Bridging the Gap: A Systematic Review on Reporting Baseline Characteristics, Process, and Outcome Parameters in Rectosigmoid Hirschsprung's Disease

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Abstract

The variation in standardized, well-defined parameters in Hirschsprung's disease (HSCR) research hinders overarching comparisons and complicates evaluations of care quality across healthcare settings. This review addresses the significant variability observed in these parameters as reported in recent publications. The goal is to compile a list of commonly described baseline characteristics, process and outcome measures, and to investigate disparities in their utilization and definitions. A systematic review of literature on the primary care process for HSCR was performed according to PRISMA guidelines. Relevant literature published between 2015 and 2021 was obtained by combining the search term "Hirschsprung's disease" with "treatment outcome," "complications," "mortality," "morbidity," and "survival" in Medline, Embase, and the Cochrane Library. We extracted study characteristics, reported process and outcome parameters, and patient and disease characteristics. We extracted 1,026 parameters from 200 publications and categorized these into patient characteristics ($n = 226$), treatment and care process characteristics ($n = 199$), and outcomes ($n = 601$). A total of 116 parameters were reported in more than 5% of publications. The most frequently reported characteristics were sex (88%), age at the time of surgery (66%), postoperative Hirschsprung-associated enterocolitis (64%), type of repair (57%), fecal incontinence (54%), and extent of aganglionosis (51%). This review underscores the pronounced variation in reported parameters within HSCR studies, highlighting the necessity for consistent, well-defined measures and reporting systems to foster improved data interpretability. Moreover, it advocates for the use of these findings in the development of a Core Indicator Set, complementing the recently developed Core Outcome Set. This will facilitate quality assessments across pediatric surgical centers throughout Europe.

Keywords
- Hirschsprung's disease
- outcome
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Introduction

Hirschsprung's disease (HSCR) is a rare congenital condition that affects 1 in 5,000 newborn children. It is characterized by the absence of ganglion cells in the enteric nervous system of the rectum, with variable involvement of the colon and small bowel. Current understanding suggests that this abnormality arises from disruptions in embryonic development stages, particularly the migration and survival of enteric nervous system precursor cells.[1,2]

Infants with HSCR present with symptoms of functional intestinal obstruction, such as delayed passage of meconium, abdominal distention, vomiting, and chronic constipation. Currently, establishing the diagnosis predominantly relies on rectal suction biopsies revealing an absence of ganglion cells. Aimed at re-establishing regular bowel functionality, the treatment generally involves resection of the aganglionic colon segment and reconnecting the ganglionic intestine segment to the anus. Surgical strategies include the Swenson,[3] Duhamel,[4] and Soave–Boley[5,6] resection techniques, fashioned as single- or multi-stage, open, laparoscopic, or total transanal procedures.[1,7]

Due to the rarity of HSCR, prospective, controlled, multicenter trials with adequate patient numbers are notably scarce. Furthermore, despite the increase in HSCR-related publications in recent decades, which could potentially contribute to more evidence-based practice, the usability of these research findings is limited. This limitation stems from the substantial variability in the reported research parameters and their respective definitions, preventing the possibility of comprehensive comparisons. Although a HSCR core outcome set has been formulated, this has not yet attained widespread recognition or utilization within the scholarly community.[7,8] These circumstances further prevent the resolution of existing evidence gaps, particularly in understanding short-term complications and long-term interactions between functional outcomes and quality of life.[9] Presently, guidelines for the management of HSCR often rely on expert opinion or consensus rather than comprehensive research, which implies that there is substantial room for improvement in the evidence-based practice.

The lack of standardized, well-defined parameters not only hinders overarching comparisons in research but also adds complexity to the evaluation and comparison of (quality of) HSCR care across hospitals, regions, or even countries. A pivotal step in advancing HSCR care has been the initiation of the European Pediatric Surgical Audit (EPSA) for several congenital malformations, including HSCR. This quality-of-care measurement tool for clinicians makes use of quality indicators to reliably identify, monitor, and evaluate variation in clinical practices and outcomes.[10–12] Improvement efforts could then be aligned with the findings from this evaluation. Indicators can be classified into three categories —structure indicators, process indicators, and outcome indicators—each evaluating different aspects of the care pathway. Examples of structure indicators are patient volumes and the availability of certain imaging techniques. Process indicators can involve factors such as the elapsed time between diagnosis and surgery, and the proportion of patients undergoing a particular diagnostic test. Outcome indicators might encompass elements such as survival rates or complication rates.[13] To develop a universally acknowledged set of quality indicators for evaluating and identifying variations in the quality of care for HSCR patients, it is essential to ascertain which parameters are considered most important by HSCR researchers and clinicians treating this disease. These identified outcomes can then serve as the basis for a consensus-driven process to delineate the new set of quality indicators. This initiative requires comprehensive understanding of all possible outcomes highlighted in HSCR research, as well as patient characteristics and treatment and care process characteristics, to adjust for case variability when comparing care quality. Such endeavors could provide an even deeper understanding of inter-hospital variability, further supporting future advancements in the care of this patient population.

The primary objective of this study was to compile a list of outcome measures described in recent peer-reviewed publications on HSCR and to explore the discrepancies in their utilization, definition, and reporting. The secondary objective was to identify patient characteristics and treatment and care process characteristics that would facilitate a more informed interpretation of future comparative analysis results.

Materials and Methods

This review was performed according to the Preferred Reporting Items for Systematic Reviews and Meta-analysis (PRISMA) statement and guidelines.[14] We developed a comprehensive search strategy in collaboration with a medical librarian of the Erasmus University Medical Center. The search was based on the search term "Hirschsprung's disease," combined with the terms "morbidity" or "mortality" or "survival" or "outcome" or "complication." The search criteria were applied to the databases Medline, Embase, and the Cochrane Library. Duplicate publications were removed and subsequently all studies published from 2015 up to and September 2021 were selected. Complete search strategies are provided in Supplementary Material 1 (available in the online version only). The search was performed in October 2021.

Inclusion and Exclusion Criteria

All publications related to any aspect of the main HSCR care process, including surgical and nonsurgical management, were included. To ensure contemporaneousness, articles published before the year 2015 were excluded. Other criteria for exclusion were the following: non-English language publications, animal research, in vitro studies, case series with fewer than 10 patients, editorials, letters, meeting abstracts, reviews, guidelines, and consensus statements.

Selection Process

Four reviewers participated in the selection process of the publications. N.T. and A.L.G. separately screened the titles and abstracts of all publications resulting from the search to determine their suitability in terms of reporting on the care

for and management of patients with HSCR. N.T., D.R., and A. L.G. then independently reviewed the full texts of the selected publications to ascertain their relevance to the study. Any disagreements were resolved by T.W., who served as a third reviewer. The reviewing authors were not blinded to the title, authors, or journal names during the screening process.

Data Collection, Analysis, and Reporting

D.R. and N.T. performed the data extraction, which included recording the origin and year of publications, study design, the number of included patients per study (as shown in ►Table 1), and all relevant parameters in a primary Excel-based framework. Parameters were categorized and, when feasible, merged based on consensus among the reviewers. For each parameter, the number of publications in which it was mentioned and the proportion relative to the total number of included publications were calculated. The parameters mentioned in more than 5% of the included publications were then recorded in a separate Excel file. Additionally, all definitions of studied parameters were extracted, as well as the types of instruments or tools, either standardized or not, with which the parameters were assessed, such as medical scores, scales, and questionnaires. This review did not include the extraction or interpretation of parameter estimates, nor did it involve the evaluation and assessment of methodological quality of the included publications. Statistical analyses were conducted utilizing a customized data computational approach implemented within the Excel environment.

Table 1 Study characteristics of included publications

		n	%
Originated in	Africa	6	3
	Asia	99	49
	Europe	62	31
	Eurasia	2	1
	North America	23	12
	Oceania	5	3
	South America	2	1
	Intercontinental	1	1
Study timing	Retrospective	130	65
	Prospective	30	15
	Cross-sectional	39	19
	Mixed design	1	1
Type of study	Observational	120	60
	Comparative	80	40
Study design	Cohort	179	90
	Case series	6	3
	Case–control	7	3
	Trial	8	4
Study scale	Single-center	150	75
	Multicenter	33	17
	National	17	8
Year of publication	2015	16	8
	2016	20	10
	2017	27	13
	2018	27	13
	2019	33	17
	2020	38	19
	2021	39	20
Number of included patients	< 25	36	18
	25–100	102	51
	101–300	34	17
	301–1,000	17	9
	> 1,000	11	5

Results

Included Publications

The search strategy resulted in 1,254 publications. After removal of duplicates and limiting the results to articles published between 2015 and 2021, a total of 828 articles remained, of which 200 met the inclusion criteria (►Fig. 1). The study characteristics of all included publications are summarized in ►Table 1. An individualized overview of included studies and corresponding study characteristics can be found in Supplementary Material 2 (available in the online version only).

Data Extraction

Full-text analysis of the 200 included publications identified 1,026 studied parameters described in at least one of the included publications. The parameters were categorized into patient characteristics ($n = 226$), treatment and care process characteristics ($n = 199$), and outcomes ($n = 601$). As several parameters could arguably be included in more than one category, categorization followed agreement between the first two authors. To facilitate the comparison of variation in studied parameters within similar subjects, we further arranged the parameters by topic, such as comorbidities, primary treatment, specific complications, and long-term outcome. A list of all identified, categorized items is attached in Supplementary Materials 3 to 5 (available in the online version only).

Measured Parameters

Of the 1,026 identified studied parameters, 116 were described in at least 5% of included publications (►Table 2). Predominantly described patient characteristics were sex (88%), age at the time of surgery (66%), and the extent of aganglionosis (histological level of transitional zone, 50.5%). Frequently studied treatment and care process characteristics included the type of repair (56.5%), duration of follow-up (39%), and preoperative stoma (35.5%). The most frequently mentioned outcome measures were postoperative Hirschsprung-associated enterocolitis (HAEC) (63.5%), incontinence (53.5%), and constipation (48%). The primarily

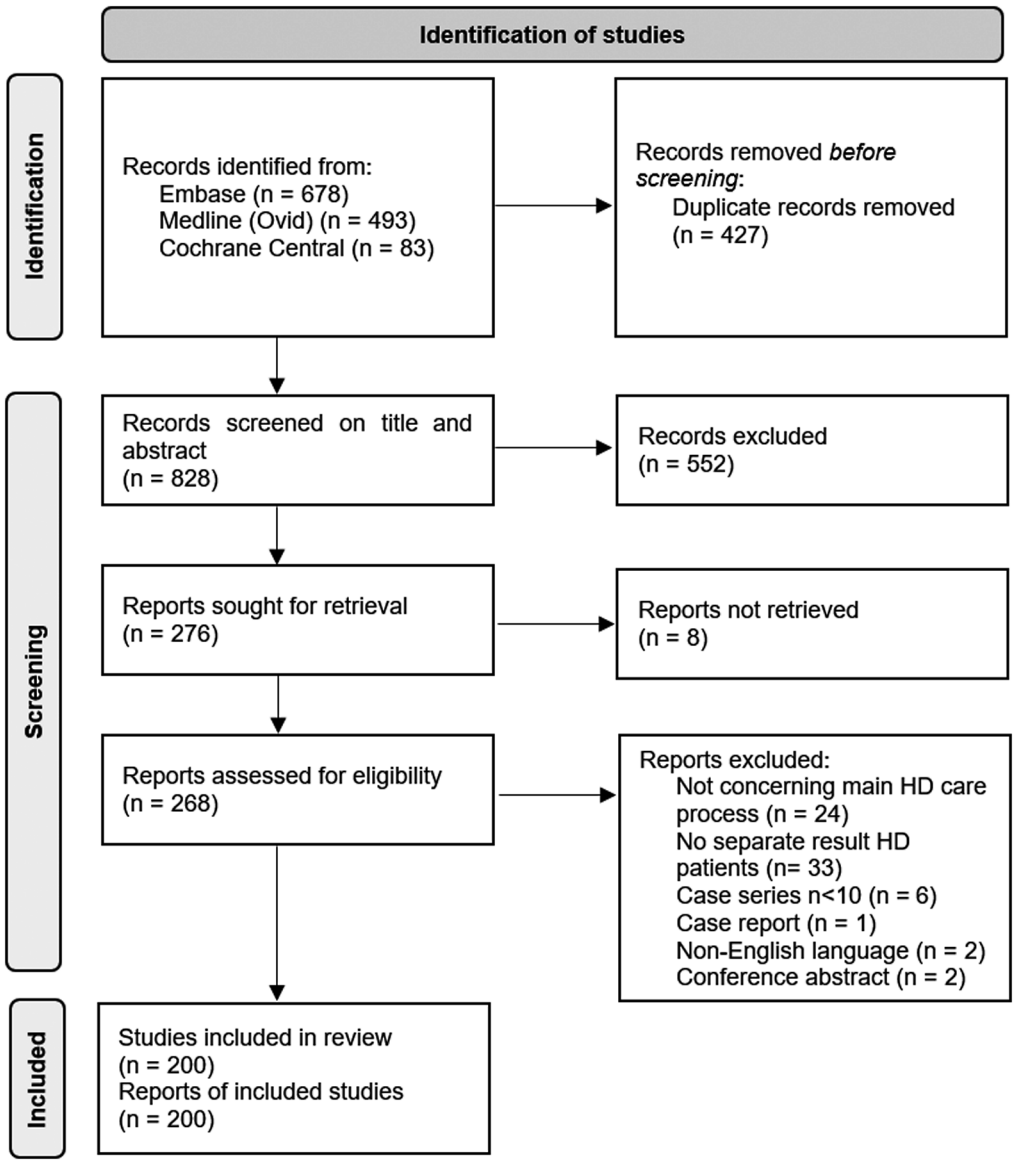

Fig. 1 PRISMA flowchart: systematic review of studied parameters in Hirschsprung's disease research.

Table 2 Identified studied parameters in HSCR research appearing in > 5% of the included articles

		n	%
Baseline characteristics	Sex	176	88
	Age at surgery	132	66
	Extent of aganglionosis[a]	101	51
	Syndromal, genetic, and chromosomal disorders	80	40
	(of which) Trisomy 21	65	33
	Age at follow-up	71	36
	Associated anomaly (cumulative)	58	29
	Initial clinical symptoms (cumulative)	55	28
	Gestational age or prematurity	45	23
	Enterocolitis (preoperative)	45	23
	Birth weight	42	21
	Cardiovascular anomalies	40	20

Table 2 (*Continued*)

		n	%
	Weight at surgery	37	19
	Age at diagnosis	34	17
	Family history of HSCR[b]	28	14
	Delay in evacuation/passage of meconium	25	13
	Congenital heart disease	25	13
	Renal/genitourinary anomalies	23	12
	Abdominal distention (preoperative)	19	10
	Constipation (preoperative)	19	10
	Neurologic anomalies	19	10
	Gastrointestinal anomaly[c]	19	10
	Ethnicity	18	9
	Vomiting (preoperative)	17	9
	Age at presenting symptoms	15	8
	Poor feeding/malnutrition/failure to thrive (preoperative)	14	7
	Congenital anomalies of the urogenital system	13	7
	Pulmonary/respiratory anomalies	12	6
	Intestinal perforation (preoperative)	10	5
	Congenital anomalies of the nervous system	10	5
Diagnostics, treatment, and care process	Type of repair	113	57
	Duration follow-up	78	39
	Preoperative/primary stoma	71	36
	Operative time (pull-through)	46	23
	Blood loss (pull-through)	41	21
	Laparoscopy or laparotomy or transanal approach	40	20
	Length of resected aganglionic and dilated segments	40	20
	Anal dilatations (preoperative)	33	17
	Contrast enema (preoperative)	27	14
	Rectal biopsies (preoperative)	26	13
	Follow-up rate	21	11
	Preoperative/primary colostomy	20	10
	Single- or multiple staged procedure	19	10
	Daily preoperative colon irrigations (mechanical bowel preparation)	18	9
	Intraoperative biopsies at the time of pull-through	18	9
	Intraoperative complications (pull-through)	18	9
	Preoperative/primary ileostomy	16	8
	Conversion laparoscopy to laparotomy (pull-through)	14	7
	ACE[d]	14	7
	Anorectal manometry (preoperative)	13	7
	Postoperative anal dilatation under general anesthesia	12	6
	Anorectal manometry: follow-up	12	6
	Time to start oral feeding	11	6
Outcome	Postoperative HAEC	127	64
	Fecal incontinence	107	54

(*Continued*)

Table 2 (*Continued*)

		n	%
	Constipation	96	48
	(Any) reoperation	85	43
	Anastomotic stricture/stenosis	76	38
	Soiling	72	36
	Length of primary hospital stay	71	36
	Anastomotic leakage	56	28
	Bowel function (standardized scoring)	54	27
	Mortality	53	27
	Defecation frequency	52	26
	Intestinal obstruction	49	25
	Gastrointestinal symptoms during follow-up	40	20
	Perianal issues (cumulative)	38	19
	Normal defecation function	37	19
	Use of bowel regulating medication	32	16
	Growth/weight/failure to thrive during follow-up	32	16
	Wound issues (cumulative)	32	16
	Permanent stoma	31	16
	Botulinum toxin	31	16
	Quality of life	28	14
	Complications (mentioned as such)	28	14
	Urinary function	27	14
	Wound infection	27	14
	Early postoperative complications (mentioned as such)	26	13
	Readmission	25	13
	Anal excoriations	24	12
	Residual aganglionosis	24	12
	Stoma at the time of follow-up	21	11
	Bowel function score, Rintala et al[15]	20	10
	Ileus/bowel obstruction due to adhesions	20	10
	Abdominal distension	18	9
	Difficulties with socializing/social adaptation/relationships	18	9
	Cause of death	17	9
	Fecal contamination/fecal accidents	17	9
	HAEC treated by conservative management	16	8
	Constipation manageable with laxatives	15	8
	Form/consistency of stool	15	8
	Rectal prolapse	15	8
	Stoma complications (cumulative)	15	8
	Constipation manageable with enema(s)	14	7
	Multiple episodes of enterocolitis	14	7
	HAEC recurrence	14	7
	Weight at the time of follow-up	13	7
	Feeling the need for defecation	13	7
	Ability to hold back defecation	13	7

Table 2 (*Continued*)

		n	%
	Use of enemas	13	7
	Use of laxatives	13	7
	Sepsis	13	7
	Incidence of HAEC	13	7
	Diarrhea	12	6
	Constipation manageable with diet	12	6
	Recurrent constipation	12	6
	Wound dehiscence	11	6
	Fever	10	5
	Problems with urinary incontinence	10	5
	Reoperation type	10	5
	Reoperation due to complications (cumulative, no timeframe)	10	5
	Reoperation due to obstruction/occlusion	10	5
	(Any) reoperation due to residual aganglionosis	10	5
	Abscess	10	5
	Infection (cumulative)	10	5
	Incomplete/transitional zone pull-through	10	5

[a]Extent of aganglionosis refers to the histological level of transitional zone, namely, rectal, sigmoid, rectosigmoid, distal descending colon.
[b]Gastrointestinal anomalies: excluding Hirschsprung's disease.
Abbreviations: ACE, antegrade colonic enema; HAEC, Hirschsprung-associated enterocolitis; HSCR, Hirschsprung's disease.

described complications after surgical treatment were anastomotic stricture and anastomotic leakage, cited in 38% and 28% of the publications, respectively. Other significant areas of focus encompassed length of primary hospital stay (highlighted in 35.5% of the studies), mortality (26.5%), reoperation rate (42.5%), and readmission rate (12.5%). Because the definitions of analyzed parameters and the used (standardized) measurement tools diverged significantly across different publications, it was challenging to derive clear-cut definitions or compare these results. Consequently, these results specifically concerning the definitions of the extracted parameters are not delineated in this report. An overview of the standardized clinical tools, scores, and scales applied in the studies is presented in ►**Table 3**.

Table 3 Identified tools/instruments utilized and reported in one or more included publications

		N	%
Incontinence scores	Wingspread criteria	4	2
	Miller incontinence score	2	1
	Baylor continence scale	2	1
	Postoperative fecal incontinence score	1	< 1
	Jorge–Wexner score	1	< 1
	Visick score	1	< 1
	Continence evaluation score	1	< 1
Scores for bowel function	Bowel function score (Rintala et al)[15]	20	10
	Bowel function score according to Holschneider	7	4
	Krickenberg classification system	6	3
	Pediatric incontinence and constipation scoring system	3	2
	Stooling survey (El Sawaf et al[16])	3	2
	Evacuation score of the Japan Society of ARM Study group	2	1
	Vancouver dysfunctional elimination syndrome survey (VDESS)	2	1
	Bowel function score according to Holschneider, adapted by Lindahl et al[17]	1	1

(*Continued*)

Table 3 (*Continued*)

		N	%
	Bowel habit diary	1	< 1
	Kelly scale score for recovery of bowel function	1	< 1
	Postoperative bowel function score	1	< 1
	HSCR anal function criteria (proposed by Reding[18])	1	< 1
	Wingspread score	1	< 1
	Rome IV criteria	1	< 1
Motor- and neurologic development and behavior	Intelligence (RAKIT[19] | WISC-III-NL)	1	< 1
	Memory (WISC-III)	1	< 1
	Attention (Dot cancellation test)	1	< 1
	Self-esteem (SPPC)	1	< 1
	WISC IV	1	< 1
	NEPSY	1	< 1
	KIDSCREEN 10	1	< 1
Sexual function	International index of erectile function	1	< 1
	Female sexual functioning index	1	< 1
	Female sexual distress scale	1	< 1
	Sexual education questionnaire	1	< 1
	Normal erectile functioning (erectile hardness score)	1	< 1
Hirschsprung's associated enterocolitis scores	HAEC score (by Pastor et al[20])	5	3
	Grading system by Elhalaby et al[21]	3	2
	Grading system by Murphy et al	1	< 1
Quality of life	Pediatric Quality of Life Inventory (PedsQL)	8	4
	Short Form 36 (SF-36)	5	3
	HSCR and ARM QoL scale (HAQL)	4	2
	Gastrointestinal Quality of Life Index (GIQLI)	4	2
	PedsQL Family Impact Module	2	1
	Fecal incontinence and constipation quality of life	2	1
	Child Health Questionnaire-Child Form 87 items (CHQ-CF87)	2	1
	World Health Organization Quality of Life 100 (WHOQOL-100)	2	1
	PedsQL Generic Core Scale	1	< 1
	PedsQL General Wellbeing Score	1	< 1
	WHO QOL-BREF	1	< 1
	Assessment of QoL in children and adolescents with fecal incontinence (AQLCAFI)	1	< 1
	Child Health Questionnaire - Parent Form 50 (CHQ-PF50)	1	< 1
	Scoring system by Bai et al[22] in patients with HD	1	< 1
	Barrena score	1	< 1
	Taiwanese Child and Caregiver Quality of Life (TCCQOL)	1	< 1
	Hirschsprung's Disease/Anorectal Malformation Quality of life Questionnaire (HAQL)	1	< 1
	KIDSCREEN 52	1	< 1
Skin and subcutaneous	Manchester scar scale	3	2

[a]Multiple instruments or tools may have been used in one publication.

Discussion

This study aimed to create a comprehensive overview of patient characteristics, treatment and care process characteristics, and outcome measures. To our knowledge, it represents the first attempt to compile such an extensive summary of parameters investigated in HSCR research. From the 200 publications on HSCR spanning from 2015 to 2021 that were included, we identified a total of 1,026 unique studied parameters, which could be categorized into 226 patient characteristics, 199 treatment and care process characteristics, and 601 outcomes. Among these, 116 parameters were described in at least 5% of the included publications, of which only 6 were mentioned in more than 50% of the included publications. These six parameters were postoperative HAEC, incontinence, type of repair, sex, age at the time of surgery, and extent of aganglionosis.

The current body of research of interest for this review primarily consists of retrospective, single-center studies with low patient volumes.[23] A substantial portion of the surveyed literature (69%) is based on data derived from cohorts comprising fewer than 100 patients, and more than three-quarters of the studies are single-center studies. This underscores the need for more multicenter and prospective studies involving larger patient cohorts to yield more robust results. Such approaches may include randomized controlled trials or the implementation of standardized data collection within patient registries, such as clinical audits. This study also confirms that contemporary research on HSCR tends to focus more on outcomes (59% of extracted variables) rather than on characteristics used to evaluate care processes (19%), and more on short-term than on long-term outcomes. Furthermore, considering the 1,026 distinct parameters mentioned in the included publications, we substantiated that data collection and reporting lacks uniformity across studies, making it harder to consolidate and analyze findings cohesively. We also demonstrated substantial variation in parameter definitions and the tools used across studies to measure important parameters such as quality of life or other patient-reported outcome measures (PROMs),[24] as illustrated in ►Table 3. This lack of uniformity hinders comparing and contrasting of results across different studies, and once more indicates an urgent need for creating and implementing standardized, reliable, and well-defined parameters and tools that are both patient-centric and disease-specific. Concurrently, it is essential to acknowledge that the NETS1HD study introduced a novel HSCR core outcome set through rigorous methods.[7] In contrast to this study, our approach not only centered on outcome measures but also encompassed a comprehensive analysis of treatment and diagnostic characteristics, as well as patients' baseline characteristics. We focused on identifying the most extensively studied parameters and did not consult a panel of experts for their ratings and definitions. Promoting the adoption of this core outcome set and similar initiatives and implementing these standardized datasets in both large and small pediatric surgical studies is crucial, as it will enhance relevance, minimizes bias, and facilitate future meta-analyses.

Standardization of research parameters is especially important in rare disease research, as the rare nature of the studied conditions often inherently complicates generating high-quality evidence, as we have confirmed with this review. Because of the demonstrated lack of high-quality evidence, current guidelines and clinical practices still predominantly hinge on expert opinions and consensus statements. Moreover, as opposed to the previously mentioned HSCR core outcome set, which was developed to standardize research, it is vital to acknowledge the current deficit in standardized measures for evaluating the quality of HSCR care and the absence of a common benchmarking system. In the modern healthcare landscape, there is a growing emphasis on the quality of provided care, and standardized measures are essential for precise benchmarking and the accurate interpretation of outcomes. Consequently, the development and implementation of standardized benchmarking through measuring predefined quality indicators could significantly enhance the overall quality of HSCR care. Current clinical guidelines and consensus statements and the list of treatment and care process characteristics and outcomes resulting from this systematic review may function as a starting point for a consensus method to develop quality indicator sets for such tools, involving both the international HSCR expert community and patient representatives. The European Reference Network for Inherited Congenital Anomalies and its registry (EPSA) have the potential to play a critical role in the more effective collection and analysis of comparative quality data, ultimately leading to the establishment of best practices and bridging the gap between short-term and long-term outcomes.[10]

By adhering to the PRISMA guidelines, we ensured a structured and transparent methodology. The extensive data extraction and categorization allowed for interpretation of the current most important parameters for clinical researchers in the field. This approach has markedly reduced the likelihood of overlooking crucial parameters, as we have included all parameters mentioned in at least 5% of the selected studies. Our findings emphasize the necessity for standardization of data collection, definitions, and use of clinical tools and instruments. By addressing a significant gap in HSCR research—namely, the lack of uniformity in parameter measurement and reporting methods—we not only bring to light the existing discrepancies but underscore the urgency for a cohesive approach in future studies. Additionally, the resulting list could lay the groundwork for determining which parameters are important for assessing the quality of HSCR care. Our research also has some limitations. First, as we pooled data from children and adults without providing a distinct analysis for each group, the findings are not age-specific. It is recommendable to study the functional changes HSCR patients undergo throughout their lifespans. Second, our research is limited by a specific time frame, focusing solely on studies published between 2015 and 2021. While this provides insight into recent clinical practices, it may overlook critical

insights from publications after 2021. Third, although we identified significant variations in definitions, the abundance of these definitions prevented us from exhaustively detailing each one.

As we are transitioning into prioritizing evidence-based practice and patient-centered outcomes, embracing a unified approach and adopting quality indicators are crucial. This approach will not only refine our understanding and management of HSCR but also fortify the foundations of pediatric surgical research, driving meaningful advancements in pediatric patient care.

Conclusion

This review highlights the substantial variation in the reporting of patient characteristics and outcomes in HSCR research, with 1,026 parameters identified in recent literature. It suggests an urgent need for adopting the newly developed core outcome set, and establishing a standardized core indicator set. The comprehensive list of studied parameters identified in our review can facilitate this process. Implementing standardized measurement and reporting systems is vital, promising enhanced interpretability of results and the potential for effective quality-of-care benchmarking across pediatric surgical centers in Europe. Looking ahead, the European Pediatric Surgery Audit (EPSA) emerges as a crucial entity to realize this vision, heralding a new era in pediatric surgical research and care.

Authors' Contributions

The study was designed by D.R., A.L.G., N.T., T.W., and R.M. H.W. Inclusion and exclusion criteria, assessment strategy, and the utilized Excel-based data extraction framework were developed by all authors. N.T. developed the search strategy, and N.T., D.R., and A.L.G. were responsible for screening, as well as data extraction and analyses. D.R. drafted the manuscript; all authors read, provided feedback, and approved the final manuscript.

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Conflict of Interest

None declared.

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