Economic evaluation of school-based caries preventive programs: A systematic review

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Objective: Assess interventions and health outcomes in studies giving data on economic evaluation (EE) of school-based caries prevention. Basic research design: Systematic review. Both partial EE that included cost description, cost-outcome description, cost analysis and full EE that included both cost and outcome of at least 2 interventions were included. Quality assessment used the Consolidated Health Economic Evaluation Reporting Standards (CHEERS) guidelines. **Results**: An electronic search of 6 databases identified 558 titles and abstracts. Paper eligibility screening identified 32 full papers which met the inclusion criteria. Most were conducted in the United States and cost effectiveness analysis was the most common type of EE. Nine were model-based studies and 17 derived their data from single studies. Sealants were most frequently evaluated followed by fluoride mouthrinse. Many CHEERS criteria were not met in the included studies. The following were found to be cost-effective: school-based, under general supervision, longer duration of program and targeting high caries risk groups. **Conclusions**: The deficiencies in the existing studies warrant more investigations of the economic aspects of school-based activities interventions to prevent caries.

Keywords: Economics, schoolchildren, dental caries prevention

INTRODUCTION

Although largely preventable, dental caries continues to be the world's most prevalent childhood disease (Ruff and Niederman, 2018). About half of five to six-year-old children experience tooth decay and the figure escalates to above 90% in some low- and middle-income countries, indicating that dental caries is an existing public health crisis (Arora et al., 2017). Poor oral health and untreated dental conditions compromise normal eating and negatively impact nutrition, self-esteem, speech, socialization, school attendance and quality of life, which may lead to overall deterioration of health (Petersen, 2003). Timely intervention has the potential to reduce overall costs associated with dental treatment since untreated dental disease increases in severity, necessitating more extensive and costly treatment secondary to postponing care (Savage et al., 2004). However, globally oral health issues continue to be among the costliest health problems to treat, resulting in high direct and indirect costs to individuals, families, and governments (Bertrand et al., 2011). According to 2015 data, the global economic burden of dental diseases for a year amounted to USD 442 billion, including both direct treatment costs and indirect costs in terms of productivity losses owing to absenteeism at school and work (Listl et al., 2015).

Schools remain an important setting not only to identify children with oral health problems, but also to bring these children into contact with oral health services (Arora *et al.*, 2017). School health programs offer an efficient and effective way to reach over 1 billion children worldwide and, through them the dissemination of information to families and community members (Petersen, 2003). Most school-based or school-linked fluoride and fissure sealant programs are effective, particularly for children with high risk of dental decay (Ruff and Niederman, 2018). However, the type of services provided, intensity of treatments (e.g., topical fluoride concentration) and frequency of care can be inconsistent across programs. This can result in disparate treatment effects. In addition to overall program-level variation, the complex relationship between oral health, the school environment, and the community may affect prevention effectiveness.

The inclusion of an economic perspective in the evaluation of health and health care is an essential component of health policy and planning (Hutubessy et al., 2003). Economic evaluation (EE) is an integral component of decision making about any oral health preventive program (Niessen and Douglass, 1984). When working under limited budget conditions, it is important to determine which intervention (or combination of interventions) maximizes results in oral health given the available resources (Marino et al., 2012). Comparable cost-effectiveness information is important to enable objective assessment of the relative return on investment of different caries prevention options (O'Neill et al., 2017). There are numerous Cochrane reviews that provide strong evidence to demonstrate the effectiveness of fluoride delivered in various guises to prevent dental caries (Marinho et al., 2004a; 2004b). However, except for water fluoridation and pit and fissure sealants, systematic reviews on the economic evaluation of other caries preventive measures remain limited. Recently, two systematic reviews reported an exhaustive quality

assessment of economic evaluations, but did not attempt to report which caries preventive method was costeffective (Marinho et al., 2013; Tonmukayakul et al., 2015). Given the limited public resources for oral health care, economic evaluations generating these conclusions should highlight that community-level preventive dental interventions can improve population oral health and be an economically feasible area for investment (Huang et al., 2019). Acknowledging the increased attention to reducing healthcare costs and the increasing emphasis on the use of interventions with evidence of effectiveness and cost-effectiveness, this systematic review aimed to identify all studies that gave data on economic evaluation of school-based caries preventive interventions and assess the various interventions they evaluated, along with the health outcomes.

METHODS

Irrespective of the study design, all published peerreviewed studies that assessed costs or/and outcomes were considered for inclusion. The other inclusion criteria were:

- Population school children aged 6 to 15 years
- Intervention: caries preventive interventions that were school-based
- Language: English
- Partial economic evaluations (EE) that included cost description, cost-outcome description, cost analysis and full EE that included both the cost and outcome of at least 2 interventions.

The exclusion criteria were

- Non-economic evaluations
- Adult population
- Literature reviews
- Methodological studies
- Non-preventive interventions or non-school based interventions

No attempt was made to search for unpublished literature or to contact the authors of such studies.

Search strategy: MEDLINE, PubMed, HTA, Web of Science, CRD, EBSCO were searched for the period from inception of the database till March 2019. The list of search words used is presented in Table 1.

The title and abstract of the identified citations were screened by the 2 authors independently and based on the inclusion and exclusion criteria, selections were made. Full texts were retrieved for relevant citations and for those that could not be finalised by only abstracts. Any disagreement between the reviewers was resolved by discussion and final consensus was reached.

The following data were extracted from the selected studies by the first author (AKM): author, year, country, type of study, target population, comparators, time horizon, perspective, discount rate, outcomes, currency, methods of managing uncertainty and key findings. The quality of the technical information was assessed using Consolidated Health Economic Evaluation Reporting Standards (CHEERS) guidelines (Husereau *et al.*, 2013). Answers were coded as 'yes', 'no' and 'not clear'. Descriptive statistics were performed to summarize the findings.
 Table 1. Search strategy.

Database: PubMed (NLM)

#1. "economic evaluation" [Title/Abstract] OR
"economics" [Subheading] OR "cost-effectiveness analysis" [MeSH Terms Terms] OR "cost-benefit analysis"
[MeSH Terms] OR "cost-utility analysis" [MeSH Terms] OR "cost-minimisation" [MeSH Terms] OR "cost"
[MeSH Terms Terms] OR "Quality Adjusted Life Years"
[MeSH Terms] OR "qalys" [All Fields]
#2. "dental caries prevention" [MeSH Terms] OR "pit and fissure sealants" [MeSH Terms] OR ("fluoride"
[MeSH Terms]) OR ("toothpaste* OR dentifrice* OR mouthrinse* OR mouthwash* OR topical* OR systemic* OR gel* OR varnish* OR foam*")
#3. "children" [Title/Abstract] OR "schoolchildren"
[MeSH Terms] OR "adolescents" [MeSH Terms]
#1 OR #2 OR #3

RESULTS

The literature search identified 558 studies (Figure 1). After examining titles and abstracts, 454 studies were excluded based on the inclusion and exclusion criteria. The full texts of the remaining articles were retrieved and screened. Of 87 sources, 55 were excluded as they were reviews, had no economic data, were not school based, or were methodological studies or policy documents. Finally, 32 studies were included in the review.

EEs were reported from 13 countries; most were from the US (n = 13), followed by England and Ireland (n=4) and Sweden (n = 3); 2 each from Australia, China and Finland. Brazil, Canada, Chile, Japan, Spain, and Thailand have one EE each reported. The first economic study on a school-based intervention was published in 1970 (Ast *et al.*, 1970). There was one in 1978 (Stephen and Campbell, 1978) followed by 8 in the 1980's and 2 in the 1990's. Most included studies were published after the year 2000 (n=22). Cost-effectiveness analysis (CEA) was most common EE (n=18), followed by cost analysis (n=8), cost-benefit analysis (CBA) (n=5) and one cost-utility analysis (CUA). Details of included studies are presented in table 2.

Seventeen studies sourced data from a single study or trial; 2 (Bergstrom *et al.*, 2016; 2019) from retrospective studies and 4 (Johnson *et al.*, 2017, Dudovitz *et al.*, 2018, Garcia, 1989, Manau *et al.*, 1987) from evaluation of dental programs. Nine studies used modelling-based EE that combined multiple data sources. While 5 did not specify the type of model used, one clearly stated that a Markov model was applied, one used a decision tree and 2 employed both Markov and a decision tree analysis. All the interventions were targeted towards school children aged between 3-17 years with children of age 5 - 6 years and 12 - 14 years more commonly studied.

Though most studies (n=27) were based in schools, one (Kay *et al.*, 2018) sourced data from school-based studies. Another (Tuominen, 2008) evaluated a program in which dental check-ups in schools was one of the interventions. One study (Bertrand *et al.*, 2011) evaluated interventions provided both at the school and private clinics.

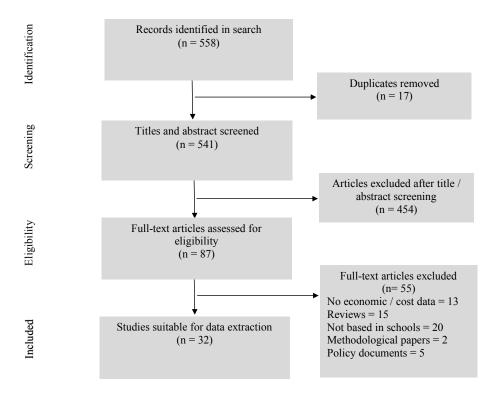


Figure 1. Flow chart of included and excluded studies

The most commonly evaluated intervention was pit and fissure sealants (n=17), followed by fluoride mouth rinsing (FMR) (n=10). The other interventions were as follows: fluoride varnish and fluoridated toothpaste (6 studies each); school water fluoridation (3 studies); milk fluoridation, fluoride gels, fluoride tablets and oral health education and dental checkup (2 each) and routine dental care, screening and referral, a comprehensive school health program and xylitol chewing gum (1 each).

The CHEERS guidelines can be used to appraise EE in dentistry (Husereau *et al.*, 2013). However, not all the CHEERS criteria were satisfied by the included studies (Table 3). While most studies identified themselves as EE in their titles, their abstracts had deficiencies of perspective and methods. The study setting and the different interventions evaluated were indicated in most studies. Nearly half failed to report the perspective used to collect the costs. Almost 80% of the studies provided information on the time horizon of the evaluation and discounting was reported in 71% of the studies. Studies that did not discount the cost or outcomes did not give the reason for not doing so.

The primary outcome measure was identified in the great majority of studies along with the sources of effectiveness data. About 35% did not provide data on the price date used and 40% did not account for currency conversion, even when indicated. More than half of the model-based studies did not report the model used for their evaluations or the assumptions on which it was based. A high proportion of these studies did not perform sensitivity analysis. However, all summarized their findings and answered the study questions framed. Furthermore, most studies did not provide data about their funding and more than three quarters did not report conflicts of interest.

The measurement of costs and outcomes in 8 studies were from a health care provider perspective and an equal number of studies adopted a societal perspective. The remaining 16 studies did not mention the perspective used. While 7 studies did not specify the time horizon of the EE, for the other studies the time horizon ranged from 2 years to 10 years, with 4 years being the most common (n=6).

The outcomes assessed varied from caries increment, averted decayed, missing, filled teeth/surfaces, restorations prevented for CEAs, disability-adjusted life years (DALY) and quality-adjusted life years (QALY) lost due to caries for CUA studies, cost of dental care or restorations prevented for CBA studies. Caries was assessed at the surface level in 8 studies and tooth/child level in 12.

Discounting is normally performed when the time horizon of a study exceeds 1 year to convert future costs and benefits to the base value (Drummond *et al.*, 2005). However, 13 studies neither used any discounting nor stated their justification for not doing so. Only one study (Goldman *et al.*, 2014) reported that the lack of discounting was because the time horizon was less than 1 year. Where studies mentioned the discount rate (n=18), it varied from 1.5% to 10%, with 9 studies using a 3% rate.

The commonly used currency for reporting the results was the US Dollar (USD) (n=14), followed by 3 studies using Pounds and Swedish Krona and 2 using Australian Dollars. One study each used Canadian Dollars, Chilean Pesos, Finnish Markkas, Irish Pounds, Brazilian Reals, Spanish Pesetas, Thai Baht, Yen and Yuan. However, half the studies did not state the price date used. Among those that mentioned the price date it was spread over the years 1978-2017. Currency conversion was mentioned in only 12 studies, using conversion to Euros in 3 studies

Name	Country	Type of EE	Single study/ model	Perspective	Comparators	Time Di horizon (years)	Discount rate (%)	Health outcomes	Effectiveness	Currency	Price date (year)	Uncertainty	Finding
Ast et al., 1970	USA	Cost Analysis	Program evaluation	NS	Fluoridated area vs Non-fluoridated area	9	NS	Cost of den- tal care	Single study based	USD	NS	Not applicable	Costs reduce with time
Stephen and Campbell, 1978	UK	CBA	Single study	NS	1mg F tabs vs placebo	3	NS	DMFT/S reduction	Single study based	Pound	NS	NS	Cost saving of 53%
Doherty et al., 1984	USA	Cost Analysis	Single study	NS	F mouth rinse	7	10	NA	Single study based	USD	1978	NS	Average total and explicit costs increase with program size
Klein <i>et al.</i> , 1985	USA	Cost Analysis	Cost Single study	NS	F mouth rinse, Lessons, Tooth brushing, Sealants, Gels in different combinations	4	NS	Difference in mean DMFS increment	Single study based	USD	1981	NS	Annual direct cost/child is \$23 -sealant/gel; \$3.29 - mouthrinse; all together - \$54.92
Doherty <i>et al.</i> , 1987	NSA	Cost Analysis	Cost Single study Analysis	NS	F mouth rinse	NS	NS	NA	Single study based	USD	1978	NS	Cost increased with paid and voluntary labour, wages
Manau <i>et al.</i> , 1987	Spain	CEA	Program	NS	Community water fluoridation, fortnightly 0.2% F mouth rinse, Supervised Tooth brushing with Fluoridated toothpaste	NS	Ľ	Surfaces saved from DMFS	Synthesis based study	Spanish Peseta	NS	NS	Community water fluoridation more cost-effective than F mouth rinse and Tooth brushing
O'Rourke <i>et</i> al., 1988	England	CBA	Single study	NS	Daily F tablet vs Nothing	3	5	Caries increment	Single study based	Pound	1983	NS	Cost benefit ratio- 1.35:1
Garcia, 1989	USA	Cost analysis	Program evaluation	NS	F mouth rinse, Sealants, School fluoridation	NS	NS	Not applicable	Program based	USD	1988	ZS	Costs F mouth rinse -\$1.3/ child/year; school fluoridation -\$4,52/child/year; Sealants -\$21.14/child/year
Crowley <i>et al.</i> , Australia 1996	Australia	CBA	Model	Societal	Sealants + F mouth rinse vs Routine dental care	10	ŝ	DMFS incre- ment	Synthesis based study	Aus. Dollar	1994	NS	Shows net economic benefits
Morgan <i>et al.</i> , 1997	Australia	CEA	Non randomised trial	Societal	Sealants + F mouth rinse vs Routine dental care	ς	Ś	DMFS incre- ment	Single study based	Aus. Dollar	1994	Discount rate at 0, 10%, extreme values of effectiveness, annual check- ups, use of dental auxiliary	ICER becomes favourable for Sealants + F mouth rinse over time
Alanen <i>et al.</i> , 2000	Finland	Cost analysis	Single study	NS	Sealants vs Xylitol chewing gums	Ś	SN	Dental caries at den- tine level	Single study based	Finnish Markka	NS	NS	No great difference in costs between sealant and xylitol gums

Table 2. Characteristics of the studies included in the systematic review of school-based caries preventive economic evaluations

Table 2 continued overleaf.....

Table 2 continued

Name	Country	Type of EE	Single study/ model	Perspective	Comparators	Time D horizon (years)	Discount rate (%)	<i>Health</i> outcomes	Effectiveness	Currency	Price date (year)	Uncertainty	Finding
Werner <i>et al.</i> , 2000	USA	CEA	Single study	NS	School sealant application vs Clinic-based sealant application	6	NS	Number of teeth without decay	Single study based	USD	1991	NS	School based - \$65, clinic based - \$42 without considering transportation, time off work, waiting time
Holland <i>et al.</i> , 2001	Ireland	Cost analysis	Single study	NS	Community water fluoridation vs School F mouth rinse	6	Ś	DMFT	Single study based	Irish Pounds	NS	NS	Community water fluoridation - Ire 0.21/person; F mouth rinse - Ire 3.26/child
Zabos <i>et al.</i> , 2002	NSA	CEA	Single study	Public	Sealants vs No sealants	5	ŝ	DMFS averted	Single study based	USD	1992	Sensitivity analysis with discount rate of 5%, filled teeth as normal	Sealants incur 44% lower costs than no sealants
Scherrer <i>et al.</i> , 2007	USA	Cost analysis	Single study	Societal	Sealant delivery by diffèrent personnel and under diffèring supervision	NS	3	Not applicable	Synthesis based study	USD	NS	Not applicable	Cost saving resulted fromreducing the level of supervision; rural programs; having dentist and dental assistant come on different days under general supervision
Skold <i>et al.</i> , 2008	Sweden	CBA	Model	Public dental care	F varnish vs F mouth rinse	×	e	Costs of restorations prevented	Synthesis based study	SEK	2006	Discount rate 0, 5%	Costs of F varnish is lesser than that of F mouth rinse
Tuominen, 2008	Finland	Evaluation of relative value	Evaluation Finland of relativeSingle study value	NS	Helicopter ambulance services, measles, parotitis and rubella (MPR) vaccination, breast cancer screen- ing, hip replacement, dental check up	NS	ZS	r Not applicable	Preference measurements using VAS, rank order (RO), willingness to pay (WTP)	Not Not applicable applicable	Not pplicable	SZ	Helicopter ambulance, MPR and breast cancer screening were preferred over hip replacement and dental check up
Sakuma <i>et al.</i> , 2010	Japan	CEA	Single study	NS	0.2% weekly F mouth rinse + targeted seal- ants vs no intervention	NS	NS	DFT averted	Single study based	Yen	NS	NS	Combined program was cost effective
Bertrand <i>et al.</i> , Canada 2011	Canada	CEA	Model	Health care system and parents	Public, private and mixed sealant application	10	3 q	Children without S decay in first permanent molars	Children without Synthesis based Canadian decay in first study Dollar molars	Canadian Dollar	NS	Discount rate 0, 5%, ex- treme values of effects and outcomes	School based intervention better than private clinic based intervention

Table 2 continued overleaf.....

Table 2 continued

Name	Country	Type of EE	Single study/ model	Perspective	Comparators	Time horizon (years)	Discount rate (%)	<i>Health</i> outcomes	Effectiveness (Currency	Price date (year)	Uncertainty	Finding
Marino <i>et al.</i> , 2012	Chile	CEA	Model	Societal	Milk fluoridation + F mouth rinse + APF + supervised toothbrushing with Fluoridated toothpaste vs no intervention (Only school-based interventions are considered)	¢	m	Dental caries averted	Synthesis based study	Chilean Peso	2009	Discount rate 0, 6%, extreme outcomes, dentist vs DA, sealing only likely carious molars	Costs saving in milk fluoridation and F mouth rinse, cost spending in APF and supervised toothbrushing
Goldman <i>et al.</i> , China 2014	China	CEA	Government Single study program providers	Government oral health program providers	Glass carbomer, conventional HVGIC with heat, without heat vs composite resin sealant	7	Not done	New caries lesions	Single study based	Yuan	2008	Varying personnel and equipment costs	LED HVGIC is \$1.11 more than composite
Neidell <i>et al.</i> , 2016	NSA	CEA	Model	NS	F varnish, Sealants	4	9	Reduction in occlusal caries	Synthesis based study	USD	NS	Non-occlusal benefits of F varnish included, sealant failure rate at 40%	Costs of F varnish < Sealants
Bergstrom <i>et</i> al., 2016	Sweden	CEA	Retrospective	S Z	F varnish + Fluoridated toothpaste + Regular dental check-ups vs Fluoridated toothpaste, Tooth brushing + Regular dental check-ups	4	NS	Prevented fillings	Single study based	SEK	2013	Not applicable	Costs and gains break-even at 15 years of age
Goldman <i>et al.</i> , China 2016	China	CEA	Government Government Single study program providers	Government oral health program providers	Glass carbomer, Conventional high viscosity glass ionomer cement (HVGIC) with heat, without heat vs Composite resin sealant	4	m	New cavitated dentine lesions	Single study based	USD	2010	Sensitivity analysis by increasing the probability of caries, changing costs of personnel, instruments, supplies	LED HVGIC is \$105 more than composite; LED thermocured HVGIC is \$115 more the composite
Griffin <i>et al.</i> , 2016	USA	CEA	Model	Societal	School based sealant program	4	ŝ	DALY	Synthesis based study	USD	2014	Sensitivity analysis- varying parameters 50% above and 50% below their base rate	Prevent 485 fillings and 1.59 daly /1000 children with sealants
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Table 2 continued

Name	Country	Type of EE	Single study/ model	Perspective	Comparators	Time D horizon (years)	Discount rate (%)	Health outcomes	Effectiveness	Currency	Price date (year)	Uncertainty	Finding
Johnson <i>et al.</i> , 2017	USA	CEA	Program evaluation	NS	General vs Direct supervision of school based dental sealant program	NS	NS	NS	Program based	USD	NS	SZ	Sealant placement under direct supervision - \$33/child, general supervision -\$14.50 / child, cost saving /child- \$28
Dudovitz <i>et al.</i> , 2017	, USA	CBA	Program evaluation	Provider	Health education + F varnish + Screening + Referral	7	NS	Reduction of caries incidence	Single study based	USD	NS	Not applicable	Larger program may result in cost saving
Goldman <i>et al.</i> , Brazil 2017	, Brazil	CEA	Governme Single study program providers	Government program providers	Composite resin sealants vs ART high viscosity GIC vs Supervised toothbrushing	ς	ς, σ	Preventing development of dentine carious lesions on first permanent molars	Single study based	Reais (Brazilian)	2012	Reducing number of visits of dental assistants to supervise toothbrushing, altering incidence of dentine cavitated lesion.	ART sealant dominant than composite sealant which is dominant on supervised toothbrushing
Kay <i>et al.</i> , 2018	England	CUA	Model	Public sector in UK	Supervised toothbrushing and F varnish	σ	1.5	QALY lost due to caries	Synthesis based study	Pound	2012	Used range of values for QALY loss per child	£ 55 /child- supervised toothbrushing, £100/child -F varnish Cost effective in 5-year-old; £23/child- supervised toothbrushing, 129/child- F varnish among 12-year-old.
Marino <i>et al.</i> , 2018	Thailand	CEA	Model	Societal	Fluoridated milk vs Milk without F	9	3.25	Difference in DMFS index	Synthesis based Thailand study Baht	Thailand Baht	2011	Sensitivity analysis- changing DMFS outcome, discounting - 0% and 5%	Milk fluoridation gives significant economic and human benefits
Bergstrom <i>et</i> al., 2019	Sweden	CEA	Retrospec- tive	Societal	F varnish application under FRAMM guideline vs No F varnish	4	NS	Approximal caries prevalence	Single study based	SEK	2017	SN	High caries group has most favourable cost-effective ratio
Huang <i>et al.</i> , 2019	USA	CEA	Model	Societal	Comprehensive school oral health program vs Sealants only vs No program	Ś	5	DALY	Synthesis based study	USD	2014	One-way sensitivity analysis-vary parameters to 50% and 150% of base value; 2-way sensitivity analysis - posterior attack rates and comprehensive program costs	Comprehensive program compared to sealants and no program becomes cost effective when extended to all the posterior teeth

Table 3. Quality of 32	included studies	s using CHEERS checklist
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Section	Item Number	Item	Studies reporting (%)
Title	1	Title identifies as EE	93.8
		Objectives in abstract	96.9
		Perspective in abstract	12.5
A la sture of	2	Setting in abstract	90.6
Abstract	2	Methods in abstract	87.5
		Results in abstract	96.9
		Conclusion in abstract	96.9
ntroduction	3	Background and objectives	100
	4	Target population and subgroup	100
	5	Setting and location	100
	6	Study perspective	50
	7	Comparators	100
	8	Time horizon	78.1
	9	Discount rate	71.9
	10	Choice of health outcome	89.7
	11a	Measurement of effectiveness (Single study-based estimates)	81
<i>x</i> (1 - 1	11b	Measurement of effectiveness (Synthesis-based estimates)	100
Methods	12	Measurement and valuation of preference-based outcomes	100
	13a	Estimating resources & costs (Single study-based EE)	100
	13b	Estimating resources & costs (Model-based EE)	100
		Currency	100
	14	Price date	65.6
		Conversion	60
	15	Choice of model	46.2
	16	Assumptions	92.3
	17	Analytical methods	36.4
Results	18	Study parameters	96.8
	19	Incremental costs and outcomes	96.8
	20a	Characterising uncertainty (Single study-based EE)	27.8
	20b	Characterising uncertainty (Model-based EE)	81.8
	21	Characterising heterogeneity	21.9
Discussion	22	Study findings, limitations, generalisability and current knowledge	e 100
	23	Source of funding	53.1
Others	24	Conflicts of interest	25

(Bergstrom et al., 2019, Bergstrom et al., 2016, Holland et al., 2001), Euro and USD in 2 studies (Skold et al., 2008, Alanen et al., 2000) and to USD in 7 studies (Marino et al., 2012, Morgan et al., 1997, Crowley et al., 1996, Goldman et al., 2014, Manau et al., 1987, Goldman et al., 2017, Marino et al., 2018).

About half of the studies did not deal with uncertainties. Among the others, altered discount rates, extreme values of effects and outcomes, use of different costs of personnel, instruments, and supplies, number of visits, annual check-ups, attack rates, or intervention methods were the methods used to check for uncertainty.

The included studies varied widely in terms of the interventions assessed, study designs, methods and outcome assessment. Additionally, few studies conducted similar comparisons of the interventions, making it difficult to draw conclusions. Nonetheless, on a broader perspective, the following findings can be extracted: when various modes of prevention were compared, fluoride mouthrinse was more cost-effective than sealants or fluoride gel (Klein *et al.*, 1985, Garcia *et al.*, 1989, Marino *et al.*, 2012, Marino *et al.*, 2018). The use of sealants was found to be more cost-effective than no sealants (Zabos *et al.*, 2002; Griffin *et al.*, 2016). Combined interventions or comprehensive programs (Crowley *et al.*, 1996, Sakuma *et al.*, 2010, Huang *et al.*, 2019) provide favourable incremental cost effectiveness ratios (ICERs) than single interventions or routine dental care.

DISCUSSION

Decision making in public health should be evidence-based. The effectiveness of school-based preventive programs has been well documented particularly in the prevention of dental caries. In a budget-constrained environment, not just the clinical effectiveness, but also the economic implications have to be considered (Tonmukayakul et al., 2015). EEs provide a range of information from the costs involved in an intervention to analyzing the costs and benefits of alternatives. Hence there exist guidelines based on economic perspectives such as the National Institute for Clinical Excellence (NICE) guidelines for the extraction of wisdom tooth, dental recall intervals etc. Similarly, it is imperative to have a clinically and economically sound framework to carry out school-based caries prevention. Systematic reviews help to conceptualize and finalize such guidelines. No existing reviews of EE in dentistry have focused specifically on school-based programs. Further, reviews by Kallestal et al. (2003) and Marinho et al. (2013) emphasized that economic analyses of caries prevention were inconclusive and need a standard platform for comparability.

The present review used the CHEERS checklist to appraise study quality, unlike the other systematic reviews that applied the Drummond checklist (Marinho *et al.*, 2013, Tonmukayakul *et al.*, 2015, Akinlotan *et al.*, 2018). Also, many previous reviews have scored the quality assessment and used medians as a cut-off for appraisal scores. Though the we included 32 studies, few contained a full EE. Hence in contrast to other reviews, we have refrained from assigning appraisal scores, as many criteria in the CHEERs checklist would not apply to partial EEs.

The number of studies has gradually increased with time. Alongside this, the type of studies has progressed from partial EEs such as cost analysis to full EEs including CEAs, CBAs, and CUAs. Additionally, most studies were based in the US, with the remainder conducted in developed countries with a few exceptions. Notwithstanding the greater quantity and quality of studies over time, some studies had questionable credibility in terms of use of terminologies, data on incremental costs and outcomes and management of uncertainties. Similar deficiencies were also pointed out by Marinho et al. (2013) and Tonmukayakul et al. (2015). Moreover, because of the limited number of similar studies, valid inferences on school-based interventions cannot be drawn. Hence, more studies comparing interventions under varying conditions are warranted to identify consistently economically viable interventions.

Most studies focused on caries prophylaxis using sealants and various fluoride vehicles, predominantly used in isolation. Since caries prevention is a package with various interventions acting together, EEs on multiple interventions should be performed. The health outcomes were the number of caries averted or caries increment as determined by DMFT/S. Despite these indices being standard caries assessment methods in epidemiology, an EE should be able to assess distinctly compendious impacts of the intervention, both health and economic (Akinlotan *et al.*, 2018). Thus, person-centred outcomes such as quality of life, QALYs and DALYs will be appropriate. However, the use of these measures of effectiveness can be challenging in children.

When cost and outcome estimates are assumed or taken from literature, the chances of uncertainty are high. Such studies warrant the use of sensitivity analysis (Marino *et al.*, 2013). Only 40% of studies had done so, which indicates a serious shortcoming in handling uncertainty. The use of mean values when the data are skewed can also be misleading.

In any EE extending for more than 1 year, the costs and outcomes incurred should be discounted to the base year (Tonmukayakul *et al.*, 2015). Many included studies did not discount the costs or outcomes. Nor did they explain this omission, even though it can result in overestimations of the value of future costs and benefits (Marino *et al.*, 2013).

One limitation of this review is the lack of grey literature. Moreover, the review might have missed relevant studies that were not written in English. The quality of the effectiveness data was not assessed. However, a review by Kallestal et al. (2003) found the studies to be of low evidence value with inconsistent results. This leads to compromised EE as a result of biased data.

Notwithstanding these concerns, the growing demand for evidence to support decision and policy making in health care has prompted the need for EE studies. However, conducting EE is highly labor-intensive and time-consuming. Hence, methods to improve the transferability of results, guidelines to standardize EE methods to enhance generalizability should be explored.

CONCLUSIONS

To the best of our knowledge, this is the first systematic review to evaluate economic evaluations of school-based caries preventive interventions and also to appraise their methodological quality. This study furnishes pragmatic data on deficiencies in the existing evaluations that might inhibit the development and implementation of school oral health policies. This review should assist oral health researchers and health economists in improving the quality of future EEs of school-based caries prevention.

REFERENCES

Akinlotan, M., Chen, B., Fontanilla, T. M., Chen, A. and Fan, V. Y. (2018): Economic evaluation of dental sealants: A systematic literature review. *Community Dentistry and Oral Epidemiology* 46, 38-46.

- Alanen, P., Holsti, M. L. and Pienihäkkinen, K. (2000): Sealants and xylitol chewing gum are equal in caries prevention. *Acta Odontologica Scandinavica* 58, 279-784.
- Arora, A., Khattri, S., Ismail, N. M., Sumanth, K. N. and Prashanti, E. (2017): School dental screening programs for oral health. *Cochrane Database of Systematic Reviews* 12, CD012595.
- Ast, D. B., Cons, N. C., Pollard, S. T. and Garfinkel, J. (1970): Time and cost factors to provide regular, periodic dental care for children in a fluoridated and nonfluoridated area: final report. *Journal of American Dental Association* **80**, 770-776.
- Bader, J. D., Shugars, D. A. and Bonito, A. J. (2001): A systematic review of selected caries prevention and management methods. *Community Dentistry and Oral Epidemiology* 29, 399-411.
- Bergström, E. K., Davidson, T. and Sköld, U. M. (2019): Costeffectiveness through the dental-health FRAMM guideline for caries prevention among 12- to 15-year-olds in Sweden. *Caries Research* 53, 339-346.
- Bertrand, E., Mallis, M., Bui, N. M. and Reinharz, D. (2011): Cost-effectiveness simulation of a universal publicly funded sealants application program. *Journal of Public Health Dentistry* 71, 38-45.
- Crowley, S. J., Campain, A. C. and Morgan, M. V. (2000): An economic evaluation of a publicly funded dental prevention programme in regional and rural Victoria: an extrapolated analysis. *Community Dental Health* **17**, 145-151.
- Doherty, N. J., Brunelle, J. A., Miller, A. J. and Li, S. H. (1984): Costs of school-based mouthrinsing in 14 demonstration programs in USA. *Community Dentistry and Oral Epidemiology* 12, 35-38.
- Doherty, N. J. and Martie, C. W. (1987): Analysis of the costs of school-based mouthrinsing programs. Community Dentistry and Oral Epidemiology 15, 67-69.
- Drummond, M., Sculpher, M., Torrance, G., O'Brien, B. and Stoddart, G. (2005): Methods for the economic evaluation of health care programmes, 3rd edn. Oxford: Oxford University Press.
- Dudovitz, R. N., Valiente, J. E., Espinosa, G., Yepes, C., Padilla, C., Puffer, M., Slavkin, H. C. and Chung, P. J. (2018): A school-based public health model to reduce oral health disparities. *Journal of Public Health Dentistry* 78, 9-16.
- Garcia, A. I. Caries incidence and costs of prevention programs. (1989): Journal of Public Health Dentistry **49**, 259-271.
- Goldman, A. S., Chen, X., Fan, M. and Frencken, J. E. (2014): Methods and preliminary findings of a cost-effectiveness study of glass-ionomer-based and composite resin sealant materials after 2 yr. *European Journal of Oral Sciences* **122**, 230-237.
- Goldman, A. S., Chen, X., Fan, M. and Frencken, J. E. (2016): Cost-effectiveness, in a randomized trial, of glass-ionomerbased and resin sealant materials after 4 yr. *European Journal of Oral Sciences* 124, 472-479.
- Goldman, A., Leal, S. C., de Amorim, R. G. and Frencken, J. E. (2017): Treating high-caries risk occlusal surfaces in first permanent molars through sealants and supervised toothbrushing: a 3-year cost-effective analysis. *Caries Research* 51, 489-499.
- Griffin, S. O., Naavaal, S., Scherrer, C., Patel, M. and Chattopadhyay, S. (2017): Evaluation of school-based dental sealant programs: an updated community guide systematic economic review. *American Journal of Preventive Medicine* 52, 407-415.
- Griffin, S., Naavaal, S., Scherrer, C., Griffin, P. M., Harris, K. and Chattopadhyay, S. (2016): School-based dental sealant programs prevent cavities and are cost-effective. *Health Affairs (Millwood)* 35, 2233-2240.
- Holland, T.J., Considine, J. and Creedon, P. (2001): The effectiveness and cost of two fluoride program for children. *European Journal of Paediatric Dentistry* **2**, 61-66.
- Huang, S. S., Ruff, R. R. and Niederman, R. (2019): An economic evaluation of a comprehensive school-based caries prevention program. *JDR Clinical and Translational Research* 22, 2380084419837587.

- Husereau, D., Drummond, M., Petrou, S., Carswell, C., Moher, D., Greenberg, D., Augustovski, F., Briggs, A. H., Mauskopf, J. and Loder, E. (2013): CHEERS Task Force. Consolidated Health Economic Evaluation Reporting Standards (CHEERS) statement. *British Journal of Obstetrics and Gynaecology* **120**, 765-770.
- Hutubessy, R., Chisholm, D. and Edejer, T. T. (2003): Generalized cost-effectiveness analysis for national-level prioritysetting in the health sector. *Cost Effectiveness and Resource Allocation* **1**, 8.
- Johnson, B., Serban, N., Griffin, P. M. and Tomar, S. L. (2017): The cost-effectiveness of three interventions for providing preventive services to low-income children. *Community Dentistry and Oral Epidemiology* 45, 522-528.
- Källestål, C., Norlund, A., Söder, B., Nordenram, G., Dahlgren, H., Petersson, L. G., Lagerlöf, F., Axelsson, S., Lingström, P., Mejàre, I., Holm, A. K. and Twetman, S. (2003): Economic evaluation of dental caries prevention: a systematic review. *Acta Odontologica Scandinavica* 61, 341-346.
- Kay, E., Owen, L., Taylor, M., Claxton, L. and Sheppard, L. (2018): The use of cost-utility analysis for the evaluation of caries prevention: an exploratory case study of two community-based public health interventions in a high-risk population in the UK. *Community Dental Health* 35, 30-36.
- Kizito, A., Caitlin, M., Wang, Y., Kasangaki, A. and Macnab, A. J. (2014): Oral health promotion in schools: rationale and evaluation. *Health Education* **114**, 293-303.
- Klein, S. P., Bohannan, H. M., Bell, R. M., Disney, J. A., Foch, C. B. and Graves, R. C. (1985): The cost and effectiveness of school-based preventive dental care. *American Journal of Public Health* **75**, 382-391.
- Listl, S., Galloway, J., Mossey, P. A. and Marcenes, W. (2015): Global economic impact of dental diseases. *Journal of Dental Research* 94, 1355-1361.
- Manau, C., Cuenca, E., Martínez-Carretero, J. and Salleras, L. (1987): Economic evaluation of community programs for the prevention of dental caries in Catalonia, Spain. *Community Dentistry and Oral Epidemiology* 15, 297-300.
- Marinho, V.C., Higgins, J.P., Sheiham, A. and Logan, S. (2004a): One topical fluoride (toothpastes, or mouthrinses, or gels, or varnishes) versus another for preventing dental caries in children and adolescents. *Cochrane Database of Systematic Reviews* 1, CD002780.
- Marinho, V.C., Higgins, J.P., Sheiham, A. and Logan, S. (2004b): Combinations of topical fluoride (toothpastes, mouthrinses, gels, varnishes) versus single topical fluoride for preventing dental caries in children and adolescents. *Cochrane Database of Systematic Reviews* 1, CD002781.
- Mariño, R., Fajardo, J. and Morgan, M. (2012): Cost-effectiveness models for dental caries prevention programmes among Chilean schoolchildren. *Community Dental Health* **29**, 302-308.
- Mariño, R. J., Khan, A. R. and Morgan, M. (2013): Systematic review of publications on economic evaluations of caries prevention programs. *Caries Research* 47, 265-272.
- Mariño, R., Traub, F., Lekfuangfu, P. and Niyomsilp, K. (2018): Cost-effectiveness analysis of a school-based dental caries prevention program using fluoridated milk in Bangkok, Thailand. *BMC Oral Health* 15, 24.
- Morgan, M. V., Campain, A. C., Crowley, S. J. and Wright, F. A. (1997): An evaluation of a primary preventive dental programme in non-fluoridated areas of Victoria, Australia. *Australian Dental Journal* 42, 381-388.
- Neidell, M., Shearer, B. and Lamster, I. B. (2016): Costeffectiveness analysis of dental sealants versus fluoride varnish in a school-based setting. *Caries Research* 50, 78-82.
- Neko-Uwagawa, Y., Yoshihara, A. and Miyazaki, H. (2011): Long-term caries preventive effects of a school-based fluoride mouth rinse program in adulthood. *Open Dentistry Journal* **5**, 24-28.

- Niessen, L. C. and Douglass, C. W. (1984): Theoretical considerations in applying benefit-cost and cost-effectiveness analyses to preventive dental programs. *Journal of Public Health Dentistry* 44, 156-168.
- O'Neill, C., Worthington, H. V., Donaldson, M., Birch, S., Noble, S., Killough, S., Murphy, L., Greer, M., Brodison, J., Verghis, R. and Tickle, M. (2017): Cost-effectiveness of caries prevention in practice: a randomized controlled trial. *Journal of Dental Research* **96**, 875-880.
- O'Rourke, C. A., Attrill, M. and Holloway, P. J. (1988): Cost appraisal of a fluoride tablet programme to Manchester primary schoolchildren. *Community Dentistry and Oral Epidemiology* **16**, 341-344.
- Petersen, P. E. (2003): The World Oral Health Report 2003: continuous improvement of oral health in the 21st century--the approach of the WHO Global Oral Health Programme. *Community Dentistry and Oral Epidemiology* **31**, 3-23.
- Ruff, R. R. and Niederman, R. (2018): Comparative effectiveness of school-based caries prevention: a prospective cohort study. *BMC Oral Health* 18, 53.
- Ruff, R. R. and Niederman, R. (2018): School-based caries prevention, tooth decay, and the community environment. *JDR Clinical and Translational Research* **3**, 180-187.
- Sakuma, S., Yoshihara, A., Miyazaki, H. and Kobayashi, S. (2010): Economic evaluation of a school-based combined program with a targeted pit and fissure sealant and fluoride mouth rinse in Japan. *Open Dentistry Journal* 4, 230-236.
- Savage, M. F., Lee, J. Y., Kotch, J. B. and Vann, W. F. Jr. (2004): Early preventive dental visits: effects on subsequent utilization and costs. *Pediatrics* 114, e418-423.
- Scherrer, C. R., Griffin, P. M. and Swann, J. L. (2007): Public health sealant delivery programs: optimal delivery and the cost of practice acts. *Medical Decision Making* 27, 762-771.

- Sköld, U. M., Petersson, L. G., Birkhed, D. and Norlund, A. (2008): Cost-analysis of school-based fluoride varnish and fluoride rinsing programs. *Acta Odontologica Scandinavica* 66, 286-292.
- Stephen, K. W. and Campbell, D. (1978): Caries reduction and cost benefit after 3 years of sucking fluoride tablets daily at school. A double-blind trial. *British Dental Journal* 144, 202-206.
- Tonmukayakul, U., Calache, H., Clark, R., Wasiak, J. and Faggion, C. M. Jr. (2015): Systematic review and quality appraisal of economic evaluation publications in dentistry. *Journal of Dental Research* 94, 1348-1354.
- Tonmukayakul, U., Sia, K. L., Gold, L., Hegde, S., de Silva, A. M. and Moodie, M. (2015): Economic models of preventive dentistry for Australian children and adolescents: a systematic review. Oral Health and Preventive Dentistry 13, 481-494.
- Tuominen, R. (2008): Evaluation of three methods assessing the relative value of a dental program. *Acta Odontologica Scandinavica* **66**, 82-87.
- Werner, C. W., Pereira, A. C. and Eklund, S. A. (2000): Costeffectiveness study of a school-based sealant program. ASDC Journal of Dentistry for Children 67, 93-97.
- World Health Organization. (2003): Oral health promotion: an essential element of a health-promoting school. Document 11. Geneva: WHO Information Series on School Health. 2003.
- Zabos, G. P., Glied, S. A., Tobin, J. N., Amato, E., Turgeon, L., Mootabar, R. N. and Nolon, A. K. (2002): Cost-effectiveness analysis of a school-based dental sealant program for lowsocioeconomic-status children: a practice-based report. *Journal* of Health Care for the Poor and Underserved 13, 38-48.