BMJ Open

Clinical Ethics Support Services in Paediatric Practice: Protocol for a mixed studies systematic review on structures, interventions and outcomes.

Journal:	*BMJ Open*
Manuscript ID	bmjopen-2021-057867
Article Type:	Protocol
Date Submitted by the Author:	20-Oct-2021
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Keywords:	PAEDIATRICS, MEDICAL ETHICS, Quality in health care < HEALTH SERVICES ADMINISTRATION & MANAGEMENT

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TITLE

Clinical Ethics Support Services in Paediatric Practice: Protocol for a mixed studies systematic review on structures, interventions and outcomes.

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KEY WORDS

Paediatrics, Clinical Ethics Committees, Clinical Ethics, Quality Indicators, Systematic Review

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ABSTRACT

Introduction: Clinical Ethics Support Services (CESS) have been developing worldwide with growing interest in evaluating their quality. Paediatric-specific CESSs (p-CESS) have received little attention, and evidence from adult services might not be generalisable. Evidence on service models and practices is crucial to inform further research and debate on quality evaluation and minimum standards for p-CESSs. We aim to systematically identify, appraise and synthesise evidence for p-CESS structures, processes, and outcomes.

Methods and analysis: We will conduct a mixed-studies systematic review including peer-reviewed empirical studies published in English or Spanish language providing data on the evaluation and/or impact on any aspect of p-CESS. We will search seven electronic databases: MEDLINE, Philosopher's Index, EMBASE, PsychINFO, LILACS, Web of Science and CINHAL, without filters applied. Search terms will be related to "clinical ethics support" AND "paediatrics" AND "structure/process/outcome". Reference and citation list of included studies will be hand-searched. A 10% random sample of retrieved titles/abstracts and all full texts will be independently dual-screened. We will conduct thematic and narrative synthesis for qualitative and quantitative data, respectively, following a sequential exploratory design guided by Donabedian's framework of structure, process and outcomes. Quality will be assessed using the Mixed-Methods Appraisal Tool (2018). The review will be reported using the 2020 Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement.

Ethics and dissemination: As a systematic review of published data, no ethical approval is necessary. Results will be published in a relevant academic peer-reviewed journal.

Registration: The protocol has been prospectively registered in PROSPERO (CRD42021280978)

STRENGTHS AND LIMITATIONS OF THIS STUDY

- We will conduct an electronic search using a wide range of electronic databases, including a Latin American database, which will broaden the coverage of publication countries.
- The use of the Donabedian framework will allow an objective assessment of the CESS contribution to the quality of medical care.
- The review will be conducted by a bilingual and international research team, contributing different experiences and perspectives on CESS structures, processes, and outcomes in different contexts (Latin America and the UK).
- However, only English and Spanish language studies will be included with the consequent potential exclusion of relevant articles.

INTRODUCTION

Clinical ethics support is the provision of support and advice to health professionals, patients and families on ethical issues arising from clinical practice or patient care.(1–3) Accordingly, Clinical Ethics Support Services (CESS) are institutionalised forms of ethics support within healthcare organisations.(2)

CESS were initially developed in the USA in 1970-80 in response to government and medical societies' recommendations,(4) and has since spread progressively worldwide, but in an uneven and very varied way.(1,5–7) Forms of, and access to, CESS varies across different centres, countries, and cultural contexts.(6,8) In some countries, the constitution of institutional CESS is recommended or required by the government and subject to official regulation, while in others, such as the UK, the organisation and function of these services lack official guidance.(1)

Traditionally, four main CESS functions have been described; clinical case consultation, education, institutional policy development, and research.(3,9) Multiple models of CESS have been described, including individual ethical case consultation, clinical ethics committees, individual ethicists, moral case deliberation, ethics rounds and ethics discussion forums.(1,10–12) Informal provision of clinical ethics support has also been reported.(10)

Unlike Research Ethics Committees (REC), CESS have been criticised for lack of standardisation, an absence of regulation of their structures, skill requirements, role and remit and the paucity of formal evaluation of their impact.(13) Even though there has always been an awareness of the need for systematic evaluation of CESS outcomes and effectiveness,(14–16) and there is a growing body of theoretical and empirical literature addressing CESS' evaluation, there remain no agreed standards or quality indicators for these services.(17–19)

Schildmann et al. defined the evaluation of CESS as "the systematic gathering of data with empirical research methods for the purpose of acquiring knowledge about the structure, functioning, quality

and results of CESS".(p681, 20) Following this definition, in line with the widely used Donabedian model for evaluating the quality of medical care,(21) a comprehensive evaluation of the quality of CESS should include three dimensions of care: structure, process and outcomes.(2,21,22) As described by Donabedian, quality measurement standards derive from both empirical and normative sources.(21) Considering CESS have an explicit normative character, identifying appropriate quality criteria is particularly complicated, and this normative feature should be reflected when defining assessment measures.(23) Widely used outcome measures such as length of stay, mortality, or financial impact are not be necessarily helpful in evaluating CESS.(22) For example, any evaluation of ethics consultation services focused on pre-established outcome measures should not ignore the consultation's central aim of responding to the ethical queries presented by those requesting support with a particular clinical case. Therefore, an appropriate evaluation system must allow for the context and particularities of each case to be considered.(24) Additionally, CESS evaluation should involve all stakeholders, including both those who receive and those who provide ethics support (i.e., healthcare professionals, patients and relatives, CESS members, hospital management).(25) Moreover, considering that CESS are, by definition, engaged in complex interventions where multiple components and interactions impact the final outcome,(2) a clear understanding of how they function is vital, before attempting any evaluation.(2)

Paediatric practice raises particular ethical challenges not frequently found in adult patient care.(26–29) The fundamental principle of respect for patient autonomy has a substantially different understanding in paediatric practice, with parents taking the responsibility of decision-making until children are afforded that possibility.(26) Generally, parents' decisions are in coherence with the child's views and the child's best interest, but conflict might arise when those involved (clinical team, parents, child) hold different views. Additionally, the child's capacity to understand the information provided and contribute to, or even make, decisions about their care increases with maturity. Because of this, careful assessment of the child's competence needs to be made, particularly for decisions with moral significance.(26) The level of involvement of parents and patients in ethics discussions is a

controversial point, particularly in a paediatric setting.(30) The paediatric landscape has changed with technological advances, lower mortality rates in many specialities and an increasing number of patients with chronic and complex conditions. Uncertainties about prognostication and treatment outcomes, overall benefits and burdens pose ethical challenges about withholding and withdrawing life-sustaining treatments.(31) The involvement of multiple teams with different perspectives and values might add further complexity to the decision-making process.(29) Finally, these significant technological advances and decreasing mortality rates might strengthen the perception of death, particularly in children, as a medical failure adding barriers to end of life discussions and decision-making.

Despite ethically challenging situations and consequent divergent opinions being common in paediatrics, the number of paediatric ethics consultations is relatively low.(32) Many of these challenges might be opportunely identified and appropriately managed by the healthcare team and the family,(33) with clinicians receiving support through alternatives to formal ethics consultation.(32) However, in complex cases, there may either be an impasse or conflict might persist. In these situations, ethical consultation has been shown to help provide a resolution.(34)

Recent controversial cases featured extensively in both print, and social media have increased international public and academic attention to the ethical challenges of paediatric practice. There has been an increased interest and debate surrounding the legal and ethical aspects of paediatric practice, such as parental and clinical teams' disagreements about the child's best interests, emerging child capacity, innovative treatments, technological dependence and end-of-life decision-making. Consultant paediatricians, trainees and residents have expressed concerns that these public cases might negatively impact their relationship with patients and families.(35) Availability of clinical ethics support services for teams facing ethical challenges has been strongly advocated by professional bodies,(31) academics(33,36,37) and clinicians.(35,38)

Interest in assessing CESS quality and effectiveness has grown in the past decades. However, most studies have focused on adult care settings, with relatively little attention paid to p-CESS.(39) Multiple systematic reviews evaluating different aspects of CESS have recently been published. Nevertheless, these focus on adult patients; ethical case intervention,(40) adult end of life context,(41) and adult ICU.(42) Other reviews that did not explicitly focused on adult patients evaluate a specific intervention (assessment tools for evaluating clinical ethics consultation(17)) or effectiveness of clinical ethics committees.(18) Generalisation of adult-focused reviews and evidence to paediatric context might not be appropriate.

To our knowledge, no systematic review on Paediatric Clinical Ethics Support Services (p-CESS) structures, processes, evaluation measures and outcomes has been published. Such a review is necessary to inform current p-CESS practice and further development. Therefore, we aim to inform further research and debate on the current quality evaluation and minimum standards for p-CESS by offering a comprehensive description of current p-CESS models and assessments by responding to this review question:

> ***"What is the range of structures, processes, and outcome measures of paediatric CESS reported in the literature?"***

Aim:

To systematically identify, appraise and synthesise evidence for p-CESS structures, processes, and outcome measures described in the literature.

Objectives:

- To identify and synthesise published data on p-CESS regarding their structures, processes, evaluation measures and outcomes.
- To explore the impact of p-CESS given the outcomes identified in the review.
- To qualitatively appraise the available evidence.

- To develop a preliminary framework for the evaluation of p-CESS based on available evidence.
- To provide recommendations for further research on CESS effectiveness and outcome measures in paediatric practice.

METHODS AND ANALYSIS

We will conduct a systematic review following the PRISMA Statement for Reporting Systematic Reviews and Meta-Analyses (PRISMA) 2020 guidelines(43) to identify and synthesise evidence for Paediatric CESS structures, interventions, evaluation measures and outcomes.

The review protocol has been developed following the recommended items included in the PRISMA-P statement(44) (See Supplemental file 1 for PRISMA-P Checklist) and has been prospectively registered in PROSPERO registry CRD42021280978

Eligibility Criteria

The inclusion and exclusion criteria are summarised in Table 1. The review will include peer-review empirical studies (qualitative, quantitative or mixed-methods), including case studies published in English or Spanish language which provide empirical data on evaluation, assessment and/or impact (i.e., effect, evaluation, importance, meaning, value)(11) of any one or more of the following aspects of paediatric CESS: service structure, constitution and membership, service's aims and functions, interventions and processes, and outcome measures of p-CESS. We will include empirical studies of qualitative, quantitative or mixed-methods design reporting both objective and/or subjective measures.

We will include studies reporting on CESS that provide services to adult and paediatric patients only if paediatric data can be extracted separately.

Non-peer-reviewed studies, reviews, theoretical works, editorials, letters, opinion pieces, book chapters will be excluded. Conference abstracts will not be included, but authors will be contacted

asking whether the relevant work has been published, with a two-week timeframe allowed for a response. There will be no timeframe or geographical restrictions.

Table 1. Eligibility Criteria

	INCLUSION CRITERIA	EXCLUSION CRITERIA
Type of participants	CESSs that serve paediatric only or paediatric and adult population, where paediatric data can be extracted separately. The paediatric population will be defined in this review as between 0-18 years old. Study participants include, but are not limited to, referring clinicians, CESS members, patients/children, parents/relatives/careers and hospital administrators.	CESSs that serve only adult patients. CESSs serving paediatric and adult populations, where paediatric data cannot be extracted and analysed
Context/ setting	Articles reporting on established CESS serving paediatric practice in any setting (hospital, community) and country.	Research ethics committees

Issues	Empirical studies reporting data on evaluation, assessment and/or impact (i.e., effect, evaluation, importance, meaning, value) of any one or more of, but not limited to the following aspects of paediatric CESS: service structure, constitution and membership, service's aims and functions, interventions and processes, outcome measures of paediatric CESS.	Theoretical analysis or narrative reviews on paediatric CESS. Studies focusing only on a description of the paediatric CESS without reporting assessment/impact data
Methods	Empirical studies of any methods (qualitative and/or quantitative), including case studies.	Theoretical reviews or analysis. Systematic reviews Case reports, narrative reviews.
Timeframe	Any time frame. Searches will be conducted from the database inception date until the search date.	
Type of publication	Peer-reviewed publications in English or Spanish Language	Non-peer-reviewed studies, reviews, theoretical works, editorials, letters, opinion pieces Conference abstracts
CESS: Clinical Ethics Support Services		

Search strategy

Electronic searches. The following databases will be searched: MEDLINE, Philosopher's Index, EMBASE, PsychINFO, LILACS, Web of Science and CINHAL. There will be no methodological, language, geographical or time filters applied to the search strategy. If a non-English paper is considered eligible for inclusion, relevant data and results will be translated to English before analysis.

The initial search strategy was developed considering previously published systematic reviews in paediatrics, clinical ethics and service evaluation. Search terms will be related to "Clinical ethics support", "paediatrics" AND "structure/process/outcome indicators" and adapted to each database requirement. Publications that would match the criteria for inclusion in the review previously known to the research team were successfully retrieved applying the search strategy in Medline online database (See Table 2). The search strategy will be checked and adjusted to the other electronic databases as appropriate.

Table 2. Medline search strategy

1. paediatric.mp. 2. paediatr*.mp. 3. pediatric.mp. or Pediatrics/ 4. pediatr*.mp. 5. child*.mp. or Child/ 6. 6Adolescent/ or adolescent.mp. 7. adolesc*.mp. 8. infant*.mp. 9. infant.mp. or Infant/ 10. kid.mp. 11. kids.mp. 12. baby.mp. 13. babies.mp. 14. toddler*.mp. 15. childhood.mp. 16. juvenil*.mp. 17. youth*.mp.	27. Ethicists/ or ethicist*.mp. 28. bioethicist*.mp. 29. medical ethics.mp. or Ethics, Medical/ 30. clinical ethics.mp. or Ethics, Clinical/ 31. clinical ethics committee.mp. or Ethics Committees, Clinical/ 32. bioethics.mp. or Bioethics/ 33. bioethical issues.mp. or Bioethical Issues/ 34. ethical issues.mp. or Ethics/ 35. ethical challenges.mp 36. moral review.mp. 37. Ethical Analysis/ or Ethics Consultation/ or moral case deliberation.mp. 38. moral case consult*.mp. 39. moral consult*.mp.	57. structure.mp. 58. model*.mp. 59. process*.mp. 60. intervention*.mp. 61. Outcome Assessment, Health Care/ or "Outcome and Process Assessment, Health Care"/ or assessment*.mp. 62. evaluation*.mp. 63. impact*.mp. 64. effectiveness.mp. 65. Medical Audit/ or Clinical Audit/ or audit.mp. 66. Outcome Assessment, Health Care/ or "Outcome and Process Assessment, Health Care"/ or outcome.mp.

18. minor.mp. or Minors/ 19. Infant, Newborn/ or infancy.mp. or Child, Preschool/ 20. Infant, Newborn/ or newborn*.mp. 21. Premature Birth/ or Infant, Premature/ or preterm*.mp. 22. prematur*.mp. 23. Puberty/ or pubert*.mp. 24. pubescen*.mp. 25. young person.mp. 26. neonatal.mp.	40. ethic* case review.mp. or Ethics Committees/ 41. ethic* deliberation.mp. 42. ethic* intervention.mp. 43. ethic* round.mp. 44. ethic* support.mp. 45. ethic* service.mp. 46. Ethical Analysis/ or ethic* analysis.mp. 47. ethic* referral.mp. 48. Ethics Committees/ or ethic* committee.mp. 49. bioethic* deliberation.mp. 50. bioethic* intervention.mp. 51. bioethic* round.mp. 52. bioethic* service.mp. 53. bioethic* support.mp. 54. bioethic* analysis.mp. 55. bioethic* referral.mp. 56. bioethic* committee.mp.	67. "Quality of Health Care"/ or quality.mp. or Quality Indicators, Health Care/ 68. indicator*.mp.
69. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23 or 24 or 25 or 26 70. 28 or 29 or 30 or 31 or 32 or 33 or 34 or 35 or 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49 or 50 or 51 or 52 or 53 or 54 or 55 or 56 or 57 71. 59 or 60 or 61 or 62 or 63 or 64 or 65 or 66 or 67 or 68 or 69 or 70 72. 69 AND 70 AND 71		

Other resources. Reference and citation list of included studies will be hand-searched. When relevant, we will contact the authors of conference abstracts identified through the search for peer-review publications. We will allow a time frame of two weeks for a response before considering the publication unavailable.

Selection process.

All retrieved records will be managed using Refworks® reference manager software. After deduplication, all titles and/or abstracts will be screened by MD to identify publications that meet the previously established inclusion and exclusion criteria. A random sample of 10% will be independently screened by a second researcher (BP) to test the reliability of the criteria. Any disagreements will be discussed within the research team until agreement, and, if required, eligibility criteria will be adjusted

and/or clarified to improve the consistency of the screening process. Selected references for assessment in the full text will be dual-screened, and any disagreements will be discussed within the research team until agreement. The screening process will be presented as a PRISMA flowchart.

Data extraction.

Data from individual studies considered relevant for the review question will be extracted to a pre-piloted Excel data extraction form by MD and checked by BP. Disagreements will be discussed within the research team. Data entries will include: Publication details (First author, year of publication, title), country, study aims, study design, sample description, CESS description (design, delivery, organisation, function, aims and interventions), study intervention, outcome measures, findings, comments. For qualitative studies, all data within the results/finding section will be considered as results.

MD and BP will independently extract the data, and any disagreements will be discussed within the team until we achieve consensus. This will include publication details (author, year, title), study design and instrument, setting (country, healthcare setting), sampling method, and sample characteristics. Primary outcomes sought in the data set will include; (i) assessed aspects of paediatric CESS as reported by study authors, including service structures, processes and outcomes; and (ii) assessed dimensions (i.e., effectiveness, safety and responsiveness) and/or subdimensions of quality of care(45) as reported by study authors.

Quality assessment.

Following our initial scoping review, we expect to find potentially eligible studies with multiple research designs, including qualitative, quantitative and mixed methods. To allow coherent and systematic critical appraisal of included studies with different designs, we will use the 2018 Mixed Methods Appraisal Tool.(46) The tool was developed specifically for the appraisal of complex

systematic reviews that include empirical qualitative, quantitative and mixed methods studies. It includes specific criteria depending on the study design category. Each criterion is rated as “yes”, “no”, “can’t tell” response. As recommended by the authors, for each study, we will present a detailed description of the rating of each criterion and calculate an overall quality score based on the number of quality criteria met. There is no recommended cut off score to exclude studies based on quality appraisal, and therefore no study will be excluded based on that criterion. Instead, we will conduct a sensitivity analysis during the data synthesis process to assess the impact of low-quality studies in the review findings and adjust recommendations accordingly.(46) Quality appraisal will be conducted by MD and checked by BP.

Data synthesis

This systematic review is not focused only on the effectiveness of a particular intervention but addresses a broader range of questions. Therefore we will follow a modified narrative synthesis approach to synthesise findings.(47) The iterative process will include developing a preliminary synthesis of findings of included studies, exploring relationships in the data, and assessing the robustness of the synthesis by considering individual studies’ quality and sensitivity analysis if possible. Data synthesis will follow a sequential exploratory design by first conducting synthesis of qualitative data followed and informing synthesis of quantitative data, looking for divergences and/or convergences and knowledge gaps across the data set. (47) Qualitative data will be analysed by the thematic synthesis approach,(48) using NVivo software for qualitative data analysis. The synthesis will include: (i) Free line-by-line coding of the primary study’s findings, and (ii) organisation of these codes into related areas to construct descriptive themes. After that, the narrative synthesis of quantitative findings will be integrated into the qualitative synthesis, followed by the development of overarching themes (48) guided by but not limited to Donabedian’s framework of structure, process and

outcomes. Thematic and narrative analysis will be conducted by MD and checked by BP. The final synthesis will be discussed within the research team.

Ethics and dissemination.

As a systematic review of published data, no ethical approval is necessary. We will present and discuss our findings in an open-access webinar, including invited experts in the field. A final report will be published in a relevant academic peer-reviewed journal.

Patient and public Involvement.

Patients and the public were not involved in the design of this systematic review protocol.

DISCUSSION

Interest in assessing CESS quality and effectiveness has grown in the past decades, with little attention paid to p-CESS. The results of this work will provide us with the first systematic review of evidence on Paediatric Clinical Ethics Support Services. We hope that our review results will allow for a better understanding of p-CESS structures, processes, and outcomes, contributing to further research exploring the normative and empirical basis of p-CESS. We plan to continue this research by conducting a modified Delphi study based on our review results to explore the most appropriate quality indicators for evaluating p-CESS. These outputs are vital if we aim to ensure CESS contribute to better quality care for patients and their families.

Author contributions: MD and JB conceived the review. MD, JB and BP developed the protocol. All authors revised and edited the manuscript and approved the final version.

Funding: This work was supported by the Institute of Medical Ethics "Research in Medical Ethics" grant (No grant number). The funder has had no involvement in the review protocol development.

Competing interests: None to declare.

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PRISMA-P (Preferred Reporting Items for Systematic review and Meta-Analysis Protocols) 2015 checklist: recommended items to address in a systematic review protocol*

Section and topic	Item No	Checklist item	Page in text
ADMINISTRATIVE INFORMATION			
Title:			
Identification	1a	Identify the report as a protocol of a systematic review	1
Update	1b	If the protocol is for an update of a previous systematic review, identify as such	n/a
Registration	2	If registered, provide the name of the registry (such as PROSPERO) and registration number	2, 8
Authors:			
Contact	3a	Provide name, institutional affiliation, e-mail address of all protocol authors; provide physical mailing address of corresponding author	1
Contributions	3b	Describe contributions of protocol authors and identify the guarantor of the review	15
Amendments	4	If the protocol represents an amendment of a previously completed or published protocol, identify as such and list changes; otherwise, state plan for documenting important protocol amendments	n/a
Support:			
Sources	5a	Indicate sources of financial or other support for the review	15
Sponsor	5b	Provide name for the review funder and/or sponsor	15
Role of sponsor or funder	5c	Describe roles of funder(s), sponsor(s), and/or institution(s), if any, in developing the protocol	15
INTRODUCTION			
Rationale	6	Describe the rationale for the review in the context of what is already known	4-7
Objectives	7	Provide an explicit statement of the question(s) the review will address with reference to participants, interventions, comparators, and outcomes (PICO)	7-8
METHODS			
Eligibility criteria	8	Specify the study characteristics (such as PICO, study design, setting, time frame) and report characteristics (such as years considered, language, publication status) to be used as criteria for eligibility for the review	8-9 Table 1
Information sources	9	Describe all intended information sources (such as electronic databases, contact with study authors, trial registers or other grey literature sources) with planned dates of coverage	11-12
Search strategy	10	Present draft of search strategy to be used for at least one electronic database, including planned limits, such that it could be repeated	11-12 Table 2

Study records:			
Data management	11a	Describe the mechanism(s) that will be used to manage records and data throughout the review	12
Selection process	11b	State the process that will be used for selecting studies (such as two independent reviewers) through each phase of the review (that is, screening, eligibility and inclusion in meta-analysis)	12-13
Data collection process	11c	Describe planned method of extracting data from reports (such as piloting forms, done independently, in duplicate), any processes for obtaining and confirming data from investigators	13
Data items	12	List and define all variables for which data will be sought (such as PICO items, funding sources), any pre-planned data assumptions and simplifications	13
Outcomes and prioritization	13	List and define all outcomes for which data will be sought, including prioritization of main and additional outcomes, with rationale	13
Risk of bias in individual studies	14	Describe anticipated methods for assessing risk of bias of individual studies, including whether this will be done at the outcome or study level, or both; state how this information will be used in data synthesis	13-14
Data synthesis	15a	Describe criteria under which study data will be quantitatively synthesised	14-15
	15b	If data are appropriate for quantitative synthesis, describe planned summary measures, methods of handling data and methods of combining data from studies, including any planned exploration of consistency (such as I^2, Kendall's τ)	14-15
	15c	Describe any proposed additional analyses (such as sensitivity or subgroup analyses, meta-regression)	14-15
	15d	If quantitative synthesis is not appropriate, describe the type of summary planned	14-15
Meta-bias(es)	16	Specify any planned assessment of meta-bias(es) (such as publication bias across studies, selective reporting within studies)	n/a
Confidence in cumulative evidence	17	Describe how the strength of the body of evidence will be assessed (such as GRADE)	n/a

*** It is strongly recommended that this checklist be read in conjunction with the PRISMA-P Explanation and Elaboration (cite when available) for important clarification on the items. Amendments to a review protocol should be tracked and dated. The copyright for PRISMA-P (including checklist) is held by the PRISMA-P Group and is distributed under a Creative Commons Attribution Licence 4.0.**

From: Shamseer L, Moher D, Clarke M, Ghersi D, Liberati A, Petticrew M, Shekelle P, Stewart L, PRISMA-P Group. Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015: elaboration and explanation. BMJ. 2015 Jan 2;349(jan02 1):g7647.

BMJ Open

Clinical Ethics Support Services in Paediatric Practice: Protocol for a mixed studies systematic review on structures, interventions and outcomes.

Journal:	*BMJ Open*
Manuscript ID	bmjopen-2021-057867.R1
Article Type:	Protocol
Date Submitted by the Author:	07-Dec-2021
Complete List of Authors:	Dittborn, Mariana; Great Ormond Street Hospital for Children, Paediatric Bioethics Centre ; Universidad del Desarrollo, Centro de Bioética Portales, Bernardita; Universidad del Desarrollo, Centro de Bioética Brierley, Joe; Great Ormond Street Hospital for Children, Paediatric Bioethics Centre
Primary Subject Heading:	Ethics
Secondary Subject Heading:	Paediatrics
Keywords:	PAEDIATRICS, MEDICAL ETHICS, Quality in health care < HEALTH SERVICES ADMINISTRATION & MANAGEMENT

1

TITLE

Clinical Ethics Support Services in Paediatric Practice: Protocol for a mixed studies systematic review on structures, interventions and outcomes.

AUTHORS

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WORD COUNT 3505

KEY WORDS

Paediatrics, Clinical Ethics Committees, Clinical Ethics, Quality Indicators, Systematic Review

ABSTRACT

Introduction: Clinical Ethics Support Services (CESS) have been developing worldwide with growing interest in evaluating their quality. Paediatric-specific CESSs (p-CESS) have received little attention, and evidence from adult services might not be generalisable. Evidence on service models and practices is crucial to inform further research and debate on quality evaluation and minimum standards for p-CESSs. We aim to systematically identify, appraise and synthesise evidence for p-CESS structures, processes, and outcomes.

Methods and analysis: We will conduct a mixed-studies systematic review including peer-reviewed empirical studies published in English or Spanish language providing data on the evaluation and/or impact on any aspect of p-CESS. We will search seven electronic databases: MEDLINE, Philosopher's Index, EMBASE, PsychINFO, LILACS, Web of Science and CINHAL, without filters applied. Search terms will be related to "clinical ethics support" AND "paediatrics" AND "structure/process/outcome". Reference and citation list of included studies will be hand-searched. A 10% random sample of retrieved titles/abstracts and all full texts will be independently dual-screened. We will conduct narrative and thematic synthesis for quantitative and qualitative data, respectively, following sequential explanatory synthesis guided by Donabedian's framework of structure, process and outcomes. Quality will be assessed using the Mixed-Methods Appraisal Tool (2018). The review will be reported using the adapted Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) for reporting systematic reviews of qualitative and quantitative evidence template. Stakeholders will be involved twice in the review process; prior to data extraction and synthesis and after preliminary results.

Ethics and dissemination: As a systematic review of published data, no ethical approval is necessary. Results will be published in a relevant academic peer-reviewed journal.

Registration: The protocol has been prospectively registered in PROSPERO (CRD42021280978)

STRENGTHS AND LIMITATIONS OF THIS STUDY

- We will conduct an electronic search using a wide range of electronic databases, including a Latin American database, which will broaden the coverage of publication countries.
- The use of the Donabedian framework will allow an objective assessment of the CESS contribution to the quality of medical care.
- The review will be conducted by a bilingual and international research team, contributing different experiences and perspectives on CESS structures, processes, and outcomes in different contexts (Latin America and the UK).
- However, only English and Spanish language studies will be included with the consequent potential exclusion of relevant articles and associated bias.

INTRODUCTION

Clinical ethics support is the provision of support and advice to health professionals, patients and families on ethical issues arising from clinical practice or patient care.(1–3) Accordingly, Clinical Ethics Support Services (CESS) are institutionalised forms of ethics support within healthcare organisations.(2)

CESS were initially developed in the USA in 1970-80 in response to government and medical societies' recommendations,(4) and has since spread progressively worldwide, but in an uneven and very varied way.(1,5–7) Forms of, and access to, CESS varies across different centres, countries, and cultural contexts.(6,8) In some countries, the constitution of institutional CESS is recommended or required by the government and subject to official regulation, while in others, such as the UK, the organisation and function of these services lack official guidance.(1)

Traditionally, four main CESS functions have been described; clinical case consultation, education, institutional policy development, and research.(3,9) Multiple models of CESS have been described, including individual ethical case consultation, clinical ethics committees, individual ethicists, moral case deliberation, ethics rounds and ethics discussion forums.(1,10–12) Informal provision of clinical ethics support has also been reported.(10)

Unlike Research Ethics Committees (REC), CESS have been criticised for lack of standardisation, an absence of regulation of their structures, skill requirements, role and remit and the paucity of formal evaluation of their impact.(13)

Even though there has always been an awareness of the need for systematic evaluation of CESS outcomes and effectiveness,(14–16) and there is a growing body of theoretical and empirical literature addressing CESS' evaluation, there remain no agreed standards or quality indicators for these services.(17–19)

Considering that CESS are, by definition, engaged in complex interventions where multiple components and interactions impact the final outcome,(2) a clear understanding of how they function is vital, before attempting any evaluation.(2) Schildmann et al. defined the evaluation of CESS as "the systematic gathering of data with empirical research methods for the purpose of acquiring knowledge about the structure, functioning, quality and results of CESS".(p681, 20) Following this definition, in line with the widely used Donabedian model for evaluating the quality of medical care,(21) a comprehensive evaluation of the quality of CESS should include three dimensions of care: structure, process and outcomes.(2,21,22) As described by Donabedian, quality measurement standards derive from both empirical and normative sources.(21) Considering CESS have an explicit normative character, identifying appropriate quality criteria is particularly complicated, and this normative feature should be reflected when defining assessment measures.(23) Widely used outcome measures such as length of stay, mortality, or financial impact are not be necessarily helpful in evaluating CESS.(22) For example, any evaluation of ethics consultation services focused on pre-established outcome measures should not ignore the consultation's central aim of responding to the ethical queries presented by those requesting support with a particular clinical case. Therefore, an appropriate evaluation system must allow for the context and particularities of each case to be considered.(24) Additionally, CESS evaluation should involve all stakeholders, including both those who receive and those who provide ethics support (i.e., healthcare professionals, patients and relatives, CESS members, hospital management).(25)

Paediatric practice raises particular ethical challenges not frequently found in adult patient care.(26–29) The fundamental principle of respect for patient autonomy has a substantially different understanding in paediatric practice, with parents taking the responsibility of decision-making until children are afforded that possibility.(26) Generally, parents' decisions are in coherence with the child's views and the child's best interest, but conflict might arise when those involved (clinical team, parents, child) hold different views. Additionally, the child's capacity to understand the information provided and contribute to, or even make decisions about their care will depend on their age,

maturity, and the presence of chronic health conditions, physical disabilities and neurodevelopmental disorders. The United Nations Convention on the rights of the Child (UNCRC) designates a duty to actively involve children in decision-making on matters that concern them, including their health and care.(30) Thus, regardless of the condition, children must always be involved in the decision-making process with a careful assessment of the child's competence needs to be made, particularly for decisions with moral significance.(26)

The paediatric landscape has changed with technological advances, lower mortality rates in many specialities and an increasing number of patients with chronic and complex conditions. Uncertainties about prognostication and treatment outcomes, overall benefits and burdens pose ethical challenges about withholding and withdrawing life-sustaining treatments.(31) The involvement of multiple teams with different perspectives and values might add further complexity to the decision-making process.(29) Finally, these significant technological advances and decreasing mortality rates might strengthen the perception of death, particularly in children, as a medical failure adding barriers to end of life discussions and decision-making.

Despite ethically challenging situations and consequent divergent opinions being common in paediatrics, the number of paediatric ethics consultations is relatively low.(32) Many of these challenges might be opportunely identified and appropriately managed by the healthcare team and the family,(33) with clinicians receiving support through alternatives to formal ethics consultation.(32) However, in complex cases, there may either be an impasse or conflict might persist. In these situations, ethical consultation has been shown to help provide a resolution.(34)Recent controversial cases featured extensively in both print, and social media have increased international public and academic attention to the ethical challenges of paediatric practice. There has been an increased interest and debate surrounding the legal and ethical aspects of paediatric practice, such as parental and clinical teams' disagreements about the child's best interests, emerging child capacity, innovative treatments, technological dependence and end-of-life decision-making. Consultant paediatricians,

trainees and residents have expressed concerns that these public cases might negatively impact their relationship with patients and families.(35) Availability of clinical ethics support services for teams facing ethical challenges has been strongly advocated by professional bodies,(31) academics(33,36,37) and clinicians.(35,38) However, there are no standards or guidance on p-CESS structure, functions and aims. Nor is the, for example, agreement on the level of involvement of patients and parents in ethics discussions, being this a particularly controversial matter in paediatric setting.(39)

Interest in assessing CESS quality and effectiveness has grown in the past decades. However, most studies have focused on adult care settings, with relatively little attention paid to p-CESS.(40) Multiple systematic reviews evaluating different aspects of CESS have recently been published. Nevertheless, these focus on adult patients; ethical case intervention,(41) adult end of life context,(42) and adult ICU.(43) Other reviews that did not explicitly focused on adult patients evaluate a specific intervention (assessment tools for evaluating clinical ethics consultation(17)) or effectiveness of clinical ethics committees.(18) Generalisation of adult-focused reviews and evidence to paediatric context might not be appropriate. Although many CESSs will support both adult and paediatric patients, their families and clinical teams, it is likely that, together with the increasing number and complexities of children hospitals around the globe,(44) many CESS will serve patients and staff of paediatric healthcare institutions.(45) Moreover, there might be a value in accumulating knowledge and expertise in an increasingly complex paediatric field. Thus, a better understanding on current models of paediatric specific CESS will inform further development and research to contribute to the provision of optimal care for paediatric patients and their families. To our knowledge, no systematic review on Paediatric Clinical Ethics Support Services (p-CESS) structures, processes, evaluation measures and outcomes has been published. Such a review is necessary to inform current p-CESS practice and further development. Therefore, we aim to inform further research and debate on the current quality evaluation and minimum standards for p-CESS by offering a comprehensive description of current p-CESS models and assessments by responding to this review question:

"What is the range of structures, processes, and outcome measures of paediatric CESS reported in the literature?"

Aim:

To systematically identify, appraise and synthesise evidence for p-CESS structures, processes, and outcome measures described in the literature.

Objectives:

- To identify and synthesise published data on p-CESS regarding their structures, processes, evaluation measures and outcomes.
- To explore the impact of p-CESS given the outcomes identified in the review.
- To qualitatively appraise the available evidence.
- To develop a preliminary framework for the evaluation of p-CESS based on available evidence.
- To provide recommendations for further research on CESS effectiveness and outcome measures in paediatric practice.

METHODS AND ANALYSIS

We will conduct a mixed studies systematic review to identify and synthesise evidence for Paediatric CESS structures, interventions, evaluation measures and outcomes. By integrating studies with diverse research methods, a mixed studies systematic review allows the compensation for the limitations of qualitative and quantitative evidence and a better the understanding of the complexities of p-CESS.(46) The review will be reported following the adapted PRISMA for reporting systematic reviews of qualitative and quantitative evidence template,(47) as recommended by Pluye et al.(48)

The review protocol has been developed following the recommended items included in the PRISMA-P statement(49) and has been prospectively registered in PROSPERO registry CRD42021280978.(50)

Eligibility Criteria

The inclusion and exclusion criteria are summarised in Table 1. The review will include peer-review empirical studies (qualitative, quantitative or mixed-methods), including case studies published in English or Spanish language which provide empirical data on evaluation, assessment and/or impact (i.e., effect, evaluation, importance, meaning, value)(11) of any one or more of the following aspects of paediatric CESS: service structure, constitution and membership, service's aims and functions, interventions and processes, and outcome measures of p-CESS. We will include empirical studies of qualitative, quantitative or mixed-methods design reporting both objective and/or subjective measures.

We will include studies reporting on CESS that provide services to adult and paediatric patients only if paediatric data can be extracted separately.

Non-peer-reviewed studies, reviews, theoretical works, editorials, letters, opinion pieces, book chapters will be excluded. Conference abstracts will not be included, but authors will be contacted asking whether the relevant work has been published, with a two-week timeframe allowed for a response. There will be no timeframe or geographical restrictions.

Table 1. Eligibility Criteria

	INCLUSION CRITERIA	EXCLUSION CRITERIA
Type of participants	CESSs that serve paediatric only or paediatric and adult population, where paediatric data can be extracted separately.	CESSs that serve only adult patients. CESSs serving paediatric and adult populations, where paediatric data cannot be extracted and analysed

	The paediatric population will be defined in this review as between 0-18 years old. Study participants include, but are not limited to, referring clinicians, CESS members, patients/children, parents/relatives/careers and hospital administrators.	
Context/ setting	Articles reporting on established CESS serving paediatric practice in any setting (hospital, community) and country.	Research ethics committees
Issues	Empirical studies reporting data on evaluation, assessment and/or impact (i.e., effect, evaluation, importance, meaning, value) of any one or more of, but not limited to the following aspects of paediatric CESS: service structure, constitution and membership, service's aims and functions, interventions and processes, outcome measures of paediatric CESS.	Theoretical analysis or narrative reviews on paediatric CESS. Studies focusing only on a description of the paediatric CESS without reporting assessment/impact data

Methods	Empirical studies of any methods (qualitative and/or quantitative), including case studies.	Theoretical reviews or analysis. Systematic reviews Case reports, narrative reviews.
Timeframe	Any time frame. Searches will be conducted from the database inception date until the search date.	
Type of publication	Peer-reviewed publications in English or Spanish Language	Non-peer-reviewed studies, reviews, theoretical works, editorials, letters, opinion pieces Conference abstracts
CESS: Clinical Ethics Support Services		

Search strategy

Electronic searches. The following databases will be searched: MEDLINE, Philosopher's Index, EMBASE, PsychINFO, LILACS, Web of Science and CINHAL. There will be no methodological, language, geographical or time filters applied to the search strategy. If a non-English paper is considered eligible for inclusion, relevant data and results will be translated to English before analysis.

The initial search strategy was developed considering previously published systematic reviews in paediatrics, clinical ethics and service evaluation. Search terms will be related to "Clinical ethics support", "paediatrics" AND "structure/process/outcome indicators" and adapted to each database requirement. Publications that would match the criteria for inclusion in the review previously known to the research team were successfully retrieved applying the search strategy in Medline online

database. Please see Table 2 for Medline search strategy and refer to Supplementary file 1 for complete search strategy for all included databases). The search strategy for all other databases is provided in Supplementary File 1.

Table 2. Medline search strategy

1. paediatric.mp. 2. paediatr*.mp. 3. pediatric.mp. or Pediatrics/ 4. pediatr*.mp. 5. child*.mp. or Child/ 6. 6Adolescent/ or adolescent.mp. 7. adolesc*.mp. 8. infant*.mp. 9. infant.mp. or Infant/ 10. kid.mp. 11. kids.mp. 12. baby.mp. 13. babies.mp. 14. toddler*.mp. 15. childhood.mp. 16. juvenil*.mp. 17. youth*.mp. 18. minor.mp. or Minors/ 19. Infant, Newborn/ or infancy.mp. or Child, Preschool/ 20. Infant, Newborn/ or newborn*.mp. 21. Premature Birth/ or Infant, Premature/ or preterm*.mp. 22. prematur*.mp. 23. Puberty/ or pubert*.mp. 24. pubescen*.mp. 25. young person.mp. 26. neonatal.mp.	27. Ethicists/ or ethicist*.mp. 28. bioethicist*.mp. 29. medical ethics.mp. or Ethics, Medical/ 30. clinical ethics.mp. or Ethics, Clinical/ 31. clinical ethics committee.mp. or Ethics Committees, Clinical/ 32. bioethics.mp. or Bioethics/ 33. bioethical issues.mp. or Bioethical Issues/ 34. ethical issues.mp. or Ethics/ 35. ethical challenges.mp 36. moral review.mp. 37. Ethical Analysis/ or Ethics Consultation/ or moral case deliberation.mp. 38. moral case consult*.mp. 39. moral consult*.mp. 40. ethic* case review.mp. or Ethics Committees/ 41. ethic* deliberation.mp. 42. ethic* intervention.mp. 43. ethic* round.mp. 44. ethic* support.mp. 45. ethic* service.mp. 46. Ethical Analysis/ or ethic* analysis.mp. 47. ethic* referral.mp. 48. Ethics Committees/ or ethic* committee.mp. 49. bioethic* deliberation.mp. 50. bioethic* intervention.mp. 51. bioethic* round.mp. 52. bioethic* service.mp. 53. bioethic* support.mp. 54. bioethic* analysis.mp. 55. bioethic* referral.mp. 56. bioethic* committee.mp.	57. structure.mp. 58. model*.mp. 59. process*.mp. 60. intervention*.mp. 61. Outcome Assessment, Health Care/ or "Outcome and Process Assessment, Health Care"/ or assessment*.mp. 62. evaluation*.mp. 63. impact*.mp. 64. effectiveness.mp. 65. Medical Audit/ or Clinical Audit/ or audit.mp. 66. Outcome Assessment, Health Care/ or "Outcome and Process Assessment, Health Care"/ or outcome.mp. 67. "Quality of Health Care"/ or quality.mp. or Quality Indicators, Health Care/ 68. indicator*.mp.

69. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23 or 24 or 25 or 26
70. 28 or 29 or 30 or 31 or 32 or 33 or 34 or 35 or 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49 or 50 or 51 or 52 or 53 or 54 or 55 or 56 or 57
71. 59 or 60 or 61 or 62 or 63 or 64 or 65 or 66 or 67 or 68 or 69 or 70
72. 69 AND 70 AND 71

Other resources. Reference and citation list of included studies will be hand-searched. When relevant, we will contact the authors of conference abstracts identified through the search for peer-review publications. We will allow a time frame of two weeks for a response before considering the publication unavailable.

Selection process.

All retrieved records will be managed using Refworks® reference manager software. After deduplication, a random sample of 10% will be independently screened by MD and BP to test the reliability of the criteria. Any disagreements will be discussed within the research team until agreement, and, if required, eligibility criteria will be adjusted and/or clarified to improve the consistency of the screening process. Thereafter, all titles and/or abstracts will be screened by MD to identify publications that meet the previously established inclusion and exclusion criteria... References selected for assessment in the full text will be independently dual-assessed by MD and BP against inclusion/exclusion criteria, and any disagreements will be discussed within the research team until agreement. Reasons for excluding articles after full-text assessment will be recorded and study references and reasons for exclusion will be provided as a supplementary file. A Cohen's Kappa score over 90% will be required at both, the title/abstract and full text screening processes. The screening process will be presented as a PRISMA flowchart.

Data extraction.

Data from individual studies considered relevant for the review question will be extracted to a pre-piloted Excel data extraction form by MD and checked by BP. Disagreements will be discussed within the research team. Data entries will include: Publication details (First author, year of publication, title), setting (country, healthcare setting), study aims, study design, sampling method and sample description. Primary outcomes sought in the data set will include; (i) assessed aspects of paediatric CESS as reported by study authors, including service structures, processes and outcomes (i.e., membership, service's activities, referrers, cases, contexts and reasons for referrals); (ii) assessed dimensions (i.e., effectiveness, safety and responsiveness) and/or subdimensions of quality of care(51) as reported by study authors; and (iii) methods and instruments used in the assessment. For qualitative studies, all data within the results/finding section will be considered as results. Characteristics of included studies will be tabulated and presented in a Table.

Quality assessment.

Following our initial literature review, we expect to find around 10-30 potentially eligible studies with multiple research designs, including qualitative, quantitative and mixed methods. To allow coherent and systematic critical appraisal of included studies with different designs, we will use the 2018 Mixed Methods Appraisal Tool.(52) The tool was developed specifically for the appraisal of complex systematic reviews that include empirical qualitative, quantitative and mixed methods studies. It includes specific criteria depending on the study design category. Each criterion is rated as "yes", "no", "can't tell" response. As recommended by the authors, for each study, we will present a detailed description of the rating of each criterion and calculate an overall quality score based on the number of quality criteria met. There is no recommended cut off score to exclude studies based on quality appraisal, and therefore no study will be excluded based on that criterion. Instead, we will conduct a sensitivity analysis during the data synthesis process to assess the impact of low-quality studies in the

review findings and adjust recommendations accordingly.(52) Quality appraisal will be conducted by MD and checked by BP.

Data synthesis

This systematic review is not focused only on the effectiveness of a particular intervention but addresses a broader range of questions. Data synthesis will follow a sequential explanatory synthesis following the two-step process described by Pluye and Hong:(46) First, a quantitative synthesis including results from quantitative studies and quantitative data from mixed-methods studies, followed by qualitative synthesis of results of qualitative studies and qualitative data form mixed-methods studies. The qualitative synthesis will be informed by the previously conducted quantitative synthesis. The integration of the qualitative synthesis into the quantitative one will allow a better understanding of the quantitative results,(46) also highlighting convergences and divergences between quantitative and qualitative synthesis to inform future research-

As we expect to find great diversity of outcome measures, quantitative data will be synthetised and tabulated using descriptive statistics where appropriate(53) guided by but not limited to Donabedian's framework of structure, process and outcomes. For qualitative data, we will conduct thematic synthesis approach,(54) using NVivo software for qualitative data analysis. The thematic synthesis will include: (i) Free line-by-line coding of the primary study's findings, (ii) organisation of these codes into related themes informed by the quantitative synthesis.(54) If the qualitative synthesis process develops additional themes that are not described in the quantitative synthesis, these will be included in the integrated synthesis as qualitative results only. To assess the robustness of the synthesis we will considering individual studies' quality and conduct sensitivity analysis if possible.

The synthesis will be conducted by MD and checked by BP. The final synthesis will be discussed within the research team.

Timeline

The protocol for this review is published in PROSPERO (date 27 September 2021).(50) Searches on the databases mentioned in the protocol were conducted in August 2021. Retrieved references were screened at title and abstract level during September-October 2021. Screening at full text level is planned for December 2021 – January 2022. We plan to proceed with further stages of the review, including stakeholder involvement, and data extraction and synthesis after the protocol is accepted for publication following the peer-review process. Data extraction and analysis are expected to take 6 months after study selection.

Patient and public Involvement.

Patients and the public were not involved in the design of this systematic review protocol, but will be involved further at two stages in the process of the research, to ensure the review outcomes are useful and relevant.(55,56) Following Cochrane good practice guidance for people involvement in systematic reviews,(55) we sought to involve views of diverse stakeholders. Since p-CESS are established within healthcare institutions, and provide support to clinicians and patients and their families in making ethically challenging decision, we defined four stakeholders' categories whose collaboration would be valuable to the research process; (i) institutional managers, (ii) p-CESS board members, (iii) clinicians and (iv) parents. We will establish two advisory groups, one Chilean and one UK-based, with one representative for each stakeholder category. We will invite representatives that are already known to the research team to join the advisory group and participate in two one-hour workshops, one before data extraction and synthesis and a second one after preliminary results. Parents' representatives will be or have been previously involved as parent representative in a p-CESS. In the first webinar, participants will be asked to share their views, thoughts, opinions or experiences to

ensure we will be looking for the appropriate data in the included studies. At the second webinar, participants will have the opportunity to provide their feedback on the preliminary findings, to add context and meaning to the findings, contributing to the overall interpretation and recommendations. Stakeholder involvement will be reported following the GRIPP (Guidance for Reporting Involvement of Patients and Public) checklist.(44)

Ethics and dissemination.

As a systematic review of published data, no ethical approval is necessary. Following Cochrane guidance,(55) ethical approval for stakeholder involvement in this review would not be required as workshops would not be audio-recorded and no vulnerable groups will participate.

We will present and discuss our findings in an open-access webinar, inviting a broad range of stakeholders to attend, including hospital managers, clinicians, academic ethicists, and patient representatives. A final report will be published in a relevant academic peer-reviewed journal.

We plan to continue this research by conducting a modified Delphi study based on our review results to further explore the most appropriate quality indicators for evaluating p-CESS.

DISCUSSION

Interest in assessing CESS quality and effectiveness has grown in the past decades, with little attention paid to p-CESS. The results of this work will provide us with the first systematic review of evidence on Paediatric Clinical Ethics Support Services.

The review team is comprised by a bilingual and international research team that includes a Paediatric Intensivist Consultant with vast experience in paediatric medical ethics leading the teaching, research and clinical activities of the p-CESS at a large tertiary children's hospital; a speech therapist, certified clinical ethics consultant and PhD student in CESSs evaluation and a medical doctor and ethicist with experience in systematic reviews and ethics research. The researchers' diverse backgrounds will contribute with their experiences and perspectives on CESS structures, processes, and outcomes in

different contexts (Latin America and the UK). This will also allow a more comprehensive review both, by searching a Latin American specific database and the inclusion papers published in English and Spanish languages. This will enhance the review comprehensiveness, as long as potential bias is given due consideration in the result interpretation and recommendation development stages. Evidence on the effect of English-restricted criteria in traditional systematic reviews of randomised controlled trials with meta-analyses has not shown to result in significant bias.(57,58). However, this review on p-CESS structures, processes and outcomes will include a broader range of study designs and therefore potential bias associated with the exclusion of studies published in languages other than English and Spanish will be considered in the interpretation of results and recommendations.

The inclusion of peer-reviewed publications only might result in the omission of relevant publications (i.e., CESS terms of references and/or reports published in institutional websites). However, focusing on peer-reviewed publications will ensure validity of data included in the synthesis and also warrant a balance between the amount of data and the capacity of the research team, without compromising the review results. Moreover, we aim at mitigating the potential exclusion of relevant data by including case reports and case studies. The use of the Donabedian model will allow a structured and objective assessment of p-CESS contribution to patients' care. This is a well-accepted and widely used framework. However, considering the normative nature of CESS and their interventions and outcomes, the framework will be used as a guide and adaptation is expected.

We hope that our review results will allow for a better understanding of p-CESS structures, processes, and outcomes, contributing to further research exploring the normative and empirical basis of p-CESS.

Author contributions: MD and JB conceived the review. MD, JB and BP developed the protocol. All authors revised and edited the manuscript and approved the final version.

Funding: This work was supported by the Institute of Medical Ethics "Research in Medical Ethics" grant (No grant number). The funder has had no involvement in the review protocol development.

Competing interests: None to declare.

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Supplementary file 1. Complete search strategy for all databases

Embase Classic+Embase

1 paediatric.mp.

2 paediatr*.mp.

3 pediatric.mp. or Pediatrics/

4 pediatr*.mp.

5 child*.mp. or Child/

6 Adolescent/ or adolescent.mp.

7 adolesc*.mp.

8 infant*.mp.

9 infant.mp. or Infant/

10 kid.mp.

11 kids.mp.

12 baby.mp.

13 babies.mp.

14 toddler*.mp.

15 childhood.mp.

16 juvenil*.mp.

17 youth*.mp.

18 minor.mp. or Minors/

19 Infant, Newborn/ or infancy.mp. or Child, Preschool/

20 Infant, Newborn/ or newborn*.mp.

21 Premature Birth/ or Infant, Premature/ or preterm*.mp.

22 prematur*.mp.

23 Puberty/ or pubert*.mp.

24 pubescen*.mp.

25 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23 or 24

26 Ethicists/ or ethicist*.mp.

27 bioethicist*.mp.

28 medical ethics.mp. or Ethics, Medical/

29 clinical ethics.mp. or Ethics, Clinical/

30 clinical ethics committee.mp. or Ethics Committees, Clinical/

31 bioethics.mp. or Bioethics/

32 bioethical issues.mp. or Bioethical Issues/

33 ethical issues.mp. or Ethics/

34 ethical challenges.mp.

35 moral review.mp.

36 Ethical Analysis/ or Ethics Consultation/ or moral case deliberation.mp.

37 moral case consult*.mp.

38 moral consult*.mp.

39 ethic* case review.mp. or Ethics Committees/

40 ethic* deliberation.mp.

41 ethic* intervention.mp.

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90 juvenil*.mp.

91 youth*.mp.

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144 effectiveness.mp.

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148	indicator*.mp.
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4	pediatr*.mp.
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7	adolesc*.mp.
8	infant*.mp.
9	infant.mp. or Infant/
10	kid.mp.
11	kids.mp.
12	baby.mp.
13	babies.mp.
14	toddler*.mp.
15	childhood.mp.
16	juvenil*.mp.
17	youth*.mp.
18	minor.mp. or Minors/
19	Infant, Newborn/ or infancy.mp. or Child, Preschool/
20	Infant, Newborn/ or newborn*.mp.
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29 clinical ethics.mp. or Ethics, Clinical/

30 clinical ethics committee.mp. or Ethics Committees, Clinical/

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Philosopher's Index

Philosophy - journal articles, books, book chapters and book reviews

Subject Area(s): Social Sciences , History , The Arts

(paediatr* OR pediatr* OR child* OR adolesc* OR infant* OR kid OR kids OR baby OR babies OR toddler* OR childhood OR juvenil* OR youth* OR minor* OR infancy OR newborn* OR prematurity OR preterm OR pubert* OR pubescen* OR "young person" OR neonatal) AND (ethicist* OR bioethicist* OR bioethic* OR "clinical ethics" OR "clinical ethics committee" OR "bioethical issue*" OR "ethical issue*" OR "ethical challenge*" OR "moral review" OR "moral case deliberation" OR "moral case consult*" OR "moral consult*" OR "ethic* case review" OR "ethic* deliberation" OR

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APA PsycInfo®

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VHL Lilacs

(paediatr* OR pediatr* OR child* OR adolesc* OR infant* OR kid OR kids OR baby OR babies OR toddler* OR childhood OR juvenil* OR youth* OR minor* OR infancy OR newborn* OR prematurity OR preterm OR pubert* OR pubescen* OR young person OR neonatal) AND (ethicist* OR bioethicist* OR bioethic* OR clinical ethics OR clinical ethics committee OR bioethical issue* OR ethical issue* OR ethical challenge* OR moral review OR moral case deliberation OR moral case consult* OR moral consult* OR ethic* case review OR ethic* deliberation OR ethic* round OR ethic* intervention OR ethic* support OR ethic* service OR ethic* analysis OR ethic* referral OR ethic* committee OR bioethic* deliberation OR bioethic* intervention OR bioethic* round OR bioethic* service OR bioethic* support OR bioethic* analysis OR bioethic* referral OR bioethic* committee) AND (structure OR model* OR process* OR intervention OR assessment* OR evaluation* OR impact* OR effectiveness OR audit OR outcome* OR quality OR indicator*) AND (db:("LILACS"))

Web of Science

(paediatr* OR pediatr* OR child* OR adolesc* OR infant* OR kid OR kids OR baby OR babies OR toddler* OR childhood OR juvenil* OR youth* OR minor* OR infancy OR newborn* OR prematurity OR preterm OR pubert* OR pubescen* OR "young person" OR neonatal) AND (ethicist* OR bioethicist* OR bioethic* OR "clinical ethics" OR "clinical ethics committee" OR "bioethical issue*" OR "ethical issue*" OR "ethical challenge*" OR "moral review" OR "moral case deliberation" OR "moral case consult*" OR "moral consult*" OR "ethic* case review" OR "ethic* deliberation" OR "ethic* round" OR "ethic* intervention" OR "ethic* support" OR "ethic* service" OR "ethic* analysis" OR "ethic* referral" OR "ethic* committee" OR "bioethic* deliberation" OR "bioethic* intervention" OR "bioethic* round" OR "bioethic* service" OR "bioethic* support" OR "bioethic* analysis" OR "bioethic* referral" OR "bioethic* committee") AND (structure OR model* OR process* OR intervention OR assessment* OR evaluation* OR impact* OR effectiveness OR audit OR outcome* OR quality OR indicator*)

CINAHL (EBSCOhost Research Databases)

S10 S7 AND S8 AND S9

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

S9 S4 OR S5 OR S6

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

S8 S2 OR S3

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

S7 strucure OR model* OR process* OR intervention OR assessment* OR evaluation* OR impact* OR effectiveness OR audit OR outcome* OR quality OR indicator*

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

S6 ethic* referral OR ethic* committee OR bioethic* deliberation OR bioethic* intervention OR bioethic* round OR bioethic* service OR bioethic* support OR bioethic* analysis OR bioethic* referral OR bioethic* committee

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

S5 ethical challenge* OR moral review OR moral case deliberation OR moral case consult* OR moral consult* OR ethic* case review OR ethic* deliberation OR ethic* intervention OR ethic* round OR ethic* support OR ethic* service OR ethic* analysis

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

S4 ethicist* OR bioethicist* OR medical ethics OR clinical ethics OR clinical ethics committee OR bioethi* OR bioethical issues OR ethical issues

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

S3 youth* OR infancy OR newborn* OR prematur* OR pubert* OR pubescen* OR young person OR neonatal

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

S2 paediatr* OR pediatr* OR child* OR adolesc* OR infant* OR kid OR kids OR baby OR babies OR toddler* OR juvenil* OR minor*

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

S1 paediatric

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

PRISMA-P (Preferred Reporting Items for Systematic review and Meta-Analysis Protocols) 2015 checklist: recommended items to address in a systematic review protocol*

Section and topic	Item No	Checklist item	Page in text
ADMINISTRATIVE INFORMATION			
Title:			
Identification	1a	Identify the report as a protocol of a systematic review	1
Update	1b	If the protocol is for an update of a previous systematic review, identify as such	n/a
Registration	2	If registered, provide the name of the registry (such as PROSPERO) and registration number	2, 9
Authors:			
Contact	3a	Provide name, institutional affiliation, e-mail address of all protocol authors; provide physical mailing address of corresponding author	1
Contributions	3b	Describe contributions of protocol authors and identify the guarantor of the review	21
Amendments	4	If the protocol represents an amendment of a previously completed or published protocol, identify as such and list changes; otherwise, state plan for documenting important protocol amendments	n/a
Support:			
Sources	5a	Indicate sources of financial or other support for the review	21
Sponsor	5b	Provide name for the review funder and/or sponsor	21
Role of sponsor or funder	5c	Describe roles of funder(s), sponsor(s), and/or institution(s), if any, in developing the protocol	21
INTRODUCTION			
Rationale	6	Describe the rationale for the review in the context of what is already known	4-8
Objectives	7	Provide an explicit statement of the question(s) the review will address with reference to participants, interventions, comparators, and outcomes (PICO)	8
METHODS			
Eligibility criteria	8	Specify the study characteristics (such as PICO, study design, setting, time frame) and report characteristics (such as years considered, language, publication status) to be used as criteria for eligibility for the review	9-10 Table 1
Information sources	9	Describe all intended information sources (such as electronic databases, contact with study authors, trial registers or other grey literature sources) with planned dates of coverage	12-14
Search strategy	10	Present draft of search strategy to be used for at least one electronic database, including planned limits, such that it could be repeated	12 Table 2

			Supplementary file 1
Study records:			
Data management	11a	Describe the mechanism(s) that will be used to manage records and data throughout the review	14-15
Selection process	11b	State the process that will be used for selecting studies (such as two independent reviewers) through each phase of the review (that is, screening, eligibility and inclusion in meta-analysis)	14
Data collection process	11c	Describe planned method of extracting data from reports (such as piloting forms, done independently, in duplicate), any processes for obtaining and confirming data from investigators	14-15
Data items	12	List and define all variables for which data will be sought (such as PICO items, funding sources), any pre-planned data assumptions and simplifications	14-15
Outcomes and prioritization	13	List and define all outcomes for which data will be sought, including prioritization of main and additional outcomes, with rationale	15
Risk of bias in individual studies	14	Describe anticipated methods for assessing risk of bias of individual studies, including whether this will be done at the outcome or study level, or both; state how this information will be used in data synthesis	15-16
Data synthesis	15a	Describe criteria under which study data will be quantitatively synthesised	16-17
	15b	If data are appropriate for quantitative synthesis, describe planned summary measures, methods of handling data and methods of combining data from studies, including any planned exploration of consistency (such as I^2, Kendall's τ)	16-17
	15c	Describe any proposed additional analyses (such as sensitivity or subgroup analyses, meta-regression)	16-17
	15d	If quantitative synthesis is not appropriate, describe the type of summary planned	16-17
Meta-bias(es)	16	Specify any planned assessment of meta-bias(es) (such as publication bias across studies, selective reporting within studies)	n/a
Confidence in cumulative evidence	17	Describe how the strength of the body of evidence will be assessed (such as GRADE)	n/a

*** It is strongly recommended that this checklist be read in conjunction with the PRISMA-P Explanation and Elaboration (cite when available) for important clarification on the items. Amendments to a review protocol should be tracked and dated. The copyright for PRISMA-P (including checklist) is held by the PRISMA-P Group and is distributed under a Creative Commons Attribution Licence 4.0.**

From: Shamseer L, Moher D, Clarke M, Ghersi D, Liberati A, Petticrew M, Shekelle P, Stewart L, PRISMA-P Group. Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015: elaboration and explanation. BMJ. 2015 Jan 2;349(jan02 1):g7647.

BMJ Open

Clinical Ethics Support Services in paediatric practice: protocol for a mixed studies systematic review on structures, interventions and outcomes

Journal:	*BMJ Open*
Manuscript ID	bmjopen-2021-057867.R2
Article Type:	Protocol
Date Submitted by the Author:	19-Jan-2022
Complete List of Authors:	Dittborn, Mariana; Great Ormond Street Hospital for Children, Paediatric Bioethics Centre ; Universidad del Desarrollo Facultad de Medicina, Centro de Bioética Portales, Bernardita; Universidad del Desarrollo, Centro de Bioética Brierley, Joe; Great Ormond Street Hospital for Children, Paediatric Bioethics Centre
Primary Subject Heading:	Ethics
Secondary Subject Heading:	Paediatrics
Keywords:	PAEDIATRICS, MEDICAL ETHICS, Quality in health care < HEALTH SERVICES ADMINISTRATION & MANAGEMENT

1

TITLE

Clinical Ethics Support Services in paediatric practice: protocol for a mixed studies systematic review on structures, interventions and outcomes

AUTHORS

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WORD COUNT 3534

KEY WORDS

Paediatrics, Clinical Ethics Committees, Clinical Ethics, Quality Indicators, Systematic Review

ABSTRACT

Introduction: Clinical Ethics Support Services (CESS) have been developing worldwide with growing interest in evaluating their quality. Paediatric-specific CESSs (p-CESS) have received little attention, and evidence from adult services might not be generalisable. Evidence on service models and practices is crucial to inform further research and debate on quality evaluation and minimum standards for p-CESSs. We aim to systematically identify, appraise and synthesise evidence for p-CESS structures, processes, and outcomes.

Methods and analysis: We will conduct a mixed-studies systematic review including peer-reviewed empirical studies published in English or Spanish language providing data on the evaluation and/or impact on any aspect of p-CESS. We will search seven electronic databases: MEDLINE, Philosopher's Index, EMBASE, PsychINFO, LILACS, Web of Science and CINHAL, without filters applied. Search terms will be related to "clinical ethics support" AND "paediatrics" AND "structure/process/outcome". Reference and citation list of included studies will be hand-searched. A 10% random sample of retrieved titles/abstracts and all full texts will be independently dual-screened. We will conduct narrative and thematic synthesis for quantitative and qualitative data, respectively, following sequential explanatory synthesis guided by Donabedian's framework of structure, process and outcomes. Quality will be assessed using the Mixed-Methods Appraisal Tool (2018). The review will be reported using the adapted Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) for reporting systematic reviews of qualitative and quantitative evidence template. Stakeholders will be involved twice in the review process; prior to data extraction and synthesis and after preliminary results.

Ethics and dissemination: As a systematic review of published data, no ethical approval is necessary. Results will be published in a relevant academic peer-reviewed journal.

Registration: The protocol has been prospectively registered in PROSPERO (CRD42021280978)

STRENGTHS AND LIMITATIONS OF THIS STUDY

- We will conduct an electronic search using a wide range of electronic databases, including a Latin American database, which will broaden the coverage of publication countries.
- The use of the Donabedian framework will allow an objective assessment of the CESS contribution to the quality of medical care.
- The review will be conducted by a bilingual and international research team, contributing different experiences and perspectives on CESS structures, processes, and outcomes in different contexts (Latin America and the UK).
- However, only English and Spanish language studies will be included with the consequent potential exclusion of relevant articles and associated bias.

INTRODUCTION

Clinical ethics support is the provision of support and advice to health professionals, patients and families on ethical issues arising from clinical practice or patient care.(1–3) Accordingly, Clinical Ethics Support Services (CESS) are institutionalised forms of ethics support within healthcare organisations.(2)

CESS were initially developed in the USA in 1970-80 in response to government and medical societies' recommendations,(4) and has since spread progressively worldwide, but in an uneven and very varied way.(1,5–7) Forms of, and access to, CESS varies across different centres, countries, and cultural contexts.(6,8) In some countries, the constitution of institutional CESS is recommended or required by the government and subject to official regulation, while in others, such as the UK, the organisation and function of these services lack official guidance.(1)

Traditionally, four main CESS functions have been described; clinical case consultation, education, institutional policy development, and research.(3,9) Multiple models of CESS have been described, including individual ethical case consultation, clinical ethics committees, individual ethicists, moral case deliberation, ethics rounds and ethics discussion forums.(1,10–12) Informal provision of clinical ethics support has also been reported.(10)

Unlike Research Ethics Committees (REC), CESS have been criticised for lack of standardisation, an absence of regulation of their structures, skill requirements, role and remit and the paucity of formal evaluation of their impact.(13)

Even though there has always been an awareness of the need for systematic evaluation of CESS outcomes and effectiveness,(14–16) and there is a growing body of theoretical and empirical literature addressing CESS' evaluation, there remain no agreed standards or quality indicators for these services.(17–19)

Considering that CESS are, by definition, engaged in complex interventions where multiple components and interactions impact the final outcome,(2) a clear understanding of how they function is vital, before attempting any evaluation.(2) Schildmann et al. defined the evaluation of CESS as "the systematic gathering of data with empirical research methods for the purpose of acquiring knowledge about the structure, functioning, quality and results of CESS".(p681, 20) Following this definition, in line with the widely used Donabedian model for evaluating the quality of medical care,(21) a comprehensive evaluation of the quality of CESS should include three dimensions of care: structure, process and outcomes.(2,21,22) As described by Donabedian, quality measurement standards derive from both empirical and normative sources.(21) Considering CESS have an explicit normative character, identifying appropriate quality criteria is particularly complicated, and this normative feature should be reflected when defining assessment measures.(23) Widely used outcome measures such as length of stay, mortality, or financial impact are not be necessarily helpful in evaluating CESS.(22) For example, any evaluation of ethics consultation services focused on pre-established outcome measures should not ignore the consultation's central aim of responding to the ethical queries presented by those requesting support with a particular clinical case. Therefore, an appropriate evaluation system must allow for the context and particularities of each case to be considered.(24) Additionally, CESS evaluation should involve all stakeholders, including both those who receive and those who provide ethics support (i.e., healthcare professionals, patients and relatives, CESS members, hospital management).(25)

Paediatric practice raises particular ethical challenges not frequently found in adult patient care.(26–29) The fundamental principle of respect for patient autonomy has a substantially different understanding in paediatric practice, with parents taking the responsibility of decision-making until children are afforded that possibility.(26) Generally, parents' decisions are in coherence with the child's views and the child's best interest, but conflict might arise when those involved (clinical team, parents, child) hold different views. Additionally, the child's capacity to understand the information provided and contribute to, or even make decisions about their care will depend on their age,

maturity, and the presence of chronic health conditions, physical disabilities and neurodevelopmental disorders. The United Nations Convention on the rights of the Child (UNCRC) designates a duty to actively involve children in decision-making on matters that concern them, including their health and care.(30) Thus, regardless of their health condition, children must always be involved in the decision-making process with a careful assessment of the child's competence needs to be made, particularly for decisions with moral significance.(26)

The paediatric landscape has changed with technological advances, lower mortality rates in many specialities and an increasing number of patients with chronic and complex conditions. Uncertainties about prognostication and treatment outcomes, overall benefits and burdens pose ethical challenges about withholding and withdrawing life-sustaining treatments.(31) The involvement of multiple teams with different perspectives and values might add further complexity to the decision-making process.(29) Finally, these significant technological advances and decreasing mortality rates might strengthen the perception of death, particularly in children, as a medical failure adding barriers to end of life discussions and decision-making.

Despite ethically challenging situations and consequent divergent opinions being common in paediatrics, the number of paediatric ethics consultations is relatively low.(32) Many of these challenges might be opportunely identified and appropriately managed by the healthcare team and the family,(33) with clinicians receiving support through alternatives to formal ethics consultation.(32) However, in complex cases, there may either be an impasse or conflict might persist. In these situations, ethical consultation has been shown to help provide a resolution.(34)Recent controversial cases featured extensively in both print, and social media have increased international public and academic attention to the ethical challenges of paediatric practice. There has been an increased interest and debate surrounding the legal and ethical aspects of paediatric practice, such as parental and clinical teams' disagreements about the child's best interests, emerging child capacity, innovative treatments, technological dependence and end-of-life decision-making. Consultant paediatricians,

trainees and residents have expressed concerns that these public cases might negatively impact their relationship with patients and families.(35) Availability of clinical ethics support services for teams facing ethical challenges has been strongly advocated by professional bodies,(31) academics(33,36,37) and clinicians.(35,38) However, there are no standards or guidance on p-CESS structure, functions or aims. Nor is there agreement about the level of involvement of patients, or in paediatrics parents and children, in ethics discussions.(39,40)

Interest in assessing CESS quality and effectiveness has grown in the past decades. However, most studies have focused on adult care settings, with relatively little attention paid to p-CESS.(41) Multiple systematic reviews evaluating different aspects of CESS have recently been published. Nevertheless, these focus on adult patients; ethical case intervention,(42) adult end of life context,(43) and adult ICU.(44) Other reviews that did not explicitly focused on adult patients evaluate a specific intervention (assessment tools for evaluating clinical ethics consultation(17)) or effectiveness of clinical ethics committees.(18) Generalisation of adult-focused reviews and evidence to paediatric context might not be appropriate. Although many CESSs will support both adult and paediatric patients, their families and clinical teams, it is likely that, together with the increasing in number and complexity of children's hospitals around the globe,(45) many CESS will serve patients and staff of paediatric healthcare institutions.(40) Moreover, there might be a value in accumulating knowledge and expertise in an increasingly complex paediatric field. Thus, a better understanding on current models of paediatric specific CESS will inform further development and research to contribute to the provision of optimal care for paediatric patients and their families. To our knowledge, no systematic review on Paediatric Clinical Ethics Support Services (p-CESS) structures, processes, evaluation measures and outcomes has been published. Such a review is necessary to inform current p-CESS practice and further development. Therefore, we aim to inform further research and debate on the current quality evaluation and minimum standards for p-CESS by offering a comprehensive description of current p-CESS models and assessments by responding to this review question:

"What is the range of structures, processes, and outcome measures of paediatric CESS reported in the literature?"

Aim:

To systematically identify, appraise and synthesise evidence for p-CESS structures, processes, and outcome measures described in the literature.

Objectives:

- To identify and synthesise published data on p-CESS regarding their structures, processes, evaluation measures and outcomes.
- To explore the impact of p-CESS given the outcomes identified in the review.
- To qualitatively appraise the available evidence.
- To develop a preliminary framework for the evaluation of p-CESS based on available evidence.
- To provide recommendations for further research on CESS effectiveness and outcome measures in paediatric practice.

METHODS AND ANALYSIS

We will conduct a mixed studies systematic review to identify and synthesise evidence for Paediatric CESS structures, interventions, evaluation measures and outcomes. By integrating studies with diverse research methods, a mixed studies systematic review allows the compensation for the limitations of qualitative and quantitative evidence and a better the understanding of the complexities of p-CESS.(46) The review will be reported following the adapted PRISMA for reporting systematic reviews of qualitative and quantitative evidence template,(47) as recommended by Pluye et al.(48)

The review protocol has been developed following the recommended items included in the PRISMA-P statement(49) and has been prospectively registered in PROSPERO registry CRD42021280978.(50)

Eligibility Criteria

The inclusion and exclusion criteria are summarised in Table 1. The review will include peer-review empirical studies (qualitative, quantitative or mixed-methods), including case studies published in English or Spanish language which provide empirical data on evaluation, assessment and/or impact (i.e., effect, evaluation, importance, meaning, value)(11) of any one or more of the following aspects of paediatric CESS: service structure, constitution and membership, service's aims and functions, interventions and processes, and outcome measures of p-CESS. We will include empirical studies of qualitative, quantitative or mixed-methods design reporting both objective and/or subjective measures.

We will include studies reporting on CESS that provide services to adult and paediatric patients only if paediatric data can be extracted separately.

Non-peer-reviewed studies, reviews, theoretical works, editorials, letters, opinion pieces, book chapters will be excluded. Conference abstracts will not be included, but authors will be contacted asking whether the relevant work has been published, with a two-week timeframe allowed for a response. There will be no timeframe or geographical restrictions.

Table 1. Eligibility Criteria

	INCLUSION CRITERIA	EXCLUSION CRITERIA
Type of participants	CESSs that serve paediatric only or paediatric and adult population, where paediatric data can be extracted separately.	CESSs that serve only adult patients. CESSs serving paediatric and adult populations, where paediatric data cannot be extracted and analysed

	The paediatric population will be defined in this review as between 0-18 years old. Study participants include, but are not limited to, referring clinicians, CESS members, patients/children, parents/relatives/careers and hospital administrators.	
Context/ setting	Articles reporting on established CESS serving paediatric practice in any setting (hospital, community) and country.	Research ethics committees
Issues	Empirical studies reporting data on evaluation, assessment and/or impact (i.e., effect, evaluation, importance, meaning, value) of any one or more of, but not limited to the following aspects of paediatric CESS: service structure, constitution and membership, service's aims and functions, interventions and processes, outcome measures of paediatric CESS.	Theoretical analysis or narrative reviews on paediatric CESS. Studies focusing only on a description of the paediatric CESS without reporting assessment/impact data

Methods	Empirical studies of any methods (qualitative and/or quantitative), including case studies.	Theoretical reviews or analysis. Systematic reviews Case reports, narrative reviews.
Timeframe	Any time frame. Searches will be conducted from the database inception date until the search date.	
Type of publication	Peer-reviewed publications in English or Spanish Language	Non-peer-reviewed studies, reviews, theoretical works, editorials, letters, opinion pieces Conference abstracts
CESS: Clinical Ethics Support Services		

Search strategy

Electronic searches. The following databases will be searched: MEDLINE, Philosopher's Index, EMBASE, PsychINFO, LILACS, Web of Science and CINHAL. There will be no methodological, language, geographical or time filters applied to the search strategy. If a non-English paper is considered eligible for inclusion, relevant data and results will be translated to English before analysis.

The initial search strategy was developed considering previously published systematic reviews in paediatrics, clinical ethics and service evaluation. Search terms will be related to "Clinical ethics support", "paediatrics" AND "structure/process/outcome indicators" and adapted to each database requirement. Publications that would match the criteria for inclusion in the review previously known to the research team were successfully retrieved applying the search strategy in Medline online

database. Please see Table 2 for Medline search strategy and refer to Supplementary file 1 for complete search strategy for all included databases.

Table 2. Medline search strategy

1. paediatric.mp. 2. paediatr*.mp. 3. pediatric.mp. or Pediatrics/ 4. pediatr*.mp. 5. child*.mp. or Child/ 6. 6Adolescent/ or adolescent.mp. 7. adolesc*.mp. 8. infant*.mp. 9. infant.mp. or Infant/ 10. kid.mp. 11. kids.mp. 12. baby.mp. 13. babies.mp. 14. toddler*.mp. 15. childhood.mp. 16. juvenil*.mp. 17. youth*.mp. 18. minor.mp. or Minors/ 19. Infant, Newborn/ or infancy.mp. or Child, Preschool/ 20. Infant, Newborn/ or newborn*.mp. 21. Premature Birth/ or Infant, Premature/ or preterm*.mp. 22. prematur*.mp. 23. Puberty/ or pubert*.mp. 24. pubescen*.mp. 25. young person.mp. 26. neonatal.mp.	27. Ethicists/ or ethicist*.mp. 28. bioethicist*.mp. 29. medical ethics.mp. or Ethics, Medical/ 30. clinical ethics.mp. or Ethics, Clinical/ 31. clinical ethics committee.mp. or Ethics Committees, Clinical/ 32. bioethics.mp. or Bioethics/ 33. bioethical issues.mp. or Bioethical Issues/ 34. ethical issues.mp. or Ethics/ 35. ethical challenges.mp 36. moral review.mp. 37. Ethical Analysis/ or Ethics Consultation/ or moral case deliberation.mp. 38. moral case consult*.mp. 39. moral consult*.mp. 40. ethic* case review.mp. or Ethics Committees/ 41. ethic* deliberation.mp. 42. ethic* intervention.mp. 43. ethic* round.mp. 44. ethic* support.mp. 45. ethic* service.mp. 46. Ethical Analysis/ or ethic* analysis.mp. 47. ethic* referral.mp. 48. Ethics Committees/ or ethic* committee.mp. 49. bioethic* deliberation.mp. 50. bioethic* intervention.mp. 51. bioethic* round.mp. 52. bioethic* service.mp. 53. bioethic* support.mp. 54. bioethic* analysis.mp. 55. bioethic* referral.mp. 56. bioethic* committee.mp.	57. structure.mp. 58. model*.mp. 59. process*.mp. 60. intervention*.mp. 61. Outcome Assessment, Health Care/ or "Outcome and Process Assessment, Health Care"/ or assessment*.mp. 62. evaluation*.mp. 63. impact*.mp. 64. effectiveness.mp. 65. Medical Audit/ or Clinical Audit/ or audit.mp. 66. Outcome Assessment, Health Care/ or "Outcome and Process Assessment, Health Care"/ or outcome.mp. 67. "Quality of Health Care"/ or quality.mp. or Quality Indicators, Health Care/ 68. indicator*.mp.
69. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23 or 24 or 25 or 26		

70. 28 or 29 or 30 or 31 or 32 or 33 or 34 or 35 or 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49 or 50 or 51 or 52 or 53 or 54 or 55 or 56 or 57
71. 59 or 60 or 61 or 62 or 63 or 64 or 65 or 66 or 67 or 68 or 69 or 70
72. 69 AND 70 AND 71

Other resources. Reference and citation list of included studies will be hand-searched. When relevant, we will contact the authors of conference abstracts identified through the search for peer-review publications. We will allow a time frame of two weeks for a response before considering the publication unavailable.

Selection process.

All retrieved records will be managed using Refworks® reference manager software. After deduplication, a random sample of 10% will be independently screened by MD and BP to test the reliability of the criteria. Any disagreements will be discussed within the research team until agreement, and, if required, eligibility criteria will be adjusted and/or clarified to improve the consistency of the screening process. Thereafter, all titles and/or abstracts will be screened by MD to identify publications that meet the previously established inclusion and exclusion criteria... References selected for assessment in the full text will be independently dual-assessed by MD and BP against inclusion/exclusion criteria, and any disagreements will be discussed within the research team until agreement. Reasons for excluding articles after full-text assessment will be recorded and study references and reasons for exclusion will be reported. A Cohen's Kappa score over 90% will be required at both, the title/abstract and full text screening processes. The screening process will be presented as a PRISMA flowchart.

Data extraction.

Data from individual studies considered relevant for the review question will be extracted to a pre-piloted Excel data extraction form by MD and checked by BP. Disagreements will be discussed within the research team. Data entries will include: Publication details (First author, year of publication, title), setting (country, healthcare setting), study aims, study design, sampling method and sample description. Primary outcomes sought in the data set will include; (i) assessed aspects of paediatric CESS as reported by study authors, including service structures, processes and outcomes (i.e., membership, service's activities, referrers, cases, contexts and reasons for referrals); (ii) assessed dimensions (i.e., effectiveness, safety and responsiveness) and/or subdimensions of quality of care(51) as reported by study authors; and (iii) methods and instruments used in the assessment. For qualitative studies, all data within the results/finding section will be considered as results. Characteristics of included studies will be tabulated.

Quality assessment.

Following our initial literature review, we expect to find around 10-30 potentially eligible studies with multiple research designs, including qualitative, quantitative and mixed methods. To allow coherent and systematic critical appraisal of included studies with different designs, we will use the 2018 Mixed Methods Appraisal Tool.(52) The tool was developed specifically for the appraisal of complex systematic reviews that include empirical qualitative, quantitative and mixed methods studies. It includes specific criteria depending on the study design category. Each criterion is rated as "yes", "no", "can't tell" response. As recommended by the authors, for each study, we will present a detailed description of the rating of each criterion and calculate an overall quality score based on the number of quality criteria met. There is no recommended cut off score to exclude studies based on quality appraisal, and therefore no study will be excluded based on that criterion. Instead, we will conduct a sensitivity analysis during the data synthesis process to assess the impact of low-quality studies in the

review findings and adjust recommendations accordingly.(52) Quality appraisal will be conducted by MD and checked by BP.

Data synthesis

This systematic review is not focused only on the effectiveness of a particular intervention but addresses a broader range of questions. Data synthesis will follow a sequential explanatory synthesis following the two-step process described by Pluye and Hong:(46) First, a quantitative synthesis including results from quantitative studies and quantitative data from mixed-methods studies, followed by qualitative synthesis of results of qualitative studies and qualitative data form mixed-methods studies. The qualitative synthesis will be informed by the previously conducted quantitative synthesis. The integration of the qualitative synthesis into the quantitative one will allow a better understanding of the quantitative results,(46) also highlighting convergences and divergences between quantitative and qualitative synthesis to inform future research.

As we expect to find great diversity of outcome measures, quantitative data will be synthetised and tabulated using descriptive statistics where appropriate(53) guided by but not limited to Donabedian's framework of structure, process and outcomes. For qualitative data, we will conduct thematic synthesis approach,(54) using NVivo software for qualitative data analysis. The thematic synthesis will include: (i) Free line-by-line coding of the primary study's findings, (ii) organisation of these codes into related themes informed by the quantitative synthesis.(54) If the qualitative synthesis process develops additional themes that are not described in the quantitative synthesis, these will be included in the integrated synthesis as qualitative results only. To assess the robustness of the synthesis we will considering individual studies' quality and conduct sensitivity analysis if possible.

The synthesis will be conducted by MD and checked by BP. The final synthesis will be discussed within the research team.

Timeline

The protocol for this review is published in PROSPERO (date 27 September 2021).(50) Searches on the databases mentioned in the protocol were conducted in August 2021. Retrieved references were screened at title and abstract level during September-October 2021. Screening at full text level is planned for December 2021 – January 2022. We plan to proceed with further stages of the review, including stakeholder involvement, and data extraction and synthesis after the protocol is accepted for publication following the peer-review process. Data extraction and analysis are expected to take 6 months after study selection.

Patient and public Involvement.

Patients and the public were not involved in the design of this systematic review protocol, but will be involved further at two stages in the process of the research, to ensure the review outcomes are useful and relevant.(55,56) Following Cochrane good practice guidance for people involvement in systematic reviews,(55) we sought to involve views of diverse stakeholders. Since p-CESS are established within healthcare institutions, and provide support to clinicians and patients and their families in making ethically challenging decision, we defined four stakeholders' categories whose collaboration would be valuable to the research process; (i) institutional managers, (ii) p-CESS board members, (iii) clinicians and (iv) parents. We decided not to involve children and young people since, to the authors' knowledge, they are rarely involved in ethics consultations themselves, but represented by their parents throughout the process. We will establish two advisory groups, one Chilean and one UK-based, with one representative for each stakeholder category. We will invite representatives that are already known to the research team to join the advisory group and participate in two one-hour workshops, one before data extraction and synthesis and a second one after preliminary results.

Parents' representatives will be or have been previously involved as parent representative in a p-CESS. In the first webinar, participants will be asked to share their views, thoughts, opinions or experiences to ensure we will be looking for the appropriate data in the included studies. At the second webinar, participants will have the opportunity to provide their feedback on the preliminary findings, to add context and meaning to the findings, contributing to the overall interpretation and recommendations. Stakeholder involvement will be reported following the GRIPP (Guidance for Reporting Involvement of Patients and Public) checklist.(45)

Ethics and dissemination.

As a systematic review of published data, no ethical approval is necessary. Following Cochrane guidance,(55) ethical approval for stakeholder involvement in this review would not be required as workshops would not be audio-recorded and no vulnerable groups will participate.

We will present and discuss our findings in an open-access webinar, inviting a broad range of stakeholders to attend, including hospital managers, clinicians, academic ethicists, and patient representatives. A final report will be published in a relevant academic peer-reviewed journal.

We plan to continue this research by conducting a modified Delphi study based on our review results to further explore the most appropriate quality indicators for evaluating p-CESS.

DISCUSSION

Interest in assessing CESS quality and effectiveness has grown in the past decades, with little attention paid to p-CESS. The results of this work will provide us with the first systematic review of evidence on Paediatric Clinical Ethics Support Services.

The review team is comprised by a bilingual and international research team that includes a Paediatric Intensivist Consultant with vast experience in paediatric medical ethics leading the teaching, research and clinical activities of the p-CESS at a large tertiary children's hospital; a speech therapist, certified clinical ethics consultant and PhD student in CESSs evaluation and a medical doctor and ethicist with

experience in systematic reviews and ethics research. The researchers' diverse backgrounds will contribute with their experiences and perspectives on CESS structures, processes, and outcomes in different contexts (Latin America and the UK). This will also allow a more comprehensive review both, by searching a Latin American specific database and the inclusion papers published in English and Spanish languages. This will enhance the review comprehensiveness, as long as potential bias is given due consideration in the result interpretation and recommendation development stages. Evidence on the effect of English-restricted criteria in traditional systematic reviews of randomised controlled trials with meta-analyses has not shown to result in significant bias.(57,58). However, this review on p-CESS structures, processes and outcomes will include a broader range of study designs and therefore potential bias associated with the exclusion of studies published in languages other than English and Spanish will be considered in the interpretation of results and recommendations.

The inclusion of peer-reviewed publications only might result in the omission of relevant publications (i.e., CESS terms of references and/or reports published in institutional websites). However, focusing on peer-reviewed publications will ensure validity of data included in the synthesis and also warrant a balance between the amount of data and the capacity of the research team, without compromising the review results. Moreover, we aim at mitigating the potential exclusion of relevant data by including case reports and case studies. The use of the Donabedian model will allow a structured and objective assessment of p-CESS contribution to patients' care. This is a well-accepted and widely used framework. However, considering the normative nature of CESS and their interventions and outcomes, the framework will be used as a guide and adaptation is expected.

We hope that our review results will allow for a better understanding of p-CESS structures, processes, and outcomes, contributing to further research exploring the normative and empirical basis of p-CESS.

Author contributions: MD and JB conceived the review. MD, JB and BP developed the protocol. All authors revised and edited the manuscript and approved the final version.

Funding: This work was supported by the Institute of Medical Ethics "Research in Medical Ethics" grant (No grant number). The funder has had no involvement in the review protocol development.

Competing interests: None to declare.

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Supplementary file 1. Complete search strategy for all databases

Embase Classic+Embase

1 paediatric.mp.

2 paediatr*.mp.

3 pediatric.mp. or Pediatrics/

4 pediatr*.mp.

5 child*.mp. or Child/

6 Adolescent/ or adolescent.mp.

7 adolesc*.mp.

8 infant*.mp.

9 infant.mp. or Infant/

10 kid.mp.

11 kids.mp.

12 baby.mp.

13 babies.mp.

14 toddler*.mp.

15 childhood.mp.

16 juvenil*.mp.

17 youth*.mp.

18 minor.mp. or Minors/

19 Infant, Newborn/ or infancy.mp. or Child, Preschool/

20 Infant, Newborn/ or newborn*.mp.

21 Premature Birth/ or Infant, Premature/ or preterm*.mp.

22 prematur*.mp.

23 Puberty/ or pubert*.mp.

24 pubescen*.mp.

25 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23 or 24

26 Ethicists/ or ethicist*.mp.

27 bioethicist*.mp.

28 medical ethics.mp. or Ethics, Medical/

29 clinical ethics.mp. or Ethics, Clinical/

30 clinical ethics committee.mp. or Ethics Committees, Clinical/

31 bioethics.mp. or Bioethics/

32 bioethical issues.mp. or Bioethical Issues/

33 ethical issues.mp. or Ethics/

34 ethical challenges.mp.

35 moral review.mp.

36 Ethical Analysis/ or Ethics Consultation/ or moral case deliberation.mp.

37 moral case consult*.mp.

38 moral consult*.mp.

39 ethic* case review.mp. or Ethics Committees/

40 ethic* deliberation.mp.

41 ethic* intervention.mp.

42 ethic* round.mp.

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45 Ethical Analysis/ or ethic* analysis.mp.

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68 Medical Audit/ or Clinical Audit/ or audit.mp.

69 Outcome Assessment, Health Care/ or "Outcome and Process Assessment, Health Care"/ or outcome.mp.

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82 infant*.mp.

83 infant/ or infant.mp.

84 kid.mp.

85 kids.mp.

86 baby.mp. or baby/

87 babies.mp.

88 toddler/ or toddler*.mp.

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90 juvenil*.mp.

91 youth*.mp.

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95 newborn/ or newborn*.mp.

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143 impact*.mp.

144 effectiveness.mp.

145 audit.mp. or clinical audit/

146 outcome assessment/ or outcome.mp.

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148	indicator*.mp.
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4	pediatr*.mp.
5	child*.mp. or Child/
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7	adolesc*.mp.
8	infant*.mp.
9	infant.mp. or Infant/
10	kid.mp.
11	kids.mp.
12	baby.mp.
13	babies.mp.
14	toddler*.mp.
15	childhood.mp.
16	juvenil*.mp.
17	youth*.mp.
18	minor.mp. or Minors/
19	Infant, Newborn/ or infancy.mp. or Child, Preschool/
20	Infant, Newborn/ or newborn*.mp.
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29 clinical ethics.mp. or Ethics, Clinical/

30 clinical ethics committee.mp. or Ethics Committees, Clinical/

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32 bioethical issues.mp. or Bioethical Issues/

33 ethical issues.mp. or Ethics/

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36 Ethical Analysis/ or Ethics Consultation/ or moral case deliberation.mp.

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71 indicator*.mp.

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Philosopher's Index

Philosophy - journal articles, books, book chapters and book reviews

Subject Area(s): Social Sciences , History , The Arts

(paediatr* OR pediatr* OR child* OR adolesc* OR infant* OR kid OR kids OR baby OR babies OR toddler* OR childhood OR juvenil* OR youth* OR minor* OR infancy OR newborn* OR prematurity OR preterm OR pubert* OR pubescen* OR "young person" OR neonatal) AND (ethicist* OR bioethicist* OR bioethic* OR "clinical ethics" OR "clinical ethics committee" OR "bioethical issue*" OR "ethical issue*" OR "ethical challenge*" OR "moral review" OR "moral case deliberation" OR "moral case consult*" OR "moral consult*" OR "ethic* case review" OR "ethic* deliberation" OR

"ethic* round" OR "ethic* intervention" OR "ethic* support" OR "ethic* service" OR "ethic* analysis" OR "ethic* referral" OR "ethic* committee" OR "bioethic* deliberation" OR "bioethic* intervention" OR "bioethic* round" OR "bioethic* service" OR "bioethic* support" OR "bioethic* analysis" OR "bioethic* referral" OR "bioethic* committee") AND (structure OR model* OR process* OR intervention OR assessment* OR evaluation* OR impact* OR effectiveness OR audit OR outcome* OR quality OR indicator*)

APA PsycInfo®

(paediatr* OR pediatr* OR child* OR adolesc* OR infant* OR kid OR kids OR baby OR babies OR toddler* OR childhood OR juvenil* OR youth* OR minor* OR infancy OR newborn* OR prematurity OR preterm OR pubert* OR pubescen* OR "young person" OR neonatal) AND (ethicist* OR bioethicist* OR bioethic* OR "clinical ethics" OR "clinical ethics committee" OR "bioethical issue*" OR "ethical issue*" OR "ethical challenge*" OR "moral review" OR "moral case deliberation" OR "moral case consult*" OR "moral consult*" OR "ethic* case review" OR "ethic* deliberation" OR "ethic* round" OR "ethic* intervention" OR "ethic* support" OR "ethic* service" OR "ethic* analysis" OR "ethic* referral" OR "ethic* committee" OR "bioethic* deliberation" OR "bioethic* intervention" OR "bioethic* round" OR "bioethic* service" OR "bioethic* support" OR "bioethic* analysis" OR "bioethic* referral" OR "bioethic* committee") AND (structure OR model* OR process* OR intervention OR assessment* OR evaluation* OR impact* OR effectiveness OR audit OR outcome* OR quality OR indicator*)

VHL Lilacs

(paediatr* OR pediatr* OR child* OR adolesc* OR infant* OR kid OR kids OR baby OR babies OR toddler* OR childhood OR juvenil* OR youth* OR minor* OR infancy OR newborn* OR prematurity OR preterm OR pubert* OR pubescen* OR young person OR neonatal) AND (ethicist* OR bioethicist* OR bioethic* OR clinical ethics OR clinical ethics committee OR bioethical issue* OR ethical issue* OR ethical challenge* OR moral review OR moral case deliberation OR moral case consult* OR moral consult* OR ethic* case review OR ethic* deliberation OR ethic* round OR ethic* intervention OR ethic* support OR ethic* service OR ethic* analysis OR ethic* referral OR ethic* committee OR bioethic* deliberation OR bioethic* intervention OR bioethic* round OR bioethic* service OR bioethic* support OR bioethic* analysis OR bioethic* referral OR bioethic* committee) AND (structure OR model* OR process* OR intervention OR assessment* OR evaluation* OR impact* OR effectiveness OR audit OR outcome* OR quality OR indicator*) AND (db:("LILACS"))

Web of Science

(paediatr* OR pediatr* OR child* OR adolesc* OR infant* OR kid OR kids OR baby OR babies OR toddler* OR childhood OR juvenil* OR youth* OR minor* OR infancy OR newborn* OR prematurity OR preterm OR pubert* OR pubescen* OR "young person" OR neonatal) AND (ethicist* OR bioethicist* OR bioethic* OR "clinical ethics" OR "clinical ethics committee" OR "bioethical issue*" OR "ethical issue*" OR "ethical challenge*" OR "moral review" OR "moral case deliberation" OR "moral case consult*" OR "moral consult*" OR "ethic* case review" OR "ethic* deliberation" OR "ethic* round" OR "ethic* intervention" OR "ethic* support" OR "ethic* service" OR "ethic* analysis" OR "ethic* referral" OR "ethic* committee" OR "bioethic* deliberation" OR "bioethic* intervention" OR "bioethic* round" OR "bioethic* service" OR "bioethic* support" OR "bioethic* analysis" OR "bioethic* referral" OR "bioethic* committee") AND (structure OR model* OR process* OR intervention OR assessment* OR evaluation* OR impact* OR effectiveness OR audit OR outcome* OR quality OR indicator*)

CINAHL (EBSCOhost Research Databases)

S10 S7 AND S8 AND S9

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

S9 S4 OR S5 OR S6

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

S8 S2 OR S3

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

S7 strucure OR model* OR process* OR intervention OR assessment* OR evaluation* OR impact* OR effectiveness OR audit OR outcome* OR quality OR indicator*

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

S6 ethic* referral OR ethic* committee OR bioethic* deliberation OR bioethic* intervention OR bioethic* round OR bioethic* service OR bioethic* support OR bioethic* analysis OR bioethic* referral OR bioethic* committee

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

S5 ethical challenge* OR moral review OR moral case deliberation OR moral case consult* OR moral consult* OR ethic* case review OR ethic* deliberation OR ethic* intervention OR ethic* round OR ethic* support OR ethic* service OR ethic* analysis

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

S4 ethicist* OR bioethicist* OR medical ethics OR clinical ethics OR clinical ethics committee OR bioethi* OR bioethical issues OR ethical issues

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

S3 youth* OR infancy OR newborn* OR prematur* OR pubert* OR pubescen* OR young person OR neonatal

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

S2 paediatr* OR pediatr* OR child* OR adolesc* OR infant* OR kid OR kids OR baby OR babies OR toddler* OR juvenil* OR minor*

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

S1 paediatric

Expanders - Apply equivalent subjects Search modes - Boolean/Phrase

PRISMA-P (Preferred Reporting Items for Systematic review and Meta-Analysis Protocols) 2015 checklist: recommended items to address in a systematic review protocol*

Section and topic	Item No	Checklist item	Page in text
ADMINISTRATIVE INFORMATION			
Title:			
Identification	1a	Identify the report as a protocol of a systematic review	1
Update	1b	If the protocol is for an update of a previous systematic review, identify as such	n/a
Registration	2	If registered, provide the name of the registry (such as PROSPERO) and registration number	2, 9
Authors:			
Contact	3a	Provide name, institutional affiliation, e-mail address of all protocol authors; provide physical mailing address of corresponding author	1
Contributions	3b	Describe contributions of protocol authors and identify the guarantor of the review	21
Amendments	4	If the protocol represents an amendment of a previously completed or published protocol, identify as such and list changes; otherwise, state plan for documenting important protocol amendments	n/a
Support:			
Sources	5a	Indicate sources of financial or other support for the review	21
Sponsor	5b	Provide name for the review funder and/or sponsor	21
Role of sponsor or funder	5c	Describe roles of funder(s), sponsor(s), and/or institution(s), if any, in developing the protocol	21
INTRODUCTION			
Rationale	6	Describe the rationale for the review in the context of what is already known	4-8
Objectives	7	Provide an explicit statement of the question(s) the review will address with reference to participants, interventions, comparators, and outcomes (PICO)	8
METHODS			
Eligibility criteria	8	Specify the study characteristics (such as PICO, study design, setting, time frame) and report characteristics (such as years considered, language, publication status) to be used as criteria for eligibility for the review	9-10 Table 1
Information sources	9	Describe all intended information sources (such as electronic databases, contact with study authors, trial registers or other grey literature sources) with planned dates of coverage	12-14
Search strategy	10	Present draft of search strategy to be used for at least one electronic database, including planned limits, such that it could be repeated	12 Table 2

			Supplementary file 1
Study records:			
Data management	11a	Describe the mechanism(s) that will be used to manage records and data throughout the review	14-15
Selection process	11b	State the process that will be used for selecting studies (such as two independent reviewers) through each phase of the review (that is, screening, eligibility and inclusion in meta-analysis)	14
Data collection process	11c	Describe planned method of extracting data from reports (such as piloting forms, done independently, in duplicate), any processes for obtaining and confirming data from investigators	14-15
Data items	12	List and define all variables for which data will be sought (such as PICO items, funding sources), any pre-planned data assumptions and simplifications	14-15
Outcomes and prioritization	13	List and define all outcomes for which data will be sought, including prioritization of main and additional outcomes, with rationale	15
Risk of bias in individual studies	14	Describe anticipated methods for assessing risk of bias of individual studies, including whether this will be done at the outcome or study level, or both; state how this information will be used in data synthesis	15-16
Data synthesis	15a	Describe criteria under which study data will be quantitatively synthesised	16-17
	15b	If data are appropriate for quantitative synthesis, describe planned summary measures, methods of handling data and methods of combining data from studies, including any planned exploration of consistency (such as I^2, Kendall's τ)	16-17
	15c	Describe any proposed additional analyses (such as sensitivity or subgroup analyses, meta-regression)	16-17
	15d	If quantitative synthesis is not appropriate, describe the type of summary planned	16-17
Meta-bias(es)	16	Specify any planned assessment of meta-bias(es) (such as publication bias across studies, selective reporting within studies)	n/a
Confidence in cumulative evidence	17	Describe how the strength of the body of evidence will be assessed (such as GRADE)	n/a

*** It is strongly recommended that this checklist be read in conjunction with the PRISMA-P Explanation and Elaboration (cite when available) for important clarification on the items. Amendments to a review protocol should be tracked and dated. The copyright for PRISMA-P (including checklist) is held by the PRISMA-P Group and is distributed under a Creative Commons Attribution Licence 4.0.**

From: Shamseer L, Moher D, Clarke M, Ghersi D, Liberati A, Petticrew M, Shekelle P, Stewart L, PRISMA-P Group. Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015: elaboration and explanation. BMJ. 2015 Jan 2;349(jan02 1):g7647.