Progress of core outcome set development in maternal and neonatal health: a systematic review using COS-STAD standards

Valerie Slavin[1], Debra Creedy[2], and Jennifer Gamble[2]

[1]GriffithUniversity
[2]Griffith University

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Abstract

Background: Methods used to develop existing core outcome sets relevant to maternal and neonatal health have not been fully evaluated. Objectives: To systematically review core outcome sets relevant to maternal and neonatal health; evaluate against minimum standards for development; and evaluate overlap between core outcome sets. Search strategy: Multi-faceted search of two core outcome set registers (COMET, CROWN) the ICHOM database of standard sets, and three electronic databases (MEDLINE, EMBASE, CINAHL) was conducted from inception to January 2020. Selection criteria: Published papers reporting completed core outcome sets relevant to maternal or neonatal health, for research or clinical use, were evaluated against COS-STAD minimum standards for development. Data collection and analysis: Descriptive statistics describe characteristics and results. Main results: Thirty-two papers relating to 26 core outcome sets were included (maternal: 18 papers: 17 COS; neonatal: 14 papers: 9 COS). Fifteen (58%) were published since 2017. No included COS met all minimum standards for development. All COS met the minimum standard for scope. Eighteen (69%) met all three minimum standards for stakeholder involvement. No included COS met all five minimum standards for consensus process. COS included between 6 and 56 outcomes. Two COS (8%) provided recommendations for how and when to measure outcomes. Conclusions: This is the first application of COS-STAD minimum standards relevant to maternal and neonatal health. Findings offer a baseline evaluation. There is an urgent need to address outcomes, measurement and timing in core outcomes to support harmonization between core outcome sets.

Introduction

A core outcome set (COS) represents an agreed set of outcomes that should be measured and reported, as a minimum, in all clinical trials in a specific area of health or health care,[1] and can be used in other research and clinical audit.[2] COS development and implementation is supported by the Core Outcome Measures in Effectiveness Trials (COMET) Initiative. Launched in 2010, COMET collates and stimulates the development and application of agreed standardised COS by maintaining a publicly available, searchable database of published and ongoing COS.[3] The Core Outcomes in Women's and Newborn Health (CROWN) Initiative strongly supports the development and dissemination of COS within women's health research.[4]

With over 330 published COS developed in several health disciplines since 1981,[5] COS related to maternal and neonatal care has seen slower development. In a 2017 systematic review, Duffy and colleagues identified only four published COS related to women's and newborn health, three of which related to pregnancy and childbirth.[6] To be effective, COS must be developed using rigorous methods that reflect outcomes important to patients and other stakeholders.[7] The recent minimum standards for COS development (Core Outcome Set-STAndards for Development: COS-STAD) facilitates the assessment of whether a COS has

been developed using a reasonable approach.[8] Core outcome set methodology is however in its infancy, and advancements in methodologic standards are likely in the future.[9] To evaluate progress, a baseline of methodological rigor is required to inform future maternal and neonatal COS development.

To date, little attention has been paid to the harmonisation of outcomes between similar COS. Without harmonisation of definitions, measures and timing, there is a danger that heterogeneity and research wastage will continue. The aim of the current study was to evaluate maternal and neonatal COS against available standards of development. The current study is guided by the following research questions:

1. Do existing completed maternal and neonatal core outcome sets meet minimum standards for development?
2. What is the extent of concordance between core outcome sets?

Methods

Protocol registration

The detailed protocol was prospectively registered with COMET (Registration Id: 1489) (http://www.comet-initiative.org/) and submitted to a peer reviewed journal. The study is reported according to the PRISMA statement (Figure S1).[10]

Study selection

Studies describing completed core outcome sets specific to maternal or neonatal health were included if they developed or applied methodology for determining which outcomes should be measured, or are important to measure in clinical trials, research or clinical practice. Studies were eligible if they related to pregnant or postpartum participants (up to 12 months postpartum), or neonates/infants (where the first outcome measurement is recommended within the first 28 days of birth), with any health condition and in any setting. Published conference papers were included if they provided adequate information about the completed COS or where a primary paper provided further information.

Excluded studies

Consistent with Gargon et al. papers were excluded if the design or rationale reported (i) a single study; (ii) related to pre-clinical or early phase trials only; (iii) reported the use of a COS; (iv) a systematic review of clinical trials; (v) studies of prognosis; (vi) studies of outcomes measured in clinical trials or quantitative descriptions of outcomes; (vii) based on the opinion of a single author; (viii) or focused on one domain/outcome only.[5] We excluded papers relating to early pregnancy loss (prior to 20 weeks gestation) as these were considered gynaecological rather than maternity.

Information sources

We searched the COMET[11] and CROWN[4]registers and ICHOM (International Consortium for Health Outcomes Measurement) list of standard sets.[12] We conducted an electronic database search of MEDLINE (via Ovid), EMBASE and CINAHL Complete (via EBSCOhost) in January 2020. Studies reported in English language were included from inception to January 2020. All ongoing COS identified in the COMET register and in previous reviews[2,5,6,13-17] were searched for updates of progress. Hand-searching reference lists complimented the search.

Search

A university health librarian helped to develop and pilot the search strategy. Our search terms combined three concepts ('core outcome set', 'methodology' and 'population'). All terms within each concept were combined with the Boolean operator OR and then the three concepts with AND. Search terms are outlined in Appendix S1.

Study selection and data management

Endnote software X8 was used to screen all citations. Duplicates were identified and removed. Titles and abstracts were screened by one author (VS). Full text papers were reviewed for all studies meeting the inclusion criteria. Papers not meeting the eligibility criteria were excluded and reason recorded. Full paper screening was conducted independently by one researcher (VS). Ten percent of included and excluded papers were assessed by a second reviewer (DC). Any disagreement between reviewers or uncertainty were resolved by consensus or by arbitration using a third reviewer (JG).

Data extraction

Data were extracted by one author (VS) using extraction forms guided by criteria outlined in previous reviews:[2,6] author(s), year of publication, COMET registration number, disease category, disease name, related papers, funder, CROWN registration, publication type, each item as defined on the COS-STAR statement[18], scope, stakeholder involvement, geographical location of stakeholders, patient participation, consensus process, final list of outcomes/domains, and measurement recommendations.

Sources of information

Supporting data was collected from primary COS papers, relevant project papers (systematic reviews and protocols) and from the COMET and CROWN registers.

Assessment of study against minimum standards

Included studies were assessed against COS-STAD minimum standards.[8] COS-STAD contains 11 standards covering three key domains (scope, stakeholders, and consensus process). Consistent with others,[9] item 9 ('*A scoring process and consensus definition is described a priori* ') was modified to include two assessment items for scoring process (termed 9a) and consensus definition (termed 9b). Each item was assessed as standard met, unclear, or not met using the assessment criteria outlined by Gargon and colleagues.[9]

Synthesis of results

Findings were described descriptively using text and tables and summarised as frequencies and percentages.

Results

Our search identified 5,227 individual citations. We excluded 5,096 records at the title and abstract screen, and a further 99 after screening the full paper (see Figure S2). A summary of reasons for exclusion of the full papers is presented in Table S1. Thirty-two papers relating to 26 individual core outcome sets met the inclusion criteria representing published COS related to maternal (n = 18 papers; 17 COS) and neonatal (n = 14 papers; 9 COS) health.

Study and registry characteristics

Of the 18 papers related to maternal COS, 17 were primary journal articles and one was a published conference paper (secondary paper) (outlined in Table S2). Of the 14 papers related to neonatal COS, 10 were primary journal articles three were published conference papers and one was a meeting abstract (secondary papers). Fifteen COS (58%) were published from 2017 onwards (range 2006 – 2020). All 26 COS projects were registered with COMET and 11 with CROWN: four as published and seven as ongoing projects.

Eighteen studies (69%) were published in free to view journals and three quarters were funded projects (n = 20). Of the eight COS with separately published protocols, all related COS were published from 2017 onwards. Fifteen individual systematic reviews were identified relating to 11 separate COS, with all but one COS published from 2016 onwards. Fourteen of the 26 COS (54%) were included in previous reviews. The current review includes twelve new COS (46%).

Methods used in COS development

The scope and methods for development are outlined in Table S3 and summarised in the following sections.

Scope

The scope of included studies is summarised in Table 1. While most COS were intended for research (81%), almost one in five were also recommended for clinical practice. One COS developed by ICHOM was designed specifically for clinical application.[19] Of the 25 COS developed for research, most cover any intervention (81%).

[Insert Table 1 about here]

Stakeholders involvement

Stakeholder groups involvement is summarised in Table 2. Clinical experts from 18 disciplines were involved in COS development. Neonatologists (54%), obstetricians (46%) and midwives (42%) represented the largest clinical expert groups involved. Public representation was sought in 18 COS (69%). Countries represented by stakeholder groups are outlined in Table S4. Of the 18 COS for which country representation data was available, each COS stakeholder group represented a median 26 countries (range 1- 36). High income countries, as defined by the World Bank (https://www.worldbank.org/), dominated representation. Developing countries were poorly represented in almost all COS.

Patient participation and retainment was reported in 18 COS projects (Table S5). Among 18 Delphi studies, half of all stakeholders were retained at final round (Mdn = 48%, range 20 - 86%). Reported median patient participation in 13 Delphi studies was 16% (range 11 - 53%) at round one and 16% (range 2 - 61%) at the final round. It was not possible to evaluate healthcare professional participation and attrition by discipline due to limitations of joint category reporting in several studies.

[Insert Table 2 about here]

Methods used in consensus process

Methods used during the consensus process are summarised in Table 3. Commonly, the initial list of outcomes was generated by literature/systematic review (n = 19; 73%). Consensus was most often reached through electronic Delphi procedure (n = 19, 73%) using either a two- (n = 8) or three-round (n = 10) process. Around half of all COS projects used a combination of Delphi with some form of final consensus meeting (n = 15). Scoring and consensus processes were employed in 18 (69%) COS. Of these, a 9-point Likert scale was

the most common procedure to score outcomes (72.2%) and the 70%/15% process (see example description in Table 3) was the most common consensus definition (67%).

[Insert Table 3 about here]

Standard of COS development

Each COS was evaluated against COS-STAD minimum standards as outlined in Table S6 and summarised in Table 4. None of the included studies met all minimum standards for COS development. Median number of standards met was 8 (range 5 – 11). For 14 COS published up to and including 2017, the medium number of standards met was 6.5 (range 5-11), while the median number of standards met for COS published from 2018 onwards was 10 (range 6 – 11).

[Insert Table 4 about here]

Scope specification (Standards 1–4)

All 26 COS (100%) described in some way the research or practice setting (*Standard 1*), health condition (*Standard 2*), population (*Standard 3*), and intervention (*Standard 4*) covered by the COS, meeting the criteria representing the scope covered by the COS. Where no specific intervention was specified, any intervention was assumed.

Stakeholders involved (Standards 5–7)

Eighteen (69%) studies met all three standards for stakeholder involvement including health professionals, researchers and patients or their representatives. Some assumptions were made regarding research and health professional stakeholder involvement. Author contribution and affiliation indicated standards were most probably met in these studies. Health care professionals and researchers were well presented in all stakeholder groups. Eighteen studies (69%) met the criteria for patient or representative involvement. While eight (31%%) did not meet this criterion, these were assumed as stakeholder involvement occurred during professional conference meetings or expert working groups.

Consensus process (Standards 8-11)

No studies met all standards for the consensus process. As such standards within this domain are addressed individually.

Standard 8: Initial list of outcomes considered both health care professionals' and patients' views'

Six studies (23%) met this standard, seven were unclear (27%) and 13 (50%) did not meet the standard. Those that met the standard demonstrated clear consideration of patient views by conducting either qualitative research studies of patients' views or conducted patient interviews to generate the initial list of outcomes. Jones et al., for example, stated, '*The list of core outcomes was developed incollaboration with the PCG consumers group. . . .outcomes that were of importance to them'* [20] whereas Van't Hooft et al.,[21] stated '*Patient representatives and parents were invited through social media. . . to share their opinions regarding outcomes relevant to preterm birth* '.

Standard 9a: a scoring process was described a priori

Eleven studies (42%) met this standard but was unclear in seven studies (27%). Eight studies (31%) did not meet this standard as they did not use a scoring process. For those that met the standard, clear evidence was provided either within a published prospective protocol or COMET protocol registry entry (n = 7) or authors reported '*a priori* ' specific to scoring process.

Standard 9b: a consensus definition was described a priori

Eleven studies (42%) met the standard, providing clear evidence that consensus methods were defined a priori. In eight studies (31%) it was unclear and seven (27%) did not use consensus methods. Those that met this criterion provided clear evidence in published protocols, COMET registry entries or authors stated '*a priori* ' consensus definition.

Standard 10: Criteria for including/dropping/adding outcomes was described a priori.

Eight studies (31%) met this standard providing clear evidence that all three elements were defined a priori through published protocols, registry entry or stated '*a priori* ' within the body of the text specific to each element. In 18 (69%) studies it was unclear if the standard was met, commonly because all three elements were not clearly described.

Standard 11: Care was taken to avoid ambiguity of language used in the list of outcomes

Ten studies (38%) met this standard if evidence was described in either the protocol or main study paper. Perry et al. for example developed '*lay definitions for individual outcomes'* which were reviewed by consumer group representatives,[22] while the study protocol of Bogdanet et al. described '*the questionnaire will contain lay terminology...* '.[23] One study (4%) did not meet the standard, describing as a limitation, '*illegible translated outcomes that were not included in the list* '.[24] In 15 studies (58%) it was unclear if language ambiguity had been considered.

Outcomes and measurement considerations

Core outcomes, definitions, and measurement considerations described in 26 included COS are outlined in Table S7. The number of outcomes included in each COS ranged from six to 56. Maternal COS included both maternal and fetal/neonatal outcomes (Mdn = 17, range = 50), while neonatal specific COS generally included only neonatal outcomes (Mdn = 8, range = 20). To aid analysis, outcomes were organised into grouping domains (i.e. survival, maternal morbidity, neonatal morbidity, resource utilisation). Survival was common across 16 separate COS, related to maternal death, fetal and neonatal loss but only clearly defined in three COS.[19,25,26] Similarly, resource utilisation was shared across 12 COS relating mainly to maternal/neonatal admission to intensive care, but definitions were only clearly defined in one COS.[22]

Significant overlap of outcomes between similar studies was evident. For example, eclampsia and pre-eclampsia are core outcomes outlined in six separate COS,[25,27-31] but only defined in one.[31] Similarly, while maternal haemorrhage is a domain shared across three separate COS, [25,32,33] a definition is only offered in one.[33] Two COS (8%), related to maternity care and gastroschisis also addressed '*how*'and '*when*' to measure outcomes.[19,26] While how to measure outcomes were considered in four additional papers,[33-36] clear recommendations were not reported. Although future work is planned by five COS developers to outline recommendations for how and when to measure outcomes,[22-24,37,38] and is acknowledged as needed by two,[30,39] 17 COS offer no guidance on how or when to measure outcomes, with no reported future plans to do so.

Search strategy analysis

Table S8 outlines the results of four individual searches in EMBASE, MEDLINE, CINAHL and COMET. Twelve studies (46%) were identified in all four individual searches. Of the 16 studies with the primary aim of developing COS, nine (56%) would have been identified in any single search. One COS study was not identifies in any electronic database search, but was identified in the COMET register.[40]Despite all 26 included studies being registered with COMET, four studies were not identified in our search, due mainly to falling outside our search parameters.

Discussion

Main findings

This comprehensive systematic review goes someway to update the previous work of Duffy et al.,[6] which included three completed core outcome sets relevant to maternal health. Our multi-faceted search strategy identified 32 unique citations relating to 26 core outcome sets: 17 maternal and 9 neonatal COS. Almost half the COS included in the current review had not been included in previous reviews (n = 12) indicating significant progress in COS development. Recently developed COS-STAD criteria were applied to evaluate each COS against minimum standards.[8] No COS met all the minimum standards. While scope was well addressed in all COS, patient participation in both stakeholder group and initial outcome generation are areas in need of greater attention. Our findings further indicated greater attention is needed to improve the methods used and reporting of the consensus process. Our findings indicate that standards of COS development have improved from 2018 onwards compared to those prior to 2018 (median standards 6.5 vs 10) likely reflecting the use of COMET guidelines,[7] COS-STAD minimum standards,[8] and COS-STAR reporting guidelines.[18]

It is important to acknowledge our findings relate to two distinct groups of COS: those specifically designed as core outcome sets and those that determined outcomes in studies with wider study aims. It is also important to acknowledge that almost half of the included COS pre-date recent methodological guidance.

Strengths and limitations

Our findings are strengthened by the comprehensive search strategy. We used the same tried and tested search strategy employed in several previous reviews relevant to maternal and newborn health.[2,5,13-16] Building on past reviews we further included COS specific to clinical practice. We believe this is the first application of the COS-STAD criteria to evaluate the COS development process relevant to maternal and neonatal health, making the findings pertinent to the CROWN Initiative. For consistency, we applied identical COS-STAD assessment criteria to that of Gargon et a., in their assessment of cancer-related COS;[9] the first report using COS-STAD criteria. Despite our efforts, our findings do have some limitations. Due to resource limitations, the search and screening process was conducted primarily by one person (VS). Although ten percent of papers were evaluated by a second person (DC), it is possible that some COS papers may have been missed. Assessing each standard against COS-STAD criteria was challenging. Some assumptions were made, particularly in terms of stakeholder involvement. For example, we assumed those who would use the COS in research were involved when participants were conference delegates or expert working groups. It is possible that this may have not been the case. In contrast to Duffy et al.,[6] we have not reported COS in development. A quick search of the COMET database identified 44 COS in development related to maternal and neonatal health. Future COS developers and users are encouraged to review the COMET database of ongoing *and* completed COS.

Interpretation

Our findings demonstrate the COMET register to be a comprehensive and up-to-date resource for COS developers and users to identify completed COS. Not all relevant COS were identified within our search which was limited to *Pregnancy and Childbirth* or *Neonatal* health categories despite being registered within other health areas. We recommend using a broad search strategy within COMET to avoid missing relevant COS. The CROWN register was less current. We hope our findings will be used to update this important discipline-specific resource.

While core outcome set methodology is in its infancy, exponential growth in COS development signifies an overwhelming commitment to the methodology as a strategy to address variation and research wastage. The recent publishing of COS-STAD minimum standards[8] and COS-STAR reporting guidelines[18] indicate a positive impact on COS development over the past three years, with COS being developed with increasing rigor. Many recent COS include published and/or registered protocols and systematic reviews improving the transparency of the project. While COMET registration is not yet mandated, prospective registration of planned COS projects outlining*a priori* the COS-STAD criteria could further improve transparency and is recommended.

Our review found similar limitations even in the most recent COS. To be relevant COS must include outcomes relevant to patients, clinicians and other stakeholders.[7] The initial list of outcomes must then be generated taking all views into account. Patient views were not included in a significant number of outcome lists. Since most COS are funded, it is important that funding be allocated to address this neglected area to ensure COS are truly relevant.

To be effective, COS must be implemented in practice.[7]To optimise implementation, COS should be generally relevant. Inclusive stakeholder participation is an important consideration in COS development to facilitate wide implementation. Similar to the most recent annual review [5] patient participation was not considered in some COS (participation: 71% vs 69%, respectively). In others, patient attrition rates were high despite large recruited samples. COS developers need to identify strategies to improve retention in this stakeholder group. Similar to the most recent review,[5] while most participants in the current study had international representation, low and middle-low income countries were poorly represented. This was despite median country representation being significantly higher in maternal and neonatal COS compared to general COS development (Mdn = 26 vs Mdn = 10).[5]Without diverse representation, COS may not be relevant in these countries. COS developers should identify strategies aimed at both improving and maintaining representation from these countries to improve global applicability of COS.[14]

Lastly, only two of the 26 COS specific to maternal and neonatal health included recommendations for measurement of outcomes. While core outcome set development focuses primarily on outcome generation, there is a danger that a lack of attention in determining how and when outcomes are measures will continue to contribute to ongoing variation in outcome reporting. A joint initiative by the COMET and COS-MIN (Consensus-based Standards for the selection of health Measurement Instruments) initiatives provides guidance for selecting outcome measures in COS.[41] COS developers are encouraged to include timely recommendations to address this gap in knowledge. Strategies to harmonise outcome measurement is an important consideration and may require concerted efforts in the future to address the issue.

Conclusions

The number of core outcome sets is growing exponentially in maternal and neonatal health. While the rigor of COS development has improved, there is room for improvement. Using minimum standards for development our findings offer a baseline to evaluate future COS.

Acknowledgements

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Disclosure of interests

None declared. Completed disclosure of interests form available to view online as supporting information

Contribution to authorship

VS designed the study and DC and JG provided feedback on the proposed design. VS conducted the primary literature search and DC and JG were involved in paper screening. VS conducted data extraction and quality evaluation. VS analysed and interpreted the data. VS drafted the manuscript. VS, DC and JG edited and approved the final manuscript.

Funding

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Details of ethics approval

Not applicable.

Supporting Information

Figure S1. PRISMA checklist

Hosted file

FIGURE_S1_SUPPINFO_PRISMA_CHECKLIST.doc available at https://authorea.com/users/302967/articles/433032-progress-of-core-outcome-set-development-in-maternal-and-neonatal-health-a-systematic-review-using-cos-stad-standards

Figure S2. Flow chart

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FIGURE_S2 _SUPPINFO_PRISMA_FLOWCHART.doc available at https://authorea.com/users/302967/articles/433032-progress-of-core-outcome-set-development-in-maternal-and-neonatal-health-a-systematic-review-using-cos-stad-standards

Appendix S1. Search strategy

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APPENDIX_S1_SUPPINFO_SEARCH.docx available at https://authorea.com/users/302967/articles/433032-progress-of-core-outcome-set-development-in-maternal-and-neonatal-health-a-systematic-review-using-cos-stad-standards

Table S1. Excluded papers

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TABLE_S1_SUPPINFO_Excluded Studies.docx available at https://authorea.com/users/302967/articles/433032-progress-of-core-outcome-set-development-in-maternal-and-neonatal-health-a-systematic-review-using-cos-stad-standards

Table S2. Study characteristics

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TABLE_S2_SUPPINFO_Characteristics included studies.docx available at https://authorea.com/users/302967/articles/433032-progress-of-core-outcome-set-development-in-maternal-and-neonatal-health-a-systematic-review-using-cos-stad-standards

Table S3. Scope and methods

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TABLE_S3_SUPPINFO_Scope and methods.docx available at https://authorea.com/users/302967/articles/433032-progress-of-core-outcome-set-development-in-maternal-and-neonatal-health-a-systematic-review-using-cos-stad-standards

Table S4. Countries represented

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TABLE_S4 _SUPPINFO_Countries represented.docx available at https://authorea.com/users/302967/articles/433032-progress-of-core-outcome-set-development-in-maternal-and-neonatal-health-a-systematic-review-using-cos-stad-standards

Table S5. Patient group involvement

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TABLE_S5_SUPPINFO_Patient involvement.docx available at https://authorea.com/users/302967/articles/433032-progress-of-core-outcome-set-development-in-maternal-and-neonatal-health-a-systematic-review-using-cos-stad-standards

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TABLE_S6_SUPPINFO_COS Minimum standards.docx available at https://authorea.com/users/302967/articles/433032-progress-of-core-outcome-set-development-in-maternal-and-neonatal-health-a-systematic-review-using-cos-stad-standards

Table S6. COS minimum standards

Table S7. Outcomes and definitions

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TABLE_S7_SUPPINFO_Outcomes.docx available at https://authorea.com/users/302967/articles/433032-progress-of-core-outcome-set-development-in-maternal-and-neonatal-health-a-systematic-review-using-cos-stad-standards

Table S8. Search strategy analysis

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TABLE_S8_SUPPINFO_Search strategy analysis.docx available at https://authorea.com/users/302967/articles/433032-progress-of-core-outcome-set-development-in-maternal-and-neonatal-health-a-systematic-review-using-cos-stad-standards

Table 1. Summary of scope specification of included studies (N = 26)

	n (%)
Study aims	
Specifically considered outcome selection and measurement	16 (61.5)
Considered outcomes while addressing wider clinical trial design issues	10 (38.5)
Intended use of recommendations	
Clinical research	21 (80.8)
Clinical research and practice	4 (15.4)
Clinical practice	1 (3.8)

	n (%)
Population characteristics	
Women	16 (61.5)
Neonates/infants	9 (34.6)
Women and neonates	1 (3.8)
Intervention characteristics	
Any / All intervention types	21 (80.8)
Management of care (labour pain)	1 (3.8)
Complimentary therapies	1 (3.8)
Drug treatments	2 (7.7)
Not applicable	1 (3.8)

Table 2. Stakeholder groups involved in selecting outcomes for inclusion in COS (N = 26)

Participant category	**n (%)**
Clinical experts	**26 (100.0)**
Neonatologist	14 (53.8)
Obstetricians	12 (46.2)
Midwife	11 (42.3)
Nurse	8 (30.8)
Family Physician/GP	7 (26.9)
Nutritionist/dietician	6 (23.1)
Paediatricians	6 (23.1)
Maternal fetal medicine specialist	3 (11.5)
Gastroenterologist	3 (11.5)
Endocrinologists	2 (7.7)
Medical specialists: Anaesthetics; Neurology; Haematology; Oncology	5 (19.0)
Allied Health: Ultrasonographers; Psychologist; Physiotherapists; Acupuncturists	5 (19.0)
Clinical research expertise; Clinical trialists/Member of a clinical trial network	2 (7.7)
Others	9 (34.6)
Public representatives	**18 (69.2)**
Patients	12 (46.2)
Patient support group representative	7 (26.9)
Parents/Carers	6 (23.1)
Relatives/advocates	4 (15.4)
Service users	2 (7.7)
Non-clinical experts	**18 (69.2)**
Researchers	13 (50.0)
Statisticians	4 (15.4)
Epidemiologists	2 (7.7)
Academic research representatives	1 (3.8)
Methodologists	2 (7.7)
Economists	1 (3.8)
Service providers	2 (7.7)
Authorities	**8 (30.8)**

Participant category	**n (%)**
Policy makers	3 (11.5)
Charities	1 (3.8)
Governmental agencies	4 (15.4)
Industry representatives	**3 (11.5)**
Pharmaceutical industry representatives	3 (11.5)
Others	**5 (19.2)**
Ethicists	3 (11.5)
Journal editors	2 (7.7)

Table 3. Methods used to develop 26 maternal/neonatal COS

Main methods	n (%)
Semi-structured group discussion only	1 (3.8)
Unstructured group discussion only	0
Consensus development conference only	4 (15.4)
Literature/systematic review only	0
Delphi only	0
Survey only	0
NGT only	0
Interview only	0
Mixed methods (as detailed below)	21 (80.8)
Literature/systematic review and Delphi	3 (11.5)
Literature/systematic review, Delphi, and Consensus meeting	12 (46.2)
Literature/systematic review, Delphi, and modified NGT	2 (7.7)
Literature/systematic review and stakeholder meeting	1 (3.8)
Literature/systematic review and survey	1 (3.8)
Survey, Delphi, consensus meeting	1 (3.8)
No methods described	1 (3.8)
Delphi	**n = 19 (100)**
Delphi process	
2-round	8 (42.1)
3-round	10 (52.6)
Not specified	1 (5.3)
Delphi software	
DelphiManager	5 (26.3)
QuestionPro	1 (5.3)
SurveyGizmo	1 (5.3)
Survey Monkey	4 (21.1)
Survey Methods	2 (10.5)
LimeSurvey	1 (5.3)
Online (not specified)	5 (26.3)
Scoring process	
5-point Likert scale	2 (7.7)
9-point Likert	13 (50.0)
Ranking of outcomes	3 (11.5)
No scoring process described	8 (30.8)

Main methods	n (%)
Consensus definition	
70/15% criteria*	12 (46.2)
70% or more participants reach consensus	3 (11.5)
Consensus across stakeholder groups	1 (3.8)
Consensus by majority of respondents	1 (3.8)
Unanimous decision from expert panel	1 (3.8)
No consensus definition	8 (30.7)

*For example: Consensus in: > 70% scoring 7-9 and <15% scoring 1-3; Consensus out: > 70% scoring 1-3 and <15% scoring 7-9; No consensus: anything else.

Table 4. Summary of COS-STAD minimum standards (N = 26)

Domain	Standard number	Standard
Scope specification	1	Research or practice setting(s) in which the COS is to be applied
	2	Health condition covered by the COS
	3	Population(s) covered by the COS
	4	Interventions covered by the COS
Stakeholders involved	5	Those who will use the COS in research
	6	Health professionals with experience of patients with the condition
	7	Patients with the condition or their representatives
Consensus process	8	Initial list of outcomes considered both health care professionals' and patients'
	9a	A scoring process was described a priori
	9b	A consensus definition was described a priori
	10	Criteria for including/dropping/adding outcomes was described a priori
	11	Care to avoid ambiguity of language used in the list of outcomes

References

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