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A Dutch paediatric palliative care guideline: a systematic review and recommendations on advance care planning and shared decision-making

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Abstract

Background In paediatric palliative care, children with life-threatening and life-limiting conditions, their families, and their health care professionals often face difficult decisions about treatment, goals of care, and delivery of care. Advance care planning and shared decision-making are strategies that can improve quality of care by discussing goals and preferences on future care. In this paper, we provide recommendations that aim to optimise advance care planning and shared decision-making in paediatric palliative care in the Netherlands.

Methods A multidisciplinary guideline panel of 20 experts in paediatric palliative care and nine (bereaved) parents was established to develop recommendations on advance care planning and shared decision-making. We performed systematic literature searches to identify quantitative and qualitative evidence and used the GRADE (CERQual) methodology for appraisal of evidence. Recommendations were formulated based on quantitative and qualitative evidence, clinical expertise, and patient and family experiences.

Results We identified 4 RCTs that reported on the effect of advance care planning interventions in paediatric palliative care and 33 qualitative studies on barriers and facilitators to advance care planning and shared decision-making. We formulated 28 strong recommendations in close collaboration with a multidisciplinary guideline panel that provide guidance to offer advance care planning and shared decision-making, involve children and their family, and communicate information about care and treatment.

Conclusion The identified evidence and recommendations support the use of advance care planning and shared decision-making in paediatric palliative care. However, we found several knowledge gaps that should be addressed.

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As advance care planning and shared decision-making require specific skills and can be time-consuming, we emphasise the importance of education, adequate staffing and sufficient funding to improve integration in clinical practice. We do believe that our recommendations can be used as a starting point to develop recommendations in other countries. However, country-specific factors should be very carefully considered before applying any recommendations in other countries.

Keywords Evidence-based guideline, Paediatric palliative care, Advance care planning, Shared decision-making

Background

Children with life-threatening and life-limiting conditions and their families need palliative care to relief physical, psychological, social, and spiritual suffering and improve quality of life during the entire disease trajectory, from diagnosis till the end-of-life and during bereavement [1]. These children often receive high complex care that can last for months or years. This care is often provided by multiple health care professionals and can be delivered at the hospital, home, or elsewhere [2]. During the disease trajectory, these children, their families, and health care professionals face difficult decisions about goals of care and related treatment, place of care, and delivery of care dependent on the situation and prognosis of the child [3, 4].

It is increasingly recognised that children, families, and health care professionals value person-centred approaches to identify goals and preferences for care and treatment to inform decision-making when receiving palliative care [3, 5]. Advance care planning and shared decision-making are conversational approaches that can be used to give substance to care and treatment together with child and family. These approaches focus on open and equivalent exchange of knowledge, experiences, values, goals, and preferences between children, families, and health care professionals [6, 7].

Advance care planning enables individuals to define goals and preferences for future medical care and treatment, to discuss these goals and preferences with the child, its family and health care professionals, to record these, and review these if appropriate [7]. The outcomes of an advance care planning conversation can guide the child, family, and health care professionals in determining what treatment policy supports the best interest of the child and aligns with the family's values and preferences. In this way, advance care planning is extremely helpful when a decision on the treatment is needed [8].

In paediatric palliative care, many decisions on care and treatment need to be made. In these decisions, there is often not one best treatment option and many uncertainties regarding the child's condition or disease evolution pertain. Each treatment option can have advantages and disadvantages, which may be experienced or valued differently among children, family, or health care professionals. Shared decision-making is a collaborative process that can support stakeholders in making these preference-sensitive decisions [9]. In this process, the child (if possible), family and health care professionals are working together to make joint decisions on the best care and treatment [9]. The goals and preferences discussed and recorded in advance care planning conversations can guide the shared decision-making process [10].

Advance care planning and shared decision-making are acknowledged as key elements of paediatric palliative care [3]. However, a recent qualitative study among parents in the Netherlands has shown that the use of these person-centred approaches in paediatric palliative care remains to be very challenging [11]. Clinical practice guidelines (CPGs) are powerful tools in which knowledge from scientific literature, clinical expertise and patient experiences are combined to provide recommendations which can enhance delivery of high quality care and contribute to the integration of care services [12–14].

As part of the revised Dutch CPG on paediatric palliative care, we provide new recommendations that focus on two key elements of paediatric palliative care: advance care planning and shared decision-making. In this paper, we present an overview of the evidence, clinical expertise, and recommendations on these topics.

Methods

The full methodology of the Dutch CPG for paediatric palliative care has been published in a separate paper [15].

Scope

This guideline provides guidance on palliative care for all children aged 0 to 18 years with life-threatening or life-limiting conditions and their caregivers, brothers, and sisters (hereafter referred to as families) throughout the entire palliative trajectory (from palliative diagnosis till after end-of-life), with the ultimate goal to improve quality of paediatric palliative care and thereby quality of life of children and their families [16]. Here, we provide recommendations for advance care planning and shared decision-making.

Multidisciplinary guideline development panel

The guideline development panel consisted of an expert panel of 20 professionals with expertise in paediatric palliative care and a panel of nine (bereaved) parents (Appendix A). Professionals from multiple disciplines such as paediatricians, paediatric nurses, medical pedagogical care providers, and specialists in intellectual disabilities, were included in the guideline development panel. Within the expert panel, a core group of 11 experts was established to ensure consistency throughout the guideline. The other nine experts were assigned to the working group advance care planning and shared decision-making. The addressed topics and clinical questions were selected based on priorities of health care professionals and parents [15]. An overview of the working structure and guideline development process is shown in Appendix B and C.

Representation of patients and their families

To ensure representation of patients and their families, different methods were used [15]. Two members of the core group were dedicated to ensure the representation of patients and their families during the entire guideline process. Additionally, a panel of 9 (bereaved) parents of children with life-threatening or life-limiting conditions reviewed the first drafts of all guideline texts and recommendations and reviewed the complete concept guideline. We ensured parents represented a broad spectrum of experiences by including parents of children with a variety of palliative conditions, ages, and stages of disease (currently receiving palliative care or deceased).

Identification of quantitative studies

The working group formulated one clinical question on the effect of advance care planning and shared decision-making interventions (Appendix D). Therefore, we updated the literature search on paediatric palliative interventions that was conducted for the former CPG (2013) until January 24, 2020 (Appendix E). Studies were selected according to inclusion criteria related to study design (randomised controlled trials (RCTs), controlled clinical trials (CCTs), and systematic reviews (SRs) of RCTs and CCTs; study population (children aged 0 to 18 with life-threatening or life-limiting conditions according to the definition of the World Health Organisation [16]) and study subject (paediatric palliative care interventions related to advance care planning and shared decision-making). Only studies published in English or Dutch language were included (Appendix F).

Included studies were summarised in evidence tables. We categorised evidence by outcome measures in summary of findings tables. Then, we formulated conclusions of evidence for each outcome measure. The quality of the total body of evidence was graded using the Grading of Recommendations Assessment, Development, and Evaluation (GRADE) method [17].

Identification of qualitative studies

Additionally, the working group formulated a clinical question to identify barriers and facilitators to advance care planning and shared decision-making (Appendix D). To find studies on this topic, we first searched for existing evidence-based guidelines that performed a systematic literature search on this topic. We found one evidence-based guideline, 'End of life care for infants, children and young people with life-limiting conditions (2016)' of the National Institute for Health and Care Excellence (NICE) [18]. We used the original systematic literature search that was conducted in this existing guideline and updated it until 16 September, 2020 (Appendix E). Studies were selected based on inclusion criteria related to study design (qualitative studies, mixed-methods observational studies with qualitative data and SRs of qualitative studies), study population (children aged 0 to 18 years old with life-threatening or life-limiting conditions according to the definition of the World Health Organisation [16]), their parents and health care professionals) and study outcomes (barriers and facilitators to advance care planning or shared decision-making). Moreover, only studies published in English or Dutch language were included (Appendix F).

We used evidence tables to extract barriers and facilitators to advance care planning and shared decision-making in themes and subthemes and formulated conclusions of evidence for each theme or subtheme. The quality of the total body of evidence was assessed with the adapted GRADE Confidence in the Evidence from Reviews of Qualitative research (GRADE CERQual) methodology [19]. As we updated the systematic search of the NICE-guideline, we integrated the conclusions of evidence in our updated conclusions of evidence. Additionally, the recommendations in the NICE-guideline were used to refine considerations and recommendations [15].

Translating evidence into recommendations

Recommendations were based on evidence from scientific literature, expert opinion, and patient and family values. With regard to the evidence from scientific literature, WG members evaluated the quality of evidence before formulating recommendations, with stronger recommendations generally supported by higher-quality evidence. Moreover, when evidence was specific to a particular group of children (e.g., children with cancer), the WG members carefully considered whether the identified evidence could be extrapolated to children with other life-threatening or life-limiting conditions. Furthermore, the WG members described other relevant considerations including patient preferences, expert opinion, legal and ethical considerations, applicability and feasibility, and potential benefits and harms [15]. We followed the criteria for the strength of recommendations according to published methods [17, 20] (Appendix G). Recommendations were categorised as strong to do (green), moderate to do (yellow) or strong not to do (red).

Results

Identification of evidence

The systematic literature search for quantitative studies on paediatric palliative care interventions yielded 5078 citations of which 168 citations were subjected to full-text screening. A total of four RCTs on the effect of advance care planning interventions in children with cancer or human immunodeficiency virus (HIV) infections were identified [21–24]. No studies on the effects of shared decision-making were identified.

The updated systematic literature search for qualitative studies on barriers and facilitators to advance care planning and shared decision-making identified 1238 eligible citations. We excluded 1147 citations based on title/abstract and 85 citations were included for full-text screening. We included a total of 33 qualitative studies on barriers and facilitators of advance care planning and shared decision-making, of which 22 studies [6, 25–45] were newly published studies and 11 studies were identified in the original search of the NICE-guideline [18]. In Fig. 1, a flow chart of the study selection process of both systematic searches is presented.

Evidence on effect of advance care planning interventions

We included a total of four RCTs that all described the effect of the Family-Centred Advance Care Planning (FACE) intervention as compared to usual care combined with information provision sessions in family format on development, safety, and school and career planning [21–24]. The FACE-intervention aims to facilitate advance care planning conversations between adolescents and their adult surrogates in three weekly sessions. The effects of the FACE-intervention were studied in adolescents with cancer [22, 24] and adolescents with an HIV-infection [21, 46]. In Table 1, the conclusions of the evidence are shown. A full overview of all conclusions of evidence corresponding evidence tables, and summary of findings tables can be found in Appendix I, J and K.

The studies reported the effects of different outcome measures, including the completion of a legal statement of treatment preferences, level of congruence in treatment preferences, anxiety, depression, quality of life, and

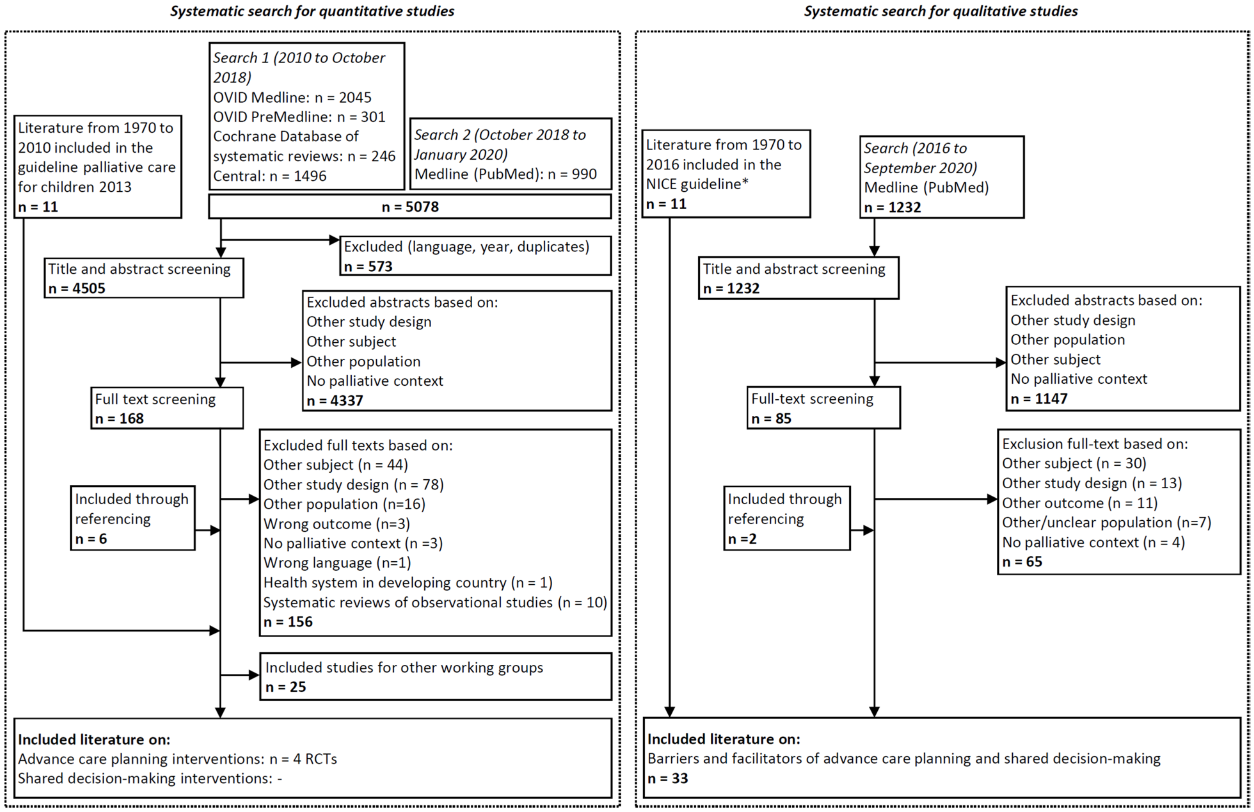
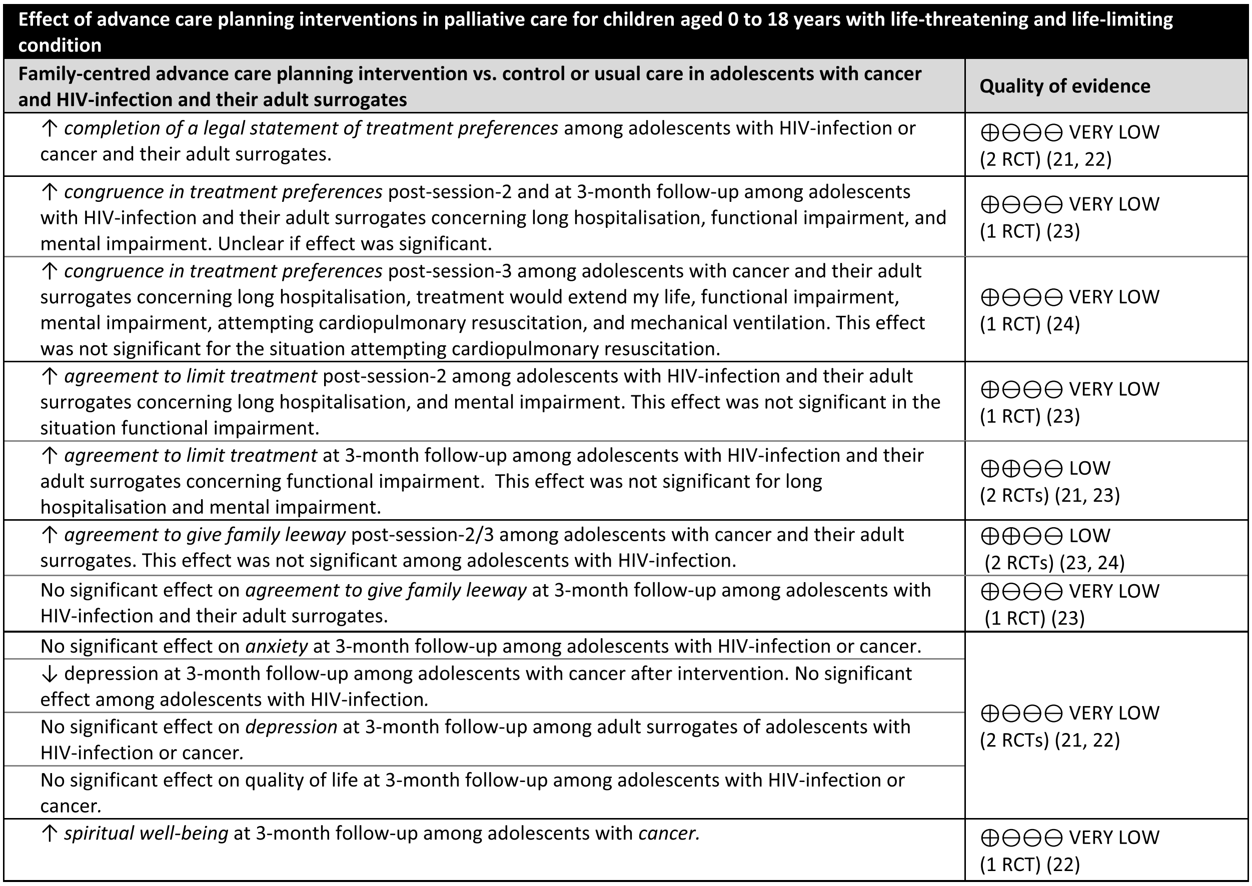


Fig. 1 Flow chart of the study selection process *We only used the conclusions of evidence from the 11 identified studies from the search of the NICE guideline

Table 1 Conclusions of evidence on advance care planning interventions in paediatric palliative care



Effect of advance care planning interventions in palliative care for children aged 0 to 18 years with life-threatening and life-limiting condition	
Family-centred advance care planning intervention vs. control or usual care in adolescents with cancer and HIV-infection and their adult surrogates	**Quality of evidence**
↑ *completion of a legal statement of treatment preferences* among adolescents with HIV-infection or cancer and their adult surrogates.	⊕⊖⊖⊖ VERY LOW (2 RCT) (21, 22)
↑ *congruence in treatment preferences* post-session-2 and at 3-month follow-up among adolescents with HIV-infection and their adult surrogates concerning long hospitalisation, functional impairment, and mental impairment. Unclear if effect was significant.	⊕⊖⊖⊖ VERY LOW (1 RCT) (23)
↑ *congruence in treatment preferences* post-session-3 among adolescents with cancer and their adult surrogates concerning long hospitalisation, treatment would extend my life, functional impairment, mental impairment, attempting cardiopulmonary resuscitation, and mechanical ventilation. This effect was not significant for the situation attempting cardiopulmonary resuscitation.	⊕⊖⊖⊖ VERY LOW (1 RCT) (24)
↑ *agreement to limit treatment* post-session-2 among adolescents with HIV-infection and their adult surrogates concerning long hospitalisation, and mental impairment. This effect was not significant in the situation functional impairment.	⊕⊖⊖⊖ VERY LOW (1 RCT) (23)
↑ *agreement to limit treatment* at 3-month follow-up among adolescents with HIV-infection and their adult surrogates concerning functional impairment. This effect was not significant for long hospitalisation and mental impairment.	⊕⊕⊖⊖ LOW (2 RCTs) (21, 23)
↑ *agreement to give family leeway* post-session-2/3 among adolescents with cancer and their adult surrogates. This effect was not significant among adolescents with HIV-infection.	⊕⊕⊖⊖ LOW (2 RCTs) (23, 24)
No significant effect on *agreement to give family leeway* at 3-month follow-up among adolescents with HIV-infection and their adult surrogates.	⊕⊖⊖⊖ VERY LOW (1 RCT) (23)
No significant effect on *anxiety* at 3-month follow-up among adolescents with HIV-infection or cancer.	⊕⊖⊖⊖ VERY LOW (2 RCTs) (21, 22)
↓ depression at 3-month follow-up among adolescents with cancer after intervention. No significant effect among adolescents with HIV-infection.	
No significant effect on *depression* at 3-month follow-up among adult surrogates of adolescents with HIV-infection or cancer.	
No significant effect on quality of life at 3-month follow-up among adolescents with HIV-infection or cancer.	
↑ *spiritual well-being* at 3-month follow-up among adolescents with *cancer.*	⊕⊖⊖⊖ VERY LOW (1 RCT) (22)

spiritual well-being. The studies showed that adolescents with cancer and HIV-infection in the intervention group were more likely to complete a legal statement of treatment preferences than the control group (very low quality of evidence) [21, 22]. Additionally, it was found that the FACE-intervention increased agreement on treatment preferences (very low quality evidence) [23, 24] and treatment restrictions (low quality evidence) [21, 23] between adolescents with cancer or HIV-infection and their adult surrogates. Moreover, the FACE-intervention increased agreement between adolescents with cancer or HIV-infection and their adult surrogates to give family leeway (low quality evidence) [23, 24]. However, this effect did not sustain at three month follow-up (low quality evidence) [23].

Furthermore, risk of depression was lower among adolescents with cancer three months after participating in the FACE-intervention (very low quality evidence). This effect was not found among adolescents with HIV-infection and adult surrogates of adolescents with cancer or HIV-infection (very low quality evidence) [21, 22]. Adolescents with cancer that participated in the FACE-intervention reported a higher spiritual well-being at three month follow-up as compared to the control group (very low quality evidence) [22]. The studies reported no significant effect on anxiety and quality of life in adolescents with cancer or HIV-infection after three months (very low quality evidence) [21, 22].

Evidence on barriers and facilitators to advance care planning and shared decision-making

We identified ten themes from the included qualitative literature that cover barriers and facilitators to advance care planning and shared decision-making: (1) information provision, (2) involvement, (3) interpersonal relations and communication, (4) holistic approach to care, (5) timing, (6) preparation, (7) documentation, (8) setting, (9) support, (10) education. Within these themes, we formulated various conclusions of very low to moderate quality evidence. For each conclusion we distinguished whether it was perceived as a facilitator or a barrier. Furthermore, we indicated whether the conclusion was reported by parents, health care professionals and/or children. Lastly, we indicated for each conclusion

Table 2 Conclusions of evidence on barriers and facilitators to advance care planning and shared decision-making in paediatric palliative care

Barriers and facilitators in advance care planning and shared decision-making for children aged 0 to 18 years with life threatening and life-limiting conditions, their families, and their health care professionals		
Theme /subtheme	**Facilitators**[#]	**Barriers**[#]
Information provision		
Information provision on treatment and prognosis	• Providing frequent complete, unbiased, and understandable information about the child's condition, likely treatment outcomes and treatment options (25, 26, 28, 29, 32, 35 ,37, 39-41, 43, 45). [*, a, b] • Acknowledging of uncertainties on diagnosis and prognosis as an aspect of the child's unique situation (31, 35, 43). [a] • HCPs prefer parents and teenagers to provide type and amount of information (26, 27). [b]	
Prognostic uncertainty		• HCPs find it complicated to give consistent and clear information due to prognostic uncertainty (28, 32, 45). [*, b] • Guesses by HCPS and disagreements among HCPs due to prognostic uncertainty (36, 38, 43). [*, a]
Involvement		
Involvement of parents	• Acknowledging parents as the expert of their child (6, 29-31, 35, 36, 38, 39, 41, 43, 45). [*, a]	• Individual variation in the preferred level of parent involvement in ACP and decision making i.e., parents should be final decision-maker [a, b], parents make decisions in collaboration with HCPs [a, b], parents do not want to be involved [a, b], parents feel like there is no choice to be made [a] (25-31, 37, 40, 41, 43, 45). [*, a, b]
Involvement of children	• Considering the child perspective during ACP and shared decision-making (6, 30, 31). [a]	• Individual variation in the preferred level of child involvement in ACP and decision making i.e., all children should be involved, level of child involvement is dependent on age, the decision that needs to be made, or the situation of the child (26-31, 34, 36, 37, 39). [*, a, b, c] • Uncertainty of HCPs on their role in communicating with the child (26, 27, 30, 31). [b]
Involvement of HCPs		• Individual variation in the preferred level of HCP involvement in decision making i.e., HCPs should solely provide information, HCPs should orient parents to the decision most beneficial for the child, HCPs should be the final decision-maker in certain situations (26, 45). [b]
Personal facilitators/barriers to advance care planning and shared decision-making	• Good experiences with care facilitate decision-making for parents (6, 35). [*, a]	• Parents experienced difficulty in advance care planning and decision-making i.e. some parents do not feel ready to make decisions, some parents do not want their child to suffer but want the child to live as long as possible, some parents cannot foresee consequences, and some parents want to keep option open (25, 26, 28, 29, 36-38, 45). [*, a, b] • HCPs experienced stress addressing sensitive themes (28, 30, 32, 40-42). [b]

Table 2 (continued)

Interpersonal relations and communication		
Communication	• Using open, honest, and clear lay language (26-29, 32, 36, 38, 41). [* a, b] • Communication and information provision by a trusted HCP (29, 38). [a] • Using of interpreters for non-native speakers (29). [a] • Using open and reassuring nonverbal cues (43). [a] • HCPs that show compassion, give support, and listen actively (28, 32). [*, a]	
Family-provider relationship	• Long-lasting, trusted relationships between parents and HCPs (26, 28, 32, 35, 38, 40, 43, 45). [*, a, b]	• Relationships between parents and HCPs are fragile and easily compromised when parents do not feel heard (29, 38, 41, 45). [*, b] • Disagreements between HCPs and parents, difficulty to reach agreement with families (27, 31, 42, 45). [*, a, b]
Holistic approach to care		
Living with illness	• Acknowledging the impact of the child's illness on all aspects of the child's and families daily live (6, 25, 30-32, 35, 36, 39, 41). [*, a, b]	
Hope	• Support parents' hope (26, 36, 37, 43). [a]	• Varied preferences on how HCPs should support hope (26, 36, 37, 43). [a]
Faith and Religion	• Hope, faith, religion empower parents to make decisions (28, 44). [*, a] • HCPs acknowledging parent's beliefs (44). [*, a]	• HCPs worry that hope, faith, and religion may allow parents to disregard medical evidence in decision-making (28, 44). [*, b]
Attention for different cultures	• Providing culturally sensitive information (39). [*, a]	• Disagreements due to differences in cultural background (28, 32, 45). [*, a, b]
Timing		
Ongoing process	• Acknowledging ACP as an ongoing process and continuous part of the child's care (31-33, 35, 36, 41). [*, a, b]	
Sufficient time for decision making	• Giving parents sufficient time between receiving information and making decisions, to think about several options prior to the actual decision-making (25, 29). [*, a]	
Preparation		
	No moderate quality evidence was found	
Documentation		
	• Personal conversation when handing out supplementary written materials (31, 36). [*, a, b]	
Setting		
	• Comfortable and appropriate setting of ACP meetings (32, 33, 35, 41, 43). [*, a, b] • Key family members and HCPs have the opportunity to be present during ACP conversations (35, 36, 41). [*, a, b]	
Support		
	• Parents connecting to family-members and other parents in similar situations (29, 36, 39, 45). [*, a]	
Education		
	• ACP and communication training for HCPs (26, 30, 36, 40, 41). [*, a, b]	

[#] all facilitators/barriers for which moderate quality evidence was found are presented in this table

[*] conclusions of evidence were presented in NICE guideline as well

[a] facilitator/barrier reported by parents

[b] facilitator/barrier reported by HCPs

[c] facilitator/barrier reported by children

Abbreviations: *ACP* Advance care planning, *HCP* Health Care Professional, *NICE* National Institute of health Care Excellence whether it was reported in the NICE-guideline [18]. In Table 2, all conclusions of moderate quality evidence are shown. A full overview of all conclusions of evidence, corresponding evidence tables, and summary of findings tables can be found in Appendix I, J and K.

Regarding the theme information provision, regular provision of complete, unbiased, and understandable information about the child's condition, likely treatment outcomes and treatment options was considered essential by parents and health care professionals [25, 26, 28, 29, 32, 35, 37, 39–41, 43, 45]. Health care professionals found it helpful when families themselves indicated their preferred type and amount of information [26, 27]. Parents reported that the acknowledgement of uncertainties about the diagnosis and prognosis was important [31, 35, 43]. Uncertainties that led to guesses and disagreements among health care professionals were considered a barrier as perceived by parents [36, 38, 43]. Health care professionals found information provision very difficult, especially due to these uncertainties in diagnosis and prognosis [28, 32, 45].

Acknowledging parents as experts of their own child was seen as an important facilitator for parental involvement in advance care planning and shared decision-making [6, 29–31, 35, 36, 38, 39, 41, 43, 45]. In addition, including the child's perspective was considered essential [6, 30, 31]. The large individual variation in the desired level of parental involvement [25–31, 37, 40, 41, 43, 45] and child involvement [26–31, 34, 36, 37, 39] in treatment decision-making was perceived as a barrier. Moreover, health care professionals reported variation in preferred level of their own involvement as a barrier as well [26, 45]. Furthermore, both parents and health care professionals experienced barriers on a personal level to participate in advance care planning or shared decision-making. For example, parents found it difficult to share their perspectives with health care professionals, as they feared this could impact their child's treatment in a unfavourable way [25, 26, 28, 29, 36–38, 45] and health care professionals found it difficult to address sensitive themes [28, 30, 32, 40–42]. Parents reported that good experiences with care could facilitate shared decision-making [6, 35].

For communication and interpersonal relations, parents and health care professionals considered the use of open, honest, and clear lay language as facilitators [26–29, 32, 36, 38, 41]. Additionally, parents acknowledged that communication by a trusted health care professional [29, 38], the use of interpreters for non-native speakers [29], reassuring non-verbal and compassionate communication, offering support, and active listening were essential facilitators [28, 32, 43]. Both health care professionals and parents reported that long-lasting trusted relationships between parents and health care professionals facilitated advance care planning and shared decision-making [26, 28, 32, 35, 38, 40, 43, 45]. However, parents did note that relationships were easily compromised when they did not feel heard [29, 38, 41, 45]. Additionally, different perspectives between health care professionals and parents were seen as hindering [27, 31, 42, 45].

Within the theme holistic approach to care, parents and health care professionals considered recognising the impact of the child's illness on all aspects of the child's and family's life as a facilitating factor [6, 25, 30–32, 35, 36, 39, 41]. In addition, parents reported that talking about their hopes, faith, and religion empowered them in their decision-making process [28, 44]. Furthermore, parents considered acknowledgement of their beliefs [44], their hopes [26, 36, 37, 43], and provision of culturally sensitive information [39] as facilitators. However, parents also reported a variety in preferences on how health care professionals should support hope [26, 36, 37, 43]. Health care professionals worried that parents' hopes, faith, and religion may lead to disregarding of medical evidence [28, 44]. Both parents and health care professionals agreed that disagreements due to cultural background hindered advance care planning and shared decision-making [28, 32, 45].

With regards to the timing, preparation, documentation and setting of advance care planning or shared decision-making conversations, both parents and health care professionals distinguished multiple facilitating factors. First of all, recognising advance care planning as a dynamic and continuous process and as a standard part of care promoted its use [31–33, 35, 36, 41]. Parents stressed that they should be given sufficient time to consider (still) possible treatment options [25, 29]. Additionally, a personal conversation when handing out supplementary written materials were considered facilitators [31, 36]. It was important that advance care planning conversations were conducted in an appropriate and comfortable setting preferably a quiet room with adequate seating, without distractors, possibly away from the hospital or at home [32, 33, 35, 41, 43]. Key family members and health care professionals should be present during these conversations [35, 36, 41]. Moreover, parents considered connections to other families in similar situations to share experiences as supportive [29, 36, 39, 45]. Finally, offering education and training to health care professionals was considered beneficial in enhancing the quality of advance care planning conversations and shared decision-making [26, 30, 36, 40, 41].

Translating evidence into recommendations

Existing studies described the effectivity of advance care planning interventions and the barriers and facilitators to advance care planning and shared decision-making. Our recommendations are based on the conclusions from the existing evidence and the consensus of the guideline development panel. The guideline development panel formulated a total of 28 recommendations on advance care planning ($n = 11$), shared decision-making ($n = 10$), roles of child, family and health care professionals in advance care planning and shared decision-making ($n = 3$), and communication skills during advance care planning and shared decision-making ($n = 4$). All recommendations are shown in Table 3.

Recommendations on advance care planning

Multiple RCTs indicated that an advance care planning intervention can enhance agreement on treatment preferences among adolescents with serious illnesses and their adult surrogates. No negative impact on quality-of-life related outcomes was reported [21–24]. Although the quality of evidence was very low to low, the guideline development panel acknowledged the benefits of advance care planning for children to be prepared for the future and to guide shared decision-making. This was supported by the identified qualitative evidence. We therefore strongly recommend that advance care planning conversations should be a standard of care for all children with a palliative disease trajectory and their families (recommendation 1). In addition, various barriers and facilitators to advance care planning were embedded in the recommendations formulated by the guideline development panel. We strongly recommend that advance care planning should be a continuous and dynamic process (moderate quality evidence) [31–33, 35, 36, 41] and conversations should be initiated early in the disease process, certainly when the need to prepare for specific scenarios increases (moderate quality evidence) [25, 29] (recommendation 2–4). Furthermore, information materials should be handed out with a personal conversation to prepare child and family for advance care planning conversations (moderate quality evidence) [31, 36] (recommendation 5). All conversations should be documented in the medical record and should be led by a trusted health care professional (moderate quality evidence) [26, 28, 29, 32, 35, 38, 40, 43, 45] (recommendation 6–7). The guideline development panel recognised that strategies to improve practical application of advance care planning are key for the integration into clinical practice. The use of a conversation guide can be a helpful strategy to facilitate integration by raising awareness among health care professionals on the advance care planning process, advance care planning topics that can be addressed, and creating a standard way of documenting advance care planning conversations [47–50]. As a result, we strongly recommend using a conversation guide to structure the advance care planning process (recommendation 8–10), keeping in mind that these conversation guides only provide guidance but are not all-encompassing. With consent of child and family, specific treatment agreements can be shared with all involved health care professionals (recommendation 11). Furthermore, the guideline development panel notes that leading advance care planning conversations requires specific communication skills, such as exploring child and family perspectives openly, neutrally, and empathetically, responding to emotions appropriately, and presenting the own professional perspectives based on scientific insights [51].

Recommendations on shared decision-making

With regards to shared decision-making, the guideline development panel acknowledged that the recommendations should be targeted at the application of shared decision-making in clinical practice and recognised this was dependent on legal and ethical considerations. Additionally, the panel concluded the identified evidence was not conclusive enough to formulate specific recommendations on the practical application of shared decision-making. According to the guideline development panel, the shared decision-making process is usually structured along four steps: (1) acknowledging that a decision needs to be made, (2) describing treatment options and (dis)advantages for each relevant option, (3) discussing the preferences, needs, and situation of child and family and possible consequences of each treatment option, (4) coming to a joint decision on the treatment policy and discussing preferences in the parents' decisional role. Although shared decision-making can often facilitate decision-making in paediatric palliative care, the guideline development panel noted that shared decision-making might not be appropriate when an emergency occurs which requires an immediate response. The guideline development panel therefore strongly recommends health care professionals to think ahead of time what treatment decision(s) must be made, consider in advance what treatment options are available, how these can best be explained, and that multiple conversations might be needed to come to a decision (recommendation 12–14). Additionally, it is strongly recommended to structure each conversation by using an agenda (recommendation 15). During each conversation health care professionals should strike a balance between the information they provide and receive, explain advantages and disadvantages clearly and concretely, give all participants the opportunity to ask questions, discuss the preferences of child and family, and provide their own preferences only if asked (recommendation 16–19).

Regarding child involvement, evidence did show that individual preferences regarding child involvement exist

Table 3 Recommendations on advance care planning and shared decision-making in paediatric palliative care

Recommendations on advance care planning and shared decision-making palliative care for children aged 0 to 18 years with life-threatening and life-limiting conditions, their families, and health care professionals (n=28)		
Advance care planning		
Do	1	Offer ACP conversations as a standard of care for all children with a palliative diagnosis and their families. In doing so, consider the specific situation and burden bearing capacity of child and family. [a]
	2	Integrate ACP as a continuous and dynamic process in the care of child and family from diagnosis through the end of life by holding regular conversations. [b]
	3	Start ACP conversations early in the disease process to encourage acceptance and allow space to prepare for the future. [b]
	4	Hold timely conversations with the child and family when the need to prepare for specific scenarios increases, as the child's condition deteriorates or when the child approaches end of life. [b]
	5	Provide explanations and written information to child and family to prepare for an ACP conversation and provide opportunities to include others they may wish to have present during the conversations. [b]
	6	Include the content of ACP conversations and any treatment arrangements in the medical record. [b]
	7	Have a health care provider trusted by the child and family lead the ACP conversations. This may be the primary caregiver, or another trusted caregiver, such as a case manager, or a caregiver outside the treatment team trained in ACP conversations. [b]
	8	When preparing and conducting ACP conversations, use a conversation guide to provide structure and to ensure relevant topics are covered.
	9	Structure the ACP process through preparation, interviewing and reporting.
	10	Discover, discuss, and note in the ACP process what the child and family's values, goals and preferences are for future care and treatment medically, psychologically, spiritually, and socially.
	11	With the consent of parents and child, share specific treatment agreements with all health care professionals involved.
Shared decision-making		
Do	12	Think ahead of time what treatment decision(s) must be made in the short and longer term and how you will explain the need for these decisions to child and/or parents.
	13	Consider in advance what (treatment) options are available and how you will explain them in a way that the child and/or parents can understand.
	14	Consider in advance how many conversations you think you will need to come to a decision and within what time frame you would prefer to have these conversations. Explain this to the child and parents.
	15	Start the conversation with an agenda and ask the child and/or his parents what they want to discuss.
	16	During the conversation, strike a good balance between the information you give and the information you want to receive from the child and/or his parents.
	17	Explain the advantages and disadvantages of the treatment options clearly and concretely. This includes the option to “wait and see” and the option to forgo further curative or life-sustaining treatments and focus entirely on comfort care.
	18	Give the child and/or his parents the opportunity to ask questions about the various treatment options and to share their views and experiences.
	19	Consult with the child and/or parents as to their preference and, if asked, explain your preference as well.
	20	Come to a decision that all involved are comfortable with and summarise it. Prevent the child and/or his parents from feeling too burdened by the responsibility of this decision.
	21	Involve the child even if he or she is under 12. Do this in a way that is developmentally appropriate. This also applies to the words you choose. [b]
Roles of child, family and health care professionals in advance care planning and shared decision-making		
Do	22	Involve child and family in framing ACP and shared decision-making in terms of form, content, preferred place, time and stakeholders and tailor this process to their needs. [b]
	23	Involve the (perspective of the) child in ACP conversations and shared decision-making in accordance with the child's developmental age. [b]
	24	In the ACP process and in shared decision-making, recognise the child and family as experts in living with illness and in assessing their quality of life. Ensure your actions reflect due consideration for the knowledge and experiences of child and family. [b]

Table 3 (continued)

Communication skills during ACP and shared decision-making		
Do	25	Regularly provide child and family with clear and honest information about diagnosis, prognosis, treatment, and uncertainties surrounding the child's situation throughout the disease process. Match this information to their (language) abilities and needs. [b]
	26	Use specific communication skills such as exploratory listening, acknowledging emotions, using concrete, appropriate and clear language, and formulating value-based goals during ACP conversations and shared decision-making. [b]
	27	During ACP conversations and shared decision-making, consider the communication preferences and cultural, religious, and philosophical beliefs of child and family. [b]
	28	Be aware in your preparation and during ACP conversations and shared decision-making that these discussions can be perceived as very difficult by the child and family. Do this by exploring child and family attitudes toward ACP and responding empathetically to their emotions without making the emotions the focus.

Abbreviations: ACP Advance care planning

[a] For this recommendation, very low to low quality evidence was identified

[b] For this recommendation, moderate quality evidence was identified

(moderate quality evidence) [26–31, 34, 36, 37, 39]. The guideline development panel considered this and recommended to always involve the child in a way that is developmentally appropriate, urging health care professionals to adapt the used language accordingly (recommendation 21).

Recommendations on roles of child, family and health care professionals in advance care planning and shared decision-making

The evidence showed that parents and health care professionals reported individual variation in preferred level of parent involvement, varying from parents as the final decision-makers, decision making by parents in collaboration with health care professionals, preference of parents not to be involved in decision-making, and sometimes parents found it was not possible to make a decision (moderate quality of evidence) [25–31, 37, 40, 41, 43, 45]. Similarly, children, parents and health care professionals reported variation in preferred level of child involvement, which was often dependent on age, the decision at hand, and the child's situation (moderate quality of evidence) [26–31, 34, 36, 37, 39]. Only two studies reported the preferences of children themselves [26, 34]. The guideline development panel acknowledged level of involvement should be tailored to the needs and preferences of child and family. We strongly recommend involving child and family in advance care planning and shared decision-making conversations (recommendation 22). Additionally, we strongly recommend that (the perspective of) the child should be involved while acknowledging the child's developmental age (recommendation 23). We found moderate quality evidence that parents should be acknowledged as the expert of the child [6, 29–31, 35, 36, 38, 39, 41, 43, 45]. The panel translated this in a strong recommendation (recommendation 24).

Recommendations on communication skills during advance care planning and shared decision-making

Relating to communication, several facilitators have been identified. The panel has used these facilitators to formulate recommendations. As a result, we strongly recommend clear and honest information about diagnosis, prognosis, treatment and uncertainties [25, 26, 28, 29, 31, 32, 35, 37, 39–41, 43, 45], the use of communication skills such as using concrete and appropriate language, explorative listening and acknowledging emotions [31, 35, 43], acknowledging communication preferences, and cultural, religious, and philosophical beliefs of child and family (moderate quality evidence) [26, 28, 36, 37, 39, 43, 44] (recommendation 25–27). Furthermore, evidence showed that parents experienced difficulties with regards to advance care planning and shared decision-making as they did not always feel ready to make decisions, could not foresee consequences or were conflicted in not wanting their child to suffer but also want their child to live as long as possible (moderate quality evidence) [25, 26, 28, 29, 36–38, 45]. Therefore, we strongly recommend health care professionals to acknowledge this when preparing for advance care planning conversations and shared decision-making (recommendation 28).

Discussion

Advance care planning and shared decision-making in children with life-threatening and life-limiting conditions and their families are essential to paediatric palliative care [2, 3]. These strategies have proven to enhance collaboration among children, families, and health care professionals, and decrease concerns about the future [52, 53]. As a result, advance care planning and shared decision-making are expected to contribute to quality of care for children and their families [52].

In the Netherlands, health care professionals, parents, and other stakeholders have expressed the need for guidance with regards to advance care planning and shared decision-making in paediatric palliative care [6, 15, 54]. We responded to this need by developing evidence-based recommendations on these topics as part of the revised Dutch CPG for paediatric palliative care. In this paper, we present the identified evidence and provide recommendations to optimise advance care planning and shared decision-making in paediatric palliative care in the Netherlands.

Our recommendations are developed by following an evidence-based approach in which we identified both quantitative and qualitative evidence. First, we systematically searched for quantitative evidence (RCTs, CCTs and SRs of RCTs) on the effects of advance care planning and shared decision-making interventions. Second, we searched for qualitative evidence on possible barriers and facilitators related to advance care planning and shared decision-making [15]. We included a total of four RCTs and 33 qualitative studies. Finally, all recommendations were formulated in close collaboration with a national multidisciplinary guideline development panel consisting of professionals from multiple disciplines and parents. Based on evidence from international literature, clinical expertise, and patient and family values, we were able to compile a comprehensive set of strong recommendations that provide guidance on advance care planning and shared decision-making in paediatric palliative care.

We identified four RCTs that reported on the effects of advance care planning interventions. These studies showed that an advance care planning intervention can enhance agreement on future treatment preferences among adolescents with cancer or an HIV-infection and their adult surrogates without negatively impacting quality-of-life related outcomes. Mainly, due to the imprecision of effects (small number of participants) and potential risk of bias, the total body of evidence was rated as low to very low quality. Despite the low to very low quality evidence, outcomes of advance care planning interventions are promising. In fact, other studies strongly indicate that advance care planning interventions can decrease feelings of stress, helplessness, anxiety, and depression among families of paediatric patients [55, 56].

Furthermore, we found a total of 33 qualitative studies that reported on barriers and facilitators to advance care planning and shared decision-making, of which 22 studies were newly published and 11 studies were identified in the original search of the NICE-guideline [18]. Meaning that, since the original search of the NICE-guideline in 2016, the total number of included qualitative studies has tripled. As the evidence on barriers and facilitators related to advance care planning and shared decision-making increased substantially, we are able to identify a large set of barriers and facilitators that were reported in various studies by multiple stakeholders. This allowed us to allocate each identified barrier and facilitator to the reporting stakeholder group, namely children, parents, and health care professionals and compare the results. Through this comparison, which was not performed in the NICE-guideline, we found that the different stakeholders mostly reported similar barriers and facilitators. After appraisal of the evidence, we found the majority of identified barriers and facilitators to advance care planning and shared decision-making were of moderate quality evidence. As the conclusions of evidence in the NICE-guideline were largely appraised as low to very low quality, we can conclude that the evidence base on barriers and facilitators related to advance care planning and shared decision-making is strengthened.

It should be noted that although we identified both quantitative evidence and qualitative evidence, there are still several knowledge gaps that should be addressed. We identified only very low to low quality of evidence on the effect of advance care planning interventions which focused on a specific group of patients diagnosed with cancer or HIV-infection. Also, the evidence did not report on the effect of advance care plans that were not followed or modified. As advance care plans can be adjusted over time due to changes in goals and preferences of children and family, investigating the effect and frequency of advance care plans that were not followed or modified, can facilitate a more comprehensive understanding on the effects of advance care planning. Furthermore, we identified no evidence on the effect of shared decision-making interventions in paediatric palliative care. Additionally, we found limited qualitative evidence on the barriers and facilitators reported by children. Only two qualitative studies included children in their study population. In these studies, children reported barriers and facilitators related to two out of ten identified themes, i.e. information provision and child involvement in advance care planning and shared decision-making. Based on the identified knowledge gaps, we call for more research on the effect of advance care planning and shared decision-making interventions in paediatric palliative care and its barriers and facilitators. Simultaneously, future research should focus on addressing the child's perspective in an age-appropriate way [57].

With regard to the recommendations a few points should be addressed. First, due to identified knowledge gaps, a large proportion of our recommendations were primarily based upon clinical expertise and the values of patients and their families. The incorporation of clinical expertise and patient and family values is considered invaluable for clinical decision-making [58, 59]. As a result, both clinical expertise and patient and family values are essential to interpret evidence and formulate recommendations which can improve the overall quality of CPGs [60]. However, it should be noted that recommendations based upon clinical expertise and patient and family values only, can be prone to personal bias [61]. To address this, recommendations were developed according to a rigorous process, in which we only approved recommendations that had group consensus. Unfortunately, clinical expertise and patient and family values were not systematically collected. To improve transparency and minimise personal bias, using surveys to systematically and independently gather the opinions of experts, patients and their families could be a useful strategy for future guideline updates [61].

Second, when translating evidence into recommendations, it became clear that advance care planning and shared decision-making can be beneficial to children with life-threatening and life-limiting conditions and their families. However, the guideline development panel emphasised that advance care planning and shared decision-making are dynamic and continuous processes that should be tailored to child and family. We recognise that provision of tailored advance care planning and shared decision-making requires specific skills such as exploring child and family perspectives openly, responding to emotions appropriately, and presenting the own professional perspectives [51]. Furthermore, we realise that both advance care planning and shared decision-making are considered time-consuming as these services often require multiple conversations and should be documented correctly. Thus, provision of tailored advance care planning and shared decision-making, requires training and education to develop specific skills, adequate staffing and funding. These are essential preconditions for the application of recommendations in clinical practice.

Finally, with regard to international use of these recommendations, it is important to note that our recommendations were informed by studies which were primarily conducted in countries associated with western culture such as the United States of America, the United Kingdom, the Netherlands, and Germany. Furthermore, all recommendations were formulated by a Dutch guideline development panel which considered essential factors, such as legal and ethical considerations, that are specific to the Netherlands. As a result, our recommendations are primarily applicable to the Netherlands. However, we believe that our recommendations can be used as a starting point to develop recommendations in other countries. We do emphasise that even between different western cultures considerable adaptations to recommendations will be required to meet the needs of a specific population [62]. Populations not associated with western culture will need even more comprehensive changes [62]. This is particularly relevant as the principles of advance care planning and shared decision-making are rooted in western culture, making it very challenging and in some cases not possible to adopt recommendations in non-western countries [57, 62–64]. Thus, country-specific factors, such as cultural background, legal and ethical considerations, and organisational infrastructure should be very carefully considered before applying any recommendations in other settings.

In conclusion, we developed recommendations based on strengthened evidence in close collaboration with a multidisciplinary guideline panel of professionals and parents. With these evidence-based recommendations, we aim to optimise advance care planning and shared decision-making in paediatric palliative care in the Netherlands. Both identified evidence as well as the formulated recommendations support the use of advance care planning and shared decision-making in paediatric palliative care. However, we identified several knowledge gaps that should be addressed to further optimise advance care planning and shared decision-making. Also, advance care planning and shared decision-making require specific skills and is time-consuming. Therefore, we emphasise the importance of education, adequate staffing and funding to improve integration in clinical practice. With regard to international use of recommendations, we believe that our recommendations can be used as a starting point to develop recommendations in other countries. However, country-specific factors should be very carefully considered before applying any recommendations in other countries.

Supplementary Information

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Supplementary Material 1

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Authors' contributions

KvT, RM, JV, HR, BB, LK, EM and EV conceived and designed the study. KvT, RM, DvH and EM performed the search, data extractions, risk of bias assessment, and GRADE (CERQual) assessment. KvT, RM, DvH, JF, MdV, LK, MK, EM, and EV interpreted the data. KvT, RM, DvH, JF, MdV, JV, HR, BB, LK, MK, EM, and EV contributed to the formulation of the recommendations. KvT, RM, LK, MK, EM, and EV drafted the manuscript; and all authors (KvT, RM, DvH, JF, MdV, JV, HR, BB, LK, MK, EM, and EV) critically revised the manuscript. All authors and the collaborators approved the final version of this paper.

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Availability of data and materials

All data analysed during this study is included in this published article and additional information file 1.

Declarations

Ethics approval and consent to participate

All methods were carried out in accordance with relevant guidelines and regulations, all guideline panel members have given consent to participate. No institutional or other licensing committee's approval is needed for guideline creation, as participants are not subjected to procedures and are not required to follow rules of behaviour. Therefore, in accordance to the Dutch law (Medical Research Involving Human Subjects Act (WMO), article 1b) ethics approval was deemed unnecessary: https://english.ccmo.nl/investigators/legal-framework-for-medical-scientific-research/your-research-is-it-subject-to-the-wmo-or-not.

Consent for publication

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Competing interests

The authors declare no competing interests.

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