eligible articles and
21
22 119 independently extracted data from the included studies; this process is also illustrated in
23
24 120 Figure 1. Consensus for data extraction was obtained as part of a group discussion between
25
26 121 the authors. Supplementary materials 2 lists the title, author(s), study country, year of
27
28 122 publication, aim, analysis, and study participants of all included studies. Most studies were
29
30 123 from Europe: Sweden (3), Great Britain (3), Denmark (1), Belgium (1), and Turkey (1);
31
32 124 seven were from Canada (3) and the USA (4), two from Australia and two from Taiwan.
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34 125 The tumor type and stage varied. For details, see supplementary materials 2.
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41 126 Figure 1 about here
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44 127 ***Quality appraisal***
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46 128 The quality of the 20 papers was evaluated using the Critical Appraisal Skills Program
47
48 129 (CASP) for qualitative studies. The first evaluation was conducted blinded and
49
50 130 independently by AWL and GR, whose CASP evaluations were then compared. Using the
51
52 131 criteria in CASP for independent assessment, the authors mutually agreed on a final quality
53
54 132 evaluation. For details, see Table 1.
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3 133 The included studies appraised according to CASP are listed in Table 2. All studies had
4
5 134 clearly stated the study aim and the qualitative methodologies were considered appropriate.
6
7 135 Furthermore, several of the studies had been published in highly ranked journals. The most
8
9 136 poorly addressed issue (criteria number 6 in the CASP list) was the influence of the
10
11 137 researcher on the research and vice versa.
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14
15 138 Table 1 about here
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17 18 139 ***Data abstraction and analyses*** 19

20
21 140 As suggested by Sandelowski and Barroso,¹⁶ two approaches to qualitative synthesis were
22
23 141 used. The first of these involved qualitative metasummaries of qualitative findings from
24
25 142 the original studies. This method is defined as qualitative, but the findings are presented
26
27 143 quantitatively. The second involved a metasynthesis that developed new interpretations of
28
29 144 the target findings from the original studies.¹⁶ The narrative analysis was inspired by
30
31 145 Lindseth and Nordberg's phenomenological–hermeneutic methods.¹⁸ Three steps were
32
33 146 followed. First, the empirical materials were read several times. Second, after extraction,
34
35 147 the target findings were imported into NVivo 11 data management software for further
36
37 148 analysis.¹⁹ The text was read line-by-line to identify meaning units, subthemes, and
38
39 149 themes. Third, the researchers aimed to achieve a comprehensive understanding of the
40
41 150 empirical materials, meaning units, and themes, and to relate these to the aim and research
42
43 151 question of the metasynthesis.¹⁸ The analytic themes were identified by AWL and
44
45 152 discussed with GR. The process of deriving the themes was inductive. The contribution of
46
47 153 targeted findings from each of the included papers is outlined, and quotations are used to
48
49 154 illustrate and support the findings, which increases the trustworthiness of the study. To
50
51 155 validate the findings, both authors participated in discussions of the empirical analysis and
52
53 156 in writing up the findings. Ethical approval was not required for the study.
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3 157 Table 2 about here
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6 158 Qualitative metasynthesis enables researchers to identify specific research questions,
7
8 159 search for, apprise, summarize and combine qualitative evidence to address the research
9
10 160 question. Metasynthesis provides novel interpretations of the target findings from original
11
12 161 studies. ¹⁶ In our methasynthesis we identified two main themes: 1) coping factors within
13
14 162 the next of kin and as external support and 2) coping strategies – tcontrol and proactivity,
15
16 163 each comprising 3-5 sub themes. For a list of the studies that generated findings for the
17
18 164 main themes and subthemes, see Table 2. When analyzing and organizing the results into
19
20 165 themes and subthemes we chose to be in line with the content and meaning of coping in the
21
22 166 original included studies, although some of the results could have been considered to also
23
24 167 contributed and organized differently. The results will be elaborated below.
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30 168 **PATIENT AND PUBLIC INVOLVEMENT**
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34 169 Patient or patient organization were not involved in the planning of the study, the analyses
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36 170 and writing of this metasynthesis which are based on published original studies and of
37
38 171 whom, some included patient involvement.
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172 RESULTS

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

176 *Metasummaries*

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

185 *Metasynthesis*

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

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

191 *Personal characteristics* such as a strong and positive personality were important coping
192 factors for next of kin in new challenging situations.^{25 29 37} Being able to show empathy for
193 the patient and the health professionals were important, as if not the situation otherwise

1
2
3 194 easily could engender feelings such as discouragement and reproach.²⁵ A positive mood
4
5 195 and humor were also emphasized as for the same reasons.²⁹
6
7

8 196 The role as next of kin was considered to be *meaningful* and important, as it made them
9
10 197 feel needed and productive in the situation.^{23 25 28 31} Engagement and commitment in the
11
12 198 care of their relatives were highlighted by many next of kin, especially when the patients
13
14 199 appreciated the help.²³ The engagement was even stronger when the emotional bond
15
16 200 between patient and next of kin was strong.^{20 21 29 35}
17
18
19

20
21 201 *“But caring for him is something I will do—it is not a burden.”*³¹ (p. 81)
22
23

24 202 However, other studies revealed less engagement and commitment, and underlined anger
25
26 203 and reluctance with the new role as the heavy responsibility and sacrifice impacted the
27
28 204 next of kin's own needs and wishes.^{21 22 25 31 33}
29
30

31 205 *External support* made the role of next of kin easier to cope with. The support was given
32
33 206 by family, friends, neighbors, colleagues and workplaces, health personnel, schools, the
34
35 207 religious community, people in the local community, and even strangers.^{2 10 20-35} The
36
37 208 support from health-care professionals was especially important. This support included
38
39 209 emotional support and assistance during patient care and treatment.^{2 10 20-27 29-35} The
40
41 210 importance of assistance such as medical supervision and nursing care was emphasized,¹⁰
42
43 211 ^{22 29} with next of kin noting that this made it possible to feel like a partner again,²³ while
44
45 212 concurrently allowing anticipated time alone.²⁴ A well-known health-care professional was
46
47 213 crucial in making this possible, because it implied that the patient received the best care as
48
49 214 they were known to the health-care professional, and also because the assistance was
50
51 215 considered to be less intrusive.^{23 24} To experience the assistance with care as a coping
52
53 216 factor, it was crucial that care be compassionate and of the best quality. These qualities
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3 217 emphasized the health professional's genuine care and gave the patients and the next of kin
4
5 218 hope and desire to fight the disease.^{10 21 23 26 27 29}

7
8
9 219 *"She (neurosurgeon) had to give us some bad news some of the time...*
10
11 220 *and you couldn't ask for a better manner in her delivery of that bad*
12
13 221 *news, or her support in what we were going through."*³⁵ (p. 8)

15
16 222 When next of kin experienced that their loved ones received a low quality of care or
17
18 223 suffered malpractice, it implied mistrust of the health-care system and weakened the
19
20 224 experience of health-care professionals as a support factor.^{10 20 23 24} Emotional support from
21
22 225 health-care professionals implied an acknowledgment that the disease affected not only the
23
24 226 patients, but also their next of kin. It also implied that the health-care professionals
25
26 227 recognized and met the wishes of the next of kin for active participation in monitoring the
27
28 228 patient's disease course.^{23 25 26 34} Next of kin who did not have such involvement felt
29
30 229 ignored, useless, and helpless.^{25 29}

31
32
33 230 Support from family and friends was invaluable in the care tasks and in coping with the
34
35 231 role of the next of kin.

36
37
38 232 *"Just support from family and friends, that was important to me, and just*
39
40 233 *knowing that I could call on them..."*²² (p. 1098)

41
42
43 234 Social, practical, and emotional support was emphasized, and included such things as
44
45 235 economic help, childcare, transport, and housekeeping.^{10 22 24 25 29-32 34 35} Some next of kin
46
47 236 would have appreciated even more support and help from family and friends, preferably
48
49 237 given on their own initiative.^{20 22 24 25 35 36}

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2
3 238 Having *interlocutors*, meaning having someone to confide in and talk to, were also
4
5 239 important in coping with the role as next of kin, as the situation, the responsibility and the
6
7 240 impressions were though. Supportive conversations with health-care professionals were
8
9 241 highly appreciated by many next of kin. However, this required the health-care
10
11 242 professional's understanding and empathy for the situation of the patient as well as of their
12
13 243 next of kin, and preferably that they should be available at all times.^{21 23 26 30 31 37}
14
15
16 244 Discussions with family and friends were also important,^{21 24 25 27} and could even produce a
17
18 245 stronger bond.²⁵ Such a bond required families and friends to understand and recognize the
19
20 246 challenges faced by the next of kin.²⁴ Support groups and conversations with other next of
21
22 247 kin were also highlighted,^{2 22 24 30 34 35 37} as it could broaden the next of kins understanding
23
24 248 of the tumor and what might to expect in the future.²⁷ These conversations could be face-
25
26 249 to-face or via the Internet.^{2 22 24 30 34 35 37}

30
31 250 *"From time to time, I need to be able to talk to someone. Because when I*
32
33 251 *lay down in the evening, then it starts to work in the inside."*²³ (p. 411)

34
35
36
37 252 On the other hand, support groups were also considered demanding because it was difficult
38
39 253 to listen to other families' stories. Furthermore, for some it was considered a waste of time
40
41 254 to spend valuable hours with people other than their closest family members.^{10 22 31}

42
43
44 255 *Hope and religion* were emphasized as important coping factors. The next of kin hoped
45
46 256 that a miraculous treatment would be developed so that their loved ones could survive the
47
48 257 disease or just have a better quality of life.^{2 10 20-23 26 33 34}

49
50
51
52 258 *You see a positive evolution, and everything that goes better is good for*
53
54 259 *her. (...) Nobody can forbid us to have hope. And miracles happen.*

1
2
3 260 *Whether we believe it or not, that's not the point, it is the only thing to*
4
5 261 *focus on.*"²³ (p. 409)
6
7

8
9 262 Hope gave a reason to fight, although it weakened in the palliative phase.^{21 26 34} Faith
10
11 263 strengthened the hope of healing during the treatment period and gave some form of peace
12
13 264 in the final palliative phase. In most cases, hope was related to faith.^{25-27 30 34 37}
14
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16
17 265 *Main theme 2: Coping strategies – control and proactivity*
18

19
20 266 Eighteen of the included studies provided data for the second main theme; *Coping*
21
22 267 *strategies – control and proactivity* (see table 2). This main theme comprised the three sub
23
24 268 themes: *regain control, fight against, and acceptance*.

25
26
27
28 269 *Regaining control* of the situation was a frequent coping strategy, and for most this
29
30 270 included gathering enough information to allow an overview of what to expect, which
31
32 271 implied some form of security.^{10 20-23 27 30 35 37}
33
34

35
36 272 *“So it's a, it's a roller coaster of emotion but for the most part I've been,*
37
38 273 *'What do we need to do? Where do we need to be?' And then just read,*
39
40 274 *read, read whatever I can find out, whatever information because I feel*
41
42 275 *like whatever I know, I can ask for.*"³⁰ (p. 34)
43
44
45

46 276 The information gathered and provided should preferably be adapted to the situation and
47
48 277 the disease trajectory, and been given by health-care professionals.^{20 22 23 25 27 29 37} The next
49
50 278 of kin often hid this information from the patients to protect them and not diminish their
51
52 279 hope.^{10 26 30 31 34}
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3 280 To regain control meant not only control of the diagnosis, but also personal control and
4
5 281 control over their own reactions. In some cases, the next of kin denied their feelings. Some
6
7 282 even denied the entire diagnosis,^{20 25 29 30} and instead focused on being strong for the
8
9
10 283 patient and the entire family.^{23 25 30 32-34 36} One next of kin in Edvardsson and Ahlstroms'
11
12 284 (2008) study²⁵ reported:

15 285 *"I've sort of stowed it all away, I suppose. It is as if I'd experienced it from the*
16
17 286 *outside or seen it on TV. It's often that way with sorrowful things."* (p. 588)

19
20 287 *Being proactive, facilitate and encouraging the patient to fight the disease were also*
21
22 288 *important coping strategies, as it felt better than accepting the morbid situation and not do*
23
24 289 *anything.*^{10 21 25 26 34}

26
27
28 290 *"People ask you how you cope. But what if you were to give up? You've*
29
30 291 *got to cope—and we do have each other! (...)."*²⁵ (p. 588)

31
32
33 292 This implied adopting a healthier lifestyle, including changing diet and exercise habits,
34
35 293 hoping that this would improve the effects of medical treatment,^{21 26} or trying alternative
36
37 294 treatments.^{10 34} However, an increasing feeling of powerlessness was emphasized if the
38
39 295 fight, in the form of these actions and treatments, did not meet the hope of a cure.^{21 23 26 34}

40
41
42 296 As the disease progressed and life went on, most next of kin accepted the diagnosis,
43
44 297 prognosis, and a new pattern to everyday life.^{10 26 28 30 34} There was a striving for normality,
45
46 298 starting with recommencing hobbies, work, and school for children.^{10 26 28 30 31 33 34 36} This
47
48 299 was particularly important within families with children. At the same time, accepting
49
50 300 disease progression or a bad diagnosis was most challenging when the patient was a
51
52 301 child.³⁴

302 **DISCUSSION**

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

309 Being the next of kin to a person with a brain tumor is considered to be a negative stressor
310 because of the challenging life situation and care tasks. Nevertheless, several next of kin
311 included in the metasynthesis expressed a desire to fight the disease and to gain control
312 over the situation. This is described by Lazarus and Folkman¹⁴ as a secondary assessment
313 of the situation, in which the next of kin decide which measures to implement. One such
314 measure could be to gain personal control—one of the most important and stress-reducing
315 personal strategies available.¹⁴

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

1
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3 325 Several studies refer to the same ambivalent experience regarding commitment and
4
5 326 attitudes toward being a next of kin.^{39 40}
6
7
8 327 External support was the factor that most relatives emphasized as promoting coping. It was
9
10 328 described as invaluable, which was also confirmed in other studies,^{41 42} and in Lazarus and
11
12 329 Folkman's transactional stress theory.¹⁴ At the same time, in both this metasynthesis and in
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14
15 330 other studies, next of kin voiced a strong desire and longing for even greater external
16
17 331 support.^{41 42} The findings of the metasynthesis also showed that the configuration and
18
19 332 arrangement of the support, especially that given by health-care providers is of great
20
21
22 333 importance. An explanation for the next of kin's experience of unmet needs might be lack
23
24 334 of knowledge among health-care providers about how to assist in due course. This may
25
26 335 indicate that in some cases health-care providers should pay more attention to offer support
27
28 336 in line with individual needs of the next of kin and for the care situations.
29
30
31 337 The findings of this metasynthesis show that several next of kin considered hope to be an
32
33 338 important coping factor, especially during the disease trajectory. Hope has also been
34
35 339 shown to be an extensional coping factor in several studies,^{43 44} and transactional stress
36
37 340 theory states that faith and hope are two of the most important personal factors in the
38
39 341 cognitive assessment of stressors.^{14 38} Furthermore, according to Lazarus and Folkman,¹⁴
40
41 342 the two factors are strongly related, which is consistent with the findings of our
42
43 343 metasynthesis. For several next of kin, hope was strongly grounded in religion. This was
44
45 344 especially prominent in the studies conducted in the palliative phase, which indicated that
46
47 345 faith is strengthened when there is no hope of curative treatment. The same pattern has also
48
49 346 been reported in other studies describing cancer patients' experiences of palliative care.^{45 46}
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3 348 As the disease progressed, several next of kin accepted the diagnosis and its burden. Their
4
5 349 fight against the disease diminished to some extent, and the relatives instead tried to
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7
8 350 “normalize” everyday life as much as possible. Similar acceptance is also reported by next
9
10 351 of kin of other cancer patients, especially in the palliative phase.^{47 48} Lazarus and Folkman
11
12 352 describe this as a reassessment, referring to a changed cognitive assessment of the stressor
13
14 353 based on new information from the environment and/or the person.¹⁴
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16
17

18 354 **Strengths and limitations**

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20
21 355 A strength of this metasynthesis is that the primary search in the databases was conducted
22
23 356 with the assistance of an experienced librarian, in an attempt to ensure that as many as
24
25 357 possible of the relevant studies were included.⁴⁹ Furthermore, most of the included studies
26
27 358 were of high methodological quality (see Table 2). Our sample was also highly
28
29 359 multicultural (see Table 1). This attribute strengthens the validity of the metasynthesis
30
31 360 since geographical origin could have affected the study sample because of different
32
33 361 participant backgrounds related to different welfare and health-care systems, cultures,
34
35 362 and/or religions.
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40 363 A limitation of our metasynthesis is that one of the 72 articles intended to be read in full
41
42 364 text could not be obtained.⁵⁰ The formation of the subthemes is also a possible limitation.
43
44 365 Some of the subthemes, or parts of their content, could have been categorized in the other
45
46 366 main theme. Both main themes and subthemes overlap in several cases, and we have read
47
48 367 similar studies^{26 30} where the findings are categorized differently than in our metasynthesis.
49
50 368 We chose to be true to the informants’ statements, the organization and meaning of the
51
52 369 original studies included, and designated the location based on the informants’ way of
53
54 370 speaking and description of the experience. Another possible limitation is that our sample
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3 371 consisted mainly of women (see supplementary materials 2). A more heterogeneous
4
5 372 sample might have revealed more nuanced findings and different experiences of the role of
6
7
8 373 the next of kin.
9

10 11 374 **CONCLUSION** 12 13

14 375 The findings of this metasynthesis show that next of kin experience and use a range of
15
16 376 coping factors and strategies in their role. Their experience is marked by individual
17
18 377 differences. It is of great importance that health-care providers offer assistance that is
19
20 378 individually adapted for these coping factors and strategies because this can reduce stress
21
22 379 among the next of kin. The coping experience seems to go through phases, and further
23
24 380 information is needed to understand fully how and when the various factors and strategies
25
26 381 are used as the disease progresses. Longitudinal studies would therefore be of particular
27
28 382 interest in this field.
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34 35 36 37 384 **Supplementary information** 38

39
40 385 The manuscript has been edited by OnLine English (<https://www.oleng.com.au>) to comply
41
42 386 with international publishing guidelines.
43
44

45 46 387 **Acknowledgments** 47

48
49 388 Librarian Ellen Sejersted at the University of Agder assisted in the development of the
50
51 389 search strategy.
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2
3 390 **Authors' contributions**
4
5

6 391 AWL and GR designed the research project and developed the research plan. Librarian
7
8 392 Ellen Sejersted at the University of Agder and AWL were responsible for the literature
9
10 393 search, while AWL and GR were responsible for the analysis. Both authors were involved
11
12 394 in the screening and inclusion of the studies, reviewed the manuscript, and contributed to
13
14 395 the revision of the paper. Both authors read and approved the final version of the paper.
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18
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21

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23
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25
26

27
28 399 **Competing interests**
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31 400 The authors declare that they have no competing interests.
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34 401 **Patient consent for publication**
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37 402 Not required
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40 403 **Ethics approval**
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43 404 Ethical approval was not required, as no primary data were collected as part of this study.
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46 405 **Data availability**
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49 406 Data are available on reasonable request.
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52 407 **References**
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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
Owensworth 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

Author	<i>Coping factors within the next of kin and as external support</i>					<i>Coping strategies – control and proactivity</i>		
	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		

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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

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

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PRISMA 2009 Flow Diagram

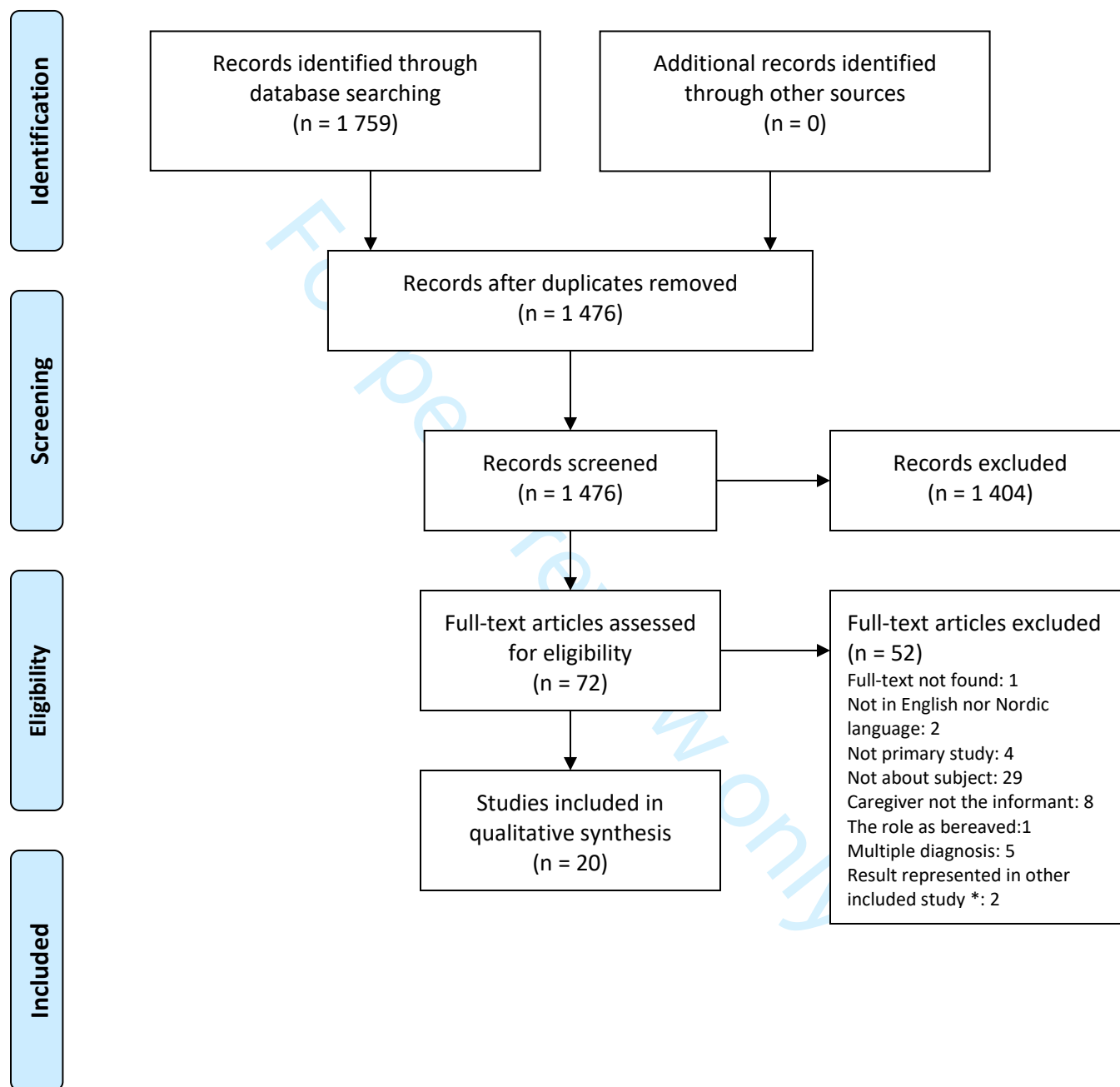


Figure 1. Flow chart of the inclusion process

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097

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3 The search strategy for the metasynthesis:
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5 To search the PsycINFO database, we used the following terms: *((qualitative adj2 (research**
6 *or design* or stud* or method*)) or hermeneutic**
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8 *or “grounded theory” or “meta syntheses” or metasynthesis* or metaethnograph* or interview**
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12 *methods or phenomenology AND (caregiver* or famil* or next of kin* or relatives or spous**
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16 *parent* or mother* or father* or carer* or care giver*).ti,ab,hw,id. AND glioma*.ti,ab,hw,id.*
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18 *OR (brain adj2 (cancer or neoplasm* or tumor*).ti,ab,hw,id.*
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26 In Medline and CHINAL, we used the following terms: *caregiver* OR famil* OR “next of*
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34 *(MH “Grounded Theory”) OR Interview* OR experienc* OR phenomenolog* OR (qualitative*
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38 *recording” OR audiorecording OR metasyntheses* OR “meta syntheses*” OR metaethnograph**
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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.
Cuttillo 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.

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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.

1 2 3 4 5 6 7 8 9 10 11	Owensworth 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.
12 13 14 15 16 17 18 19 20 21 22 23 24	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.
25 26 27 28 29 30 31	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.
32 33 34 35 36 37 38 39 40	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.

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

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1 2 3 4 5 6 7 8 9 10 11 12	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 Ricoeur.
13 14 15 16 17 18 19 20 21 22	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.
23 24 25 26 27 28 29 30 31 32 33	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.
34 35 36 37 38 39 40 41 42 43 44 45 46	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.

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				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

For peer review only



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 <i>The page number refers to the copy without track changes</i>	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

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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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BMJ Open

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

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Secondary Subject Heading:	Health policy
Keywords:	MEDICAL ETHICS, ONCOLOGY, Head & neck tumours < ONCOLOGY

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

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16 **Word Count:** 3981

17
18 **ABSTRACT**

19 **Objective:** Being the next of kin of a person with a brain tumor is a stressful experience.

20 For many, being a next of kin involves fear, insecurity, and overwhelming responsibility.

21 The purpose of this study was to identify and synthesize qualitative original studies that

22 explore coping in the role as next of kin of a person with a brain tumor.

23 **Methods:** A qualitative metasynthesis guided by Sandelowski and Barroso's guidelines

24 was used. The databases Medline, CHINAL, and PsycINFO were searched for studies

25 from January 2000 to January 18, 2022. Inclusion criteria were qualitative original studies

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3 26 that aimed to explore coping experience by the next of kin of a person with brain tumor.

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5 27 The next of kin had to be 18 years of age or older.

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8 28 **Results:** Of a total of 1 476 screened records data from 20 studies, including 342
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10 29 participants (207 female, 81 male, and 54 unclassified) were analyzed into metasummaries
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12 30 and a metasyntesis. The metasyntesis revealed that the next of kin coping experiences
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14 31 were characterized by two main themes: 1) Coping factors within the next of kin and as a
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16 32 support system, such as their personal characteristics, perceiving the role as meaningful,
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18 33 having a support system, and hope and religion. 2) Coping strategies – control and
19
20 34 proactivity, including regaining control, being proactive, and acceptance.

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24 25 **Conclusion:** Next of kin of patients with brain tumors used coping factors and coping
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27 36 strategies gathered within themselves and in their surroundings to handle the situation and
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29 37 their role. It is important that health-care professionals suggest and facilitate these coping
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31 38 factors and strategies because this may reduce stress and make the role of next of kin more
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33 39 manageable.

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37 40 **Keywords:** brain tumor; coping factors; coping strategies; metasyntesis; next of kin;
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39 41 review; qualitative studies

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43 **Strengths and limitations of the study**

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48 44 • The qualitative approach makes an important contribution to the research field by
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50 45 providing a deeper understanding of coping factors and strategies used by the next
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52 46 of kin of a person with a brain tumor.
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55 47 • Most of the included studies in this metasyntesis were high-quality studies.
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3 48 • Our sample is highly multicultural with different geographical origins represented
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5 49 and includes different welfare and health-care systems, and different cultures and
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7 50 religions.
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10 51 • The majority of the sample were women, and a more heterogeneous sample might
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12 52 have revealed more nuanced findings regarding the role of next of kin.
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54 INTRODUCTION

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

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3 70 All these strains can lead to next of kin experiencing stress and lack of coping. Lazarus and
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5 71 Folkman define coping as a cognitive and behavioral endeavor under constant change,
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7 72 dealing with external and/or internal demands that a cognitive assessment indicates as
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9 73 stressful or exceeding personal resources. When dealing with these demands, the next of
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11 74 kin has to review available coping strategies to be able to make the situation more
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13 75 manageable, meaning active actions the next of kin use to cope in the situation. ^{9 13 14}
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17 76 There are some original qualitative studies that have explored coping in the role as next of
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19 77 kin of a person with a brain tumor. To our knowledge, this research has not been
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21 78 synthesized. Such information is of great importance, especially for health-care providers
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23 79 working with this group of caregivers. With improved understanding, they may be better
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25 80 equipped to facilitate a more manageable everyday life among the next of kin. Previous
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27 81 quantitative research directed at these aspects exist,^{8-11 15} but we were interested in studies
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29 82 that were personal and focused on the lived experience of next of kin, hence the choice of
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31 83 qualitative studies. Therefore, the purpose of this metasynthesis was to identify and
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33 84 synthesize evidence from original qualitative studies regarding the experience of coping in
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35 85 the role as next of kin of a person with a brain tumor. The findings are discussed in the
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37 86 context of Lazarus' and Folkman's stress theory¹⁴ and their approach to coping with stress
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39 87 in order to interpret our findings in a theoretical context.
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46 **METHODS**

47 ***Design***

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50 89 The study is a metasynthesis within the interpretative paradigm. It was inspired by a
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52 90 phenomenological–hermeneutic design because the aim was to identify and synthesize
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54 91 qualitative original studies that explored next of kin attitudes and experiences.¹⁶ The
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3 93 metasynthesis process consisted of five steps: (1) formulating the purpose and rationale of
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5 94 the study; (2) searching for and retrieving relevant qualitative research studies; (3)
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7 95 critically appraising the included studies; (4) classifying the findings, and finally; (5)
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10 96 synthesizing the findings.

11 12 13 97 ***Search strategy***

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15 98 In collaboration with an experienced librarian, we conducted a systematic search in the
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17 99 PsycINFO, OVID, CHINAL, and Medline databases via the EBSCO host from January
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20 100 2000 until 18 January 2022. For search strategy see supplementary materials 1.

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23 101 The inclusion criteria were qualitative original studies published in English, Norwegian,
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25 102 Swedish or Danish that aimed to explore coping experience by the next of kin of a person
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27 103 with a brain tumor, regardless of tumor type and stage which enhanced their role as next of
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29 104 kin. The next of kin had to be 18 years of age or older. The exclusion criteria were studies
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31 105 that did not clearly identify coping, coping that included the participants' experiences in
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33 106 the role of bereaved and not next of kin, and studies including diagnoses other than a brain
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35 107 tumor.

36 37 38 39 108 ***Search outcome***

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42 109 The search strategy generated 1 476 unique citations. Titles and abstracts were screened by
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44 110 the authors using Rayyan, a systematic review management software.¹⁷ A final consensus
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46 111 regarding the eligible articles was obtained through a group discussion between the
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48 112 authors. Seventy-two papers were read in full and evaluated against the inclusion criteria
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50 113 by both authors; 20 of these were included in the metasynthesis. Figure 1 shows the
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52 114 Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA)
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54 115 flowchart with a full overview of the screening process. The search output is presented in

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3 116 the PRISMA flowchart. The authors read the full text of the eligible articles and
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5 117 independently extracted data from the included studies; this process is also illustrated in
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7 118 Figure 1. Consensus for data extraction was obtained as part of a group discussion between
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10 119 the authors. Supplementary materials 2 lists the title, author(s), study country, year of
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12 120 publication, aim, analysis, and study participants of all included studies. Most studies were
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14 121 from Europe: Sweden (3), Great Britain (3), Denmark (1), Belgium (1), and Turkey (1);
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16 122 seven were from Canada (3) and the USA (4), two from Australia and two from Taiwan.
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18 123 The tumor type and stage varied. For details, see supplementary materials 2.
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22 124 Figure 1 about here
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25 125 ***Quality appraisal***

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27 126 The quality of the 20 papers was evaluated using the Critical Appraisal Skills Program
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29 127 (CASP) for qualitative studies. The first evaluation was conducted blinded and
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31 128 independently by AWL and GR whose CASP evaluations were then compared. Using the
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33 129 criteria in CASP for independent assessment, the authors mutually agreed on a final quality
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35 130 evaluation. For details, see Table 1.
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39 131 The included studies that were appraised according to CASP are listed in Table 2. All
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41 132 studies had clearly stated the study aim and the qualitative methodologies were considered
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43 133 appropriate. Furthermore, several of the studies had been published in highly ranked
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45 134 journals. The most poorly addressed issue (criteria number 6 in the CASP list) was the
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47 135 influence of the researcher on the research and vice versa.
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3 137 ***Data abstraction and analyses***
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6 138 As suggested by Sandelowski and Barroso,¹⁶ two approaches to qualitative synthesis were
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8 139 used. The first of these involved qualitative metasummaries of qualitative findings from
9
10 140 the original studies. This method is defined as qualitative, but the findings are presented
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12 141 quantitatively. The second involved a metasynthesis that developed new interpretations of
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14 142 the target findings from the original studies.¹⁶ The narrative analysis was inspired by
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16 143 Lindseth and Nordberg's phenomenological–hermeneutic methods.¹⁸ Three steps were
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18 144 followed. First, the empirical materials were read several times. Second, after extraction,
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20 145 the target findings were imported into NVivo 11 data management software for further
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22 146 analysis.¹⁹ The text was read line-by-line to identify meaning units, subthemes, and
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24 147 themes. Third, the researchers aimed to achieve a comprehensive understanding of the
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26 148 empirical materials, meaning units, and themes, and to relate these to the aim and research
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28 149 question of the metasynthesis.¹⁸ The analytic themes were identified by AWL and
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30 150 discussed with GR. The process of deriving the themes was inductive. The contribution of
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32 151 targeted findings from each of the included papers is outlined, and quotations are used to
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34 152 illustrate and support the findings, something which increases the trustworthiness of the
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36 153 study. To validate the findings, both authors participated in discussions of the empirical
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38 154 analysis and in writing up the findings. Ethical approval was not required for the study.
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45 155 Table 2 about here

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48 156 Qualitative metasynthesis enables researchers to identify specific research questions,
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50 157 search for, appraise, summarize, and combine qualitative evidence to address the research
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52 158 question. Metasynthesis provides novel interpretations of the target findings from the
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54 159 original studies. ¹⁶ In our methasynthesis we identified two main themes: 1) coping factors
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56 160 within the next of kin themselves and as a support system and 2) coping strategies –
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3 161 control and proactivity, each comprising 3-4 subthemes. For a list of the studies that
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5 162 generated findings regarding the main themes and subthemes, see Table 2. When
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7 163 analyzing and organizing the results into themes and subthemes we chose to be in line with
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9 164 the content and meaning of coping in the original included studies, although some of the
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11 165 results could have been considered to also contributed and organized differently. The
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13 166 results will be elaborated below.
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18 167 **PATIENT AND PUBLIC INVOLVEMENT**

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22 168 No patients or patient organizations were involved in the planning of the study, the
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24 169 analyses or the writing of the metasynthesis. These were based on published original
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26 170 studies some of which included patient involvement.
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30 171 **RESULTS**

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34 172 The results are presented as metasummaries supported by tables and figures, and as a
35
36 173 metasynthesis containing two main themes. The themes are supported by illustrative
37
38 174 quotes from the included original studies.
39
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41 175 *Metasummaries*

42
43
44 176 The 20 studies that were included comprised 342 participants (207 women, 81 men, and 54
45
46 177 not classified). The focus was on the following themes: the needs of the next of kin;^{2 20-24}
47
48 178 their overall experiences as next of kin;^{10 25-27} coping and coping strategies;²⁸⁻³⁰
49
50 179 postoperative caregiving;^{31 32} being a next of kin in the palliative phase;^{33 34} experienced
51
52 180 support factors;³⁵ how the caregiving changed over time;³⁶ and factors influencing
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54 181 treatment choice in the palliative phase.³⁷ Three of the studies were undertaken six months
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3 182 after diagnosis,^{27 30 31 36} and three in the patients' palliative phase or postmortem.^{33 34 37} In
4
5 183 six studies the patients were children of the informants.^{10 29 30 33 34}
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7

8 184 ***Metasynthesis***
9

10 185 *Main theme 1: Coping factors within the next of kin and as a support system*
11

12 186 Nineteen of the included studies provided data regarding the first main theme; *Coping*
13
14 187 *factors within the next of kin and as external support* (see table 2). This main theme
15
16 188 comprised the following four sub themes: *personal characteristics, perceiving the role as*
17
18 189 *meaningful, having a support system, and hope and religion.*
19
20
21
22

23 190 *Personal characteristics* such as a strong and positive personality were important coping
24
25 191 factors for next of kin in new challenging situations.^{25 29 37} Being able to show empathy for
26
27 192 the patient and the health professionals was important, if not the situation could easily
28
29 193 engender feelings such as discouragement and reproach.²⁵ A positive mood and a sense of
30
31 194 humor were also emphasized for the same reasons.²⁹
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35 195 To perceive the role as next of kin as *meaningful* was important, as it made the next of kin
36
37 196 feel needed and productive in the situation.^{23 25 28 31} Engagement and commitment in the
38
39 197 care of their relatives were highlighted as important by many next of kin, especially when
40
41 198 the patients appreciated the help.²³ The engagement was even stronger when the emotional
42
43 199 bond between patient and next of kin was strong.^{20 21 29 35}
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45
46

47 200 *"But caring for him is something I will do—it is not a burden."*³¹ (p. 81)
48
49

50 201 However, other studies revealed less engagement and commitment, and underlined anger
51
52 202 and reluctance with the new role as the heavy responsibility and sacrifice impacted the
53
54 203 next of kin's own needs and wishes.^{21 22 25 31 33}
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3 204 *Having a support system* made the role of next of kin easier to cope with. The support was
4
5 205 given by family, friends, neighbors, colleagues and workplaces, health personnel, schools,
6
7 206 the religious community, people in the local community, and even strangers.^{2 10 20-35} The
8
9
10 207 support from health-care professionals was especially important. This support included
11
12 208 emotional support and assistance during patient care and treatment.^{2 10 20-27 29-35} The
13
14 209 importance of assistance such as medical supervision and nursing care was emphasized,¹⁰
15
16 210 ^{22 29} with next of kin noting that this made it possible to feel like a partner again,²³ while at
17
18 211 the same time allowing for anticipated time alone.²⁴ A familiar health-care professional
19
20 212 was crucial in making this possible, because it implied that the patient would receive the
21
22 213 best care as they were known to the health-care professional, and also because the
23
24 214 assistance was considered to be less intrusive.^{23 24} To experience the assistance with care as
25
26 215 a coping factor, it was crucial that the care was compassionate and of the best quality.
27
28 216 These qualities emphasized the health professionals genuine care and gave the patients and
29
30 217 the next of kin hope and a desire to fight the disease.^{10 21 23 26 27 29}

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35
36 218 *“She (neurosurgeon) had to give us some bad news some of the time...*
37
38 219 *and you couldn’t ask for a better manner in her delivery of that bad*
39
40 220 *news, or her support in what we were going through.”³⁵ (p. 8)*

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42
43
44 221 When next of kin experienced that their loved ones received a low quality of care or
45
46 222 suffered malpractice it caused mistrust of the health-care system and weakened the
47
48 223 experience of health-care professionals as a support factor.^{10 20 23 24} Emotional support from
49
50 224 health-care professionals implied an acknowledgment that the disease affected not only the
51
52 225 patients, but also their next of kin. It also implied that the health-care professionals
53
54 226 recognized and met the wishes of the next of kin for active participation in monitoring the
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1
2
3 227 patients disease course.^{23 25 26 34} Next of kin who did not have such involvement felt
4
5 228 ignored, useless and helpless.^{25 29} Supportive conversations with health-care professionals
6
7 229 were highly appreciated by many next of kin. However, this required the health-care
8
9 230 professional's understanding and empathy for the situation of the patient as well as of their
10
11
12 231 next of kin, and preferably that they should be always available.^{21 23 26 30 31 37}

13
14
15 232 Support from family and friends was invaluable in the care tasks and in coping with the
16
17 233 role of next of kin.

18
19
20 234 *“Just support from family and friends, that was important to me, and just*
21
22 *knowing that I could call on them... ”*²² (p. 1098)

23
24
25
26 236 Social, practical, and emotional support was emphasized, and included such things as
27
28 237 economic help, childcare, transport and housekeeping.^{10 22 24 25 29-32 34 35} Some next of kin
29
30 238 would have appreciated even more support and help from family and friends, preferably
31
32 239 given on the family and friends' own initiative.^{20 22 24 25 35 36}

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35
36 240 Discussions with family and friends were also important,^{21 24 25 27} and could even create a
37
38 241 stronger bond.²⁵ Such a bond required families and friends to understand and recognize the
39
40 242 challenges faced by the next of kin.²⁴ Support groups and conversations with other next of
41
42 243 kin were also highlighted as important,^{2 22 24 30 34 35 37} as they might broaden the next of
43
44 244 kin's understanding of the tumor and what they might expect in the future.²⁷ These
45
46 245 conversations could be face-to-face or via the Internet.^{2 22 24 30 34 35 37}

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49
50 246 *“From time to time, I need to be able to talk to someone. Because when I*
51
52 *lay down in the evening, then it starts to work in the inside.”*²³ (p. 411)

1
2
3 248 On the other hand, support groups were also considered demanding because it was difficult
4
5 249 to listen to other families' stories. Furthermore, for some it was considered a waste of time
6
7
8 250 to spend valuable hours with people other than their closest family members.^{10 22 31}

9
10 251 *Hope and religion* were emphasized as important coping factors. The next of kin hoped
11
12 252 that a miraculous treatment would be developed so that their loved ones could survive the
13
14
15 253 disease or just have a better quality of life.^{2 10 20-23 26 33 34}

16
17
18 254 *You see a positive evolution, and everything that goes better is good for*
19
20 255 *her. (...) Nobody can forbid us to have hope. And miracles happen.*
21
22
23 256 *Whether we believe it or not, that's not the point, it is the only thing to*
24
25 257 *focus on.*"²³ (p. 409)

26
27
28 258 Hope gave a reason to fight, although it weakened in the palliative phase.^{21 26 34} Faith
29
30 259 strengthened the hope of healing during the treatment period and gave some form of peace
31
32
33 260 in the final palliative phase. In most cases, hope was related to faith.^{25-27 30 34 37}

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37 261 *Main theme 2: Coping strategies – control and proactivity*

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40 262 Eighteen of the included studies provided data regarding the second main theme; *Coping*
41
42 263 *strategies – control and proactivity* (see table 2). This main theme comprised the three
43
44 264 subthemes: *regaining control, being proactive and acceptance.*

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46
47
48 265 *Regaining control* of the situation was a frequent coping strategy, and for most this
49
50 266 included gathering enough information to allow an overview of what to expect, something
51
52 267 which implied some form of security.^{10 20-23 27 30 35 37}

1
2
3 268 *“So it’s a, it’s a roller coaster of emotion but for the most part I’ve been,*
4
5 269 *‘What do we need to do? Where do we need to be?’ And then just read,*
6
7 270 *read, read whatever I can find out, whatever information because I feel*
8
9
10 271 *like whatever I know, I can ask for.”*³⁰ (p. 34)
11
12

13 272 The information that was gathered and provided should preferably be adapted to the
14
15 273 situation and the disease trajectory, and had been given by health-care professionals.^{20 22 23}
16
17 274 ^{25 27 29 37} The next of kin often hid this information from the patients to protect them and
18
19
20 275 not diminish their hope.^{10 26 30 31 34}
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22

23 276 To regain control meant not only control of the diagnosis, but also personal control and
24
25 277 control over own reactions. In some cases, the next of kin denied their feelings. Some even
26
27 278 denied the entire diagnosis,^{20 25 29 30} and instead focused on being strong for the patient and
28
29 279 the entire family.^{23 25 30 32-34 36} One next of kin in Edvardsson and Ahlstroms’ (2008)
30
31 280 study²⁵ reported:
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34

35 281 *“I’ve sort of stowed it all away, I suppose. It is as if I’d experienced it from the*
36
37 282 *outside or seen it on TV. It’s often that way with sorrowful things.”* (p. 588)
38
39

40 283 *Being proactive*, facilitating and encouraging the patient to fight the disease were also
41
42 284 important coping strategies, as it felt better than accepting the morbid situation and not do
43
44 285 anything.^{10 21 25 26 34}
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47

48 286 *“People ask you how you cope. But what if you were to give up? You’ve*
49
50 287 *got to cope—and we do have each other! (...).”*²⁵ (p. 588)
51
52

53 288 This implied adopting a healthier lifestyle, including a change in diet and exercise habits,
54
55 289 hoping that this would improve the effects of medical treatment,^{21 26} or trying alternative
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1
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3 290 treatments.^{10 34} However, an increasing feeling of powerlessness was emphasized if the
4
5 291 fight, in the form of these actions and treatments, did not meet the hope of a cure.^{21 23 26 34}
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7
8 292 As the disease progressed and life went on there was a strive for normality, particularly in
9
10 293 families with children. This lead most next of kin into a strategy of *acceptance*, as
11
12 294 everyday life continued. This involved work, school for children and hobbies.^{10 26 28 30 34 10}
13
14
15 295 ^{26 28 30 31 33 34 36} Although this was an important and expected strategy, accepting disease
16
17 296 progression or a bad diagnosis was challenging, especially when the patient was a child.³⁴
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19

20 21 297 **DISCUSSION**

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23
24 298 This metasynthesis aimed to explore coping in the role as next of kin of a person with a
25
26 299 brain tumor. This generated two main themes: 1) coping factors within the next of kin and
27
28 300 as a support system, 2) and coping strategies – control and proactivity. Valuable coping
29
30 301 factors included personal characteristics, perceiving the role as next of kin as meaningful,
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32 302 having a support system, and hope and religion. Active strategies to manage the situation
33
34 303 involved regaining control, being proactive, and acceptance.^{14 38}
35
36
37 304 Being the next of kin to a person with a brain tumor is considered to be a negative stressor
38
39 305 because of the challenging life situation and care tasks. Nevertheless, several next of kin
40
41 306 who were included in the metasynthesis expressed a desire to be proactive, fight the
42
43 307 disease and to gain control over the situation. This is described by Lazarus and Folkman¹⁴
44
45 308 as a secondary assessment of the situation, in which the next of kin decide which measures
46
47 309 to implement. One such measure could be to gain personal control—one of the most
48
49 310 important and stress-reducing personal strategies available.¹⁴
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3 311 A possible explanation for the proactive attitude of next of kin toward the disease may be
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5 312 their obligation and commitment to the patient. Commitment is an expression of something
6
7 313 of great importance and can cause some to be willing to meet threats and challenges that he
8
9
10 314 or she would otherwise avoid.¹⁴ However, our findings revealed that the experience of
11
12 315 contributing to something meaningful, not the obligation to do so, promoted coping in the
13
14 316 situation. We consider that this is caused by the fact that obligation does not automatically
15
16 317 make an action meaningful, but rather that it can be experienced as a compulsion. This
17
18 318 assumption is strengthened by the findings that the tasks as next of kin may arouse
19
20 319 emotions such as anger and aversion towards the patient and the diagnosis, rather than
21
22 320 coping. Several studies refer to the same ambivalent experience regarding commitment and
23
24 321 attitudes toward being a next of kin.^{39 40}

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29 322 Having a support system was the factor that most relatives emphasized as promoting
30
31 323 coping. It was described as invaluable something which was also confirmed in other
32
33 324 studies,^{41 42} and in Lazarus and Folkman's transactional stress theory.¹⁴ At the same time,
34
35 325 in both this metasynthesis and in other studies, next of kin voiced a strong desire and
36
37 326 longing for even greater external support.^{41 42} The findings of the metasynthesis also
38
39 327 showed that the configuration and arrangement of the support, especially that given by
40
41 328 health-care providers are of great importance. An explanation for the next of kin's
42
43 329 experience of unmet needs might be lack of knowledge among health-care providers about
44
45 330 how to assist at the right time. This may indicate that in some cases health-care providers
46
47 331 should pay more attention to offering support in line with the individual needs of the next
48
49 332 of kin and the care situations.

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53
54 333 The findings of this metasynthesis show that several next of kin considered hope to be an
55
56 334 important coping factor, especially during the disease trajectory. Hope has also been

1
2
3 335 shown to be an strengthening coping factor in several studies,^{43 44} and transactional stress
4
5 336 theory states that faith and hope are two of the most important personal factors in the
6
7 337 cognitive assessment of stressors.^{14 38} Furthermore, according to Lazarus and Folkman,¹⁴
8
9 338 the two factors are strongly related, which is consistent with the findings in our
10
11 339 metasynthesis. For several next of kin, hope was strongly grounded in religion. This was
12
13 340 especially prominent in the studies conducted in the palliative phase, which indicated that
14
15 341 faith is strengthened when there is no hope of curative treatment. The same pattern has also
16
17 342 been reported in other studies describing cancer patients' experiences of palliative care.^{45 46}
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19 343
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24 344 As the disease progressed, several next of kin chose an acceptance strategy toward the
25
26 345 diagnosis and its burden. Their fight against the disease diminished to some extent, and
27
28 346 instead the relatives tried to "normalize" everyday life as much as possible. A similar
29
30 347 strategy is also reported by next of kin of other cancer patients, especially in the palliative
31
32 348 phase.^{47 48} Lazarus and Folkman describe this as a reassessment, referring to a changed
33
34 349 cognitive assessment of the stressor based on new information from the environment
35
36 350 and/or the person himself or herself.¹⁴
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42 **Strengths and limitations**

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45 352 A strength of this metasynthesis is that the primary search in the databases was conducted
46
47 353 with the assistance of an experienced librarian in an attempt to ensure that as many as
48
49 354 possible of the relevant studies were included.⁴⁹ Furthermore, most of the included studies
50
51 355 were of high methodological quality (see Table 2). Our sample was also highly
52
53 356 multicultural (see Table 1). This attribute strengthens the validity of the metasynthesis
54
55 357 since geographical origin might have affected the study sample because of different
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3 358 participant backgrounds related to different welfare and health-care systems, cultures,
4
5 359 and/or religions.
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7

8 360 A limitation of our metasynthesis is that one of the 72 articles that was intended to be read
9
10 361 in full text could not be obtained.⁵⁰ The formation of the subthemes is also a possible
11
12 362 limitation. Some of the subthemes, or parts of their content, could also have been
13
14 363 categorized in the other main theme. Both main themes and subthemes overlap in several
15
16 364 cases, and we have read similar studies^{26 30} where the findings are categorized differently
17
18 365 than in our metasynthesis. We chose to be true to the informants' statements, the
19
20 366 organization and meaning of the original studies that were included, and allocated the
21
22 367 findings based on the informants' way of speaking and description of the experience.
23
24 368 Another possible limitation is that our sample consisted mainly of women (see
25
26 369 supplementary materials 2). A more heterogeneous sample might have revealed more
27
28 370 nuanced findings and different experiences of the role of the next of kin.
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33 371 **CONCLUSION**

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38 372 The findings of this metasynthesis show that next of kin experience and use a range of
39
40 373 coping factors and strategies in their role. Their experience is marked by individual
41
42 374 differences. It is of great importance that health-care providers offer assistance which is
43
44 375 individually adapted to these coping factors and strategies because this may reduce stress
45
46 376 among the next of kin. The coping experience seems to go through phases, and further
47
48 377 information is needed to fully understand how and when the various factors and strategies
49
50 378 are used as the disease progresses. Longitudinal studies would therefore be of particular
51
52 379 interest in this field.
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3 381 **Supplementary information**
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6 382 The manuscript has been edited by OnLine English (<https://www.oleng.com.au>) to comply
7
8 383 with international publishing guidelines.
9
10

11
12 384 **Authors' contributions**
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14

15 385 AWL and GR designed the research project and developed the research plan. AWL was
16
17 386 responsible for the literature search, while AWL and GR were responsible for the analysis.
18
19 387 Both authors were involved in the screening and inclusion of the studies, reviewed the
20
21 388 manuscript, and contributed to the revision of the paper. Both authors read and approved
22
23 389 the final version of the paper.
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27
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35

36
37 393 **Competing interests**
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40 394 The authors declare that they have no competing interests.
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43 395 **Patient consent for publication**
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46 396 Not required
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48

49 397 **Ethics approval**
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51

52 398 Ethical approval was not required, as no primary data were collected as part of this study.
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54

55 399 **Data availability**
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58 400 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
Cuttilo 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
Owensworth 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

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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

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

Author	<i>Coping factors within the next of kin and as a support system</i>				<i>Coping strategies – control and proactivity</i>		
	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		
Cuttillo 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
Owensworth 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

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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

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3 Figure legend: Figure 1. Flow chart of the inclusion process
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PRISMA 2009 Flow Diagram

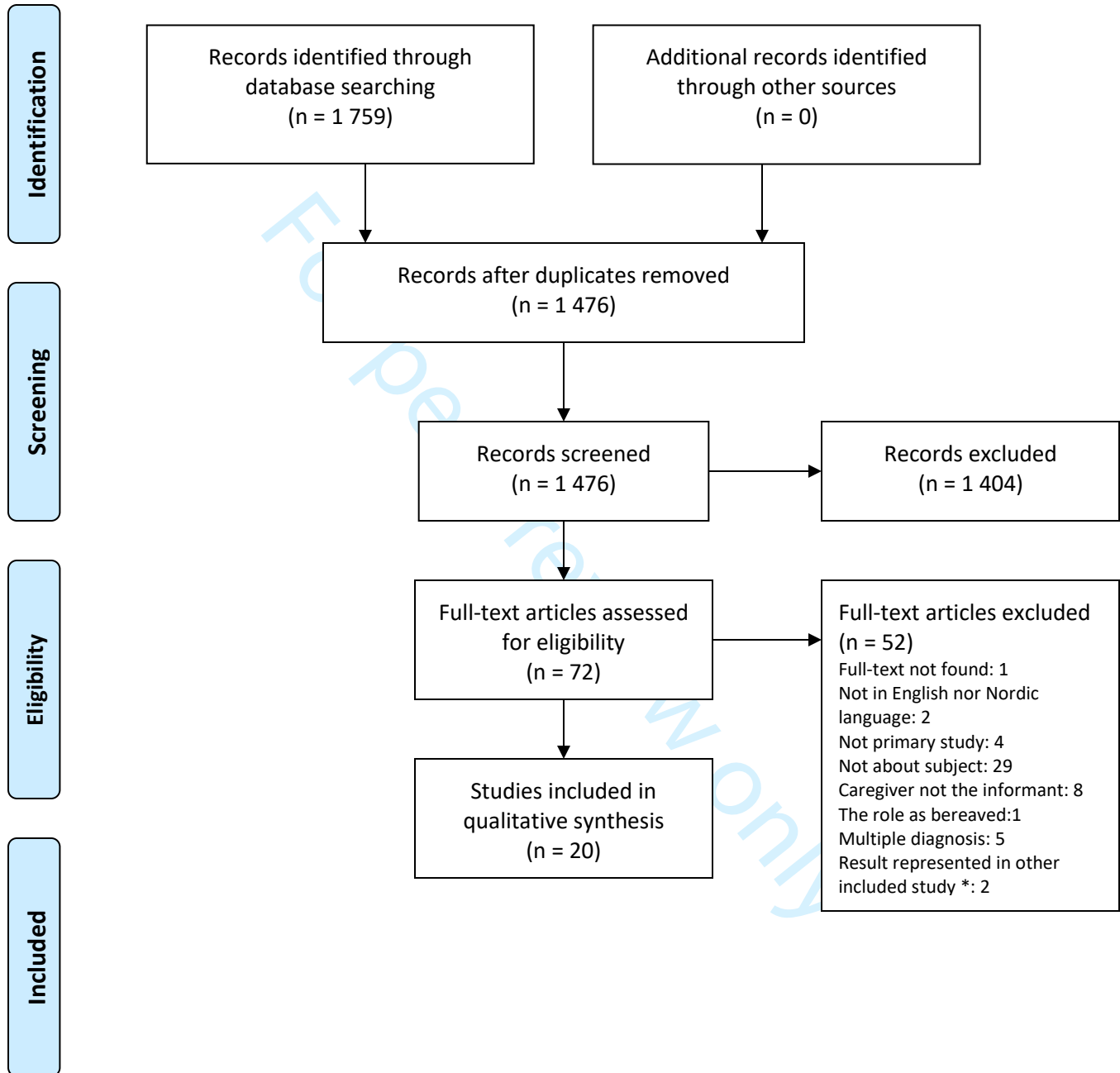


Figure 1. Flow chart of the inclusion process

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097

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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 WI (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 WI (cancer OR tumor* or neoplasm*).*

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4 **Supplementary materials 2: Characteristics of the included studies.**
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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.
Cuttillo 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.

1 2 3 4 5 6 7 8 9 10 11 12 13 14	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.
15 16 17 18 19 20 21 22 23 24	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.
25 26 27 28 29 30 31	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.
32 33 34 35 36 37 38 39 40 41 42 43 44 45 46	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.

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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 Ricoeur.
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 <i>The page number refers to the copy without track changes</i>	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

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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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