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A quality appraisal of economic evaluations of community water fluoridation: A systematic review

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Objectives: To critically appraise the methodological conduct and reporting quality of economic evaluations (EE) of community water fluoridation (CWF). ***Methods***: A systematic literature search was conducted in general databases and specialist directories of the economic literature. The Consensus on Health Economic Criteria list (CHEC) appraised the methodological quality while the Consolidated Health Economic Evaluation Reporting Standards (CHEERS) assessed the reporting quality of included studies. ***Results***: A total of 1,138 records were identified, of which 18 met the inclusion criteria. Cost analysis emerged as the most prevalent type of EE, though a growing trend towards conducting full EEs is observed. CHEC revealed the items most frequently unfulfilled were the study design, measurement and valuation of costs and outcomes, while CHEERS also identified reporting deficiencies in these aspects. Furthermore, the review highlights subtleties in methodological aspects that may not be discerned by CHEC, such as the estimation of the impact of fluoridation and the inclusion of treatment savings within cost estimates. ***Conclusions***: While numerous studies were conducted before publication of these assessment instruments, this review reveals that a noteworthy subset of studies exhibited good methodological conduct and reporting quality. There has been a steady improvement in the methodological and reporting quality over time, with recently published EEs largely adhering to best practice guidelines. The evidence presented will assist policymakers in leveraging the available evidence effectively to inform resource allocation decisions. It may also serve as a resource for researchers to enhance the methodological and reporting standards of future EEs of CWF.

Keywords*: systematic review, oral health, fluoridation, cost-effectiveness analysis, cost-benefit analysis, costs and cost analysis*

Introduction

Oral diseases pose a major health and economic burden on individuals, communities, and the wider society (WHO, 2023). The 2019 Global Burden of Disease study shows that oral diseases are the most widespread non-communicable diseases affecting 3.47 billion people worldwide with caries (decay) of the permanent teeth being the most prevalent of these. It is estimated that over 2 billion people had decay in permanent teeth and 520 million children experienced decay in the deciduous dentition in 2019 (Murray *et al*., 2020). The global economic impact of dental diseases was estimated at $554.41 billion in 2015, of which 65.5% was attributed to treatment costs (Righolt *et al*., 2018). A comparison of expenditures amongst EU states, revealed that the direct cost of dental diseases (€90 billion) ranked third behind cardiovascular diseases (€111 billion) and diabetes (€119 billion) in 2015 (Peres *et al*., 2019).

Dental decay is preventable and may be reversible if detected and addressed in the early stages (Watt *et al*., 2019). The current system of oral healthcare delivery is focused on clinical intervention, resulting in a cycle of repeat restorations with increasing complexity and costs over a lifetime. This form of provision, often isolated from general healthcare services, is predominantly demand-led and consequently children, low-income families, and marginalised groups are generally underserved and disproportionally affected (Watt *et al*., 2019). In 2021, the World Health Assembly, approved a resolution on oral health (OH) promoting a re-orientation of services towards a preventive model of care to achieve improved and equitable OH for all (World Health Organization, 2021, 2022; Eaton *et al*., 2023).

Community water fluoridation (CWF), the controlled addition of fluoride to the public water system, is an approved preventive OH intervention to reduce the prevalence and severity of decay. As CWF does not require active participation, it can equitably reduce decay across all socio-economic groups (Harding and O'Mullane, 2013). Since its introduction to Grand Rapids, Michigan in 1945, it has been adopted by 26 countries as the bedrock of their preventive OH strategy (Mariño and Zaror, 2020; Centers for Disease Control and Prevention, 2021). CWF is recognised as one of the ten great public health promotion measures of the 20th Century (Centers for Disease Control and Prevention, 1999).

While numerous studies have confirmed the safety and effectiveness of CWF (Truman *et al*., 2002; Iheozor-Ejiofor *et al*., 2015; National Health and Medical Research Council, 2017; Lambe *et al*., 2022), the recent research, conducted in an environment with multiple fluoride sources, demonstrates a continued benefit. However it also indicates a reduction in relative disease levels between populations with and without exposure to CWF (Jackson *et al*., 1985; Murray, 1993; Whelton *et al*., 2006a, 2007) along with an increased prevalence in dental fluorosis (Clark, 1994; Whelton *et al*., 2006b; Chankanka *et al*., 2010; Browne, 2012). The cost-effectiveness of CWF has also been confirmed and continues to yield positive results (Akehurst and Sanderson, 1993; Ran *et al*., 2016; Mariño and Zaror, 2020). Although studies

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appraising previous economic evaluations (EEs) of OH programmes have highlighted methodological weaknesses (White *et al*., 1989; Källestål *et al*., 2003; Tonmukayakul *et al*., 2015; Eow *et al*., 2019; Rogers *et al*., 2019; Mariño and Zaror, 2020; Nguyen *et al*., 2023) and concerns regarding the reporting quality of studies (Mariño *et al*., 2013, 2020; Hettiarachchi *et al*., 2018; Rogers *et al*., 2019; Anopa *et al*., 2020). Moreover, a number of these reviews incorrectly classified cost-analyses of CWF as full EEs, raising concerns about the credibility of their findings (Mariño and Zaror, 2020; Nguyen *et al*., 2023). This is of concern, especially in the climate of pressures on public health systems, where government stakeholders are increasingly using EEs to inform policy and guide resource allocation. Furthermore, the users of these evaluations are frequently not health-economists and thus, are ill equipped to assess the methodological validity and reliability of these studies. Considering the emergent shift towards a prevention-based approach to the management of OH (Pitts and Zero, 2016; Vernazza *et al*., 2021; World Health Organization, 2021), and the increasing role of EE to support decision making (Watt *et al*., 2019; Anopa *et al*., 2020), it is opportune to conduct a comprehensive assessment of the methodological and reporting quality in EEs of CWF. Our intention is to maximize the usefulness of the existing economic evidence to inform decisions about whether to implement or continue CWF programmes. Additionally, the review will serve as a reference for future research, contributing to the improvement and refinement of methodological and reporting standards for EEs. The review will also examine whether the reduction in relative disease levels between fluoridated and non-fluoridated populations has affected the cost-effectiveness of CWF.

Methods

This review, guided by principles of conducting reviews of the EE evidence (Carande-Kulis *et al*., 2000; Gomersall *et al*., 2015; Thielen *et al*., 2016; van Mastrigt *et al*., 2016; Wijnen *et al*., 2016; Aluko *et al*., 2021), used the preferred reporting items for systematic reviews and meta-analyses (PRISMA) statement (Page *et al*., 2021) to facilitate reporting.

The PICOS (population, interventions, comparators, outcomes, and study type) framework (Amir-Behghadami and Janati, 2020) supported the inclusion criteria. The review considered full and partial EEs that evaluated CWF for populations in countries with a very high human development index (HDI) (≥ 0.9) (UNDP, 2022). A full list of the inclusion/exclusion criteria is presented in the online appendix Table A1 (available at; https://www.ucc.ie/en/media/academic/centreforpolicystudies/AppendixTableA1_EligibilityCriteria.pdf).

General databases and specialist directories of health economics literature were searched between the 9th and 19th of January 2023. A snowball search also used the bibliographies of eligible publications to identify relevant studies. Details of the general databases consulted are available in the online appendix Table A2 (available at: https://www.ucc.ie/en/media/academic/centreforpolicystudies/AppendixTableA2_DetailsofdatabasesconsultedpdfDetailsofdatabasesconsulted.pdf).

To develop the search strategy, an initial Medline search identified words in the titles and abstracts of identified EEs and their index terms. The final search used a broad set of terms based on text words and standardised subject terms for two elements of the PICOS framework ((i) intervention and (ii) type of study) (Higgins *et al*., 2019). The initial strategy was translated, reviewed, and approved for each database in accordance with PRESS guidelines (McGowan *et al*., 2016) by a subject librarian (DOD) and the research team that comprised three health economists (JC, SM and NW) and two dentists (MH and HW) with systematic review experience. The general search strategy is available in the online appendix Figure A1 (https://www.ucc.ie/en/media/academic/centreforpolicystudies/AppendixFigureA1_Generalsearchstrategy.pdf).

All retrieved records were imported into Rayyan (Ouzzani *et al*., 2016). Two reviewers (JC, SM) independently assessed the titles and abstracts against the inclusion criteria. Any disagreements were resolved through discussion with a third reviewer (NW). The full texts of all potentially relevant articles were retrieved and screened independently by the reviewers (JC, SM). To identify whether the decrease in relative disease levels between populations with and without CWF was evident in the results of the EEs, studies were included if there was at least one other qualifying study that had conducted an EE in the same jurisdiction for a different period of analysis (≥ 8 years).

A data extraction form, guided by previous guidelines on the methodological quality and reporting standards for EEs (Drummond and Jefferson, 1996; Evers *et al*., 2005; Guide to Community Preventive Services, 2010; Wijnen *et al*., 2016; Husereau *et al*., 2022) was designed to extract data systematically from included studies. Two instruments were employed to evaluate the methodological rigour and reporting quality of the included studies. The Consensus on Health Economic Criteria (CHEC)-list (Evers *et al*., 2005), providing a core set of criteria for assessing the methodological quality of EEs, was used to determine the methodological quality of the studies. The updated Consolidated Health Economic Evaluation Reporting Standards (CHEERS) statement (Husereau *et al*., 2022) served as a framework for evaluating the reporting quality of studies. The methodological and reporting quality of the studies was appraised independently by two reviewers (JC, SM). In addition, each item of the CHEC and CHEERS checklist was scored as per Anopa et al. (2020) to represent the quality of each study and highlight any recurring shortcomings. Meta-analysis was not undertaken due to the heterogeneity of study designs and methods. The methodological and reporting quality of the main study elements are discussed within the results section and are also presented both graphically and in tabular format in the online appendix.

Results

A total of 1,138 records were identified, of which, 18 met the inclusion criteria. The PRISMA flow diagram (Figure 1) shows the number of records that were included and excluded throughout the different phases of the review.

Of the included studies, nine were full EEs in the form of cost-utility (CUA, n=5) and cost-effectiveness analyses (CEA, n=4), and nine studies were partial EEs in the form of cost-analyses. The full EEs covered five countries (New Zealand, Australia, Canada, England, and USA), published between 1984 through to 2022, while the cost-analyses

considered four jurisdictions (USA, Australia, Canada, and England) published between 1976 and 2016. Seventeen EEs compared the costs, or the costs and consequences, associated with CWF to the scenario of no CWF intervention in either the total population (full n=3 (Wright *et al*., 2001; Fyfe *et al*., 2015; Moore *et al*., 2017), partial n=6 (Dowell, 1976; Griffin *et al*., 2001; O'Connell *et al*., 2005, 2016; Campain *et al*., 2010; Tchouaket *et al*., 2013)) or a child only population (full n=5 (Niessen and Douglass, 1984; O'Keefe, 1994; Ciketic *et al*., 2010; Cobiac and Vos, 2012; Goodwin *et al*., 2022), partial n=3 (Nelson and Swint, 1976; Carr *et al*., 1980; Doessel, 1985)). The remaining EE performed a CEA of CWF for child communities with differing levels of underlying decay (Birch, 1990). Of note with regard to CHEC, three studies were presented as full EEs despite only conducting a partial analysis (Nelson and Swint, 1976; Doessel, 1985; Tchouaket *et al*., 2013) and two cost analyses failed to state the type of EE performed (Dowell, 1976; Carr *et al*., 1980). In addition, six studies didn't explicitly label their study as an EE (Dowell, 1976; O'Connell *et al*., 2005, 2016; Campain *et al*., 2010; Tchouaket *et al*., 2013; Moore *et al*., 2017) which does not align with CHEERS guidance on this study aspect.

Table 1 summarises study characteristics and methodological aspects of the studies reviewed. Six EEs adopted the societal perspective (full n=1 (Fyfe *et al*., 2015), partial n=5 (Griffin *et al*., 2001; O'Connell *et al*., 2005, 2016; Campain *et al*., 2010; Tchouaket *et al*., 2013) while the remaining evaluations took the health/public payer's perspective. It is important to note that in three studies, the chosen perspective did not align with the analysis performed and thus they fail to meet CHEC criteria for the study item (full n=1 (Wright *et al*., 2001), partial n=2(Nelson and Swint, 1976; Doessel, 1985)). While in the context of the CHEERS guidelines, five EEs had not specified a study perspective (full n =3 (Niessen and Douglass, 1984; Birch, 1990; Ciketic *et al*., 2010), partial n= 2 (Dowell, 1976; Carr *et al*., 1980)) and a further three studies failed to provide a rationale supporting the viewpoint adopted (full n=2 (Fyfe *et al*., 2015; Moore *et al*., 2017), partial n=1 (Campain *et al*., 2010).

Six evaluations examined either the costs, or costs and consequences, associated with CWF exposure for one year (full n=1 (Fyfe *et al*., 2015), partial n=5 (Griffin *et al*., 2001; O'Connell *et al*., 2005, 2016; Campain *et al*., 2010; Tchouaket *et al*., 2013)). The timeframe of eleven (full n=7, partial n=4) EEs varied from 5 (Goodwin *et al*., 2022) to 35-years (Doessel, 1985). However, the period covered by one CUA was not reported (Ciketic *et al*., 2010), and a further two studies (full n=1 (Fyfe *et al*., 2015), partial n=1 (Tchouaket *et al*., 2013)), failed to justify their timeframe of analysis. Hence, these studies do not meet the requisite methodological and reporting standard for this study component.

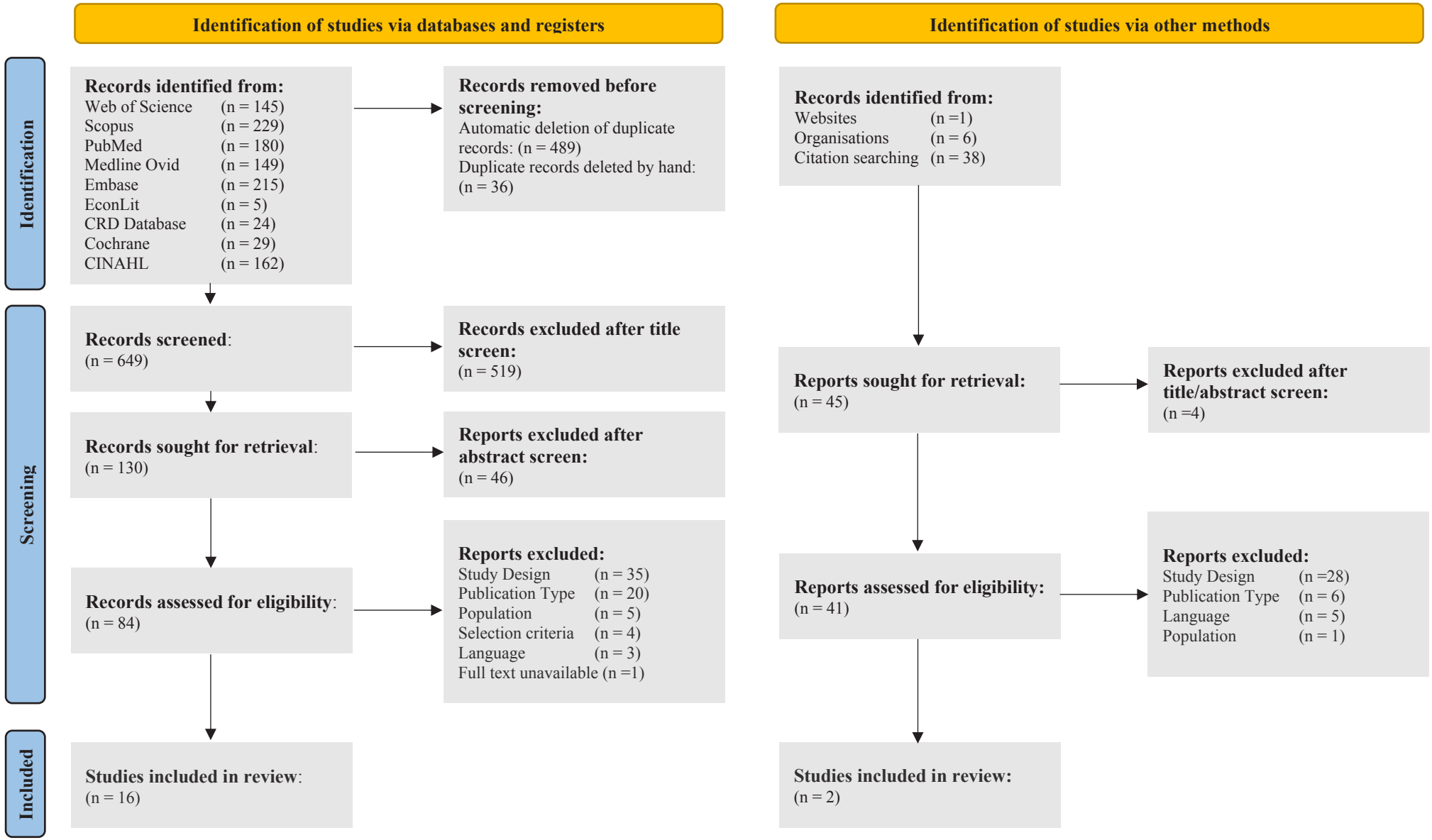
All the full EEs effectively reported their selected primary health outcomes. In the context of CHEC, only five studies (Wright *et al*., 2001; Ciketic *et al*., 2010; Fyfe *et al*., 2015; Moore *et al*., 2017; Goodwin *et al*., 2022) identified fluorosis as an adverse outcome associated with CWF. However, these studies did not include fluorosis in their analyses. Four CUAs estimated outcomes using a generic measure of effect in the form of quality-adjusted life years (QALYs) (Moore *et al*., 2017; Goodwin *et al*., 2022), or disability-adjusted life years (DALYs) (Ciketic *et al*., 2010; Cobiac and Vos, 2012), while one CUA used a measure specific to OH outcomes, the quality-adjusted tooth years (QATYs) (O'Keefe, 1994). The CEAs considered either the decay-missing-filled index for teeth (dmft/DMFT) (Birch, 1990; Fyfe *et al*., 2015) or tooth surfaces (dmfs/DMFS) (Niessen and Douglass, 1984; Wright *et al*., 2001).

All the full EEs reported how outcomes were measured, albeit with varying levels of detail. With regard to CHEC, six of the nine studies applied an effect of CWF based on previous research to the decay outcomes of populations without CWF exposure to estimate the associated health benefits (Niessen and Douglass, 1984; O'Keefe, 1994; Wright *et al*., 2001; Ciketic *et al*., 2010; Cobiac and Vos, 2012; Moore *et al*., 2017). However, the data informing the effect size in three studies (Wright *et al*., 2001; Ciketic *et al*., 2010; Cobiac and Vos, 2012) was more than a decade old, and consequently these studies do not meet CHEC criteria for this element. In contrast, two studies measured benefits using differences in the decay outcomes between populations with and without CWF exposure (Birch, 1990; Fyfe *et al*., 2015). The remaining CUA (Goodwin *et al*., 2022), used a validated instrument, the Child Health Utility 9D questionnaire (CHU9D) (Stevens, 2011), to measure the differences in health states between exposed and unexposed study populations, though concerns have been raised regarding the suitability of the CHU9D to assess health related quality of life (HRQoL) related to OH (Foster Page *et al*., 2015; Rogers *et al*., 2019).

Given the absence of a preference-based measure of decay, a decision about whether the CUAs adhere to CHEC guidelines regarding the methods used to value health outcomes is difficult. The five CUAs assigned utility weights informed by previous research (Ciketic *et al*., 2010; Cobiac and Vos, 2012; Goodwin *et al*., 2022) or expert opinion (O'Keefe, 1994; Moore *et al*., 2017) to health state information as defined by levels of decay or collected by the study itself. It is important to highlight that in two studies, the preference weights used to value health states were not appropriate for the study population (Ciketic *et al*., 2010; Goodwin *et al*., 2022), indicating a deviation from the CHEC standard. Two other CUAs did not clearly describe how the underlying components of the outcome measure contributed to the health outcome (O'Keefe, 1994; Ciketic *et al*., 2010). Consequently, neither study conformed to CHEC or CHEERS guidelines for this aspect of the research.

We evaluated the quality dimensions of both the positive (CWF provision costs, fluorosis etc.) and the negative costs (treatment savings, productivity losses avoided etc.) associated with CWF. However, the quality assessment instruments were applied to the positive costs only.

While all studies identified and reported the direct costs associated with CWF provision, only five EEs, providing detail on the methods employed to measure and value CWF resource items (full n=3 (Wright *et al*., 2001; Moore *et al*., 2017; Goodwin *et al*., 2022), partial n=2 (Tchouaket *et al*., 2013; O'Connell *et al*., 2016)) adhered to both CHEC and CHEERS standards. The remainder explained how they either adjusted (full n=3 (Niessen and Douglass, 1984; O'Keefe, 1994; Fyfe *et al*., 2015), partial n=2 (Griffin *et al*., 2001; O'Connell *et al*., 2005)), or, assumed (full n=3



Source: Page *et al.,* (2021)

Figure 1. *PRISMA 2020 flow diagram for new systematic reviews which includes searches of databases, registers, and other sources.*

Table 1. Characteristics of included studies.

Characteristic	*No of studies (n=18)*	*Type of EE*
Type of EE		
CUA	5	
CEA	4	
Cost analysis*	9	
Unreported study type	*2*	2 partial
Mislabelled study design	*3*	3 partial
Year of publication		
1975-1979	2	2 partial
1980-1989	3	1 full, 2 partial
1990-1999	2	2 full
2000-2009	3	1 full, 2 partial
2010-2019	7	4 full, 3 partial
2020 to date	1	1 full
Study Country		
Australia	5	2 full, 3 partial
USA	5	1 full, 4 partial
England	3	2 full, 1 partial
New Zealand	3	3 full
Canada	2	1 full, 1 partial
Population		
Children	9	6 full, 3 partial
Children and adults	9	3 full, 6 partial
Type of Intervention		
Community Water Fluoridation (CWF)	17	8 full, 9 partial
CWF and high levels of underlying caries	1	1 full
Target CWF level reported	*12*	8 full, 4 partial
Fluoride chemical reported	*7*	5 full, 2 partial
Comparator		
No CWF Intervention	17	8 full, 9 partial
CWF and high levels of underlying caries	1	1 full
Underlying natural water fluoride level reported	*4*	2 full, 2 partial
Study Perspective		
Societal perspective	6	1 full, 5 partial
Public payers perspective*	12	8 full, 4 partial
Unreported study perspective	*5*	3 full, 2 partial
Incorrect perspective stated	*3*	1 full, 2 partial
Analytical Timeframe		
1 year	6	1 full, 5 partial
> 1 year	12	8 full, 4 partial
Justification for analytical timeframe		
Previous research	5	1 full, 4 partial
Useful life of capital equipment	5	4 full, 1 partial
Study period	2	1 full, 1 partial
Public spending code	1	1 partial
Effects of discounting	2	2 partial
Not stated	3	3 full
Full EE outcome measures (n=9)		
QALYs gained	2	
DALY losses prevented	2	
QATYs gained	1	
DMFT/dmft prevented	2	
DFS prevented	1	
Carious surfaces prevented	1	

Table 1 continued overleaf.....

Table 1. Characteristics of included studies continued.

Health outcome measure to determine resources saved		
DMFT/dmft prevented	5	3 full, 2 partial
DMFT/dft prevented	1	1 partial
DMFT prevented	1	1 partial
DMFS/dmfs prevented	1	1 partial
DFS/dmfs prevented	1	1 full
DMFS prevented	1	1 partial
DFS prevented	1	1 partial
dmfs prevented	1	1 full
Carious surfaces prevented	1	1 full
Not applicable	5	3 full, 2 partial
Measurement of resources saved		
Initial restoration	8	5 full, 3 partial
Initial and replacement restorations	7	2 full, 5 partial
Assumed reduction in treatment costs	2	1 full, 1 partial
Not applicable	1	1 full
Sensitivity analyses		
Deterministic S/A	11	6 full, 5 partial
Probabilistic S/A	5	3 full, 2 partial
No S/A	2	2 partial

(Birch, 1990; Ciketic *et al*., 2010; Cobiac and Vos, 2012), partial n=5 (Dowell, 1976; Nelson and Swint, 1976; Carr *et al*., 1980; Doessel, 1985; Campain *et al*., 2010)) these costs from previous research. Aside from the five full EEs that identified fluorosis as an adverse outcome, only three partial evaluations acknowledged the costs associated with treating fluorosis (Griffin *et al*., 2001; O'Connell *et al*., 2005, 2016). However, like the full EEs, the partial evaluations excluded these costs from their analyses.

All but one study (Birch, 1990) included the expected treatment savings associated with the reduced need for treatment due to CWF within their cost calculation. These studies reported how they identified, measured, and valued the treatment costs prevented in accordance with CHEERS criteria. In fifteen studies, the treatment costs averted were estimated using either differences in the decay outcomes (full n=6 (Niessen and Douglass, 1984; Wright *et al*., 2001; Ciketic *et al*., 2010; Cobiac and Vos, 2012; Fyfe *et al*., 2015; Moore *et al*., 2017), partial n=7 (Nelson and Swint, 1976; Doessel, 1985; Griffin *et al*., 2001; O'Connell *et al*., 2005, 2016; Campain *et al*., 2010; Tchouaket *et al*., 2013)), or the treatment requirements (full n=1 (Goodwin *et al*., 2022), partial n=1(Carr *et al*., 1980)) between populations with and without exposure to CWF. Eight studies considered initial treatments while seven studies (full n=2 (Wright *et al*., 2001; Moore *et al*., 2017), partial n=5 (Griffin *et al*., 2001; O'Connell *et al*., 2005, 2016; Campain *et al*., 2010; Tchouaket *et al*., 2013)) also included the replacement restorations to maintain the decay over a lifetime. Studies conducted from the societal perspective also incorporated the costs associated with obtaining treatment (Griffin *et al*., 2001; O'Connell *et al*., 2005, 2016; Campain *et al*., 2010; Tchouaket *et al*., 2013; Fyfe *et al*., 2015). Savings were valued using national cost data. The two remaining studies applied an effect of CWF to the treatment costs of populations without CWF exposure to measure and value the treatment savings associated with CWF (full n=1 (O'Keefe, 1994), partial n=1 (Dowell, 1976)). Like the limitations of some full EEs regarding the measurement of the health benefits, several partial EEs also relied on an effect of CWF derived from older studies to estimate the decay prevented and the subsequent treatment savings associated with the intervention (Dowell, 1976; Nelson and Swint, 1976; Griffin *et al*., 2001; O'Connell *et al*., 2005).

Tables A3 and A4 in the online appendix summarise the resource items consumed and saved by the CWF intervention (https://www.ucc.ie/en/media/academic/centreforpolicystudies/AppendixTables_A3andA4_AnoverviewofresourceitemsconsumedandsavedbytheCWFprogramme.pdf)

All studies reported the discount rate, though it was not justified in five studies (full n=4 (Niessen and Douglass, 1984; Birch, 1990; O'Keefe, 1994; Ciketic *et al*., 2010), partial n=1 (O'Connell *et al*., 2005)). Consequently, these studies fail to meet the relevant CHEC and CHEERS standard.

All full evaluations reported incremental analyses of costs and consequences of the alternatives, thereby meeting CHEC guidelines. However, four full EEs (O'Keefe, 1994; Ciketic *et al*., 2010; Moore *et al*., 2017; Goodwin *et al*., 2022) summarised the study result using the incremental cost-effectiveness ratio (ICER), while the remaining five full EEs reported the average cost-effectiveness ratio (ACER) (Niessen and Douglass, 1984; Birch, 1990; Wright *et al*., 2001; Cobiac and Vos, 2012; Fyfe *et al*., 2015). Typically, cost-effectiveness ratios (CERs) that have been calculated against a baseline of no intervention without reference to an alternative are referred to as ACERs (Gray *et al*., 2010).

Ten studies assessed how their results varied among different population subgroups (full n=6 (Birch 1990; Wright *et al*., 2001; Cobiac and Vos 2012; Fyfe *et al*., 2015; Moore *et al*., 2017; Goodwin *et al*., 2022), partial n=4 (Griffin *et al*., 2001; O'Connell *et al*., 2005, 2016; Campain *et al*., 2010)), thus adhering to both the methodological and reporting quality standards.

Except for two cost-analyses (Dowell, 1976; Nelson and Swint, 1976), all studies described how variations in their

input parameters or assumptions would have influenced their results. Sensitivity analysis, in the form of deterministic or probabilistic analyses, examined how discount rate assumptions (Carr *et al*., 1980; Niessen and Douglass, 1984; Doessel, 1985; Birch, 1990; Griffin *et al*., 2001; Wright *et al*., 2001; Campain *et al*., 2010; Ciketic *et al*., 2010; Tchouaket *et al*., 2013; Fyfe *et al*., 2015; O'Connell *et al*., 2016; Moore *et al*., 2017), parameters influencing costs (O'Keefe, 1994; Griffin *et al*., 2001; Wright *et al*., 2001; O'Connell *et al*., 2005, 2016; Ciketic *et al*., 2010; Cobiac and Vos, 2012; Fyfe *et al*., 2015; Moore *et al*., 2017; Goodwin *et al*., 2022), estimates affecting resources saved (Carr *et al*., 1980; Doessel, 1985; Griffin *et al*., 2001; Wright *et al*., 2001; O'Connell *et al*., 2005, 2016; Campain *et al*., 2010; Ciketic *et al*., 2010; Cobiac and Vos, 2012; Tchouaket *et al*., 2013; Fyfe *et al*., 2015; Moore *et al*., 2017), the inclusion of missing data and consideration of an alternative clinical outcome measure (Goodwin *et al*., 2022) influenced the study results. Six studies quantified the uncertainty of their estimated CERs (O'Connell *et al*., 2005, 2016; Ciketic *et al*., 2010; Cobiac and Vos, 2012; Fyfe *et al*., 2015; Goodwin *et al*., 2022), of which, four provided a visual representation (Griffin *et al*., 2001; O'Connell *et al*., 2005; Ciketic *et al*., 2010; Cobiac and Vos, 2012). These studies demonstrated strong adherence to both the CHEC and CHEERS guidelines.

Only three EEs defined the model type and justified its use within the study (full n= 2 (Moore *et al*., 2017; Goodwin *et al*., 2022), partial n=1 (O'Connell *et al*., 2016)). Of the remaining studies, only nine provided comprehensive information about the input parameters selected for analysis (full n=5 (Birch, 1990; Wright *et al*., 2001; Cobiac and Vos, 2012; Moore *et al*., 2017; Goodwin *et al*., 2022), partial n= 4 (Griffin *et al*., 2001; O'Connell *et al*., 2005, 2016; Tchouaket *et al*., 2013)). This is important as economic models synthesize data from several sources, involving some level of analyst discretion in terms of data selection, methods, and assumptions. In the absence of transparent information about these choices, it is unfeasible to verify the study's results and conclusions.

Only six studies confirmed there were no conflicts of interest in accordance with CHEC (full n=4 (Cobiac and Vos, 2012; Fyfe *et al*., 2015; Moore *et al*., 2017; Goodwin *et al*., 2022), partial n=2 (O'Connell *et al*., 2005; Tchouaket *et al*., 2013)), however, just one study explicitly met guidelines for reporting such conflicts (Goodwin *et al*., 2022). Furthermore, ten studies (full n=5 (Wright *et al*., 2001; Cobiac and Vos, 2012; Moore *et al*., 2017; Goodwin *et al*., 2022) partial n=5 (Doessel, 1985; O'Connell *et al*., 2005, 2016; Campain *et al*., 2010; Tchouaket *et al*., 2013)) adhered to reporting guidelines and addressed how the study was funded and the role of the funder within the context of their analysis. Four studies considered the distributional impact of CWF (full n= 3 (Birch, 1990; Moore *et al*., 2017; Goodwin *et al*., 2022), partial n=1 (O'Connell *et al*., 2016)) despite having been conducted before the updated CHEERS statement (Husereau *et al*., 2022). However, no study reported on items concerning patient or stakeholder engagement and the availability of a health economics plan.

A visual overview of fulfilled CHEC and CHEERS criteria for each study is presented in Figures 2 and 3 and in tabular format in Tables A5 and A6 in the online appendix (https://www.ucc.ie/en/media/academic/centreforpolicystudies/AppendixTables_A5andA6_StudyperformanceagainsttheCHEC-listandCHEERSstatementcriteria.pdf).

A visual summary of the performance of the studies in relation to checklist criteria is presented online in Figures A2 and A3 (https://www.ucc.ie/en/media/academic/centreforpolicystudies/AppendixFiguresA2andA3_OverviewofperformanceofstudiesinrelationtotheindividualcriteriaoutlinedintheCHECandCHEERS.pdf).

Discussion

The review identifies the most widely used type of EE of CWF as cost-analysis. As cost-analysis only considers the costs of the alternatives and does not evaluate the consequences, it is a partial EE. While information on costs is valuable, cost-analysis does not provide the information necessary to guide decision-makers on the efficient use of scarce resources (Drummond *et al*., 2015). Of particular concern is that several cost-analyses were presented as full EEs in the form of CBA (Nelson and Swint, 1976; Doessel, 1985) or CEA (Tchouaket *et al*., 2013). Despite comprehensive guidelines on the conduct of EEs (Drummond and Jefferson, 1996; Drummond and McGuire, 2001; Evers *et al*., 2005; Philips *et al*., 2006; Centre for Reviews and Dissemination, 2009; Drummond *et al*., 2015; Husereau *et al*., 2022), and documented concerns around the mislabelling of cost-analyses (White *et al*., 1989; Birch and Gafni, 1996; Zarnke *et al*., 1997; Mariño *et al*., 2013; Drummond *et al*., 2015; Tonmukayakul *et al*., 2015), misunderstanding persists on the methods required to value consequences in CBAs. This is evident in a recent review of EEs of CWF (Mariño and Zaror, 2020), where nine cost-analyses were incorrectly referred to as CBAs, even though some of them were not originally presented as CBAs. Furthermore, another review appraising the evidence of OH interventions (Nguyen *et al*., 2023) also described several cost analyses of CWF as CBAs.

The study perspective, an important feature of EE, defines the costs relevant for the funding decision. This review highlights shortcomings in both the methodological and reporting quality with respect to this study element. Two studies published after the original CHEERS statement (Husereau *et al*., 2013) did not adhere to the reporting standard. Previous reviews also observed issues with the reporting quality of the perspective, although this was mainly in the context of older EEs of OH interventions (Mariño *et al*., 2013, 2020; Rogers *et al*., 2019; Anopa *et al*., 2020).

All full EEs adhered to CHEERS guidelines in identifying their primary health outcomes. Apart from two studies that were published before the original CHEERS statement, all the others described the methods used to measure and value outcomes. However, the quality of these components in certain EEs did not meet CHEC standards. Only five of the full EEs identified fluorosis, a cosmetic condition that affects teeth, as a relevant health outcome, despite it being an established risk associated with CWF. However, these studies did not account for the condition within their analysis, with only the most recent EE providing a valid justification for its exclusion (Goodwin *et al*., 2022). Also of note, was that some studies applied an effect of CWF informed by previous research to the decay outcomes of

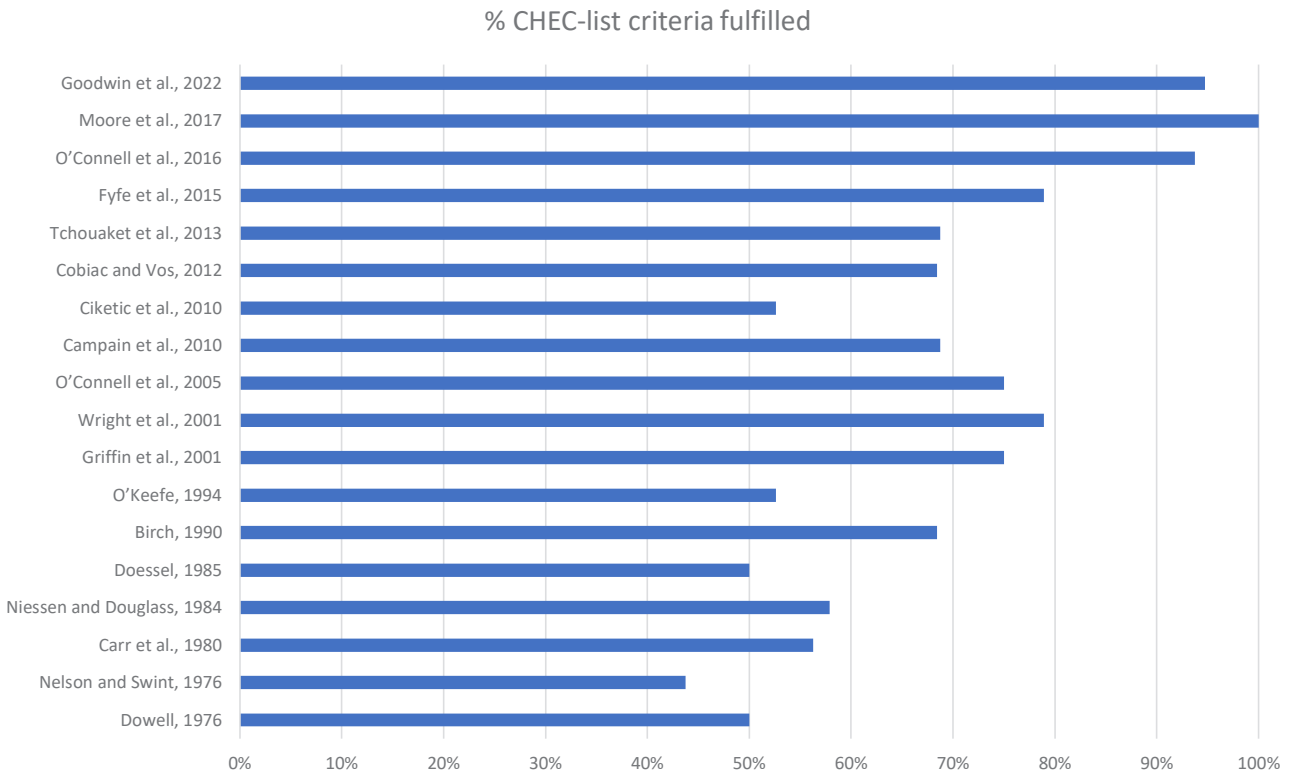
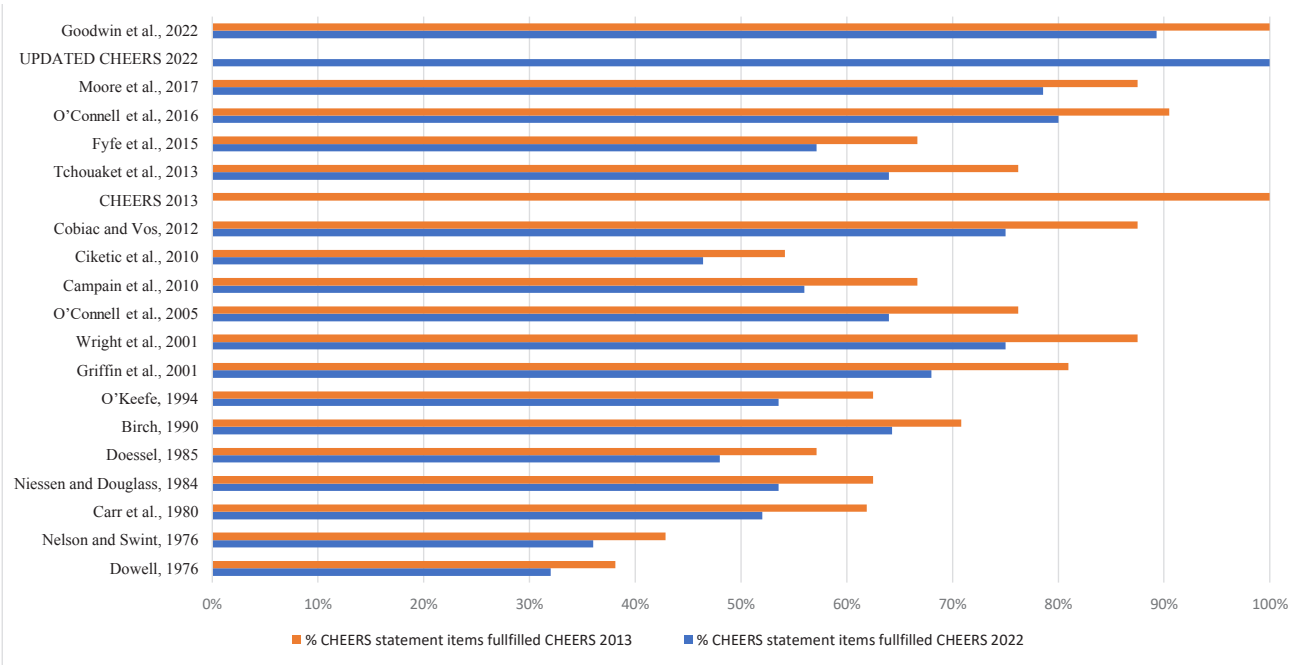


Figure 2. *An overview of the percentage of fulfilled CHEC-list criteria for each study.*



Note: Full EEs are assessed on 28/24 study criteria of the updated/original CHEERS statement, while partial EEs are assessed on 25/21 criteria of the updated/original CHEERS statement.

Figure 3. *An overview of the percentage of fulfilled CHEERS statement criteria for each study.*

non-fluoridated populations to determine the reduction in decay attributed to the intervention. Consequently, these results were dependent on the initial caries levels and the presumed effect of CWF, which in some cases, was based on data that was over a decade old. This makes it difficult to ascertain whether the reported reduction in disease between fluoridated and non-fluoridated populations has affected the cost-effectiveness of CWF. This issue has been noted in a previous review of preventive OH interventions (Nguyen *et al*., 2023). Subsequent research should consider contemporary epidemiological differences in decay outcomes between populations with and without lifetime exposure to establish the actual effect of CWF. Furthermore, various methods and outcome measures were used to identify and quantify the reduction in decay. To enhance interstudy comparability and broader generalisability, future research should report both dmfs/DMFS and dmft/DMFT outcomes when assessing the impact of CWF on decay experience.

Four CUAs estimated the expected HRQoL benefits associated with CWF using a generic outcome measure, while one reported outcomes in terms of QATYs, which limits interstudy comparability. Only one CUA employed a validated preference-based instrument, the CHU9D to measure the impact of CWF on children's HRQoL. However, concerns have been raised about the capacity of this instrument to capture the dynamic nature of decay on quality of life (Foster Page *et al*., 2015; Rogers *et al*., 2019). Additionally, the preference weights used to value the health states described by the CHU9D were derived from an adult rather than a child population. The remaining CUAs, depending on their chosen outcome measure, assigned either utility values, or disutility weights, to estimate preferences for health states defined by levels of decay. However, methodological weaknesses are noted with the use of arbitrary utility weights based on expert opinion. In addition, one study used a disutility weight for symptomatic caries to value surface decay outcomes. There is an obvious need for the development of a preference-based measure of decay to enable greater use of QALYS in EEs of OH programmes (Rogers *et al*., 2019).

A notable paucity of detail surrounding the costs of CWF provision persists (White *et al*., 1989), with only some recent evaluations adhering to methodological and reporting standards. Further to identifying relevant costs, evaluation requires that all costs are measured in physical units and subsequently valued (Drummond *et al*., 2015). However, this process was not followed in most included studies. Furthermore, none of the studies accounted for the costs associated with the adverse effects of CWF, and only one assessed the costs associated with higher levels of tooth retention due to CWF exposure (Campain *et al*., 2010). Considering increasing life expectancy and advancements in treatment options, future evaluations of CWF need to examine these costs. While the quality aspects of the negative costs associated with CWF were considered as part of the narrative, they did not feature in the output of the quality assessment instruments.

All but one CEA (Birch, 1990) incorporated savings within their cost estimates despite documented concerns around the realisation of these savings (Grembowski and Milgrom, 1988; Birch, 1990). Moreover, only two studies acknowledged concerns about their inclusion (Griffin *et al*., 2001; Tchouaket *et al*., 2013). Within this context, it is concerning that recent EEs have expanded the scope of savings to include a broader and more complex range of treatments over a lifetime. Future EEs need to emphasize that the study results are contingent on the assumption that savings are used to offset costs (Birch, 1990). Authors should also report the study outcomes exclusive of the expected treatment savings to provide a more transparent and accurate representation of the findings.

Policymakers should recognise the limitations of quality assessment instruments, as the outlined criteria may not be sensitive to the nuances of study elements specific to the evaluation of CWF, such as estimating the impact of fluoridation and the inclusion of treatment savings within cost estimates. To enhance transparency around the methodological and reporting quality of studies, subsequent EEs of CWF should also include a concise explanation for each checklist item, accompanied by references to the corresponding sections containing relevant information along with the data sources used to inform the study element.

While this review highlights shortcomings in the quality of the economic evidence of CWF, it also identifies considerable improvement in both their methodological and reporting quality over time. A subset of studies demonstrated commendable methodological rigor (Griffin *et al*., 2001; Wright *et al*., 2001; O'Connell *et al*., 2005, 2016; Fyfe *et al*., 2015; Moore *et al*., 2017; Goodwin *et al*., 2022) and reporting quality (Griffin *et al*., 2001; Wright *et al*., 2001; O'Connell *et al*., 2005, 2016; Cobiac and Vos, 2012; Moore *et al*., 2017; Goodwin *et al*., 2022). Many studies were conducted before the publication of the CHEC-list in 2005 and the original CHEERS statement in 2013, while all but one study predate the updated version of CHEERS published in 2022. It is encouraging to note that the three most recently published studies meet all or almost all the essential criteria specified by both assessment instruments.

This assessment of the methodological and reporting quality of the EE studies of CWF will empower future researchers to avoid the common pitfalls highlighted in previous research.

This review was unable to consider non-English language EEs and studies were included only if at least one other qualifying study from the same jurisdiction within a specific timeframe could be determined by the authors. Despite these limitations, the findings provide valuable insights into the economic evidence of CWF.

In conclusion, robust EEs can support policymakers in identifying cost-effective strategies to improve oral health. This review used the CHEC-list to assess the quality of the methods employed in the EEs and the CHEERS statement examined the transparent communication and completeness of the studies. Whilst many studies were published before best practice guidelines for EEs, a subset of studies demonstrated commendable methodological rigour and reporting quality. Furthermore, there is an upward trajectory in methodological and reporting standards over time, with the latest studies meeting all or almost all the criteria of both quality assessment instruments. This review offers valuable insights into the existing economic evidence of CWF and will assist policymakers in leveraging the available evidence effectively to inform resource allocation decisions. It may serve as a resource for researchers, enabling them to build upon previous studies and enhance the methodological and reporting standards of future EEs of CWF.

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