






# Unmet supportive care needs of patients with rare cancer: A systematic review

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## Abstract

**Objective:** Patients with rare cancers may experience different unmet needs than those with common cancer. The objective of this systematic review was to (1) investigate unmet supportive care needs of rare cancer patients throughout the disease trajectory and (2) identify predictive factors for these unmet needs.

**Methods:** PubMed, PsycINFO and CINAHL were searched for publications (January 2011 to March 2021) focusing on unmet needs of patients with rare cancer. Two reviewers independently selected studies, extracted data and performed quality assessment. Findings were synthesised.

**Results:** The search yielded 4,598 articles, of which 59 articles met eligibility criteria and 57 were of medium or high quality. Rare cancer patients most frequently reported unmet needs in the healthcare system and information domain (up to 95%), followed by the psychological domain (up to 93%) and the physical and daily living domain (up to 80%). Unmet needs were mainly reported in the posttreatment phase. The most frequently identified predictors were higher anxiety, younger age and higher neuroticism.

**Conclusion:** Patients with rare cancer have unmet needs throughout their disease trajectory. Supportive care needs of rare cancer patients should be addressed individually, depending on the rare cancer subdomain and phase of disease and from diagnosis onwards.

## KEYWORDS

rare cancer, supportive care, systematic review, unmet needs

## 1 | INTRODUCTION

In Europe, 3.9 million people are diagnosed with cancer each year (Ferlay et al., 2018), of whom 22% are affected by a rare cancer (G. Gatta et al., 2011). Rare cancers are defined by the Surveillance of Rare Cancer in Europe (RARECARE) as those with an incidence of <6/100,000 people per year (G. Gatta et al., 2011). Patients who have been diagnosed with a rare cancer have lower survival rates than those with common cancers, i.e., 49% versus 63%, respectively

(G. Gatta et al., 2017), experience lower quality of life (QoL) and higher levels of distress compared to common cancer patients (Bergerot et al., 2018). Therefore, delivering high-quality cancer care for both rare and common cancer patients involves not only anti-cancer treatment but also attention for patients' supportive care needs.

Supportive care needs can be defined as care that helps the patient and his/her family to cope with cancer and its treatment, from pre-diagnosis through the process of diagnosis and treatment to cure,

continuing illness or death and into bereavement (Gysels et al., 2004). Supportive care needs are broad in dimension and range from psychological to sexual needs of an individual cancer patient (M. Fitch, 1994). Unmet needs are those supportive care needs that are not addressed and require additional service or support for an individual in order to achieve optimal well-being (M. Fitch, 2000; Sanson-Fisher et al., 2000). By assessing unmet needs, it can be determined how well needs have been met. Needs that remain unmet can be identified among a variety of domains, indicating the multidimensional impact of cancer (M. Fitch, 2008; Sanson-Fisher et al., 2000; Wen & Gustafson, 2004). Cancer patients reported high levels of unmet needs regarding information provision throughout the cancer pathway (Faller et al., 2016; Sanson-Fisher et al., 2000; Sutherland et al., 2009), psychological and psychosocial support (Sanson-Fisher et al., 2000; Sothill et al., 2001; Sutherland et al., 2009; Zabora et al., 2001), coping with the physical effects of the disease and treatment (Newell et al., 1999; Sanson-Fisher et al., 2000; Waller et al., 2012) and practical issues (e.g., transportation; Davis et al., 2002; Zebrack et al., 2013). The presence and broad range of unmet supportive care needs among cancer patients stress the importance of addressing these needs.

Patients with rare cancer face difficulties during their disease trajectory, including delayed and/or incorrect diagnosis, lack of disease-specific information and limited access to clinical expertise, treatment options and (pre)clinical research (Drabbe et al., 2021; Pillai & Jayasree, 2017; Ray-Coquard et al., 2017; The Lancet Oncology, 2001). These difficulties might differ between rare cancer subdomains, e.g., rare digestive cancer, rare gynaecological cancer and rare haematological cancer (EURACAN, 2021; EuroBloodNet, 2021). Moreover, rare cancer patients might experience more, but also different unmet needs, compared to patients with common cancer. In previous studies on rare cancers, a high prevalence of unmet supportive care needs was shown among patients with sarcomas and with brain tumours (Janda et al., 2008; Weaver et al., 2020). For example, in the study focusing on patients with sarcomas, patients reported a lack of information about their disease and treatment and expressed a need for a community and contact with fellow sufferers (Weaver et al., 2020).

In patients with common cancers, highest unmet needs were reported in the daily living, psychological, information and physical domains (Harrison et al., 2009; Sanson-Fisher et al., 2000). For example, patients with colon and/or rectum cancer indicated a high need for emotional support and reassurance, particularly regarding cancer recurrence (Kotronoulas et al., 2017). It has been shown that unmet needs of common cancer patients appear to be highest during the treatment and posttreatment phase (Harrison et al., 2009). This might have to do with the intensive phase of active treatment for the patient, or because most studies on unmet needs have been conducted in these phases. Further, in a previous systematic review, it was shown that predictors of unmet needs in common cancer patients include advanced disease stage, poor health status, geographical isolation from health services and lack of social support networks (Harrison et al., 2009). However, there is a lack of information about unmet needs in the specific disease trajectory phases, per rare cancer

subdomain and about predictors of these needs in patients who have been diagnosed with a rare cancer type.

To provide optimal supportive care for rare cancer patients, it is necessary that healthcare professionals have knowledge regarding the differences of unmet needs between rare and common cancer patients. Since one out of five cancer patients are diagnosed with a form of rare cancer, providing an overview of the unmet needs of rare cancer patients is relevant as healthcare professionals are likely to encounter this patient group. To our knowledge, no systematic review on unmet supportive care needs of patients with rare cancer at different stages of the disease trajectory has been conducted so far. Therefore, the aim of the current systematic review was to (1) explore unmet supportive care needs of rare cancer patients during the phases of their disease trajectory, for each rare cancer subdomain, and (2) identify predictors of these unmet needs.

## 2 | METHODS

### 2.1 | Protocol registration and report

This systematic review was registered in the 'International Prospective Register of Systematic Reviews' (PROSPERO) in 2020 (registration number CRD42020183601), and the protocol is available upon request. The review was performed in accordance with the recommendations of the 'Preferred Reporting Items For Systematic Reviews and Meta-Analyses' (PRISMA) statement (Moher, Liberati, Tetzlaff, & Altman, 2010).

### 2.2 | Search strategy

A systematic search was performed in the databases PubMed, PsycINFO and CINAHL, restricted to studies published from January 2011 until March 2021. This period was chosen because of the definition of rare cancers, which has been published and adopted in the European Union since 2011 (G. Gatta et al., 2011). Studies were identified using search strings based on the PubMed strategy, which uses a combination of MeSH terms and free text terms. The terms used were related to cancer (e.g., neoplasms, cancer and tumour) and unmet supportive care needs (e.g., psychological, physical and information needs). The search string included broad terms on cancer to cover all studies that included data on both rare and common cancers. Subsequently, the search syntax was adapted per database, including different or additional search terms where necessary (Appendix A).

Both quantitative and qualitative studies were eligible for inclusion if they evaluated unmet supportive care needs for rare cancer patients of adult age (i.e.,  $\geq 18$  years) regardless of cancer site and stage of disease. Rare cancers were defined as those with an incidence of  $< 6/100,000$  people per year (G. Gatta et al., 2011). An overview of the rare cancer types, based on an updated version (February 2019) of the RARECARENet list (RARECARENet, 2019), can be found in Appendix B. Studies were excluded if (1) the study population

consisted of mixed rare and common cancer types, i.e., information on unmet needs could not be distinguished between rare and common cancer patients; (2) the unmet needs of children, adolescents and young adults (<18 years), caregivers or those at high risk of developing cancer were explored; (3) nutritional needs only were explored and/or (4) other reasons for ineligibility were present (e.g., full-text not available and studies published in other language than English).

All titles and abstracts were screened by two involved researchers (EdH and AF), and papers considered as irrelevant to this review (i.e., out of scope or not meeting the inclusion criteria) were eliminated. If title and abstract did not fully provide information for enabling selection, full-text articles were retrieved and screened. The remaining studies were assessed on eligibility, and disagreements about the selection of articles were discussed until consensus was reached. In case there was no consensus between the two authors, a third author (SD) was involved to decide if the article should be included in the review. Reference lists of relevant articles were checked to identify additional studies.

## 2.3 | Data extraction

Two researchers (EdH and MR) independently extracted data from each publication, including (1) general information (e.g., year of publication and country); (2) study characteristics (e.g., design and setting); (3) study population characteristics (e.g., number of participants, age and gender); (4) disease and treatment characteristics (e.g., tumour type, cancer stage and treatment); (5) unmet needs (e.g., measurement and domain) and (5) predictors. Tumour types were categorised into cancer subdomains according to EURACAN and EuroBloodNet (EURACAN, 2021; EuroBloodNet, 2021), with the exception of male breast cancer, which is included separately, as it is not represented in one of these rare cancer subdomains (Appendix B). Unmet needs were classified into fourteen predominantly studied domains: communication, disease-specific, economic, emotional, family-oriented, healthcare system and information, patient care and support, physical and daily living, psychological, psychosocial, supportive care, sexuality, transportation and work-related (Boyes et al., 2009; Harrison et al., 2009; Holm et al., 2012). Phases of the disease trajectory were divided into diagnostic, treatment and posttreatment phase. The two researchers (EdH and MR) compared the extracted data and discussed findings until consensus was reached. In case of no consensus, disagreements were resolved by arbitration with a third researcher (SD).

## 2.4 | Quality assessment

The quality of the included studies was assessed independently by two researchers (EdH and MR), using checklists from the Critical Appraisal Skills Program (CASP; Critical Appraisal Skills Programme, 2017; for qualitative and cohort studies) and Strengthening the Reporting of Observational Studies in Epidemiology (STROBE, 2007; for cross-sectional studies). The quality of the studies

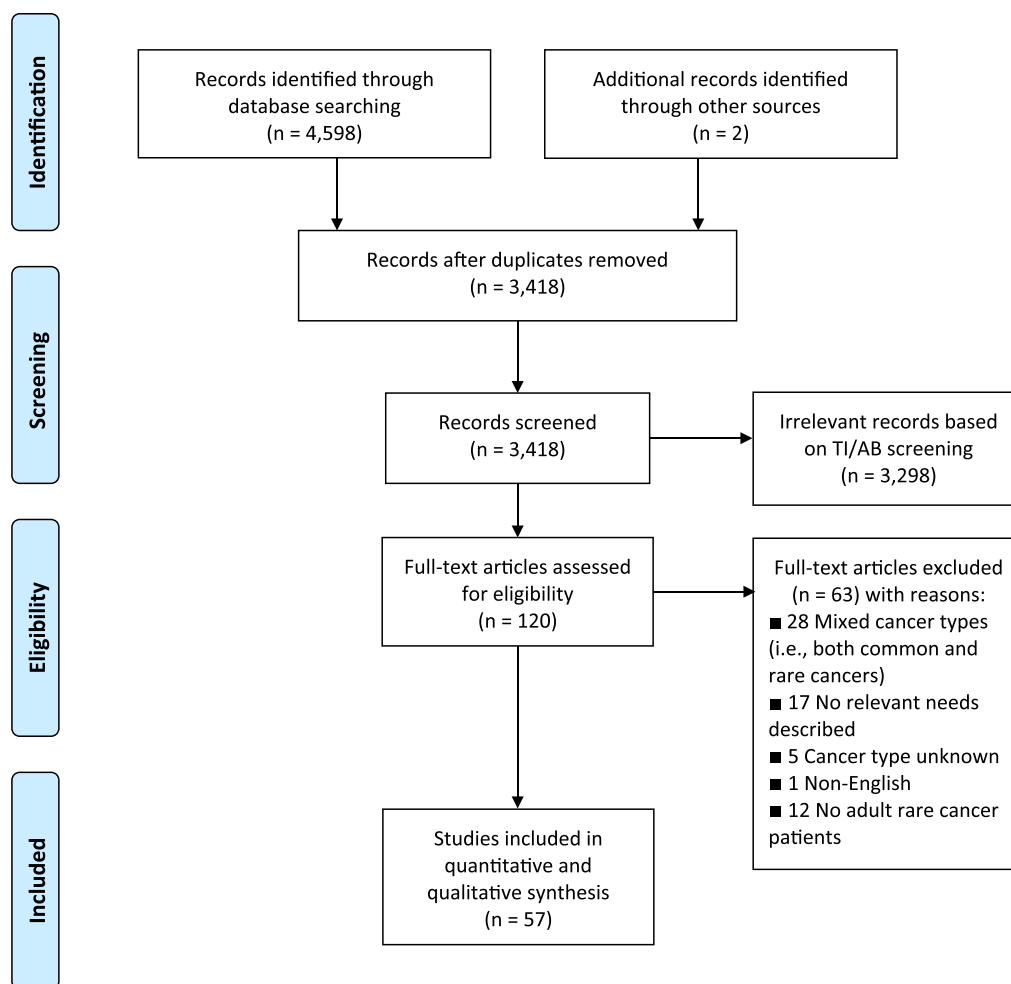
was indicated as low, medium or high quality, based on items from the checklist, e.g., a clear statement of aims, appropriate research design, appropriate recruitment strategy, description of the method of analysis and a clear data description. Qualitative and cohort studies were classified as 'high quality', if scores were  $\geq 80\%$  on CASP criteria, 'medium quality' if scores were between 60% and 79% and 'low quality' if scores were  $< 60\%$  (Al Dweik et al., 2017; Habens et al., 2020). Cross-sectional studies were considered to be of 'high quality', if scores were  $\geq 75\%$  on STROBE criteria, 'medium quality' if scores were between 50% and 74%, and 'low quality' if scores were  $< 50\%$  (De Carlo et al., 2016; Ghiasi, 2021). Inconsistencies in scoring were discussed and resolved by consensus by two researchers (EdH and MR). If consensus was not achieved, the opinion of another involved researcher (SD) was inquired for final assessment.

## 3 | RESULTS

### 3.1 | Study characteristics

After initial screening of 4,598 articles, 120 potentially eligible articles were retrieved and examined in full text (Figure 1). Finally, 57 articles met the inclusion criteria, including two additional articles that were identified after screening the reference lists of included articles (Badr et al., 2016; V.L. Beesley et al., 2013, 2018; Bender et al., 2012; Boland et al., 2014; Bootsma et al., 2020; Brennan et al., 2019; Chen et al., 2013; Feinberg et al., 2013; Gillespie et al., 2017; Giuliani et al., 2016; Goldfarb & Casillas, 2014; Halkett et al., 2015; Hansen et al., 2013; M. Henry et al., 2018; M. Henry et al., 2020; Henselmans et al., 2012; Holm et al., 2012; Husson et al., 2014; Jabbour et al., 2017; Jansen et al., 2018; Kebede & Kebede, 2017; Langbecker & Yates, 2016; M.S. Lee, Nelson, et al., 2016; B. Lee, Park, et al., 2016; Lin et al., 2020; Long et al., 2016; Manne et al., 2016; Molassiotis et al., 2011; Moore et al., 2014; Nørskov et al., 2019; D. Oberoi, White, Seymour, Prince, et al., 2017a; D.V. Oberoi, White, Seymour, Prince, et al., 2017b; O'Brien et al., 2017; Pateman et al., 2015; Peeters et al., 2018; Philp et al., 2017; Piil et al., 2018; Pongthavornkamol et al., 2019; Putri et al., 2018; Reinert et al., 2018; Renovanz et al., 2017; Richardson et al., 2015; Rietveld et al., 2018; Saroa et al., 2018; Shun et al., 2018; Singh et al., 2017; Skoogh et al., 2013; Smith et al., 2013; W.K. So et al., 2019; Sperling et al., 2014; Vermeer et al., 2015; Wells et al., 2015; Williamson et al., 2018; Wolin et al., 2017; Yu et al., 2017; Zeng et al., 2017).

Studies were conducted in Europe ( $N = 19$ ), Australia/New Zealand ( $N = 13$ ), Asia ( $N = 9$ ), Canada ( $N = 9$ ), United States ( $N = 5$ ) and Africa ( $N = 2$ ). Twenty-eight studies had a cross-sectional design, 14 had a qualitative design, 4 had a mixed methods design and 11 had a prospective cohort design, with follow-up periods ranging from 3 months to 2 years. Two mixed method studies (Bootsma et al., 2020; W.K. So et al., 2019) used two designs (i.e., quantitative and qualitative design), which are separately presented in Table 1. The other two mixed method studies



**FIGURE 1** PRISMA flow diagram of systematic search and selection procedure

(Piil et al., 2018; Saroa et al., 2018) presented findings on unmet needs only in the qualitative section. One study on haematological cancer patients was reported in two articles, with the first focusing on the course of unmet needs (D. Oberoi, White, Seymour, Prince, et al., 2017a) and the other reporting the influence of unmet needs on QoL (D.V. Oberoi, White, Seymour, Prince, et al., 2017b). In total, 12,399 patients (aged 18–94 years) were included across all studies. Cancer subdomains of research were rare HNC ( $N = 22$ ), rare gynaecological cancer ( $N = 8$ ), rare haematological cancer ( $N = 6$ ), rare CNS cancer ( $N = 5$ ), NET ( $N = 4$ ), endocrine cancer ( $N = 4$ ), rare digestive cancer ( $N = 3$ ), rare male genital and urogenital cancer ( $N = 3$ ), rare skin cancer/eye melanoma ( $N = 1$ ) and male breast cancer ( $N = 1$ ). Most patients received surgery, radiotherapy, chemotherapy or a combination of these treatments. The majority of the studies used questionnaires ( $N = 43$ ) to gather data on unmet needs, including the Supportive Care Needs Survey (SCNS;  $N = 15$ ) and the Cancer Survivors Unmet Needs (CASUN) instrument ( $N = 6$ ), but also, interviews ( $N = 18$ ) were conducted. Further details on the characteristics of the included studies have been provided in Table 1.

### 3.2 | Quality assessment

With regard to the methodological quality assessment, 45 articles were assessed as being of ‘high quality’ and 12 articles of ‘medium quality’ (Table 1). Two studies (Kebede & Kebede, 2017; Lin et al., 2020) were considered as ‘low quality’ and were excluded from further review. Criteria less described in the included studies were the adequate consideration of the relationship between the researcher and the participants (i.e., in qualitative studies), potential sources of bias (i.e., in cohort studies), the lack of indication of the study design in the title or abstract and potential sources of bias (i.e., in cross-sectional studies).

### 3.3 | Quantitative studies

A total of 42 studies reported quantitative results regarding unmet needs of patients with rare cancer (Table 2). Unmet needs are presented per cancer subdomain, beginning with ‘general supportive care needs’, as these needs are broad in dimension. The subsequent unmet

**TABLE 1** Characteristics of included studies on unmet needs in rare cancer patients

Author, year [ref]	Country	Design (follow-up)	Cancer subdomain (type) <sup>a</sup>	N	Age	Gender, % male	Cancer stage	Time since diagnosis	Treatment <sup>b</sup>	Measurement <sup>c</sup>	QA <sup>d</sup>
<b>Rare CNS cancer</b>											
Halkett et al., 2015 [1]	Australia	Cross-sectional	Rare CNS (glioma)	116	Mean 56 (SD 13.3) (range 18–86)	71%	High grade (III and IV)	ns	CRT	Questionnaire; SCNS-SF34, BrTSCNS	H
Langbecker & Yates, 2016 [2]	Australia	Prospective cohort (3 mo)	Rare CNS (diverse brain tumours)	40	Median 55 (42–65)	58%	Low grade (I or II) and high grade (III or IV)	3 mo	Surgery, RT, CT	Questionnaire; SCNS-SF34, BrTSCNS	M
Pijl et al., 2018 [3]	Denmark	Mixed methods (1 y)	Rare CNS (glioma)	30	Median 57.9 (range 29–79)	63%	High grade	1 y	Surgery, CT, RT	Interview	M
Reinhart et al., 2018 [4]	Germany	Cross-sectional	Rare CNS (diverse brain tumours)	172	Mean 51.4 (range 20–84)	44%	Low grade/WHO I/WHO II, WHO III and high grade/WHO IV	ns	ns	Questionnaire; self-developed	M
Renovan et al., 2017 [5]	Germany	Cross-sectional	Rare CNS (glioma)	173	Mean 50.9 (SD 13.9) (range 21–78)	53%	Low grade (I or II) and high grade (III or IV)	Median 33 mo (range 2–287)	Surgery, CT	Questionnaire; SCNS-SF34-G	H
<b>Rare digestive cancer</b>											
Gillespie et al., 2017 [6]	Canada	Cross-sectional	Rare digestive (HPB cancer)	36	72% 61–80 y	58%	56% no metastatic disease	ns	Surgery	Questionnaire; adapted from Papadakis et al.	H
Henselmans et al., 2012 [7]	The Netherlands	Qualitative	Rare digestive (oesophageal cancer)	20	Mean 62	70%	Stage I-IVA	ns	Surgery	Interview	M
Shun et al., 2018 [8]	Taiwan	Prospective cohort (2 mo)	Rare digestive (HCC)	90	Mean 62.5 (SD 10.6)	72%	BCLC stage (B, 1; A, 0) (C, 1; A, 0)	Mean 43.8 mo (SD 36.54) (range 1–149)	RFA, TACE	Questionnaire; SCNS-SF34	H
<b>Endocrine cancer</b>											
Goldfarb & Casillas, 2014 [9]	USA	Cross-sectional	Endocrine (thyroid cancer)	673	40+	17%	ns	Mean 5.11 y (SD 7.02)	Surgery, radioactive iodine	Questionnaire; self-developed	M
M. Henry et al., 2018 [10]	Canada	Qualitative	Endocrine (thyroid cancer)	17	Mean 45.2 (SD 16.5)	29%	ns	ns	Surgery	Interview	H
Husson et al., 2014 [11]	The Netherlands	Cross-sectional	Endocrine (thyroid cancer)	306	Mean 56.4 (SD 14.5)	25%	Stage I-IV	Mean 9.6 y (SD 5.5)	Surgery, RT, radioactive iodine, other (ns)	Questionnaire; EORTC QLQ-INFO25	H
Lee, Park, et al., 2016 [12]	Korea	Qualitative	Endocrine (thyroid cancer)	29	Range 30–69	0%	ns	ns	Surgery	Interview	M
<b>Rare gynaecological cancer</b>											
V.L. Beesley et al., 2013 [13]	Australia		Rare gyn (ovarian cancer)	185	Mean 59 (SD 10)	0%	Early (I-II) and late (III-IV) FIGO	Range 6–12 mo	ns	Questionnaire; SCNS-SF34	H

(Continues)

TABLE 1 (Continued)

Author, year [ref]	Country	Design (follow-up)	Cancer subdomain (type) <sup>a</sup>	N	Age	Gender, % male	Cancer stage	Time since diagnosis	Treatment <sup>b</sup>	Measurement <sup>c</sup>	QA <sup>d</sup>
		Prospective cohort (2 y)									
Kebede & Kebede, 2017 [14]	Ethiopia	Qualitative	Rare gyn (cervical cancer)	12	Mean 42.3 (range 25–67)	0%	ns	ns	RT, CT, CRT, surgery	Interview	L
Long et al., 2016 [15]	South Africa	Qualitative	Rare gyn (cervical cancer)	28	Range 30–73	0%	Stage I–III	ns	RT	Interview	H
Philp et al., 2017 [16]	Australia	Qualitative	Rare gyn (vulvar cancer)	12	Mean 60 (range 51–60)	0%	ns	ns	Surgery	Interview	H
Putri et al., 2018 [17]	Indonesia	Cross-sectional	Rare gyn (cervical, ovarian cancer)	153	118 ≤ 55 y, 35 > 55 y	0%	Stage I–IV	ns	CT	Questionnaire; SCNS-SF34	M
Rietveld et al., 2018 [18]	The Netherlands	Cross-sectional	Rare gyn (ovarian cancer)	185	Mean 63 (range 28–91)	0%	FIGO stage I–IV	Mean 6.2 y (range 1.9–12.3)	Surgery, CT, other (ns)	Questionnaire; CaSUN	H
Vermeer et al., 2015 [19]	The Netherlands	Cross-sectional	Rare gyn (cervical cancer)	343	Mean 48.7 (SD 8.9)	0%	85% FIGO ≤ stage IIA	Ns	RT, CT, surgery, CRT	Questionnaire; self-developed	H
Zeng et al., 2017 [20]	China	Qualitative	Rare gyn (cervical cancer)	31	Range 18–60; female only	0%	Stage IA–IVA	ns	Surgery, CT, RT, CRT	Interview	H
<b>Rare HNC</b>											
Badr et al., 2016 [21]	USA	Qualitative	Rare HNC (diverse)	6	Mean 55.3 (SD 5.9) (range 51–67)	83%	Stage I and IV	ns	RT, surgery, CRT	Interview	H
Brennan et al., 2019 [22]	Canada	Prospective cohort (1 y)	Rare HNC (diverse)	175	Mean 62.5 (SD 10.2)	83%	TNM stage T1–T4 and N0–N3	ns	Surgery, RT, CRT	Questionnaire; adapted from de Bock et al.	H
Chen et al., 2013 [23]	Taiwan	Prospective cohort (6 mo)	Rare HNC (oral cavity cancer)	82	Mean 50.1 (SD 10.83)	98%	Stage II, III and IV	ns	RT, CRT	Questionnaire; CNQ-SF-hn	H
Giuliani et al., 2016 [24]	Canada	Cross-sectional	Rare HNC (diverse)	158	Median 64 (range 19–89)	63%	ns	ns	Surgery, RT, CT	Questionnaire; CaSUN	H
Hansen et al., 2013 [25]	Denmark	Prospective cohort (14 mo)	Rare HNC (ns)	125	ns	ns	ns	ns	ns	Questionnaire; self-developed	H
M. Henry et al., 2020 [26]	Canada	Prospective cohort (3 mo)	Rare HNC (diverse)	145	Mean 63.3 (SD 10.2)	70%	Stage I–IV	ns	CT, RT, surgery	Questionnaire; SCNS-SF34	H

TABLE 1 (Continued)

Author, year [ref]	Country	Design (follow-up)	Cancer subdomain (type) <sup>a</sup>	N	Age	Gender, % male	Cancer stage	Time since diagnosis	Treatment <sup>b</sup>	Measurement <sup>c</sup>	QA <sup>d</sup>
Holm et al., 2012 [27]	Denmark	Prospective cohort (14 mo)	Rare HNC (ns)	125	ns	ns	ns	ns	ns	Questionnaire; self-developed	H
Jabbour et al., 2017 [28]	Australia	Cross-sectional	Rare HNC (diverse)	597	Mean 58, median 62 (range 21–94)	48%	Stage I–IV	ns	Surgery, RT, CRT, radioactive iodine, CT	Questionnaire; self-developed	H
Jansen et al., 2018 [29]	The Netherlands	Cross-sectional	Rare HNC (ns)	283	Mean 70 (SD 9)	84%	ns	ns	Surgery, RT, CRT	Questionnaire; SCNS-SF34, SCNS-HNC	H
Lin et al., 2020 [30]	Taiwan	Cross-sectional	Rare HNC (nasopharyngeal cancer)	145	Mean 49.2 (SD 10.3)	68%	Stage I–IV	ns	59% in-treatment	Interview	L
Lee, Nelson, et al., 2016 [31]	USA	Cross-sectional	Rare HNC (oral cancer)	342	Mean 56.4 (SD 10.5)	53%	Stage I–IV	Mean 5.56 y (SD 4.15)	RT, surgery, CT	Questionnaire; SUNS	H
Manne et al., 2016 [32]	USA	Cross-sectional	Rare HNC (oral, oropharyngeal cancer)	92	Mean 62.1 (SD 8.9) (range 33.4–79.0)	74%	Stage I–IV	Mean 41.3 mo (SD 6.4) (range 23.1–70.0)	Surgery, CT, RT	Questionnaire; SCNS-SF34	H
Moore et al., 2014 [33]	Australia	Qualitative	Rare HNC (diverse)	8	Mean 60 (range 51–60)	88%	ns	Mean 4.25 y (range 1–8)	CRT, RT, surgery, RT	Interview	H
O'Brien et al., 2017 [34]	Ireland	Cross-sectional	Rare HNC (diverse)	583	Mean 62.9 (SD 11.3)	67%	Early and late stage	50% less than 5 y, 50% 5+ y	CT, RT, surgery	Questionnaire; SCNS-SF34	H
Pateman et al., 2015 [35]	Australia	Qualitative	Rare HNC (diverse)	6	Mean 61.3 (range 50–72)	67%	ns	ns	CRT, surgery, RT	Interview	H
Peeters et al., 2018 [36]	The Netherlands	Qualitative	Rare HNC (diverse)	13	Median 60 (range 48–73)	67%	Stage II–IV	ns	CT, RT	Interview	H
Pongthavornkamol et al., 2019 [37]	Thailand	Cross-sectional	Rare HNC (ns)	26	Mean 60.5 (SD 10.1)	81%	ns	2.7 y (SD 1.8)	CT, surgery, RT, hormone therapy	Questionnaire; CaSUN	H
Richardson et al., 2015 [38]	New Zealand	Qualitative (6 mo)	Rare HNC (diverse)	83	Mean 61 (SD 13)	75%	Stage T1–T4	ns	ns	Interview	M
Saroo et al., 2018 [39]	Canada	Mixed methods	Rare HNC (oral, oropharyngeal cancer)	205	Mean 58 (SD 6.5) (range 35–71)	85%	ns	ns	ns	Questionnaire; HaNIQ	H
W.K. So et al., 2019 [40]	China	Mixed methods	Rare HNC (diverse)	285	Mean 55.3 (SD 12.3)	77%	Stage I–III	8.0 mo (SD 3.8)	RT, surgery, CT	Questionnaire; SCNS-SF34	H
			Rare HNC (diverse)	53	Mean 51.8 (SD 10.5)	76%	Stage I–III	8.1 mo (SD 3.4)	Surgery, CT, RT	Interview	H

(Continues)



TABLE 1 (Continued)

Author, year [ref]	Country	Design (follow-up)	Cancer subdomain (type) <sup>a</sup>	N	Age	Gender, % male	Cancer stage	Time since diagnosis	Treatment <sup>b</sup>	Measurement <sup>c</sup>	QA <sup>d</sup>
Sperling et al., 2014 [41]	Denmark	Cross-sectional	Rare HNC (ns)	177	ns	ns	ns	ns	Surgery, RT, CT	Questionnaire; self-developed	H
Wells et al., 2015 [42]	UK	Cross-sectional	Rare HNC (diverse)	280	Mean 64.5 (SD 11.4)	73%	Stage I-IV	9% <1y, 66% 1-4 y, 25% 4-5 y	Combination therapy, surgery	Questionnaire; PCI	H
<b>Rare male genital and urogenital cancer</b>											
Bender et al., 2012 [43]	Canada	Cross-sectional	Rare male genital and urogenital (testicular cancer)	204	Mean 35.6 (SD 10.5)	100%	ns	Mean 2.2 y (SD 1.1)	Surveillance, CT, RT, RPLND	Questionnaire; CaSUN	H
Skooqh et al., 2013 [44]	Sweden	Cross-sectional	Rare male genital and urogenital (testicular cancer)	974	Mean 41, median 39	100%	Stage I-IV	Mean 11 y, median 9 y (range 3-26)	Surgery, RT, other	Questionnaire; self-developed	M
Smith et al., 2013 [45]	Australia	Cross-sectional	Rare male genital and urogenital (testicular cancer)	244	Mean 38.3 (SD 10.3) (range 21-68)	100%	36% metastatic	ns	Surveillance, surgery, RT, CT	Questionnaire; CaSUN	H
<b>NET</b>											
V.L. Beesley et al., 2018 [46]	Australia	Cross-sectional	NET (diverse sites)	111	39% <60 y, 61% 60+y	56%	Grade 1-2	Range 2 mo-27 y	Surgery, RT, hormone therapy, CT, targeted therapy	Questionnaire; SCNS-SF34	H
Feinberg et al., 2013 [47]	Canada	Qualitative	NET (diverse sites)	18	Median 63 (range 45-77)	54%	ns	Range 0-12 y	ns	Interview	H
Singh et al., 2017 [48]	Canada	Cross-sectional	NET (diverse sites)	1928	Mean 56.8	26%	Grade 1-3, 58% metastatic disease at time of diagnosis	Mean 5.2 y	Surgery	Questionnaire; self-developed	M
Wolin et al., 2017 [49]	Australia	Cross-sectional	NET (diverse sites)	758	Mean 57	28%	Grade 1-3	Mean 5.3 y	Surgery, CT, other	Questionnaire; self-developed	M
<b>Rare skin cancer/eye melanoma</b>											
Williamson et al., 2018 [50]	USA	Prospective cohort (3 mo)	Rare skin/eye melanoma (eye melanoma)	107	Mean 59.0 (SD 12.8)	54%	ns	1 week and 3 mo	RT	Questionnaire; CNQ-SF	H
<b>Rare haematological cancer</b>											
Boland et al., 2014 [51]	UK	Cross-sectional	Rare haema (MM)	32	Median 60 (range 41-71)	53%	ns	Median 5.5 y (range 2-12)	SCT, antineoplastic therapy	Questionnaire; SPARC	M
Molassiotis et al., 2011 [52]	UK	Cross-sectional	Rare haema (MM)	132		61%	ns		CT, SCT	Questionnaire; CaSUN	H



TABLE 1 (Continued)

Author, year [ref]	Country	Design (follow-up)	Cancer subdomain (type) <sup>a</sup>	N	Age	Gender, % male	Cancer stage	Time since diagnosis	Treatment <sup>b</sup>	Measurement <sup>c</sup>	QA <sup>d</sup>
Nørskov et al., 2019 [53]	Denmark	Qualitative	Rare haema (AL)	18	Mean 62 (SD 8.8) (range 35–83)	44%	ns	Mean 67.7 mo (SD 52.2) (range 12–269)	CT	Interview	H
Oberoi et al., 2017a [54]	Australia	Prospective cohort (15 mo)	Rare haema (DLBCL, MM)	414	Mean 64.4 (SD 10.44) MM, mean 63.3 (SD 11.38) DLBCL	57%	ns	Mean 6.9 mo (SD 1.9) MM; Mean 6.5 mo (SD 1.9) DLBCL	RT, CT, SCT; RT, CT	Questionnaire; SCNS-SF34	H
Oberoi et al., 2017b [55]	Australia	Prospective cohort (15 mo)	Rare haema (DLBCL, MM)	414	Mean 63.82 (SD 11.08) DLBCL	57%	ns	Mean 6.71 mo (SD 1.98)	CT, RT, SCT	Questionnaire; SCNS-SF34	H
Yu et al., 2017 [56]	China	Cross-sectional	Rare haema (AL)	311	Median 36.4 (SD 14.7) (range 18–78)	60%	ns	81.2% <12mo, 5.1% 13–18mo, 12.7% >18mo	ns	Questionnaire; SCNS-SF34	H
<b>Male breast cancer</b>											
Bootsma et al., 2020 [57]	The Netherlands	Mixed methods	Male breast cancer	12	Mean 66 (range 49–88)	100%	8% in situ, 92% invasive	Range 2–25y	Surgery, RT, CT, anti-hormonal therapy	Interview	H
			Male breast cancer	77	Mean 66.9 (10.9) (range 44–89)	100%	7% in situ, 94% invasive	Mean 66.9 mo (SD 10.8) (range 41–86)	Surgery, RT, CT, anti-hormonal therapy, immuno-therapy	Questionnaire; EORTC QLQ-BR23, B-force, EORTC QLQ-INFO25	H

Abbreviations: H, high quality; Mo, months; M, medium quality; ns, not specified; QA, quality assessment; SD, standard deviation; y, year

<sup>a</sup>Cancer subdomain: Rare haema, rare haematological cancer; rare HNC, rare head and neck cancer; rare gyn, gynaecological cancer; NET, neuroendocrine tumours.

Cancer subdomains are presented in concordance with the domains of EURACAN and EuroBloodNet, except for male breast cancer.

Tumour types: AL, acute leukaemia; DLBCL, diffuse large B-cell lymphoma; HCC, hepatocellular carcinoma; HPB cancer, hepatopancreato-biliary cancer; MM, multiple myeloma.

<sup>b</sup>Treatment: Given treatments are listed here, but it can also involve combinations of these treatments. CRT, chemoradiotherapy; CT, chemotherapy; RFA, radiofrequency ablation; RLND, retroperitoneal lymph node dissection; RT, radiotherapy; SCT, stem cell transplantation; TACE, transarterial chemoembolization.

<sup>c</sup>Measurement: BrTSCNS, Brain Tumour Specific Supportive Care Needs Scale; CASUN, Cancer Survivors Unmet Needs Instrument; CNQ-SF, Cancer Needs Questionnaire Short Form; EORTC QLQ-BR23, EORTC Quality of Life Questionnaire – breast cancer specific; EORTC QLQ-INFO25, EORTC Quality of Life Questionnaire – Information Module; HaNIQ, Head and Neck Information Needs Questionnaire; PCI, Patient Concerns Inventory; SCNS-HNC, Supportive Care Needs Survey-Head & Neck Cancer; SCNS-SF, Supportive Care Needs Survey-Short Form; SPARC, Sheffield Profile for Assessment and Referral for Care; SUNS, Survivors Unmet Needs Survey.

<sup>d</sup>QA, quality assessment; H, high quality; M, medium quality.

**TABLE 2** Unmet supportive care needs in rare cancer patients during the diagnostic, treatment and posttreatment phase of their disease trajectory

Unmet needs (# studies)	Items	Prevalence rates per phase [ref]		
		Diagnostic	Treatment	Posttreatment
Rare CNS cancer				
Supportive care (1)	General: At least one unmet need		91% [1]	
Psychological (3)	General	45.8 (25.1) <sup>a</sup> [1]	35.6 (27.5) <sup>a</sup> [1]	
	Fears about the cancer spreading		56% [5]	
	Uncertainty about the future		45% [5]	
		27% [2]	30–44% [1, 2]	
	Worry that results of treatment are beyond your control		37% [5]	
	Learning to feel in control of your situation		32% [5]	
	Feelings about death and dying		31% [5]	
	Feeling down or depressed		28% [5]	
			28% [1]	
	Concerns about the worries of those close to you		27% [5]	
		27% [2]	39% [1]	
	Feelings of sadness		26% [5]	
			24% [1]	
	Anxiety		25% [5]	
			28–47% [1, 2]	
Physical and daily living (3)	Keeping a positive outlook		24% [5]	
	General	47.9 (26.3) <sup>a</sup> [2]	38.9 (27.6) <sup>a</sup> [2]	
	Lack of energy/tiredness		31% [5]	
		24% [2]	26–50% [1, 2]	
	Pain		19% [5]	
	Not being able to do the things you used to do		18% [5]	
		25% [2]	36–50% [1, 2]	
	Work around the home		17% [5]	
	Feeling unwell most of the time		15% [5]	
			24% [1]	
Healthcare system and information (4)	General	37.9 (17.4) <sup>a</sup> [2]	27.8 (16.5) <sup>a</sup> [2]	
	Information about medical and supportive treatment		30% [4]	
	Information about things to help get well		28% [5]	
			24% [1]	
	Access to professional counselling		28% [5]	
	Information about cancer under control or diminishing		24% [5]	
			23% [1]	
	Personalised treatment		18% [5]	
	Information about test results as soon as feasible		17% [5]	
			27% [1]	
	Information about important aspects of care		17–19% [4, 5]	
	Explanations of tests		17% [5]	
	One member of hospital staff to talk to		16–25% [4, 5]	
	Information about benefits and side-effects of treatment		16% [5]	
	Information about managing illness at home		15–20% [4, 5]	
	Information about legal issues, funding, and career		15% [4]	

TABLE 2 (Continued)

Unmet needs (# studies)	Items	Prevalence rates per phase [ref]		
		Diagnostic	Treatment	Posttreatment
Sexuality (1)	General	34.8 (28.6) <sup>a</sup> [2]	29.2 (35.3) <sup>a</sup> [2]	
	Changes in sexual feelings		23% [5]	
	Changes in sexual relationships		18% [5]	
	Information about sexual relationships		14% [5]	
Transportation (1)	Easy car parking at the hospital		36% [1]	
	Access to transport service to and from the hospital		10% [1]	
Disease specific (2)	General	32.5 (19.6) <sup>a</sup> [2]	21.9 (16.1) <sup>a</sup> [2]	
	Not feeling like the same person before the tumour	50% [2]	47% [2]	
	Information on developments in research and treatment		28% [1]	
	Physical side effects from tumour and/or treatment		24% [1]	
	Financial assistance/advice		21% [1]	
	Testing and advice about mental thinking abilities		17% [1]	
	Rehabilitation services		16% [1]	
	Internet/email to receive information and/or support		16% [1]	
	Legal assistance/advice		14% [1]	
	Change in mental or thinking ability		14% [1]	
	Talking to other people with a similar experience		12% [1]	
	Allowance for travel, treatment, equipment expenses		22% [1]	
	General	30.4 (13.2) <sup>a</sup> [2]	13.0 (16.4) <sup>a</sup> [2]	
	Hospital staff attending promptly to physical needs		20% [5]	
	Reassurance that the way of feeling is normal		14% [5]	
Economic (1)	Acknowledgement/sensitivity to emotional needs		13% [5]	
	More choice about which cancer specialists to see		11% [5]	
Patient care and support (1)	More choice about which hospital to attend		8% [5]	
Rare digestive cancer				
Healthcare system and information (2)	General		28.9 (15.0) <sup>a</sup> [8]	16.2 (12.6) <sup>a</sup> [8]
	Information about likelihood of a cure	94% [6]		
	Information about dealing with pain	92% [6]		
	Information about whether family members are at risk of this cancer	91% [6]		
	Information about surgical procedure	91% [6]		
	Information about possible side effects after surgery	89% [6]		
	Information about caring for yourself after surgery	89% [6]		
	Information about managing possible bowel changes	89% [6]		
	General information about your cancer	86% [6]		

(Continues)

TABLE 2 (Continued)

Unmet needs (# studies)	Items	Prevalence rates per phase [ref]		
		Diagnostic	Treatment	Posttreatment
	Information about results of medical tests	86% [6]		
	Information about managing changes in daily activities	86% [6]		
	Information about likely outcomes of surgery	86% [6]		
	Information about symptoms	83% [6]		
	Information about other types of treatment	83% [6]		
	Information about possible risks associated with surgery	82% [6]		
	Information about possible side effects of other treatment	82% [6]		
	Information about coping with fears of recurrence	81% [6]		
	Information about managing nausea and vomiting	80% [6]		
	Information about managing feeling tired	78% [6]		
	Information about preparing for surgery	77% [6]		
	Information about drug coverage options	74% [6]		
	Information about how your relationship with your partner may be affected	73% [6]		
	Information about managing illness at home	72% [6]		
	Information about legal issues	71% [6]		
	Information about returning to work	71% [6]		
	Information about help from healthcare workers	69% [6]		
Physical and daily living (1)	General		27.4 (21.7) <sup>a</sup> [8]	20.1 (18.1) <sup>a</sup> [8]
Psychological (1)	General		33.0 (18.1) <sup>a</sup> [8]	15.1 (18.1) <sup>a</sup> [8]
Patient care and support (1)	General		22.1 (20.7) <sup>a</sup> [8]	4.5 (14.5) <sup>a</sup> [8]
Sexuality (1)	General		3.4 (9.6) <sup>a</sup> [8]	0.6 (4.0) <sup>a</sup> [8]
<b>Endocrine cancer</b>				
Psychological (1)	General	>80% [9]		
Physical and daily living (1)	General	>65 [9]		
Healthcare system and information (1)	General			34% [11]
	Information about complications and long-term effects of treatment and medication use			67% [11]
	Information about aftercare and rehabilitation options			19% [11]
	General information about thyroid cancer			18% [11]
	Information about the cause of their cancer			11% [11]
Patient care and support (1)	General	<30% [9]		
<b>Rare gynaecological cancer</b>				
Supportive care (2)	General: At least one unmet need		59% [13]	65% [13]
Patient care and support (3)	General		75% [17], 15 <sup>b</sup> [13]	
	Addressing of any complaints			61% [18]

TABLE 2 (Continued)

Unmet needs (# studies)	Items	Prevalence rates per phase [ref]		
		Diagnostic	Treatment	Posttreatment
	Knowledge about intercollegial consultation between caregivers			52% [18]
	Access to healthcare services when required			47% [18]
	Best medical care			43% [18]
	Access to complementary or alternative therapy services			34% [18]
Physical and daily living (2)	General		80% [17], 15 <sup>b</sup> [13]	15 <sup>b</sup> [13]
	Lack of energy/tiredness		18% [13]	15% [13]
Sexuality (3)	General		35% [17], 8 <sup>b</sup> [13]	51% [19]
Psychological (2)	Information about sexuality and cancer			83% [19]
	General		72% [17], 25 <sup>b</sup> [13]	23 <sup>b</sup> [13]
	Fears about the cancer spreading		25% [13]	21% [13]
	Concerns about the worries of those close to you		20% [13]	18% [13]
Healthcare system and information (2)	Uncertainty about the future		19% [13]	19% [13]
	Anxiety		17% [13]	15% [13]
	Worry that results of treatment are beyond your control		16% [13]	18% [13]
	Keeping a positive outlook		15% [13]	10% [13]
	Feelings of sadness		15% [13]	12% [13]
	General		71% [17], 20 <sup>b</sup> [13]	11 <sup>b</sup> [13]
	Information about things to help get well		20% [13]	6% [13]
	Information about cancer under control or diminishing		16% [13]	5% [13]
	One member of hospital staff to talk to		16% [13]	3% [13]
	Information about test results as soon as feasible		16% [13]	16% [13]
	Personalised treatment		15% [13]	4% [13]
<b>Rare HNC</b>				
Supportive care (8)	General: At least one unmet need		62% [25]	
		40.9 (12.4) <sup>a</sup> [23]	39.7(13.3) <sup>a</sup> [23]	30.2 (9.5) <sup>a</sup> [23], 11.0 (10.4) <sup>a</sup> [37], 61–82% [24, 26, 29, 39, 42]
Disease-specific (5)	General	48.8 (19.5) <sup>a</sup> [23]	46.3 (19.4) <sup>a</sup> [23]	38.7 (13.4) <sup>a</sup> [23], 53% [29]
	Information on side effects of oral cancer treatment			59% [32]
	Information on oral cancer follow-up tests			58% [32]
	Information on prompting oral cancer symptoms			58% [32]
	Information on problems with chewing and/or swallowing			34% [32]
	Information on getting or maintaining insurance			33% [32]
	Information on maintaining good oral health			33% [32]
	Information on managing anxiety about recurrence			32% [32]

(Continues)

TABLE 2 (Continued)

Unmet needs (# studies)	Items	Prevalence rates per phase [ref]		
		Diagnostic	Treatment	Posttreatment
	Problems with social eating			30% [29]
	Problems with mobility of neck or shoulders			28% [29]
	Difficulty eating			26% [29]
	Information on dealing with changes in appearance			24% [32]
	Problems with coughing			23% [29]
	Information on dealing with social life			20% [32]
	Information on dealing with effects on relationships			19% [32]
	Feeling better about appearance			19% [26]
	Problems with chewing and/or swallowing			18–26% [29, 42]
	Difficulty speaking			17–28% [29, 42]
	Problems with hearing			17% [29]
	Problems with chewing/eating			14% [42]
	Information on nutrition			13–41% [29, 32]
	Problems with dry mouth and/or sticky saliva			12–27% [29, 42]
	Loss of appetite			12% [29]
	Problems with weight			11–19% [29, 42]
	Pain in head and neck/elsewhere			10% [42]
	Problems with taste and olfaction			9–35% [29, 42]
	Problems with breathing			9–24% [29, 42]
	Problems with memory			9% [42]
	Oral hygiene/dental health			8–18% [29, 42]
	Smoking cessation			5% [26]
	Alcohol cessation			2% [26]
Healthcare system and information (8)	General	48.9 (21.0) <sup>a</sup> [23]	38.5 (21.6) <sup>a</sup> [23]	31.3 (14.0) <sup>a</sup> [23], 21–63% [26,29, 34,40]
	Routine blood tests			69% [22]
	Imaging			66% [22]
	Information on risk factors			57% [22]
	Information on heredity			51% [22]
	Information on fatigue/pain			43% [22]
	Information of long-term effects of treatment	33% [28]		84% [22]
	Information on type and stage of cancer	31% [28]		
	Information on prognosis	28% [28]		95% [21]
	Information on staying healthy		23% [28]	27–67% [22, 28]
	Information on treatment options	26% [28]		
	Information on changes in eating and speaking			26% [28]
	Information on length of recovery			25% [28]
	Ongoing case manager to go to for services			21% [24]
	Up-to-date information			21% [24]

TABLE 2 (Continued)

Unmet needs (# studies)	Items	Prevalence rates per phase [ref]		
		Diagnostic	Treatment	Posttreatment
Psychological (8)	Understandable information			21% [24]
	Information on patient support groups		20% [28]	
				36% [22]
	Information on coping with stress and anxiety		20% [28]	
	Information on nutrition and maintaining weight		20% [28]	
	Information on coping with side effects of treatment			20-30% [23, 26]
	Information on coping with cosmesis			20% [28]
	One member of hospital staff to talk to			18-41% [26, 29, 32, 40]
	Information on effects of treatment on ability to work		18% [28]	
	Information on psychosexual health	17% [28]		
	Information relevant for family and/or partners			16% [24]
	Information about things to help get well			15-41% [26, 29, 32, 40]
	Information about cancer under control or diminishing			15-33% [26, 29, 40]
	Information about test results as soon as feasible			15-29% [26, 29, 32, 40]
	Information about benefits and side-effects of treatment			14-24% [26, 29, 32, 40]
	Explanations of tests			14-22% [26, 29, 32, 40]
	Access to professional counselling			14-17% [26, 29, 32, 40]
	Information about important aspects of care			13-16% [26, 29, 32, 40]
	Information on how to support family and/or partner			13% [24]
	Brochures about services and benefits for patients			13% [40]
	Personalised treatment			11-20% [26, 29, 40]
	Treatment in a physically pleasant hospital			11-17% [26, 29, 32, 40]
	Information about managing illness at home			11-16% [29, 32, 40]
	General	43.2 (15.1) <sup>a</sup> [23]	43.0 (15.8) <sup>a</sup> [23]	33.7 (15.4) <sup>a</sup> [23], 32-57% [26, 29, 34, 40]
	Concerns about cancer coming back			26% [24, 42]
	Fears about the cancer spreading			19-37% [26, 29, 32, 34, 40]
	Reducing stress			17-22% [24, 34]
	Coping with others not acknowledging the impact of cancer had on their lives			17% [24]
	Concerns about the worries of those close to you			16-28% [26, 29, 34, 40]
	Coping with changes to belief about life			16% [24]
	Finding meaning and purpose in life			16% [26]
	Dealing with expectations as a "cancer survivor"			15% [24]
	Worry that results of treatment are beyond your control			13-41% [26, 29, 34, 40]
	Uncertainty about the future			13-36% [24, 26, 29, 32, 34, 40]
	Learning to feel in control of your situation			13-27% [26, 29, 40]

(Continues)



TABLE 2 (Continued)

Unmet needs (# studies)	Items	Prevalence rates per phase [ref]		
		Diagnostic	Treatment	Posttreatment
Physical and daily living (9)	Making life count			13% [24]
	Anxiety			12–21% [26, 29, 32, 40, 42]
	Feeling down or depressed			10–23% [26, 29, 32, 34, 40, 42]
	Feelings about death and dying			9–16% [26, 29, 32, 40]
	Keeping a positive outlook			8–32% [26, 29, 32, 40]
	Feelings of sadness			7–23% [26, 29, 34, 40]
	General			38% [25]
		30.2 (11.0) <sup>a</sup> [23]	34.9 (13.3) <sup>a</sup> [23]	26.7 (8.3) <sup>a</sup> [23], 31–55% [26, 27, 29, 34, 39, 41]
	Not being able to do the things you used to do			14–28% [26, 29, 32, 34, 40]
	Problems with sleeping			14% [42]
	Lack of energy/tiredness			11–22% [26, 29, 32, 34, 40, 42]
	Pain			9–38% [26, 29, 32, 34, 40]
	Feeling unwell most of the time			9–26% [26, 29, 32, 40]
	Work around the home			3–20% [26, 29, 32, 40]
Economic (6)	General			31% [25]
				50% [27]
	Allowance for travel, treatment, equipment expenses			24% [40]
	Worry about earning money			23% [31]
	Obtaining life and/or travel insurance			21% [24]
	Help with financial support and/or state benefits			14–18% [24, 42]
	General	34.8 (18.4) <sup>a</sup> [23]	37.2 (18.8) <sup>a</sup> [23]	25.7 (14.2) <sup>a</sup> [23], 14–35% [26, 29, 34, 40]
	Knowing that all my doctors talk to each other to coordinate my care			31% [24]
	Access to healthcare services when required			23% [24]
	Feeling like managing health together with medical team			22% [24]
Patient care and support (7)	Managing side effects and/or complications of treatment			20% [24]
	Addressing of any complaints			20% [24]
	Best medical care			20% [24]
	Changes in quality of life as a result of cancer			19% [24]
	Provision of emotional support			19% [24]
	Access to complementary or alternative therapy services			19% [24]
	24-hr telephone support and cancer advisory service			19% [40]
	Adjusting to the body changes			17% [24]
	Talking to other people with a similar experience			17–21% [24, 31]
	Handling cancer in social and/or work situations			15% [24]
	Moving on with life			14% [24]

TABLE 2 (Continued)

Unmet needs (# studies)	Items	Prevalence rates per phase [ref]		
		Diagnostic	Treatment	Posttreatment
	Reassurance that the way of feeling is normal			10–16% [26, 29, 32, 40]
	Impact of cancer on relationship with partner			12% [24]
	Help with developing new relationships after cancer			12% [24]
	More choice about which cancer specialists to see			10–24% [26, 29, 40]
	Help with access to legal services			10% [24]
	Acknowledgement/sensitivity to emotional needs			9–16% [26, 29, 32, 40]
	Hospital staff attending promptly to physical needs			9–15% [26, 29, 32, 40]
	Help with exploring spiritual beliefs			9% [24]
	Assistance with getting and/or maintaining employment			7% [24]
	Help with having a family due to fertility problems			5% [24]
	More choice about which hospital to attend			4–13% [26, 29, 32, 40]
Family-oriented (2)	General			32% [25]
				42% [27]
Communication (1)	General	35.8 (18.7) <sup>a</sup> [23]	33.2 (16.9) <sup>a</sup> [23]	31.3 (13.9) <sup>a</sup> [23]
Emotional (4)	General			47% [25]
				61% [42]
	Dealing with feeling tired			35% [27]
	Dealing with changes in physical ability			23% [31]
	Dealing with feeling stressed			22% [31]
	Dealing with feeling worried/anxious			21% [31]
	Dealing with not feeling able to set future goals			21% [31]
	Dealing with losing confidence in abilities			20% [31]
	Coping with having a bad memory or lack of focus			20% [31]
Work-related (2)	General			28% [25]
				29% [27]
Sexuality (9)	General			38% [25]
				5–64% [26, 27, 29, 34, 40]
	Information about sexual relationships			9–15% [26, 29, 40]
	Changes in sexual feelings			3–20% [26, 29, 40]
	Changes in sexual relationships			1–19% [24, 26, 29, 32, 40, 42]
Transportation (2)	Easy car parking at the hospital			8–23% [24, 40]
	Transport service to and from hospital			7% [40]
<b>Rare male genital and urogenital cancer</b>				
Supportive care (2)	General: At least one unmet need			63–66% [43, 45]
Healthcare system and information (1)	Information about crisis and stress after diagnosis	59% [49]		
Psychological (3)	Access to psychological counselling	42% [49]		
	Reducing stress			24–27% [43, 45]
	Concerns about cancer coming back			16–25% [43, 45]

(Continues)

TABLE 2 (Continued)

Unmet needs (# studies)	Items	Prevalence rates per phase [ref]		
		Diagnostic	Treatment	Posttreatment
Economic (2)	Coping with others not acknowledging the impact of cancer had on their lives			13–26% [43, 45]
	Dealing with expectations as a “cancer survivor”			12% [45]
	Help with financial support and/or state benefits			16–28% [43, 45]
	Help with obtaining life and/or travel insurance			15% [45]
Patient care and support (2)	Adjusting to the body changes			28% [43]
	Access to complementary or alternative therapy services			22% [43]
	Access to community support services			21% [43]
	Handling cancer in social and/or work situations			21% [43]
Sexuality (1)	Changes in quality of life as a result of cancer			19% [43]
	Moving on with life			19% [43]
	Talking to other people with a similar experience			12–20% [43, 45]
	Provision of emotional support			12% [45]
Transportation (1)	Changes in sexual relationships			18% [45]
	Easy car parking at the hospital			12% [45]
<b>NET</b>				
Supportive care (1)	General: At least one unmet need			63% [46]
Healthcare system and information (3)	General			28% [46]
	Clearer information on longer-term impact of disease	66% [48]		
	More immediate access to NET experts	63% [48]		
	Wider range of treatment options	60% [48]		
			68% [49]	
	Better direction on where to find NET information	58% [49]		
	Better access to NET experts/centre of expertise	56% [48]		
			62% [49]	
	Clearer idea of treatment options available	50% [49]		
	Information about/opportunity to participate in trials	48% [48]		
	More knowledgeable NET medical providers	47% [48]		
		58% [49]	59% [5149]	
	Understanding test results	46% [49]		
	More treatments available “in my country that I see in other countries”	45% [48]		
			60% [49]	
	Better coordinated/aligned NET medical team	45% [48]		
		39% [49]	50% [49]	
	Immediate transfer to a centre of expertise	42% [49]		
	More information brochures from NET medical providers	37% [48]		
			37% [49]	
	Clearer information on the diagnostics tests	32% [49]		

TABLE 2 (Continued)

Unmet needs (# studies)	Items	Prevalence rates per phase [ref]		
		Diagnostic	Treatment	Posttreatment
Physical and daily living (1)	General			38% [46]
	Lack of energy/tiredness			33% [46]
	Not being able to do the things you used to do			24% [46]
	Work around the home			17% [46]
Patient care and support (2)	General			11% [46]
	Immediate access to NET patient support groups	48% [49]		
	Provision of emotional support	31% [49]		
	More positive outlook from the HCPs during diagnosis	18% [49]		
Transportation (1)	Less travel to different healthcare professionals	27% [49]		
Psychological (1)	General			44% [46]
	Feelings of sadness			28% [46]
	Fears about the cancer spreading			26% [46]
	Concerns about the worries of those close to you			26% [46]
	Uncertainty about the future			26% [46]
	Feeling down or depressed			19% [46]
	Anxiety			17% [46]
	Worry that results of treatment are beyond your control			17% [46]
Sexuality (1)	General			17% [46]
<b>Rare skin cancer/eye melanoma</b>				
Supportive care (1)	General: At least one unmet need	99% [50]	86% [50]	
Healthcare system and information (1)	General	92% [50]	77% [50]	
	Information about cancer remission	87% [50]	70% [50]	
	Information about things to help get well	87% [50]	66% [50]	
	Information about test results as soon as feasible	86% [50]	65% [50]	
	Information about the odds of treatment success	83% [50]	66% [50]	
	Information about benefits and side-effects of treatment	80% [50]	63% [50]	
	Explanations of tests	79% [50]	61% [50]	
Psychological (1)	General	93% [50]	69% [50]	
	Fears about the cancer spreading	85% [50]	61% [50]	
	Fears about further physical disability or deterioration	73% [50]	48% [50]	
	Fears about the possible pain and suffering	72% [50]	41% [50]	
Patient care and support (1)	General	70% [50]	52% [50]	
Physical and daily living (1)	General	67% [50]	54% [50]	
Communication (1)	General	49% [50]	23% [50]	

(Continues)

TABLE 2 (Continued)

Unmet needs (# studies)	Items	Prevalence rates per phase [ref]		
		Diagnostic	Treatment	Posttreatment
Rare haematological cancer				
Supportive care (1)	General: At least one unmet need			27% [52]
Healthcare system and information (4)	General		56% [56]	
		24–42% [54, 55]		32–42% [54]
	Information about test results as soon as feasible		66% [56]	
	Information about things to help get well		64% [56]	
	Information about cancer under control or diminishing		63% [56]	
	Information about benefits and side-effects of treatment		62% [56]	
	Information about managing illness at home		60% [56]	
	One member of hospital staff to talk to		60% [56]	
	Explanations of tests		56% [56]	
	Provision of materials about nursing service for disease		53% [56]	
	Access to professional counselling		52% [56]	
	Treatment in a physically pleasant hospital		40% [56]	
	Personalised treatment		35% [56]	
	Ongoing case manager to go to for services			28% [52]
	Information relevant for family and/or partners			24% [52]
	Information on how to support family and/or partner			21% [52]
Psychological (5)	General		46% [56]	
		30–55% [54, 55]		42–47% [54]
	Concerns about the worries of those close to you		64% [56]	
				40% [51]
	Fears about the cancer spreading		63% [56]	
				30% [52]
	Worry that results of treatment are beyond your control		61% [56]	
	Uncertainty about the future		53% [56]	
				24% [52]
	Feeling down or depressed		46% [56]	
	Feelings of sadness		42% [56]	
	Anxiety		42% [56]	
				9% [51]
	Feelings about death and dying		34% [56]	
	Learning to feel in control of your situation		32% [56]	
	Maintaining decent appearance		26% [56]	
	Feeling like everything is an effort			25% [51]
	Reducing stress			25% [52]
	Dealing with expectations as a “cancer survivor”			23% [52]
	Coping with others not acknowledging the impact of cancer had on their lives			21% [52]

TABLE 2 (Continued)

Unmet needs (# studies)	Items	Prevalence rates per phase [ref]		
		Diagnostic	Treatment	Posttreatment
Physical and daily living (4)	General		37% [56]	
			30–48% [54,55]	32–42% [54]
	Feeling unwell most of the time		45% [56]	
	Lack of energy/tiredness		42% [56]	
				50% [51]
	Not being able to do the things you used to do		40% [56]	
	Daytime somnolence			37% [51]
	Pain		31% [56]	
				44% [51]
	Problems sleeping at night			31% [51]
	Work around the home		28% [56]	
				25% [51]
	Worries about long-term effects of treatment			28% [51]
	Feeling weak			25% [51]
Patient care and support (5)	General		36% [56]	
			11–24% [54,55]	20–34% [54]
	More choice about which cancer specialists to see		40% [56]	
	More choice about which hospital to attend		38% [56]	
	Acknowledgement/sensitivity to emotional needs		38% [56]	
	Managing side effects of treatment			34% [51]
	Hospital staff attending promptly to physical needs		33% [56]	
	Reassurance that the way of feeling is normal		33% [56]	
	Knowing that all my doctors talk to each other to coordinate my care			24% [52]
	Talking to other people with a similar experience			23% [52]
	Changes in quality of life as a result of cancer			21% [52]
	Provision of emotional support			6% [51]
	Help with personal affairs			6% [51]
Sexuality (5)	General		16% [56]	
			9–23% [54,55]	14–17% [54]
	Information about sexual relationships		18% [56]	
	Changes in sexual relationships		15% [56]	
				31% [51]
	Changes in sexual feelings		15% [56]	
Economic (1)	Obtaining life and/or travel insurance			21% [52]
	Help with financial support and/or state benefits			39% [52]
Transportation (1)				21% [52]
	Easy car parking at the hospital			40% [52]

(Continues)

TABLE 2 (Continued)

Unmet needs (# studies)	Items	Prevalence rates per phase [ref]		
		Diagnostic	Treatment	Posttreatment
Male breast cancer				
Healthcare system and information (1)	Information on acute effects of treatment		65% [57]	
	Information on late effects of treatment			56% [57]
	Information on psychosocial problems		20% [57]	
	Information on sexuality		17-23% [57]	23-25% [57]
	Information on treatment side effects		17-18% [57]	23% [57]
Disease-specific (1)	Male-specific information on symptoms and diagnosis, treatment, side effects, psychosocial impact, peer group support, and research			
	Diagnostic			
	Treatment			
	Post-treatment			
	Diagnostic/treatment			
	Treatment/posttreatment			
	Diagnostic/treatment/posttreatment			

<sup>a</sup>Presented as Mean (SD).

<sup>b</sup>Presented as Median.

need domains are presented in order from most prevalent unmet needs to least prevalent unmet needs, based on mean percentages.

### 3.4 | Unmet needs of patients with rare CNS cancer

Four studies (Halkett et al., 2015; Langbecker & Yates, 2016; Reinert et al., 2018; Renovanz et al., 2017) were identified that quantified unmet supportive care needs of persons diagnosed with rare CNS cancer (i.e., primary brain tumour). Unmet needs were highest in the psychological domain (24–56%), followed by the physical and daily living domain (15–50%) and the healthcare system and information domain (15–30%), and these needs were reported throughout the whole disease trajectory. The highest score was found for needing help, in the diagnostic phase, with ‘uncertainty about the future’ (27%), in the treatment phase with ‘not being able to do the things you used to do’ (50%) and with ‘fears about the cancer spreading’ (56%) over all phases. In the treatment phase, 91% of the patients reported at least one unmet need. No unmet needs of these rare cancer patients were specifically reported in the posttreatment phase only.

### 3.5 | Unmet needs of patients with rare digestive cancer

Two studies (Gillespie et al., 2017; Shun et al., 2018) reported the presence of unmet needs of patients with rare digestive cancer. The unmet needs were most prevalent in the healthcare system and information domain (69–94%; mean 28.9 and 16.2, in, respectively,

the treatment and posttreatment phase), followed by the physical and daily living domain (mean 27.4 and 20.1, in, respectively, the treatment and posttreatment phase) and the psychological domain (mean 33.0 and 15.1, in, respectively, the treatment and posttreatment phase), and these needs were reported throughout the whole disease trajectory. In the diagnostic and treatment phase, the need for ‘information about the likelihood of a cure’ (94%) had the highest score. No specific unmet needs were reported in one of the separate phases only.

### 3.6 | Unmet needs of patients with endocrine cancer

In two studies (Goldfarb & Casillas, 2014; Husson et al., 2014) in this review, unmet needs of patients with endocrine cancer were described. Unmet needs were highest in the psychological domain (>80%), followed by the physical and daily living domain (>65%) and the healthcare system and information domain (11–67%), and these needs were reported throughout the whole disease trajectory. In the posttreatment phase, ‘information about complications and long-term effects of treatment and medication use’ (67%) was the most prominent need. No specific unmet needs of these rare cancer patients were reported in the diagnostic and treatment phase only.

### 3.7 | Unmet needs of patients with rare gynaecological cancer

Three studies (V.L. Beesley et al., 2013; Putri et al., 2018; Rietveld et al., 2018) reported the presence of unmet supportive care needs of



patients with rare gynaecological cancer. Patients most frequently reported unmet needs in the patient care and support domain (34–75%), followed by the sexuality domain (35–83%) and the physical and daily living domain (15–80%), and these needs were reported in the treatment and posttreatment phase. In the treatment phase, needing help with ‘fears about cancer spreading’ (25%) was the highest ranked need, and in the posttreatment phase, ‘information about sexuality and cancer’ (83%) scored highest. Of all patients with rare gynaecological cancer, 59% reported at least one unmet need in the treatment phase, and 65% at least one unmet need in the posttreatment phase. No unmet needs of these rare cancer patients were specifically reported in the diagnostic phase only.

### 3.8 | Unmet needs of patients with rare HNC

Fifteen studies (Brennan et al., 2019; Chen et al., 2013; Giuliani et al., 2016; Hansen et al., 2013; M. Henry et al., 2020; Holm et al., 2012; Jabbour et al., 2017; Jansen et al., 2018; M.S. Lee, Nelson, et al., 2016; Manne et al., 2016; O'Brien et al., 2017; Pongthavornkamol et al., 2019; W.K. So et al., 2019; Sperling et al., 2014; Wells et al., 2015) assessed unmet needs of patients with rare HNC. Unmet needs were highest in the disease-specific domain (2–59%), followed by the healthcare system and information domain (11–84%) and the psychological domain (7–41%), and these needs were reported throughout the whole disease trajectory. The highest need score, in the diagnostic phase, was reported for ‘information on type and stage of cancer’ (31%), in the treatment phase for ‘information on staying healthy’ (23%), in the posttreatment phase for ‘information on prognosis’ (95%) and for ‘information of long-term effects of treatment’ (33%) over all phases. During the treatment and posttreatment phase, 64% of the rare HNC patients reported at least one unmet need, and 61–82% reported at least one unmet need during the posttreatment phase.

### 3.9 | Unmet needs of patients with rare male genital and urogenital cancer

In three studies (Bender et al., 2012; Skoogh et al., 2013; Smith et al., 2013) in this review, unmet needs of patients with rare male genital and urogenital cancer were described. Unmet needs were most prevalent in the healthcare system and information domain (59%), followed by the psychological domain (12–42%) and the economic domain (15–28%), and these needs were reported throughout the whole disease trajectory. The highest reported need, in the diagnostic and treatment phase, was found for ‘information about crisis and stress after diagnosis’ (59%) and for ‘help with financial support and/or state benefits’ (16–28%) in the posttreatment phase. In the posttreatment phase, 63–66% of the patients reported at least one unmet need. No specific unmet needs of these male genital and urogenital cancer patients were reported in the diagnostic or treatment phase only.

### 3.10 | Unmet needs of patients with NET

Three studies (V.L. Beesley et al., 2018; Singh et al., 2017; Wolin et al., 2017) assessed unmet needs of patients with NET. Unmet needs were highest in the healthcare system and information domain (32–68%), followed by the physical and daily living domain (17–38%) and the patient care and support domain (11–48%), and these needs were reported throughout the whole disease trajectory. The highest need score, in the diagnostic phase, was reported for ‘better direction on where to find NET information’ and ‘more knowledgeable NET medical providers’ (both 58%), in the treatment phase for ‘wider range of treatment options’ (68%), in the diagnostic and treatment phase for ‘clearer information on longer-term impact of disease’ (66%) and in the posttreatment phase, for help with ‘lack of energy/tiredness’ (38%). At least one unmet need was reported by 63% of the patients in the posttreatment phase.

### 3.11 | Unmet needs of patients with rare skin cancer/eye melanoma

One study (Williamson et al., 2018) was identified that quantified the unmet needs of patients with eye melanoma. Unmet needs were highest in the healthcare system and information domain (61–92%), followed by the psychological domain (41–93%) and the patient care and support domain (52–70%), and these needs were reported in the diagnostic and treatment phase. The highest levels of need, in the diagnostic phase, were found for ‘information about cancer remission’ and ‘information about things to help get well’ (both 87%) and ‘information about cancer remission’ (70%) in the treatment phase. At least one unmet need was reported by 99% of the patients in the diagnostic phase and by 86% of the patients in the treatment phase. No unmet needs of these rare cancer patients were specifically reported in the posttreatment phase only.

### 3.12 | Unmet needs of patients with rare haematological cancer

Five studies (Boland et al., 2014; Molassiotis et al., 2011; D. Oberoi, White, Seymour, Prince, et al., 2017a; D.V. Oberoi, White, Seymour, Prince, et al., 2017b; Yu et al., 2017) were found that assessed unmet supportive care needs of patients with rare haematological cancer. Unmet needs were most frequently reported in the healthcare system and information domain (21–66%), followed by the psychological domain (9–64%) and the physical and daily living domain (6–50%), and these needs were reported in the treatment and posttreatment phase. In the treatment and posttreatment phase, ‘information about test results as soon as feasible’ (66%) was the most prominent need and in the posttreatment phase, need for help with ‘lack of energy/tiredness’ (50%) scored highest. In the posttreatment phase, 27% of the rare haematological patients reported

at least one unmet need. No unmet needs of these rare cancer patients were specifically reported in the diagnostic or treatment phase only.

### 3.13 | Unmet needs of patients with male breast cancer

One study (Bootsma et al., 2020) was found that assessed unmet needs in patients with male breast cancer. Unmet needs were reported in the healthcare system and information domain (17–65%) and in the disease-specific domain (36%). The highest need score, in the treatment phase, was reported for ‘information on acute effects of treatment’ (65%), in the posttreatment phase for ‘information on late effects of treatment’ (56%) and for ‘male-specific information on symptoms and diagnosis, treatment, side effects, psychosocial impact, peer group support, and research’ (36%) over all phases. No specific unmet needs of these male breast cancer patients were reported in the diagnostic treatment phase only.

### 3.14 | Qualitative studies

A total of 17 studies reported qualitative results regarding unmet needs of patients with rare cancer.

In one study in patients with rare CNS cancer (Piil et al., 2018), supportive care, informational and rehabilitation needs in the diagnostic and treatment phase were described. Needs included, e.g., strategies for managing psychological symptoms, the exchange of experiences with other patients and better access to specialists throughout the disease trajectory.

Posttreatment information needs regarding health-related QoL, medical care and prognosis were reported in a single study for patients with rare digestive cancers (Henselmans et al., 2012).

In two studies in patients with endocrine cancer (M. Henry et al., 2018; B. Lee, Park, et al., 2016), information needs in the treatment and posttreatment phase were found, i.e., detailed information on treatment procedures and on QoL after treatment.

High unmet supportive care and information needs were reported in three studies in patients with rare gynaecological cancer throughout the disease trajectory (Long et al., 2016; Philp et al., 2017; Zeng et al., 2017). Needs were reported regarding the provision of disease- and treatment-related information, but also concerns regarding sexuality, support from other fellow sufferers and strategies for managing symptoms were mentioned.

Seven studies in patients with rare HNC (Badr et al., 2016; Moore et al., 2014; Pateman et al., 2015; Peeters et al., 2018; Richardson et al., 2015; Saroa et al., 2018; W.K. So et al., 2019) reported supportive care, information, psychological and psychosocial needs mainly during the posttreatment phase. Needs frequently described were related to managing the side effects of treatment, support for dealing with posttreatment consequences, organised patient care and the need for returning to a normal life. The most frequently reported

healthcare system and information needs were related to information on symptoms of recurrence, information on cure, information on adverse treatment effects, access to health resources (e.g., dental oncology services), timeframes for treatment and recovery and post-treatment rehabilitation. Social support and sharing experiences and emotions with fellow sufferers were mentioned as psychosocial needs, and psychological needs included managing concerns about recurrence, managing concerns about the future and strategies for coping with HNC.

In a study in patients with NET (Feinberg et al., 2013), patients reported difficulties with access to appropriate care, and the need for disease-specific support and information on the disease and treatment.

Patients with haematological cancer (Nørskov et al., 2019) mentioned a need for social support from healthcare professionals, their social network and other fellow sufferers in the posttreatment phase.

Finally, in the male breast cancer study (Bootsma et al., 2020), information needs were reported regarding symptoms and (delay of) diagnosis, treatments, psychological impact and coping, genetics and family, research and raising awareness.

### 3.15 | Predictive factors for unmet needs throughout the rare cancer disease trajectory

In the diagnostic phase, predictive factors associated with a higher number of unmet needs in patients with rare cancers included higher neuroticism, lower instrumental social support and greater social network (Williamson et al., 2018). Predictive factors for unmet needs during treatment and posttreatment phase were somewhat inconsistent and included both younger age (Shun et al., 2018) and older age (V.L. Beesley et al., 2013), but also higher anxiety score (V.L. Beesley et al., 2013), high uncertainty (Shun et al., 2018), advanced disease (V.L. Beesley et al., 2013) and depression (V.L. Beesley et al., 2013). Predictors for unmet needs posttreatment included higher anxiety score (Brennan et al., 2019; M. Henry et al., 2020), younger age (M. Henry et al., 2020), higher neuroticism (M. Henry et al., 2020), lower QoL score (Brennan et al., 2019), more stressful life events in the year pre-diagnosis (M. Henry et al., 2020) and being female (Holm et al., 2012). Predictive factors for unmet needs throughout the disease trajectory included higher anxiety score, higher educational level and higher overall physical symptom severity (Chen et al., 2013). In Table 3, the overview of all predictive factors can be found.

## 4 | DISCUSSION

### 4.1 | Main findings

Prevalence of unmet supportive care needs in rare cancer patients varied strongly over all need domains, across all rare cancer subdomains and throughout all phases of disease. Unmet needs were most frequently reported in the healthcare system and information domain,

**TABLE 3** Predictive factors for unmet supportive care needs in rare cancer patients during the diagnostic, treatment and posttreatment phase of their disease trajectory

Predictive factors <sup>a</sup> (# studies)	# Studies per phase [ref]		
	Diagnostic	Treatment	Posttreatment
Higher anxiety score (4)		1 [23]	
			1 [13]
			2 [22, 26]
Younger age (2)		1 [8]	
			1 [26]
Higher neuroticism (2)	1 [50]		1 [26]
Higher educational level (1)		1 [23]	
Higher overall physical symptom severity (1)		1 [23]	
Higher functional level (1)		1 [23]	
Without religious beliefs (1)		1 [23]	
High uncertainty (1)			1 [8]
Advanced disease (1)			1 [13]
Depression (1)			1 [13]
Less social support (1)			1 [13]
Older age (1)			1 [13]
Insomnia (1)			1 [13]
Receiving chemotherapy (1)			1 [54]
Greater social network (1)	1 [50]		
Lower instrumental social support (1)	1 [50]		
Lower QoL score (1)			1 [22]
Lower performance status (ECOG) scores (1)			1 [22]
More stressful life events in the year pre-diagnosis (1)			1 [26]
Being female (1)			1 [27]
	Diagnostic		
	Treatment		
	Post-treatment		
	Diagnostic/treatment		
	Treatment/posttreatment		
	Diagnostic/treatment/posttreatment		

<sup>a</sup>The predictive factors are presented in order from most studies to least studies reporting on the predictive factor.

followed by the psychological domain and the physical and daily living domain. Specific unmet needs were present in patients with rare female genital organ cancer, namely, in the sexual domain; in patients with rare male genital and urogenital cancer, namely, in the economic domain, and in patients with rare HNC, namely, in the disease-specific domain. Unmet needs were primarily reported in the posttreatment phase, and over all phases, the most frequently identified predictors were higher anxiety score, younger age and higher neuroticism.

## 4.2 | Interpretation of findings

In this study, highest unmet needs of rare cancer patients were identified, throughout the disease trajectory, in the healthcare

system and information domain. This contrasts with previous findings in patients with common cancer (Harrison et al., 2009; Lisy et al., 2019; Wang et al., 2018). That is, in several reviews focusing on common cancer types, e.g., breast and lung cancer, needs in the psychological domain and physical and daily living domain were most frequently reported. Due to the lack of a clear cancer care pathway in some rare cancer types, it is not surprising that patients with these diagnoses request more information about their disease and treatment and enhanced organisation of care. Related to this, a plea for personalised treatment and one member of hospital staff to talk to has been made. In addition, in this review, high prevalence rates were found for rare cancer patients reporting at least one unmet need. The lowest rate, for at least one unmet need, was found in rare haematological cancer patients (i.e., 27%), and rates in

all other cancer subdomains ranged from 59% to 99%. A possible explanation for the low rate in rare haematological cancer patients might be that needs were only assessed in the posttreatment phase, while unmet needs during treatment, when symptom burden is expected to be higher, were not examined. Further, in another systematic review in cancer survivors posttreatment (Mirošević, Prins, Selič, Zaletel Kragelj, & Klemenc Ketiš, 2019), prevalence rates of at least one unmet need alternated from 24% to 88%, but no rates of haematological cancer patients were described here. The fact that rare cancer patients more often express at least one unmet need than common cancer patients might be explained by healthcare providers being more alert and responsive to patients' needs they encounter regularly, whereas the needs of rare cancer patients might be recognised to a lesser extent and herewith potentially remain unmet.

Regarding the identified needs in specific rare cancer subdomains, patients with rare female genital organ cancer, patients with rare male genital and urogenital cancer and patients with rare HNC should be noted. That is, the first group of patients reported high unmet needs in the sexuality domain, which is in concordance with previous studies (Gilbert et al., 2011; Stead et al., 2007). A diagnosis and treatment of gynaecological cancer significantly affects sexual functioning. Especially, women with vulvar cancer, undergoing extensive surgery, experience the most sexual problems (Stead et al., 2007). Reasonably, the sexual function of patients with this rare form of gynaecological cancer is often impaired by the removal of, or changes to, their reproductive organs, resulting in high unmet sexuality needs. In addition, patients with rare male genital and urogenital cancer reported high unmet needs in the economic domain, i.e., help with financial support and/or state benefits, and with obtaining insurance. Since this type of cancer, specifically testicular cancer, mainly affects young men under the age of 40, these patients are at working age and potentially the breadwinner of the family, which may result in financial worries due to impaired work ability. This has been described by Gupta et al. (2020) as well, explaining the negative impact of cancer-related financial stress on patient's overall well-being during emerging adulthood. Furthermore, rare HNC patients reported high unmet needs in the disease-specific domain, namely, needs for help with and information on chewing, swallowing, eating and speaking. Similarly, it has been shown in previous studies (Rathod et al., 2015; W. So et al., 2012) that this specific patient group is highly and negatively affected by these short- and long-term effects in the head and neck area, significantly impacting their physical functioning and QoL.

As previously reported for common cancer patients (Harrison et al., 2009), in our review, it has been confirmed that unmet needs of patients with rare cancer are predominantly identified in the treatment and posttreatment phase. That said, it is rather surprising that studies focusing on the unmet needs early in the disease trajectory of rare cancer patients are lacking. Moreover, patients with rare cancer are likely to experience high unmet needs, in the diagnostic phase, regarding (the delay in) obtaining the correct diagnosis and

finding information about (specific treatment for) their rare cancer type. While the implementation of psychosocial supportive care in common cancer patients has progressed over the last decades and is integrated from diagnosis up to long-term cancer survivorship (L. Grassi & Watson, 2012), this seems to lag behind in rare cancer patients.

Finally, in this study, it has been found that higher anxiety scores, younger age and higher neuroticism can be predictive factors for increased unmet supportive care needs. High level of anxiety and younger age were also found to be predictive for unmet needs in patients with common cancer (Harrison et al., 2009; Okediji et al., 2017). Moreover, in both cancer patient groups, advanced disease, higher educational level, higher physical symptom severity and low social support have been found to influence unmet needs (Harrison et al., 2009; Okediji et al., 2017). These similarities between groups are not unsurprising as sociodemographic, clinical and psychosocial factors are found to influence QoL in cancer patients (Allart et al., 2013; de Melo et al., 2019; Konieczny et al., 2020). Further, while higher neuroticism was found to be predictive for increased unmet needs in rare cancer patients, this has not been found in common cancer patients. This is in line with the finding that patients with rare cancer, in general, report higher distress levels and impaired QoL than common cancer patients (Bergerot et al., 2018). These findings highlight the relevance of addressing unmet needs of rare cancer patients throughout the disease trajectory. Also, screening for predictive factors, such as anxiety, depression, age and neuroticism, may facilitate the identification of high-risk patients, likely to benefit most from early targeted psychosocial care.

### 4.3 | Strengths and limitations

The main strength of this systematic review is that this is the first review exploring the unmet supportive care needs, and related predictive factors, in rare cancer patients at different phases of their disease trajectory. Also, it can be considered a strength that both quantitative and qualitative studies were included. This resulted in an overview of all available studies on unmet needs in adult patients within different rare cancer subdomains.

A limitation of this systematic review is that findings on unmet needs are difficult to compare between countries due to possible differences in the provision of supportive care during the disease trajectory. Although psychosocial oncology is seen as an integral part of comprehensive cancer care, it has not been fully integrated into oncological care worldwide yet (L. Grassi et al., 2016; Mehnert & Koch, 2005). In addition, in this review, a proportionate distribution of rare cancer types is missing. That is, the majority of the studies reported unmet needs within the rare HNC domain. Some studies (e.g., in sarcoma patients) might have been excluded based on our applied criteria. Lastly, specific unmet needs might have been missed due to the general measures used to assess the unmet needs in patients with a rare cancer type. Because of these limitations, results should be interpreted with caution.

#### 4.4 | Implications for practice and research

The findings of this review may guide healthcare professionals, working in the field of rare cancers, in their support of patients within a specific rare cancer subdomain during each phase of the disease trajectory. Specifically, healthcare professionals should be aware that patients with rare cancer require information about their disease and treatment and transparency about the organisation of care for their specific type of rare cancer. Knowledge in healthcare professionals should be increased, and access to clinical expertise for rare cancer patients should be improved. By establishing regional clinical networks for rare cancers, including centres of expertise, unmet needs can be identified and purposely addressed from diagnosis onwards. Such a network could play an important role in ensuring guidance and support throughout the cancer care pathway for patients with rare cancer. Since patients with rare cancer are known to face difficulties especially during diagnosis and treatment (Pillai & Jayasree, 2017; Ray-Coquard et al., 2017), more attention should be given to and research should be performed on unmet needs during these phases of the disease trajectory. Further, there is a lack of literature on unmet needs in several rare cancer subdomains, specifically in adult patients with sarcomas, rare urinary tract cancer, rare thoracic cancer and rare skin cancer. Research on the unmet needs in those specific rare cancer subdomains is warranted, since those patients may have different unmet needs compared to patients from the other rare cancer subdomains. In addition, only a few studies used disease-specific questionnaires, which implies the need for development of disease-specific instruments, measuring unmet needs of patients with a rare cancer. Finally, appropriate supportive care strategies within patients with common cancer might also be applicable to patients with rare cancer. However, those strategies are likely to be different per cancer type and therefore should be tailored according to the patient group and should be further investigated.

## 5 | CONCLUSION

Patients with rare cancer have unmet supportive care needs throughout the disease trajectory, with the highest reported needs in the healthcare system and information domain. The most frequently identified predictors of unmet needs in rare cancer patients were higher anxiety, younger age and higher neuroticism. Healthcare professionals should be aware of the different unmet needs per rare cancer subdomain, and these unmet needs should be recognised and individually addressed starting from diagnosis onwards. Future studies are needed to determine further unmet needs of patients in all rare cancer subdomains, in order to help healthcare professionals in providing tailored supportive care and improving QoL in rare cancer patients.

#### ACKNOWLEDGEMENTS

We thank Olga van der Hel for her assistance with developing the search strategy and Milou Reuvers for her help with the data extraction and quality assessment.

#### FUNDING INFORMATION

This research received no specific grant from any funding agency in the public, commercial or not-for-profit sectors.

#### CONFLICT OF INTEREST

All authors have reported no conflict of interest.

#### AUTHOR CONTRIBUTIONS


All authors have made substantial contributions to conception and design, or acquisition of data, or analysis and interpretation of data.

#### DATA AVAILABILITY STATEMENT

The data supporting this systematic review are from previously reported studies, which have been cited.

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**How to cite this article:** de Heus, E., van der Zwan, J. M., Husson, O., Frissen, A.-R., van Herpen, C. M. L., Merkx, M. A. W., & Duijts, S. F. A. (2021). Unmet supportive care needs of patients with rare cancer: A systematic review. *European Journal of Cancer Care*, e13502. <https://doi.org/10.1111/ecc.13502>

## APPENDIX A: SEARCH STRATEGY PER DATABASE

## PubMed Search Strategy

Search	Query	Hits
#1	Neoplasms[Mesh]	3,432,815
#2	Cancer*[tiab] OR tumor*[tiab] OR tumour*[tiab] OR carcinoma*[tiab] OR neoplasm*[tiab] OR adenocarcinoma*[tiab] OR malignan*[tiab] OR sarcoma*[tiab] OR oncolog*[tiab]	3,547,933
#3	#1 OR #2	4,511,020
#4	Needs Assessment[Mesh]	31,125
#5	Unmet[ti] AND need*[ti]	3,408
#6	Need*[ti] AND assess*[ti]	6514
#7	Perceived[ti] AND need*[ti]	950
#8	Support*[ti] AND care[ti] AND need*[ti]	795
#9	Psycho*[ti] AND need*[ti]	2,359
#10	Physical[ti] AND need*[ti]	728
#11	Information[ti] AND need*[ti]	2,927
#12	(Patient[ti] AND need*[ti]) OR (patient[ti] AND experience*[ti])	10,128
#13	#4 OR #5 OR #6 OR #7 OR #8 OR #9 OR #10 OR #11 OR #12	53,425
#14	#3 AND #13	5,920
#15	Search (#4) Filters: Humans, Published since 01-01-2011	3,007

## PsycINFO Search Strategy

Search	Query	Hits
#1	exp neoplasms/	51,488
#2	(Cancer* or tumor* or tumour* or carcinoma* or neoplasm* or adenocarcinoma* or malignan* or sarcoma* or oncolog*).ti,ab.	79,834
#3	1 or 2	83,086
#4	Needs assessment/	4,224
#5	Unmet need*.ti.	598
#6	Need* assess*.ti.	1,055
#7	Perceived need*.ti.	340
#8	Support* care need*.ti.	118
#9	Psycho* need*.ti.	1,069
#10	Physical need*.ti.	5
#11	Information need*.ti.	414
#12	(Patient need* or patient experience*).ti.	410

Search	Query	Hits
#13	4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12	7,251
#14	3 and 13	615
#15	Limiters: Human, Publication year: 2011 – Current	392

## CINAHL Search Strategy

Search	Query	Hits
#1	MH neoplasms	81,657
#2	TI (Cancer* OR tumor* OR tumour* OR carcinoma* OR neoplasm* OR adenocarcinoma* OR malignan* OR sarcoma* OR oncolog*)	422,559
#3	S1 OR S2	451,806
#4	MH needs assessment	18,603
#5	TI unmet need*	2,131
#6	TI need* assess*	3,749
#7	TI perceived need*	753
#8	TI support* care need*	534
#9	TI psycho* need*	1,238
#10	TI physical need*	412
#11	TI information need*	2,037
#12	TI (Patient need* OR patient experience*)	5,370
#13	S4 OR S5 OR S6 OR S7 OR S8 OR S9 OR S10 OR S11 OR S12	30,805
#14	S3 AND S13	2,757
#15	Limiters: Human, Published Date: 20110101 – 20200518	1,199

## APPENDIX B: RARE CANCER SUBDOMAINS AND CANCER TYPES

Rare cancer subdomain	Rare cancer type <sup>a</sup>
Rare CNS cancer	Tumours of central nervous system (CNS) Embryonal tumours of CNS
Rare digestive cancer	Epithelial tumours of oesophagus Epithelial tumours of small intestine Epithelial tumours of anal canal Epithelial tumours of liver and intrahepatic bile tract Epithelial tumours of gallbladder and extrahepatic bile tract
Endocrine cancer	Carcinoma of pituitary gland Carcinoma of thyroid gland Carcinoma of parathyroid gland Carcinoma of adrenal cortex
Rare gynaecological cancer	Epithelial tumours of cervix uteri Epithelial tumours of ovary and fallopian tube Non epithelial tumours of ovary and fallopian tube Epithelial tumours of vulva and vagina Trophoblastic tumours of placenta
Rare head and neck cancer (HNC)	Epithelial tumours of nasal cavity and sinuses Epithelial tumours of nasopharynx Epithelial tumours of major salivary glands and salivary-gland type tumours Epithelial tumours of hypopharynx and larynx Epithelial tumours of oropharynx Epithelial tumours of oral cavity and lip Epithelial tumours of eye and adnexa Epithelial tumours of middle ear
Rare male genital and urogenital cancer	Testicular and paratesticular cancers Epithelial tumours of penis Epithelial tumours of pelvis and ureter Epithelial tumours of urethra
Neuroendocrine tumours	NET GEP NET lung NET other sites
Sarcomas	Soft tissue sarcomas Bone sarcoma Gastrointestinal stromal sarcoma
Rare skin cancer/eye melanoma	Malignant melanoma of mucosa and extracutaneous Malignant melanoma of eye Adnexal carcinomas of skin Kaposi's sarcoma
Rare thoracic cancers	Epithelial tumours of trachea Epithelial tumours of thymus Malignant mesothelioma

Rare cancer subdomain	Rare cancer type <sup>a</sup>
Rare haematological cancer	Lymphoid diseases ( <i>except other non-Hodgkin, Mature B cell lymphoma</i> )
	Acute myeloid leukaemia and related precursor neoplasms
	Myeloid and lymphoid neoplasms
	Myeloproliferative neoplasms
	Myelodysplastic syndrome and myelodysplastic/myeloproliferative diseases
	Histiocytic and dendritic cell neoplasms
Male breast cancer	Male breast cancer

<sup>a</sup> Based on an updated version (February 2019) of the list of cancer types from RARECARENet.