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REVIEW ARTICLE

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Scoping review of outcome measures in cleft care used in research and reports

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Abstract

Treatment outcome measures are critical in the decision making of best practices in the OFC field. OFC consortium working groups provided standardization of outcome measures based on previous treatment outcome studies. However, the implementation of such standardization in OFC centres worldwide is unknown. This study presented mapped outcome measures in cleft care using a structured review method complemented by quantitative overview of the relevant published research to provide initial guidelines for the implementation of treatment outcome standardization. A scoping review of the literature of treatment outcomes in cleft care following Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews guidelines was performed. The selected indexed paper in outcome measures mapped following the international consortium in standard set of outcome measures in cleft care. Three hundred and sixty-five articles were filtered. The most discussed domains of cleft care were dental and oral health, appearance and speech/communication. Overall, the majority of publications were produced in high-income countries. The current review indicates that there are inequalities of treatment outcome studies among the domain of cleft care. In addition, there are also inequalities of published articles from HIC versus LMIC in treatment outcomes. This information can be used to develop targeted interventions aimed at encouraging cleft centres worldwide to adapt standardized outcome measures.

KEYWORDS
orofacial cleft, review, treatment outcome

1 | INTRODUCTION

Treatment for cleft patients requires multidisciplinary and longitudinal care due to impairments caused by cleft conditions affecting feeding, breathing, speech, aesthetics, and psychological problems.[1,2] To ensure best practices and serve as an auditing process of the treatment itself as well as informing decision-making in cleft care, outcome measures are undoubtedly necessary. Depending on the types of outcome measures, clinicians, patients/family and or lay persons perspectives may be used.[3] Treatment outcome measures are vital for comparative effectiveness of particular interventions and treatment protocols.

A previous paper provided an in-depth review in the outcome measures of cleft care covering primary surgery and facial growth; speech assessment for cleft palate; nasolabial appearance; dental health assessments; secondary alveolar bone grafting; orthodontic treatment; and patient satisfaction and quality of life.[1] Another

study in 2017 suggested standardization by grouping the measures into eight domains including the instruments used and timepoints.[3] The International Consortium for Health Outcomes Measurement (ICHOM) set up a consensus on the outcomes that should be measured routinely as a standard part of cleft care covering diverse treatments and perspectives.[4]

The international standardization of treatment outcomes has been highlighted by several cleft research groups. Nevertheless, standardization of treatment outcomes was aggravated by the multidisciplinarity of treatment, diverse type of clefts, variability of methods available, differences of cleft team specialties and perspectives, as well as the unequal ability of all cleft centres to perform such outcome measures. Without standardized outcome measures which are agreed globally, data match and comparison from one cleft centre to another are hindered, resulting in the inability of practitioners to decide the gold standard in one field of cleft treatment.

As previous studies provided guidelines of standardized treatment outcomes in cleft care, it is necessary to relate it with the pre-existing outcome measures in the published literature as a preliminary study to predict what may come after such implementation of an international standard of outcome measures is enforced globally. Through scoping review and mapping the relevant indexed publications, the aims of this study are:

- to report the number of research studies conducted in the field of OFC reporting outcome measures.
- to provide suggestions for the implementation of standardization of treatment outcome measures in OFC, with special emphasis on low-, and low-middle-income countries.

2 | MATERIALS AND METHODS

2.1 | Protocol

The published scoping review protocol by Arksey and O'Malley (2005) and the Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) guidelines was set as the framework of this study.[5,6] Preliminary search was conducted using PubMed in designing the study protocol. The data extraction forms were built after the results of the preliminary search. The search was based on the Population, Concept, and Context (PCC) elements which were recommended by the Joanna Briggs Institute as being less restrictive inclusion criteria suitable for scoping reviews[7] (Table 1).

2.2 | Literature search

The search was performed using PubMed, LILACS and CINAHL. Two layers of search were employed, in which the first was orofacial clefts and its synonyms, and the second was outcome measures or treatment outcome (Table S1). No limitation on time in this protocol. The first level of article identification was executed by the first author in August 2021 and exported to spreadsheet using Citation management (Mendeley™).

2.3 | Eligibility criteria

The results of the search engine were screened for their eligibility based on the titles and abstracts. Articles of outcome measures of field other than cleft lip and palate were excluded. Relevant studies with inadequate information in the title and abstract to judge the eligibility were obtained in full text. The second level of eligibility-included screening was conducted by the research team composed of three investigators. Any disagreements were resolved by the first author. A hand search of the references of the included studies was not performed.

2.4 | Results extraction

The intended variables were extracted from the final included articles and tabulated in the data extraction form. This paper adapted variables from Allori et al.[3] in terms of the targeted variables which were sought from the full text articles such as the classification of treatment domain, sub domain, instruments, and perspective. This study also noted the instruments used in the selected articles when they were different than mentioned by Allori et al.[3]

3 | RESULTS

3.1 | Search and study selection

Following the search, all collected citations were exported into Microsoft Excel for data extraction and analysis. The PRISMA-ScR flowchart depicted the filtering process (Figure 1). The electronics databases search all combined yielding 956 articles. After duplicate articles were removed 782 papers were screened for eligibility. After careful assessment, 417 were excluded due to being animal studies

TABLE 1 Description of PCC elements.

Study question	What are the available pieces of evidence of treatment outcomes used in the orofacial cleft field in the world?
Population	Orofacial cleft cases; human participants, any age, any gender
Concept	Treatment outcomes used in the orofacial cleft field
Context	All settings considered; original research articles and case reports; mention the instruments used, either practitioners or patients perspectives

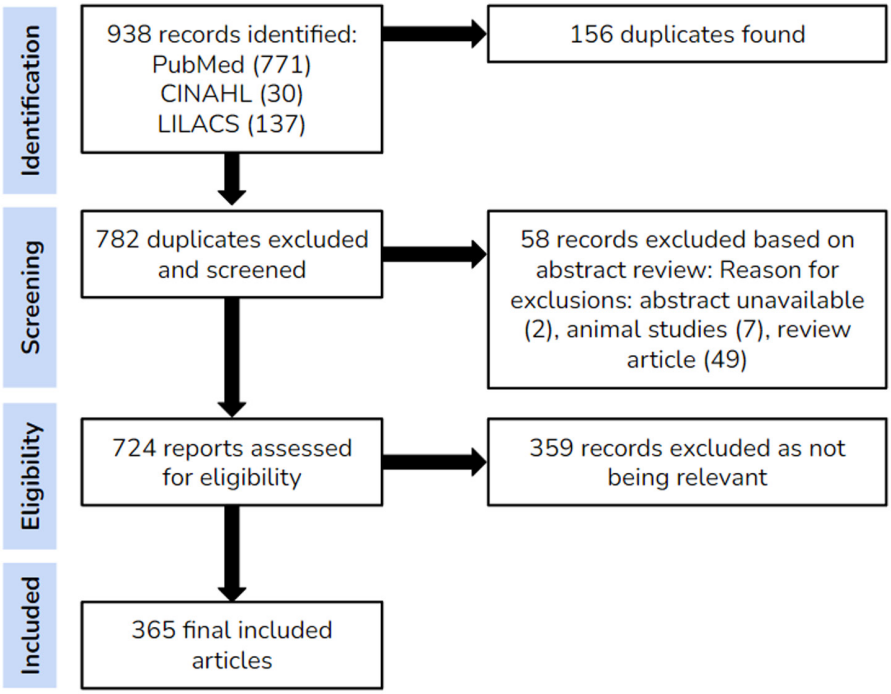

FIGURE 1 Scoping review flow chart of article filtration process adopted from PRISMA-ScR.[4]

(7), irrelevant topics (359), abstracts unavailable (2), and review (49) articles, resulting in 365 articles included for this study (Appendix S1).

3.2 | Outcome domains of included studies

This study adapted classification of treatment domains formulated by the consensus of international multidisciplinary experts in the cleft field in which presented a standard set of outcomes in cleft care grouped into eight major outcome domains and 22 subdomains.[3] The 367 included studies were allocated to the suitable domains and subdomains (Table 2).

The top three domains of outcome measures presented in the existing literature were dental and oral health, appearance and speech/communication. Eating and drinking domain apparently received little attention. Within the top three domains, not all subdomains presented an equal number of publications. Occlusions were the most discussed in the dental and oral health domain. Nasolabial appearance and facial profile were most explored by researchers in the appearance domain. Velopharyngeal competence and overall speech were the most frequently observed in speech/communication domain. Only a few subdomains such as growth chart, dmft and DMFT scores, and COHIP Oral Symptoms subscale, which utilized instruments as per the Allori et al,[3] and the rest of the subdomains employed both the recommended instruments and others.

Allori et al. has set the intended perspectives as data source for each domain outcome measures procedure whether from a single perspective (clinician or patient or family) or combination. This study noted that most publications adhered to the suggested perspectives. Only 16 out of 365 publications used different perspectives compared to the suggested perspectives by Allori et al[3] (Table 3). These differences were found in breathing (clinician), appearance (lay person), psychosocial development (clinician), and burden of care (patient). The speech/communication domain comprises several outcomes which are measured by clinicians, patients, families. What is interesting about the data in Table 3 is that 81 out of 86 published papers in the speech/communication domain used the clinician perspective while only four used the patient perspective and only one used the family perspective. This study also marked 20 articles with multiple data sources in one publication, whereas the rest of 345 publications only used a single perspective.

The year of publications of the final included articles ranged from 1992 to 2021, in which the number of articles per year ranged from one to 36 per year. The graph shows that there has been a gradual increase in the number of publications (Figure 2). The highest volume of articles was 36 in the year 2021. It is important to bear in mind that the search protocol was executed in August 2021, so the number of publications in 2021 until December may actually have risen. Another important finding was that the top six contributors of outcome measure research were USA (77), UK (36), Japan (31), Sweden (24), Netherlands (16) and Brazil (15) who yielded 54.22% of total included articles, whereas 16 articles were multi- setting studies. This study also noted the instruments employed in outcome measures. Over two-thirds of the publications (86%) utilized different instruments than advised by the standard set of treatment outcomes.

4 | DISCUSSION

4.1 | Study limitations

This study opted for a scoping review in study design, in which the intended results are to provide an overview of the existing relevant literature, to detect patterns in publications, and to identify gaps,

TABLE 2 Profile of published literature of treatment outcome in OFC based on 8 domains, 22 sub-domains and instruments.

Domains[a]	Sub-domains[a]	Instruments[a]	#publications
Eating and drinking (7)	Body weight	Growth chart	2
	Change in weight centile	Growth chart	0
	Eating and drinking	CLEFT-Q eating-and-drinking subscale	5
Dental and oral health (121)	Dental health	dmft and DMFT scores	14
	Oral health	COHIP oral symptoms subscale	5
	Occlusion	Overjet assessment (GOSLON)	100
		Lateral cephalogram	
	Mastication	CLEFT-Q eating-and-drinking subscale	2
Speech/communication (86)	Intelligibility	Intelligibility-in-context scale	5
	Articulation	Percent consonants correct scale	6
	Velopharyngeal competence	VPC graded rating scale	33
	Overall speech	CLEFT-Q speech-and-speaking subscale	42
	Documentation	Standardized speech and language sample	0
Otologic health (14)	Hearing	Puretone average	4
	Otologic health	Otologic health screening questions	10
Breathing (12)	Nasal breathing	NOSE questionnaire	12
Appearance (98)	Nasolabial appearance	CLEFT-Q face subscale	61
	Facial profile	CLEFT-Q jaw subscale	31
	Smile	CLEFT-Q dental subscale	2
	Documentation	Standardized series of facial photographs	4
Psychosocial development (16)	Sociometrics	CLEFT-Q social Life subscale	5
		CLEFT-Q school Life subscale	
	Psychometrics	CLEFT-Q feelings subscale	11
Burden of care (11)	Total number of interventions requiring anaesthesia	Medical record	11
Total			365

[a]Allori et al., (2017).[3]

but not to answer specific questions nor provide information that such outcome measure is better than the others.

Embase was not utilized as one of the databases due to its unavailability at our institution. Considering the extensive search in PubMed, the authors concluded that conducting a hand search would not significantly improve the identification of relevant studies. However, it is important to emphasize that the review methodology has been thoroughly documented, and rigorous search strategies and inclusion criteria were implemented to minimize the possibility of excluding pertinent studies.

4.2 | Previous studies in outcome measures

The number of publications in each domain was not equally distributed. In addition, there were a handful of unlisted instruments used in the published papers, varied from what was set by the international consortium.[3] There are several possible explanations for these results. First, there were perhaps some domains of cleft treatment which were unavailable in some cleft centres therefore outcome measures were unable to be performed. Secondly, it is more likely that some cleft centres, although performing a wide range of cleft care, did not have the necessary skilled researchers to conduct such intricate study. Approaches which should be executed are to encourage cleft centres to provide holistic cleft care covering all domains, while also giving research capacity building for those centres which are in need.

As the standard set by Allori et al. was established in 2017, this study included articles for scoping review since 1992. Therefore, non-compliance research articles regarding the perspective used in outcome measure to the standard set prior to 2017 can only be discussed for descriptive purposes. The majority of the articles matched with the recommended perspectives by Allori et al.[3] These minor varieties can be managed by dissemination of the standard set agreed by the international consortium in cleft to all cleft centres in the world by also involving the WHO and governments. The finding

regarding the quantity of research grouped by perspective (Table 3) has important implications for developing future research in perspectives and domains less explored such as: dental and oral health using patient perspective; speech/communication using patient and family perspectives; otologic health using family perspective; and breathing using patient perspective.

The increasing trend of indexed literature in treatment outcomes is definitely a good message that more and more cleft centres are aware of the importance of outcome measures in cleft care that they perform as a way of auditing care in order to prepare for better treatment for the patient. In order to support implementation of standardization of outcome measures, these increasing numbers of publications should be followed by the matching instruments, domains, timepoint(s), and perspective(s) of the paper with the global standard.

The top five countries who provided research in outcome measures were indeed high-income countries (HIC). We believe joint research partnerships are essential to allow researchers accessing a wider range of resources such as facilities, funding and expertise's. In addition, research collaboration promotes network expansion as well as increasing impact of the research by reaching a wider audience. Multi-setting studies are also encouraged in treatment outcomes as it enables comparative observation between cleft centres and learns best practice from one to another.

Previous study exhibited great inequalities in HIC versus LMIC peer-reviewed publications in cleft field over the last 10 years.[8] Like other areas of medical research, cleft research is also characterized by inequalities between HIC and LMIC due to differences in various aspects but mainly in research capacity and infrastructure of comprehensive data of birth defect surveillance systems. This fact highlighted the tremendous needs of research capacity building for Indonesia and other LMICs in general.

Two previous studies in HIC settings performed analysis of implementation of standardized outcome measures in cleft care.[9,10] These studies found that such implementations were challenged by team-specific constraints such as availability of specialized clinicians and disparity of interpretation between staff. Furthermore, these studies mentioned that adopting such a standard set requires significant cost which may be even more problematic for LMIC cleft centres. We suggest collaborative workshops by HIC trainers with standard set implementation experience to train a few leading cleft centres in LMIC for the first batch and may be followed by the rest of cleft centres after the first workshop achieve the desired results. However, forming an optimal dataset to initiate a treatment outcome measure, which should be done in the first place, can become more complicated in low-resource settings.

TABLE 3 Perspectives gathered from as the data source for outcome measures.

Outcome domains	Data source	#publications
Eating and drinking	Clinician	7
Dental and oral health	Clinician	123
	Patients	1
Speech/Communication	Clinician	81
	Patients	4
	Family	1
Otologic health	Clinician	13
	Family	1
Breathing	Clinician[a]	11
	Patients	1
Appearance	Clinician	84
	Lay person[a]	2
	Patients	9
Psychosocial development	Clinician[a]	2
	Patients	14
Burden of care	Clinician	10
	Patients[a]	1

[a]Marked the varied data source with the Allori et al[3] recommendation.

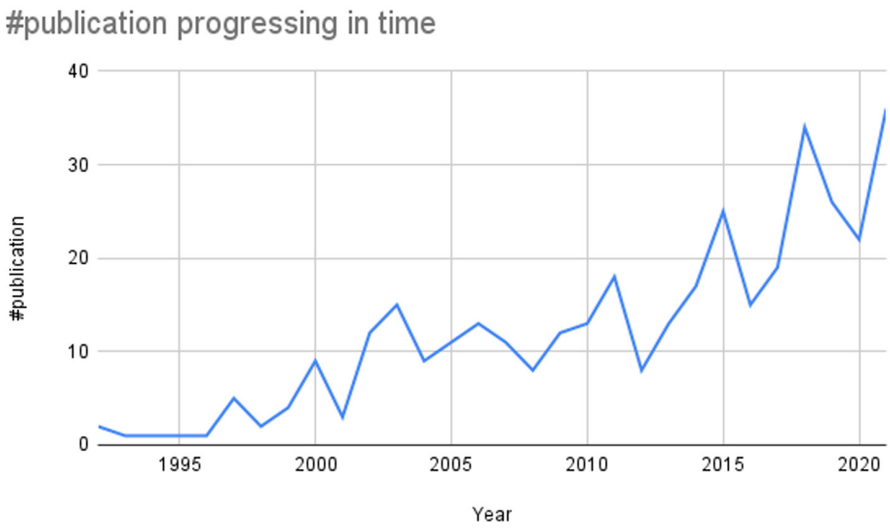

FIGURE 2 Frequencies of indexed publications of outcome measures in cleft care between 1992 and 2021.

Consensus on a set of core outcome measures or minimum dataset for CLP outcomes is a highly desirable objective as this would allow inter-centre comparisons which underpins quality improvement research. In addition to the ERN CRANIO interest in having core outcomes ERN CRANIO registry and outcome measurement (ern-cranio.eu), two recent publications, on this topic, Allori et al, 2017[3] and Mossey et al., 2023[11] both reviewed the current situation with CLP core outcomes and have provided further momentum on gaining consensus. These recent publications reveal the complexities but also areas of convergence and of agreement on a core data set for all disciplines involved in cleft care.

It is important to acknowledge that the discussions around adoption of core outcome measures in the various CLP MDT disciplines has come from the Global North (high-income countries, HICs). With respect to LMICs, this requires co-development to ensure it is implementable. This not only encourages alignment with HIC core outcomes and would encourage the adoption of a multidisciplinary approach in LMICs, but would also be a significant step towards universal health coverage (UHC) as part of the sustainable development goals (SDGs).

4.3 | Future research

Several consortium studies in outcome measures published in the literature were performed in high-income countries such as Americleft, Eurocleft, Scandcleft and ERN CRANIO studies. These studies set up good practices in treatment outcomes in cleft care. However, high-volume OFC cases occur in LMICs and yet low count of treatment outcome studies from LMICs have been produced. Although pooled in the same income category, not all LMIC research capacity is at the same level. Three LMIC (China, Brazil, and India) managed to produce a high volume of cleft research recorded in the indexed database.[7] Moreover, several LMIC have established national birth defect surveillance systems.[12–14] These facts illustrate that there are differences in research capacity and infrastructures among LMIC.

These findings suggest challenges in regards to treatment outcome standardization. Greater efforts are needed to encourage cleft centres to perform treatment outcome assessment as per international guidelines. A reasonable approach to tackle this issue could be a systematic dissemination of the treatment outcome standard. The present study lays the groundwork for future implementation of the standard set in outcome measures in cleft care with consideration of cleft centres with low-resource settings.

The standard set of outcome measures across all domains of cleft care encourages consensus which is geared towards quality improvement, research and ultimately health care reform. This study was able to highlight the inequality of research and publications in LMICs. Identification of the volume of literature on outcome measures coming from low-resource settings is scarce and there is a need to focus attention on those parts of the world where this is lacking and where future efforts will be required to achieve the SDG and UHC. Based on this study, it is imperative to gather researchers from low-resource settings to collect information on how to reduce inequalities through research capacity building and frugal innovations, tools and technologies in LMIC.

5 | CONCLUSIONS

The purpose of the current study was to map the existing literature on outcome measures in cleft care. In general, it seems that there is lots of interest in OFC across the world with copious research activity. This study has identified the greatest number of publications from the US and UK but fewer from LMICs. The research has also shown that there was notable variability in the measurement of OFC treatment outcomes compared to the established standard set of outcome measures. These variations are barriers to inter-centre comparisons, audit and research. The most frequent outcome domains focus on oral health, function and appearance. The findings of this research provide useful data to inform the Global Burden of Disease (GBD) discussions. Standardization would enhance the opportunities to improve quality of care. Consensus on outcome measures is essential in aspiring to SDG3/Universal Health Coverage.

AUTHOR CONTRIBUTIONS

Study design: ES (1), PM. Data collection: ES (1). Data analysis: ES (1), ES (2), PM. Manuscript preparation: ES (1), ES (2), PM. All authors contributed to critical revision of the manuscript. All authors read and approved the final manuscript.

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CONFLICT OF INTEREST STATEMENT

There is no conflict of interest in this project.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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