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Economic evaluation methods used in home visiting interventions:

A systematic search and review

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Author contributions:

CB and ZA conceived the study, CB wrote the protocol and conducted the database search, CB, HM and RO conducted the article search and data extraction, CB performed the analysis and wrote the first draft and final edits, HS, HM, RO, BH and ZA had input to drafting and critically reviewed the manuscript.

Abstract

Home-visiting interventions are used to improve outcomes for families experiencing disadvantage. As scarce resources must be allocated carefully, appropriate methods are required to provide accurate information on the effect of these programs. We aimed to investigate: economic evaluation/analysis methods used in home-visiting programs for children, young people and families, study designs and methods suitable in situations where randomised-controlled-trials are not feasible, and type of costs included in analyses, including any implementation costs stated. A systematic search and review was conducted of existing full economic evaluation/analysis methods in home-visiting programs for children, young people and/or families. We included studies published in English between January 2000 and mid-November 2020. Of 4,742 papers sourced, 60 were retained for full-text review, and 21 included. Economic-analysis methods found in the included studies were: within trial economic evaluation, economic evaluation using decision analytic modelling (i.e. cost-utility, cost-benefit analysis), cost comparison and cost-consequence. Studies incorporating return on investment and budget impact analysis were also found. Study designs suitable when randomisation was not feasible included parallel cluster randomised trials and using pre-post intervention data. Costs depended mainly on study context and only one study reported implementation costs. We hope this information will help guide future economic evaluations of home-visiting interventions.

(202/300)

Keywords: Cost-effectiveness; Economic evaluation; Methodology; Home Visiting Programs; Social Care

What is known about this topic?

- 1) Home-visiting programs are widely used to improve family welfare and reduce incidence of child maltreatment.
- 2) Measuring the cost-effectiveness of these interventions is important to allocate scarce health and social care resources.

- 3) Determining the study design and methods for measuring economic outcomes in the social care sector is challenging, especially when interventions are already implemented and/or there is no control group.

What this paper adds

- 1) We documented study designs and economic evaluation methodologies used in home-visiting programs
- 2) Where a control group was not implemented due to ethical/feasibility issues, other study designs/methods were identified and discussed.
- 3) This study should help guide researchers to choose appropriate research design and methods for evaluating home-visiting and similar programs from a health economic perspective.

1.0 Introduction

Early life experiences play a critical role in children's development, influencing health outcomes, educational achievements and well-being (Christakis and Rivara, 2010; Merrick et al., 2019). The developmental and epigenetic effects of these early childhood experiences appear to influence brain functions, thereby generating long-term effects on the individual's welfare (Panksepp, 2013). Traumatic stress associated with child maltreatment can be ongoing and, if left untreated, impacts the lives of children into adolescence and adulthood with potential for progression to intergenerational trauma once that child becomes a parent (Hatzis et al., 2017; Ringel et al., 2017). Interventions to prevent child maltreatment, support child and family welfare and resilience, and reverse negative outcomes are therefore critical, and home-visiting interventions have been widely employed in this context (Peacock et al., 2013).

Services in home visiting programs are usually provided by a multi-disciplinary team including child and family welfare professionals trained in visiting families. Many programs have been shown to influence parenting practices positively, thereby improving the child's home environment (Howard and Brooks-Gunn, 2009). Home-visiting programs can function as a preventative tool for families experiencing disadvantage by improving family functioning and outcomes for children who are considered to be at high risk of impaired

social, emotional and behavioural development (Dalziel and Segal, 2012; Peacock et al., 2013).

The evidence regarding which outcomes are positively impacted by home-visiting programs, however, is mixed. In two meta-analyses of home-visiting program efficacy, researchers found small but significant and positive results, but with high variability between studies in terms of outcomes and populations (Sweet and Appelbaum, 2004; Filene et al., 2013). For instance, outcomes from one meta-analysis found that home-visiting programs had a positive effect on maternal life-course, parent skills and behaviours, and child cognition but did not show an effect for child maltreatment or physical health (Filene et al., 2013). In contrast, other reviews found that the benefits of these programs were in mitigating child maltreatment and disadvantage alongside improvements in children's cognition and behaviour (Dalziel and Segal, 2012; Avellar and Supplee, 2013; Peacock et al., 2013). Peacock and colleagues found that dose (for instance, early initiation in pregnancy lowering incidence of low birth-weight) (Lee et al., 2009) and retention (many studies had attrition of over 18% of participants) still needed to be understood (Peacock et al., 2013).

Whilst there has been a significant investment globally in home-visiting programs, research on the return-on-investment from these programs has been limited (Dalziel and Segal, 2012). In their systematic review of the cost-effectiveness of home-visiting programs, Dalziel and Segal found high variability, and some programs were costly with little benefit. In programs deemed not cost-effective, program components were variable, such as length of service delivery, provision by layperson or professional, and commencement in the pre- or post-natal period (Dalziel and Segal, 2012). The more cost-effective programs targeted high-risk populations (such as young, first-time mothers experiencing disadvantage) used trained home-visiting professionals and delivered services in addition to the home-visiting program.

In a review of the implementation aspects of home-visiting programs, important characteristics found were including a comparator, program fidelity, and training and supervision (Casillas et al., 2016). Many of the programs included in this review were successful in improving outcomes for families, but the way programs were implemented was crucial in determining their success. High dropout rates significantly affected implementation of home-visiting programs (Brand and Jungmann, 2014), and an investigation of study fidelity found that programs work differently for different clients, and that it is important to understand which program component is working for whom (Goldberg, Bumgarner and

Jacobs, 2016). Adaption of the intervention to the needs of the individual is another a critical function performed by home-visitors; and thus, effective staff training was an important aspect of achieving intervention fidelity (Saïas et al., 2012).

1.1 Methodological considerations in economic evaluation

Methodologically rigorous economic evaluation is required to make optimal decisions about which programs to fund. Evaluating the costs and benefits in the social care sector, though, is relatively uncommon, and methodology has mostly been taken from healthcare (Olsson, 2011; Weatherly et al., 2017; Deidda et al., 2019). The term ‘Social care’ (or social welfare) is used here to delineate all types of personal care and/or practical assistance or support for children, young people and adults.

Study design and methods issues in economic evaluation may be apparent at the trial stage (in determining efficacy) and at the implementation stage (in determining effectiveness). At the trial stage, it is important that cost-effectiveness evaluations of home-visiting programs can produce robust estimates of effect, as although randomised controlled trials (RCT) are generally considered a preferred form of evidence, they are often difficult to conduct in public health or social care settings (Edmunds et al., 2018). Randomisation of individual participants to intervention and control groups is sometimes neither feasible (where randomisation is not possible at an individual level) nor ethical (where withholding treatment from one group is not considered appropriate), and alternate methods may need to be employed (Deaton and Cartwright, 2018). Programs that are poorly implemented (or differently implemented compared to initial trial) may not produce the expected benefits, and a rigorous understanding of how research is applied into practice and policy is essential. At the implementation stage, building the evidence of program effectiveness is critical; however, in published evaluations, implementation quality is often absent, under-reported or highly variable (Kemp et al., 2019).

Resource limitations on health and social care budgets can be a powerful motivator to encourage the use of economic analysis (Hoomans and Severens, 2014); however, just as aspects of implementation are often missing from evaluations, economic evaluations are often missing from implementation studies (Eisman et al., 2019). Given that economic evaluation is integral to understanding the costs and consequences of patient outcomes, it is important that implementation scientists work closely with health economists to consider the economic aspects of public health interventions (Roberts, Healey and Sevdalis, 2019). Thus, an

understanding of the separate costs incurred in implementation is valuable. It is also important that health economists determine appropriate research design and methods that are fit-for-purpose for evaluation in social care situations. Guidance on methodology for measuring economic outcomes in social care settings must reflect what is practical and appropriate (Weatherly et al., 2017). Further, it is important that researchers have a good understanding of what costs are important to include in an economic analysis in this field, including the cost of implementation, if the intervention is to be employed in a real world setting. It is within this context that we aimed to investigate suitable research designs and methods for the economic evaluation of home-visiting interventions.

1.2 Aims

The overall aim of the current study was to investigate study design and methods that have been used in economic evaluations of home-visiting interventions for children, young people and families, so as to provide guidance for researchers working in this area. Our specific aims were to: 1) determine study designs and types of economic evaluation methods used in home-visiting interventions for children, young people and families; 2) investigate what study designs were used in circumstances where it was not possible to randomise participants; 3) describe types of cost data collected; and 4) describe costs relating to implementation of home-visiting programs that may have been included (implementation costs defined here as the actual costs of program implementation).

2.0 Methodology

This study used a systematic search strategy to source suitable studies for the review. A “systematic search and review” approach is suitable when the focus is on determining what is known on a subject as well as recommendations for practice, whilst using a comprehensive search strategy (Grant and Booth, 2009). Prior to starting the review, a detailed protocol was developed and adhered to by the authors. Because we were interested in the study design and methods used for economic analysis and did not include clinical outcomes, the review was not eligible for PROSPERO registration (Chandler et al., 2017; Luhn et al., 2017).

2.1 Information sources

Articles were sourced through a systematic search of five academic databases: EMBASE, Medline and PsycINFO via Ovid, and Econlit and CINAHL via EBSCO. A grey

literature search was conducted on the Cochrane library, PDQ-evidence (PDQ- Evidence Database, 2018), Epistemonikos (El-Khayat, 2017) and Google. These databases have been considered suitable for retrieving papers on economic evaluations (Thielen et al., 2016). The search strategy was designed to detect all studies published in English from January 2000 to mid-November 2020. Search terms were developed collaboratively (CB, HM, RO, and ZA) for keywords and medical subject heading terms relating to four concepts: 1) evaluation type; 2) intervention topic; 3) population; and 4) paper presented an intervention or program (see Appendix A).

2.2 Eligibility criteria

For inclusion, studies needed to be: 1) a full economic evaluation including within trial or economic evaluation using decision analytic modelling (cost-effectiveness, cost-utility, cost-benefit and cost minimisation analysis), budget impact analysis (or return-on-investment), or any type of cost comparison analysis); 2) a home-visiting program or intervention (a program or intervention that was delivered in the service user's home); 3) a population of children, young people and/or families; and 4) a program or intervention. In brief, cost-effectiveness analysis estimates how much it costs to gain a unit of health outcome (e.g. cost per event prevented or averted), cost-utility analysis estimates how much it costs to gain a quality-adjusted-life-year (QALY), cost-benefit analysis is a method that values outcomes in monetary units, and cost-minimisation analysis select the least costly alternatives when the outcomes of two or more interventions are identical. Budget impact analysis is an economic assessment to estimate the financial consequences of implementing an intervention, and return-on-investment analysis measures the efficiency of an investment, where the benefit is divided by the intervention cost.

2.3 Study selection

Articles were extracted from databases into the EndNote referencing program, and screened using the Covidence tool for systematic review screening program (Veritas Health Innovation, 2018). Titles and abstracts were screened against the inclusion criteria by two of three authors (CB, HM, RO). Eligible studies were accessed in full text, and read and discussed by authors until included papers were mutually agreed upon. Any differences of opinion were resolved through discussion and consensus, and if required, authors could consult with a third person.

2.4 Quality of reporting

The Consolidated Health Economic Evaluation Reporting Standards (CHEERS) checklist (Husereau et al., 2013) was used to determine quality of reporting. Using a standard test of reporting standards for health economics papers is recommended for systematic reviews (Wijnen et al., 2016). We expected that three items from the CHEERS checklist (population type, intervention/comparator and outcome measure) would be met by all studies. For studies without modelling, items pertaining to modelling studies were excluded, and percentage scores were calculated for relevant questions.

2.5 Data extraction and analysis

In this study, we were interested in which study designs and methods were presented, rather than in the study outcomes; therefore, narrative synthesis was the most suitable analysis technique. We began this process by extracting relevant data into a table that included aims, population, outcomes, intervention, comparator, perspective and study design (as shown in Table 1). We then extracted data from each study specific to the study design, (use of modelling, study design, time horizon and main costs) by type of economic evaluation method chosen (as shown in Table 2). Subsequently, we narratively synthesised papers by economic analysis method type (Ebied et al., 2013).

3.0 Results

3.1 Study selection

After duplicates were removed, 4,134 title/abstracts were assessed, and 60 studies were retained for full-text review, of which 21 met the inclusion criteria. The PRISMA chart is presented in Figure 1. Reasons for exclusion at the full-text review stage were: not a home-visiting program, outcomes were healthcare rather than social care, not an economic evaluation (See Appendix B).

3.2 Study and patient characteristics of included papers

Most of the included papers were from the United Kingdom (UK) (seven papers) (Barlow et al., 2007, 2019; McIntosh et al., 2009; Sharac et al., 2011; Deidda et al., 2018; Sonuga-Barke et al., 2018; Bell et al., 2019) , Canada (three papers) (Browne et al., 2001; Au et al., 2006; Thanh et al., 2014) the United States (US) (four papers) (Olds et al., 2011;

Burwick et al., 2014; Wilson et al., 2016; Ammerman et al., 2017; Kuklinski, Crowley, et al., 2020; Kuklinski, Oxford, et al., 2020). There were two studies from South America (Chile and Bolivia) (Aracena et al., 2009; Barger et al., 2017), and one each from Africa (Ethiopia) (Mathewos et al., 2017) Australia (Dalziel et al., 2015) and the Netherlands (Dijkstra et al., 2018).

Home-visiting programs were aimed at mothers (Au et al., 2006; Barlow et al., 2007; Aracena et al., 2009; McIntosh et al., 2009; Olds et al., 2011; Ammerman et al., 2017; Barger et al., 2017; Mathewos et al., 2017), parents (including adoptive and sole-parents) (Browne et al., 2001; Sharac et al., 2011; Thanh et al., 2014; Dalziel et al., 2015; Deidda et al., 2018; Dijkstra et al., 2018; Sonuga-Barke et al., 2018; Barlow et al., 2019), families (Burwick et al., 2014; Bell et al., 2019; Kuklinski, Crowley, et al., 2020; Kuklinski, Oxford, et al., 2020) or young people (Wilson et al., 2016). All articles were peer reviewed except for two studies, a US Department of Justice report by Olds et al, 2001 (Olds et al., 2011), and a report by Mathematica Policy Research (Burwick et al., 2014). All studies except one (Aracena et al., 2009) performed some type of sensitivity analysis. Three studies used modelling, including Markov modelling (Ammerman et al., 2017), decision tree modelling (Thanh et al., 2014) and a decision support tool for determining cost to benefit of programs (Wilson et al., 2016). Information on study aim, population, outcomes, intervention and comparator, study perspective and study design in the included studies is presented in Table 1.

3.3 Quality of reporting

Thirteen of the included papers had CHEERS checklist (Husereau et al., 2013) scores of 90% or more (McIntosh et al., 2009; Thanh et al., 2014; Burwick et al., 2014; Dalziel et al., 2015; Ammerman et al., 2017; Mathewos et al., 2017; Sonuga-Barke et al., 2018; Deidda et al., 2018; Dijkstra et al., 2018; Barlow et al., 2019; Bell et al., 2019; Kuklinski, Crowley, et al., 2020; Kuklinski, Oxford, et al., 2020), suggesting that most of the important elements had been reported. In seven papers, scores ranged between 65% and 89% (Browne et al., 2001; Au et al., 2006; Barlow et al., 2007; Olds et al., 2011; Sharac et al., 2011; Wilson et al., 2016; Barger et al., 2017), suggesting that some reporting elements were lacking, and one study scored just lower than 50% (Aracena et al., 2009), suggesting low reporting of economic information. Studies with lower scores tended to have not measured uncertainty

(sensitivity analysis etc.) and many papers did not include a statement addressing conflict of interest.

3.4 Economic evaluation methods used in the studies

Information on the methodological aspects of the included papers including whether the study was within trial economic evaluation or used decision modelling techniques, type of economic method, the study design, time horizon and main types of cost are presented in Table 2. In line with our first aim, types of economic methods as pertaining to home-visiting programs are then discussed.

3.4.1 Cost comparison

Cost comparison studies are a relatively simple form of economic evaluation, where costs are compared between groups, but without reference to outcomes (Drummond et al., 2015) Four included studies presented results as a comparison of costs. Au and colleagues' (Au et al., 2006) aimed to compare costs for three trial arms: standard care costs plus added costs from the two interventions (standard care plus nurse visits; standard care plus nurse visits plus home-visits). Groups were compared regarding medical costs (with and without hospital costs) forming a relatively simple calculation.

The second cost comparison study investigated costs of supplying home-visiting support to families where the implementing agency considered a child to be at risk of maltreatment (Burwick et al., 2014). Costs were assessed for five home-visiting programs over 25 agencies and 13 states in the US. Resource costs to deliver the program were determined for total agency costs and per family. These costs included resources utilised, staff time, and services used per family. By comparing resource use between different types of home-visiting program and by agency this report was able to identify cost differences relevant to policy decision makers. The methodology was appropriate for this analysis which examined multiple interventions and providers.

In a third paper, the costs of three scale-up scenarios were assessed (Barger et al., 2017). Barger and colleagues found that high costs per home-visitor combined with low coverage of the mother/child dyad impeded the study's aim to lowering infant mortality. These examples show how cost comparison methodology can be usefully applied in circumstances where cost differences alone will enable effective decision-making. The paper

by Kuklinski and colleagues (Kuklinski, Crowley, et al., 2020) presented a detailed cost-analysis of a four-year, home-visiting intervention for high-risk families from a societal perspective. The total cost of delivering the program were calculated, and main drivers of costs were disaggregated by age, major intervention activity and key resources used. Key components were varied in sensitivity analysis. The detailed breakdown of costs and across more- and less-responsive families allowed an in-depth look at cost drivers.

3.4.2 Cost-consequence analysis

A cost-consequence analysis (Gray et al., 2011; Drummond et al., 2015), is a form of economic evaluation in which outcomes are reported separately from costs, and there is no specific preference for one outcome measure over another. A paper by Bell and colleagues (Bell et al., 2019) examined the costs and consequences of an intervention where participants received home visiting services from a nurse during pregnancy, with a two year follow up period. Resource use was averaged and compared between trial arms for each item, and consequences were the specified outcome measures. Costs and consequences were then presented together in a table and discussed separately without presenting an overall outcome statistic.

A paper by Browne and colleagues (Browne et al., 2001) investigated the provision of care to sole-support parents requiring social assistance. This study presented differences between effect and cost at various follow-up times over a range of outcome measures, but did not directly compare costs to effects. The paper could thus be considered to be a cost-consequence analysis, though it was not stated as such. Costs measured included the costs of proactively providing a comprehensive support package and reduction of costs associated with higher rates of families exiting social-support per annum. Although the reader is guided to a conclusion in the discussion, there is not one clear outcome measure that dominates this evaluation.

In the protocol paper by Diedda and colleagues (Deidda et al., 2018) the authors set out their future analysis plans. This paper presented a range of proposed economic analyses aimed at effective understanding of a complex intervention to improve infant mental health, demonstrating how these methodologies could work together to support a comprehensive understanding of the costs and consequences of the intervention. The economic evaluation planned for this paper encompassed cost-consequence, cost-utility, cost-effectiveness as well as a longer-term decision model.

3.4.3 Within-trial cost-effectiveness

We found that more than half the included papers were within-trial cost-effectiveness analyses (Barlow et al., 2007, 2019; Aracena et al., 2009; McIntosh et al., 2009; Sharac et al., 2011; Thanh et al., 2014; Dalziel et al., 2015; Mathewos et al., 2017; Dijkstra et al., 2018; Sonuga-Barke et al., 2018). Cost effectiveness analysis is one of the most commonly used economic analysis methodologies in healthcare, and is concerned with the differential comparison of costs and effects by calculating an incremental cost-effectiveness ratio (ICER) (Elliot and Payne, 2005). Time-horizons in the included cost-effectiveness studies were generally short: three studies were for six months (Sharac et al., 2011; Dalziel et al., 2015; Sonuga-Barke et al., 2018), four studies were for 12-months, (McIntosh et al., 2009; Mathewos et al., 2017; Dijkstra et al., 2018; Barlow et al., 2019), one study was for 15-months (Aracena et al., 2009) and one for 18-months (Barlow et al., 2007). Short time-horizons make it difficult to determine longer-term benefits. Most studies did not require discounting, being less than 12 months. Six studies were from a societal perspective (Barlow et al., 2007; Dalziel et al., 2015; Dijkstra et al., 2018; Bell et al., 2019; Kuklinski, Crowley, et al., 2020; Kuklinski, Oxford, et al., 2020), three studies included both societal and healthcare payer perspectives (McIntosh et al., 2009; Sonuga-Barke et al., 2018; Barlow et al., 2019), one study was from a healthcare provider perspective only (Mathewos et al., 2017) and two did not state the perspective (Sharac et al., 2011; Ammerman et al., 2017).

Interventions in most studies were compared to standard care, apart from Barlow and colleagues 2019 (Barlow et al., 2019) where treatment as usual was not standardised in order to better represent real world services, and Mathewos and colleagues (Mathewos et al., 2017) where standard care was improved, and then compared to improved standard care plus home-visiting. In the paper by Sharac and colleagues (Sharac et al., 2011), two interventions were compared to each other rather than care as usual. The paper by Sonuga-Barke and colleagues (Sonuga-Barke et al., 2018) was planned as a cost-effectiveness study, but as there was no statistically significant difference in outcomes between groups, only a comparison of costs was documented. When effects are not included because there are no difference in outcomes between groups, this type of study is generally termed a cost-minimisation study. In the study by Kuklinski and colleagues (Kuklinski, Crowley, et al., 2020), only the intervention group was investigated.

3.4.4 Cost-utility

A cost-utility analysis is a form of cost-effectiveness analysis where health outcome is captured in terms of quality-adjusted-life-years (QALYs), a preference-based outcome which incorporates both the quantity and quality of life. QALYs are calculated by multiplying utility scores (generated from quality of life questionnaires) by years of life lived to account for impairment in quality of life due to ill health. Although this is a widely used economic evaluation technique as it enables comparison across different interventions, only one study here used cost-utility analysis, reflecting the possible limitations of using this method in social care settings. Ammerman and colleagues (Ammerman et al., 2017) investigated the effectiveness of an in-home cognitive behavioural therapy program for low-income mothers diagnosed with a major depressive disorder. Inputs were collected from a clinical trial, QALYs were measured using the EuroQoL (EQ-5D), and data were analysed over a three-year time horizon using Markov modelling. Markov modelling is used to represent changes over time in and out of health states. ICERs were compared to three willingness-to-pay thresholds per QALY, and one-way and probabilistic sensitivity analyses were performed to test the robustness of the base-case results.

3.4.5 Cost-benefit Analysis

In cost-benefit analysis (term used here interchangeably with benefit-cost analysis), a monetary value is estimated for the both resource use and outcomes. For instance, researchers may attempt to place a monetary value per case avoided, and whether this monetary value is compensated for by the resource costs used (Briggs, Sculpher and Claxton, 2006). Cost-benefit analysis can permit comparisons between different areas of expenditure such as health, education, environment and transport (Gray et al., 2011). A cost-benefit study is differentiated from cost-utility and cost-effectiveness analysis as it measures inputs and outputs in the same units (currency), and thus ICERs are not required (Elliot and Payne, 2005).

The study by Wilson and colleagues (Wilson et al., 2016) presented a decision support tool that could be used to determine the cost-benefit of a range of interventions. The paper included a family-centred in-home program for at risk young people as an illustrative case-study. The primary focus of this paper was on the development of the decision support tool methodology, and the illustrative case study was only briefly discussed. Costs were compared in terms of cost-offsets from health and justice outcomes. The paper by Thanh and colleagues (Thanh et al., 2014) also incorporated a cost-benefit analysis. Here, the number of

cases prevented was monetized using the incremental lifetime cost per case. This study employed decision-tree modelling to estimate the harm reduction of a home-visiting intervention for reducing cases of alcohol exposed births amongst high-risk women. Effect inputs were from program evaluations, and costing was calculated for the cost per case of foetal alcohol spectrum disorder. Of note, all three of the cost-benefit analysis studies included here were modelling studies.

Kuklinski and colleagues (Kuklinski, Oxford, et al., 2020) used benefit-cost analysis to assess a strengths-based, 10-session home visiting program. Costs included the costs of implementation in the intervention arm of the trial compared to the resource and referral arm. Monetised effects were the cost of out-of-home placements and the implicit effects of child abuse and neglect. Benefit-cost results were presented as a ratio of expected benefits to expected costs, reflecting the probable monetary return per investment. Variation was calculated using Monte Carlo simulations by varying input parameters. Child abuse and neglect effect size was plotted against costs and investment risk percentages. Outcomes compared the costs of the intervention to a range of systems-level savings.

3.5 Expanded economic analysis

To estimate the financial consequences of adopting an intervention, further analysis in the form of budget impact analysis or return-on-investment analysis can be performed in addition to any of the previously discussed economic analysis methodologies. The first part of the economic evaluation investigates the relative value of an intervention, whilst a secondary analysis (such as budget impact analysis) will determine whether an intervention that has been deemed to be high value is affordable within the available budget (U.S. Department of Veterans Affairs, 2019).

3.5.1 Return-on-investment

Return-on-investment can be expressed as a net return (positive or negative) or as a ratio for comparison between various interventions (Masters et al., 2017). The included paper by Olds and colleagues (Olds et al., 2011) considered the net impact on costs from both societal and healthcare payer perspectives. The authors considered a range of outcome measures (such as smoking during pregnancy, complications/miscarriages, maternal earnings, domestic violence, maternal depression and services to children with developmental difficulties) expressed in monetary values and obtained from published literature. The Present Net Value of estimated total savings per mother was calculated, compared to the program

cost for each participant, and presented as a benefit-cost ratio (suggesting that the cost-benefit and return-on-investment ratio terms tend to be used interchangeably). Sensitivity analyses were used to confirm the base-case return-on-investment. Wilson and colleagues (Wilson et al., 2016) calculated the program costs associated with provision of the program to 1000 families, and then compared these to the price of juvenile detention and out-of-home placement. Predictions were then calculated for inclusion of all families that were involved with juvenile detention or out-of-home placement in the program, and further scenario analyses for reduced cases of school absence, school drop-out and measure of health.

3.5.2 Budget impact analysis

A budget impact analysis assesses the financial consequences of choosing an intervention, and, like return-on-investment, is usually performed in addition to a cost-effectiveness analysis, from the perspective of the payer. The actual number of people affected by a program is not measured during a cost-effectiveness analysis, but a budget impact analysis can determine whether the intervention is affordable at the population level.

Whilst none of the included papers explicitly state that they were presenting a budget impact analysis, in the paper by Barger and colleagues (Barger et al., 2017) the additional annual costs to the municipality of an intervention to improve services to an under-served population of women during pregnancy in Bolivia were presented. Annualised average costs per mother for the intervention arms were compared with costs of the comparison group, and the feasibility of the study was measured at scale in terms the implications on finances and human resources. By varying service levels, three scale-up scenarios were modelled to determine costs per mother, and cost per home-visit from the perspective of the country's Ministry of Health. These costs were further disaggregated by calculating the percentages accountable for intervention design, set-up and implementation. The main stated aim for the intervention was to reduce neonatal mortality; however, this effect was not measured, and the paper mainly focused on the budgetary impacts of the intervention.

There was some mention of budget impact analysis in the other included papers. In the paper by Mathewos and colleagues (Mathewos et al., 2017) public health expenditure of GDP per capita by World Health Organization thresholds was measured. The economic costs of the intervention were expressed as a dollar price per capita, and as a percentage of the public health expenditure per capita, to indicate the implication of the program on the jurisdiction's budget. Deidda and colleagues (Deidda et al., 2018) briefly mentioned that the

‘social rate of return’ of the intervention studied was likely to be high and to justify additional costs required by the intervention, but this analysis was not included in this protocol paper.

3.6 Study design

Our second aim was to investigate study designs used where randomisation was not appropriate. Fifteen of the 21 studies used data from RCTs where participants were randomised into intervention or control groups at recruitment (Browne et al., 2001; Au et al., 2006; Barlow et al., 2007, 2019; Aracena et al., 2009; McIntosh et al., 2009; Sharac et al., 2011; Dalziel et al., 2015; Ammerman et al., 2017; Sonuga-Barke et al., 2018; Deidda et al., 2018; Dijkstra et al., 2018; Bell et al., 2019; Kuklinski, Oxford, et al., 2020). In Kuklinski et al (Kuklinski, Crowley, et al., 2020) the study was based on an RCT; however only the intervention group was investigated. In the study by Old and colleagues (Olds et al., 2011) it was unclear whether randomisation had occurred; the paper was not peer reviewed and lacked much methodological information. Five studies supplied information on situations where randomisation was not feasible. In the study by Burwick and colleagues, cost comparisons only were conducted directly between agencies and interventions without control groups, hence randomisation did not occur (Burwick et al., 2014). (The Burwick study was also not peer reviewed, but was a useful overview and comparison of costs that would have been useful for program administrators).

Two studies used parallel cluster RCT designs where randomisation was by geographical area (Barger et al., 2017; Mathewos et al., 2017). The study by Mathewos and colleagues used a cluster randomised trial, and randomly allocated 22 clusters in two regions to intervention and control (Mathewos et al., 2017). The economic evaluation method used in this paper was cost-effectiveness analysis. In the paper by Barger and colleagues, interventions were conducted in two municipalities, and were compared to two control municipalities. The authors do not state how the regions were chosen, and there was no information supplied in the paper on the comparability of the different regions (Barger et al., 2017). The economic evaluation method used in this paper was a cost-comparison analysis.

Two studies used information from pre-post intervention study designs to estimate the impact of the intervention (Thanh et al., 2014; Wilson et al., 2016). Thanh and colleagues (Thanh et al., 2014) utilised pre- and post-intervention comparisons across several sites; however, information on the inputs in this paper was patchy, and source material for effects could not be found. In the paper by Wilson and colleagues (Wilson et al., 2016), a pre- and

post-intervention follow-up design was used sourcing data from a range of previous studies that had implemented the intervention. Again, information on the how source studies were conducted was sparse and we were unable to locate source documents. Neither paper provided a commentary on the validity of using pre-post intervention data for this input parameter. Both studies used economic modelling.

3.7 Comparison groups

Fourteen included studies compared the intervention to care-as-usual or standard care (Browne et al., 2001; Au et al., 2006; Barlow et al., 2007, 2019; Aracena et al., 2009; McIntosh et al., 2009; Olds et al., 2011; Martin and Lotspeich, 2014; Thanh et al., 2014; Dalziel et al., 2015; Barger et al., 2017; Sonuga-Barke et al., 2018; Deidda et al., 2018; Bell et al., 2019; Kuklinski, Crowley, et al., 2020). Four studies compared two different home intervention strategies (Sharac et al., 2011; Ammerman et al., 2017; Mathewos et al., 2017; Dijkstra et al., 2018). One study compared to a group receiving resources and referral (Kuklinski, Oxford, et al., 2020). One study only investigated costs, and comparisons were made across a range of interventions and agencies, but without direct control groups of standard or usual care (Burwick et al., 2014). One study compared an intervention to cost offsets from detention and out-of-home care placement (Wilson et al., 2016). Of note, the intervention group in the study by Barlow and colleagues (Barlow et al., 2019) was compared to a treatment as usual group intended to reflect a real world scenario. As this was a pragmatic trial, the researchers did not attempt to standardise the control group, allowing a comparison between the intervention and currently offered services.

3.8 Costs

Our third aim was to examine included costs. Overall, the two main costs types were staffing and hospital costs. Highest costs related to employment of therapists/staff were found in nine papers (McIntosh et al., 2009; Sharac et al., 2011; Burwick et al., 2014; Dalziel et al., 2015; Barger et al., 2017; Mathewos et al., 2017; Sonuga-Barke et al., 2018; Kuklinski, Crowley, et al., 2020; Kuklinski, Oxford, et al., 2020). For instance, in the Kuklinski et al benefit-cost study (Kuklinski, Oxford, et al., 2020), over half of costs were for intervention delivery, followed by training and ongoing support. The study by Burwick and colleagues found that the three top main expense were practitioner salaries, fringe benefits, and indirect (overhead) costs. Hospital related costs (e.g. entering treatment for major depressive disorder, postpartum overnight stays or emergency admission) were the highest cost groups found in

four papers (Au et al., 2006; Ammerman et al., 2017; Dijkstra et al., 2018; Bell et al., 2019). Thanh and colleagues identified the main costs as cost of a case of foetal alcohol spectrum disorder avoided (Thanh et al., 2014), and Olds and colleagues as loss of earnings for mothers (Olds et al., 2011). Many of the papers were a write up of an RCT with a brief economic analysis tagged on, and these studies tended to lack specific information on cost breakdowns (Browne et al., 2001; Barlow et al., 2007, 2019; Aracena et al., 2009; Wilson et al., 2016). The paper by Deidda and colleagues was only a protocol paper, and therefore did not present cost data (Deidda et al., 2018).

3.9 Implementation costs

Information on implementation costs were minimal in most of the included papers, as per our final aim, with only two papers directly discussing implementation costs. The paper by Barger and colleagues specifically calculated implementation costs, dividing intervention costs into design, set up and implementation (Barger et al., 2017). Kuklinski and colleagues (Kuklinski, Crowley, et al., 2020) discuss implementation at some length, firstly by describing how interventionists received continuing technical and consultation support as well a training update every year. Importantly, this study separates out costs that would not occur in 'real-world implementation, breaking down total costs into four categories: implementation, oversight, overhead, and training. Implementation was also broken into five categories: direct contact, collateral contact, preparation, scheduling and failed contact.

Minor references to implementation in the other studies included the following. Au and colleagues cautioned against widespread implementation of an intervention based on a single evaluation study (Au et al., 2006). Ammerman and colleagues stated in the paper's conclusion that the intervention was economically sound, and warranted implementation (Ammerman et al., 2017). Mathewos and colleagues also mentioned implementation in the conclusion of the paper, stating that implementation on a larger scale would be valuable, but that different contexts needed to be taken into account (Mathewos et al., 2017). Sonuga-Barke and colleagues noted that some of the elements of the implementation of the intervention may not have been optimal (Sonuga-Barke et al., 2018). Burwick and colleagues discussed variations in implementation costs regarding whether the intervention was new or ongoing (Burwick et al., 2014).

4.0 Discussion

We aimed to investigate economic evaluation methods that have been used in home-visiting programs for children, young people and families in social care. Whilst within-trial cost-effectiveness studies were the most utilised methods, there were valid reasons for using other methods, dependent on the type of intervention and population. The advantages of using cost-consequence and cost-benefit analysis, for instance, were that a broad range of outcomes could be assessed and interpreted. Cost-benefit analysis was cited by authors Wilson and colleagues (Wilson et al., 2016) as being a suitable tool specifically for families and children, and authors referred to the National Research Council workshop's increasing focus on cost-benefit analysis for influencing policy decisions regarding children, young people and families, despite general preferences by health economists to use cost-effectiveness and cost-utility methodologies (National Research Council, 2014). In addition, including a return-on-investment or budget impact analysis as part of the overall methodology provided an understanding of the effect of financial consequences on the available budget; however only one study included this information (Barger et al., 2017).

Our second aim was to investigate methods used in situations where it was not reasonable to randomise participants at recruitment. We found two pragmatic study designs that may be used when a traditional RCT design may not be possible: parallel cluster RCTs and analysing using pre- and post-intervention data. Another methodology type that may be used, though not found in any of the included papers, is the step-wedge design, which is mainly used for randomisation by clusters. In a step-wedged design, there is a preliminary period where no interventions are implemented, then the intervention is introduced one group at a time at regular intervals (steps) so that the control groups become intervention groups as the intervention is introduced (Hemming et al., 2015). Eventually all groups have become intervention groups, and data is collected throughout this process. This design is good for equity, as everyone receives intervention at the end. Step-wedged cluster RCTs have generally been used for service delivery evaluation or to test policy changes (Hemming et al., 2015) and have been effective in evaluating system-wide trauma informed care models in out-of-home-care settings, where effective methodologies for analysis had otherwise been lacking (Bailey et al., 2019)].

Another methodology that could be considered when lacking a control group is to model the intervention effect, then work out the threshold improvement needed for the intervention to be cost-effective. For instance, in a study that was located in the course of the review but subsequently excluded as it was not strictly a home-visiting program, control

group estimates were modelled. This paper analysed the effects of an intervention to prevent sex trading in adolescents using a benefit-cost framework whereby inputs were estimated from a case study (Martin and Lotspeich, 2014). The authors account for ‘empirical ambiguity’ by modelling a range of intervention effects. These modelled inputs were utilised to determine the likely outcomes from the intervention compared to potentially avoided harms. Similarly, the included study by Kuklinski and colleagues (Kuklinski, Oxford, et al., 2020) modelled a range of possible child abuse and neglect outcomes and the associated benefits and costs to account for uncertainty.

Natural experiments, defined here as naturally occurring situations where a subsection of the population has had different exposure to a particular factor, also offer an alternative to RCTs (Deidda et al., 2019). Natural experiments are not randomised, as the groups are defined by observable factors associated with the outcome of concern, and methodologies to address issues of selection bias in natural experiments require consideration (Deidda et al., 2019). Unfortunately, there were no natural experiment examples in this review.

Our third and fourth aims were to investigate costs of the interventions and implementation costs where/if stated. Costing interventions in social care can be highly complex, due to the multiple difficulties families under duress may experience (Mcdermid and Holmes, 2013). Main cost drivers varied considerably and were dependent on the outcomes measured. Staff employment was often the overall biggest driver of costs in studies where staff delivered the intervention. Despite the strong need for good evidence on implementation, we found little evidence of the separate costs of implementation in the included studies. Although implementation science has made significant progress in recent years in developing strategies to improve the uptake of evidence-based practices, progress on the consideration of economic outcomes in implementation has been slow (Eisman et al., 2019).

4.1 Strengths and limitations

This review used rigorous systematic search methodology to search for papers in multiple academic and grey literature databases; however, relevant papers may have been missed. Although we used rigorous systematic search methodology, followed recommendations for reviews of health economics papers (Thielen et al., 2016), and searched five academic databases and four grey literature databases, it is possible that we have missed relevant papers. We aimed to only source social care papers and exclude papers on health

topics, however the line between health and social care is unclear. We excluded interventions where the main focus was on health outcomes in hospital and general practice settings and these papers may have included relevant social care outcome measures. Quality of reporting was reasonable, with only one paper scoring significantly low and others missing several quality metrics. It was often hard to access the source documents referenced in the cost-effectiveness papers for parameters that were included in modelling studies, meaning that it was more difficult to assess the study inputs in these papers.

4.2 Conclusion

There are a number of valid research designs for economic evaluations of home-visiting programs, and some of these options are amenable to situations when a randomised controlled trial may not be feasible or ethical. There is a need for well-blended approaches that include healthcare, societal and patient perspectives and serve diverse stakeholders. Evaluating the economic impact of an intervention is important so that policy makers can make effective decisions on where to direct funds when resources are scarce. Home-visiting programs have been found to be effective in a range of social care situations, and an understanding of how to evaluate these programs effectively from an economic perspective is critical. This study should help researchers to improve their understanding of some of the possible approaches when conducting an economic evaluation in the area of home-visiting programs or related interventions and guide them to choose a methodology that is appropriate for their specific intervention.

References

Ammerman, R. T. et al. (2017) 'Cost-effectiveness of In-Home Cognitive Behavioral Therapy for low-income depressed mothers participating in early childhood prevention programs', *Journal of Affective Disorders*. Elsevier, 208(November 2016), pp. 475–482. doi: 10.1016/j.jad.2016.10.041.

Aracena, M. et al. (2009) 'A cost-effectiveness evaluation of a home visit program for adolescent mothers', *Journal of Health Psychology*, 14(7), pp. 878–887. doi: 10.1177/1359105309340988.

Au, F. et al. (2006) 'Does supplementary prenatal nursing and home visitation reduce healthcare costs in the year after childbirth?', *Journal of Advanced Nursing*, 56(6), pp. 657–

668. doi: 10.1111/j.1365-2648.2006.04036.x.

Avellar, S. A. and Supplee, L. H. (2013) 'Effectiveness of home visiting in improving child health and reducing child maltreatment', *Pediatrics*, 132(SUPPL.2). doi: 10.1542/peds.2013-1021G.

Bailey, C. et al. (2019) 'Systematic review of organisation-wide, trauma-informed care models in out-of-home care (OoHC) settings', *Health and Social Care in the Community*, 27(3), pp. e10–e22. doi: 10.1111/hsc.12621.

Barger, D. et al. (2017) 'Bolivia programme evaluation of a package to reach an underserved population: Community-based maternal and newborn care economic analysis', *Health Policy and Planning*, 32, pp. i75–i83. doi: 10.1093/heapol/czv133.

Barlow, J. et al. (2007) 'Role of home visiting in improving parenting and health in families at risk of abuse and neglect: Results of a multicentre randomised controlled trial and economic evaluation', *Archives of Disease in Childhood*, 92(3), pp. 229–233. doi: 10.1136/adc.2006.095117.

Barlow, J. et al. (2019) 'A randomized controlled trial and economic evaluation of the Parents Under Pressure program for parents in substance abuse treatment', *Drug and Alcohol Dependence*. Elsevier, 194(November 2018), pp. 184–194. doi: 10.1016/j.drugalcdep.2018.08.044.

Bell, K. et al. (2019) 'Costs and consequences of the Family Nurse Partnership (FNP) programme in England: Evidence from the building blocks trial', *F1000Research*, 8, pp. 1–16. doi: 10.12688/f1000research.20149.1.

Brand, T. and Jungmann, T. (2014) 'Participant characteristics and process variables predict attrition from a home-based early intervention program', *Early Childhood Research Quarterly*. Elsevier Inc., 29(2), pp. 155–167. doi: 10.1016/j.ecresq.2013.12.001.

Briggs, A., Sculpher, M. and Claxton, K. (2006) *Decision modelling for health economic evaluation*. Oxford: Oxford University Press.

Browne, G. et al. (2001) 'When the bough breaks: Provider-initiated comprehensive care is more effective and less expensive for sole-support parents on social assistance', *Social Science and Medicine*, 53(12), pp. 1697–1710. doi: 10.1016/S0277-9536(00)00455-X.

Burwick, A. et al. (2014) 'Costs of Early Childhood Home Visiting: An Analysis of

Programs Implemented in the Supporting Evidence-Based Home Visiting to Prevent Child Maltreatment Initiative’, p. 108. Available at: http://www.mathematica-mpr.com/~media/publications/PDFs/earlychildhood/EBHV_costs.pdf.

Casillas, K. L. et al. (2016) ‘Implementation of evidence-based home visiting programs aimed at reducing child maltreatment: A meta-analytic review’, *Child Abuse and Neglect*. Elsevier Ltd, 53, pp. 64–80. doi: 10.1016/j.chiabu.2015.10.009.

Chandler, J. et al. (2017) ‘Chapter 1: Introduction’, in Higgins, J. P. T. et al. (eds) *Cochrane Handbook for Systematic Reviews of Interventions*. Version 5. Cochrane.

Christakis, D. A. and Rivara, F. P. (2010) ‘Influence of experiences from birth to 5 years of age on emotional and psychological health’, *Archives of Pediatrics and Adolescent Medicine*, 164(5), pp. 491–492. doi: 10.1001/archpediatrics.2010.67.

Dalziel, K. et al. (2015) ‘Cost-Effectiveness Analysis of the Parents under Pressure Programme for Methadone-Maintained Parents’, *Child Abuse Review*, 24(March), pp. 317–331. doi: 10.1002/car.2371.

Dalziel, K. and Segal, L. (2012) ‘Home visiting programmes for the prevention of child maltreatment: Cost-effectiveness of 33 programmes’, *Archives of Disease in Childhood*, 97(9), pp. 787–798. doi: 10.1136/archdischild-2011-300795.

Deaton, A. and Cartwright, N. (2018) ‘Understanding and misunderstanding randomized controlled trials’, *Social Science and Medicine*. Elsevier, 210(December 2017), pp. 2–21. doi: 10.1016/j.socscimed.2017.12.005.

Deidda, M. et al. (2018) ‘Protocol for the economic evaluation of a complex intervention to improve the mental health of maltreated infants and children in foster care in the UK (The BeST? services trial)’, *BMJ Open*, 8(3), pp. 1–9. doi: 10.1136/bmjopen-2017-020066.

Deidda, M. et al. (2019) ‘A framework for conducting economic evaluations alongside natural experiments’, *Social Science and Medicine*. Elsevier, 220(October 2017), pp. 353–361. doi: 10.1016/j.socscimed.2018.11.032.

Dijkstra, S. et al. (2018) ‘Cost-effectiveness of Family Group Conferencing in child welfare: a controlled study’, *BMC Public Health*, 18(1), pp. 1–14. doi: 10.1186/s12889-018-5770-5.

- Drummond, M. F. et al. (2015) *Methods for the economic evaluation of health care programmes*. 4th edn. Oxford: Oxford University Press.
- Ebied, A. et al. (2013) 'How to cite this document', *Orthopaedic Surgery*, 6(6), pp. 1–5. doi: 10.1097/TME.000000000000194.
- Edmunds, K. et al. (2018) 'Systematic review of economic evaluations of interventions for high risk young people', *BMC Health Services Research*. *BMC Health Services Research*, 18(1), pp. 1–10. doi: 10.1186/s12913-018-3450-x.
- Eisman, A. B. et al. (2019) 'Economic evaluation in implementation science: Making the business case for implementation strategies', *Psychiatry Research*, (June). doi: 10.1016/j.psychres.2019.06.008.
- El-Khayat, Y. M. (2017) 'Resource review', *Journal of the Medical Library Association*, 105(4), pp. 431–432. doi: dx.doi.org/10.5195/jmla.2017.260.
- Elliot, R. and Payne, K. (2005) *Essentials of Economic Evaluation in Healthcare*. Padstow, Cornwall: Pharmaceutical Press.
- Filene, J. H. et al. (2013) 'Components associated with home visiting program outcomes: A meta-analysis', *Pediatrics*, 132(SUPPL.2). doi: 10.1542/peds.2013-1021H.
- Goldberg, J., Bumgarner, E. and Jacobs, F. (2016) 'Measuring program- and individual-level fidelity in a home visiting program for adolescent parents', *Evaluation and Program Planning*. Elsevier Ltd, 55, pp. 163–173. doi: 10.1016/j.evalprogplan.2015.12.007.
- Grant, M. J. and Booth, A. (2009) 'A typology of reviews: An analysis of 14 review types and associated methodologies', *Health Information and Libraries Journal*, 26(2), pp. 91–108. doi: 10.1111/j.1471-1842.2009.00848.x.
- Gray, A. M. et al. (2011) *Applied methods of cost-effectiveness analysis in healthcare*. Oxford: Oxford University Press.
- Hatzis, D. et al. (2017) 'Quality of caregiving in mothers with illicit substance use: A systematic review and meta-analysis', *Substance Abuse: Research and Treatment*, 11. doi: 10.1177/1178221817694038.
- Hemming, K. et al. (2015) 'The stepped wedge cluster randomised trial: Rationale, design, analysis, and reporting', *BMJ (Online)*, 350, pp. 1–7. doi: 10.1136/bmj.h391.

Hoomans, T. and Severens, J. L. (2014) 'Economic evaluation of interventions in health care', *Implementation Science*, 9(168). doi: 10.1186/s13012-014-0168-y.

Howard, K. S. and Brooks-Gunn, J. (2009) 'The role of home-visiting programs in preventing child abuse and neglect', *Future of Children*, 19(2), pp. 119–146. doi: 10.1353/foc.0.0032.

Husereau, D. et al. (2013) 'Consolidated Health Economic Evaluation Reporting Standards (CHEERS)--Explanation and elaboration: A report of the ISPOR Health Economic Evaluations Publication Guidelines Task Force', *Value in Health*, 16, pp. 231–250. doi: 10.1016/j.jval.2013.02.002.

Kemp, L. et al. (2019) 'Quality of delivery of "right@home": Implementation evaluation of an Australian sustained nurse home visiting intervention to improve parenting and the home learning environment', *PloS one*, 14(5), p. e0215371. doi: 10.1371/journal.pone.0215371.

Kuklinski, M. R., Oxford, M. L., et al. (2020) 'Benefit-cost analysis of Promoting First Relationships®: Implications of victim benefits assumptions for return on investment', *Child Abuse and Neglect*. Elsevier, 106(January), p. 104515. doi: 10.1016/j.chiabu.2020.104515.

Kuklinski, M. R., Crowley, D. M., et al. (2020) 'Supporting Strategic Investment in Social Programs: a Cost Analysis of the Family Check-Up', *Prevention Science*. *Prevention Science*, 21(2), pp. 256–267. doi: 10.1007/s11121-019-01077-3.

Lee, E. et al. (2009) 'Reducing Low Birth Weight Through Home Visitation. A Randomized Controlled Trial', *American Journal of Preventive Medicine*, 36(2), pp. 154–160. doi: 10.1016/j.amepre.2008.09.029.

Luhnen, M. et al. (2017) 'Systematic reviews of health economic evaluations: A protocol for a systematic review of characteristics and methods applied', *Systematic Reviews*. *Systematic Reviews*, 6(1), pp. 1–5. doi: 10.1186/s13643-017-0639-8.

Martin, L. and Lotspeich, R. (2014) 'A benefit-cost framework for early intervention to prevent sex trading', *Journal of Benefit-Cost Analysis*, 5(1), pp. 43–87. doi: 10.1515/jbca-2013-0021.

Masters, R. et al. (2017) 'Return on investment of public health interventions: A

systematic review', *Journal of Epidemiology and Community Health*, 71(8), pp. 827–834. doi: 10.1136/jech-2016-208141.

Mathewos, B. et al. (2017) 'Community-Based Interventions for Newborns in Ethiopia (COMBINE): Cost-effectiveness analysis', *Health Policy and Planning*, 32(August), pp. i21–i32. doi: 10.1093/heapol/czx054.

Mcdermid, S. and Holmes, L. (2013) 'The cost effectiveness of Action for Children's Intensive Family Support Services Final Report', (June), pp. 1–57.

McIntosh, E. et al. (2009) 'Economic evaluation of an intensive home visiting programme for vulnerable families: A cost-effectiveness analysis of a public health intervention', *Journal of Public Health*, 31(3), pp. 423–433. doi: 10.1093/pubmed/fdp047.

Merrick, M. T. et al. (2019) 'Vital Signs: Estimated Proportion of Adult Health Problems Attributable to Adverse Childhood Experiences and Implications for Prevention - 25 States, 2015-2017', *MMWR. Morbidity and mortality weekly report*, 68(44), pp. 999–1005. doi: 10.15585/mmwr.mm6844e1.

National Research Council (2014) *Considerations in Applying Benefit-Cost Analysis to Preventive Interventions for Children, Youth, and Families: Workshop summary*. Washington DC: National Academies Press. doi: 10.17226/18708.

Olds, D. et al. (2011) 'Impact of the nurse-family partnership on neighborhood context, government expenditures, and children's school functioning', *Medicine*. Available at: <https://www.ncjrs.gov/pdffiles1/ojjdp/grants/233277.pdf>.

Olsson, T. M. (2011) 'Comparing top-down and bottom-up costing approaches for economic evaluation within social welfare', *European Journal of Health Economics*, 12(5), pp. 445–453. doi: 10.1007/s10198-010-0257-z.

Panksepp, J. (2013) 'How primary-process emotional systems guide child development.', in Narváez, D., Panksepp, J., Schore, A. N., & Gleason, T. R. (ed.) *Evolution, early experience and human development: From research to practice and policy*. Oxford: Oxford University Press, pp. 74–94.

PDQ- Evidence Database (2018) PDQ-Evidence. Available at: <https://www.pdq-evidence.org/>.

Peacock, S. et al. (2013) 'Effectiveness of home visiting programs on child outcomes:

a systematic', *BMC Public Health*, 13, p. 17.

Ringel, J. et al. (2017) *Improving Child Welfare Outcomes: Balancing Investments in Prevention and Treatment*. Santa Monica, Calif.

Roberts, S. L. E., Healey, A. and Sevdalis, N. (2019) 'Use of health economic evaluation in the implementation and improvement science fields - A systematic literature review', *Implementation Science*. *Implementation Science*, 14(1), pp. 1–13. doi: 10.1186/s13012-019-0901-7.

Saias, T. et al. (2012) 'Evaluating fidelity in home-visiting programs a qualitative analysis of 1058 home visit case notes from 105 families', *PLoS ONE*, 7(5). doi: 10.1371/journal.pone.0036915.

Sharac, J. et al. (2011) 'Enhancing Adoptive Parenting: A Cost-Effectiveness Analysis', *Child and Adolescent Mental Health*, 16(2), pp. 110–115. doi: 10.1111/j.1475-3588.2010.00587.x.

Sonuga-Barke, E. J. S. et al. (2018) 'A comparison of the clinical effectiveness and cost of specialised individually delivered parent training for preschool attention-deficit/hyperactivity disorder and a generic, group-based programme: a multi-centre, randomised controlled trial of the New F', *European Child and Adolescent Psychiatry*. Springer Berlin Heidelberg, 27(6), pp. 797–809. doi: 10.1007/s00787-017-1054-3.

Sweet, M. A. and Appelbaum, M. I. (2004) 'Is home visiting an effective strategy? A meta-analytic review of home visiting programs for families with young children', *Child Development*, 75(5), pp. 1435–1456. doi: 10.1111/j.1467-8624.2004.00750.x.

Thanh, N. X. et al. (2014) 'An Economic Evaluation of the Parent–Child Assistance Program for Preventing Fetal Alcohol Spectrum Disorder in Alberta, Canada', *Administration and Policy in Mental Health and Mental Health Services Research*, 42(1), pp. 10–18. doi: 10.1007/s10488-014-0537-5.

Thielen, F. W. et al. (2016) 'How to prepare a systematic review of economic evaluations for clinical practice guidelines: database selection and search strategy development (part 2/3)', *Expert Review of Pharmacoeconomics and Outcomes Research*. Taylor & Francis, 16(6), pp. 705–721. doi: 10.1080/14737167.2016.1246962.

U.S. Department of Veterans Affairs (2019) *Budget Impact Analysis*, HERC -

Analysis Methods. Available at:

<https://www.herc.research.va.gov/include/page.asp?id=budget-impact-analysis> (Accessed: 4 December 2019).

Veritas Health Innovation (2018) 'Covidence systematic review software. Available at www.covidence.org'. Melbourne. Available at: www.covidence.org.

Weatherly, H. et al. (2017) 'Scoping review on social care economic evaluation methods', *CHE research paper*; . Available at: https://www.york.ac.uk/media/che/documents/papers/researchpapers/CHERP150_social_care_evaluation_methods.pdf <https://www.york.ac.uk/che/publications/in-house/>.

Wijnen, B. et al. (2016) 'How to prepare a systematic review of economic evaluations for informing evidence-based healthcare decisions: data extraction, risk of bias, and transferability (part 3/3)', *Expert Review of Pharmacoeconomics and Outcomes Research*. Taylor & Francis, 16(6), pp. 723–732. doi: 10.1080/14737167.2016.1246961.

Wilson, F. A. et al. (2016) 'A decision support tool to determine cost-to-benefit of a family-centered in-home program for at-risk adolescents', *Evaluation and Program Planning*. Elsevier Ltd, 56, pp. 43–49. doi: 10.1016/j.evalprogplan.2016.03.003.

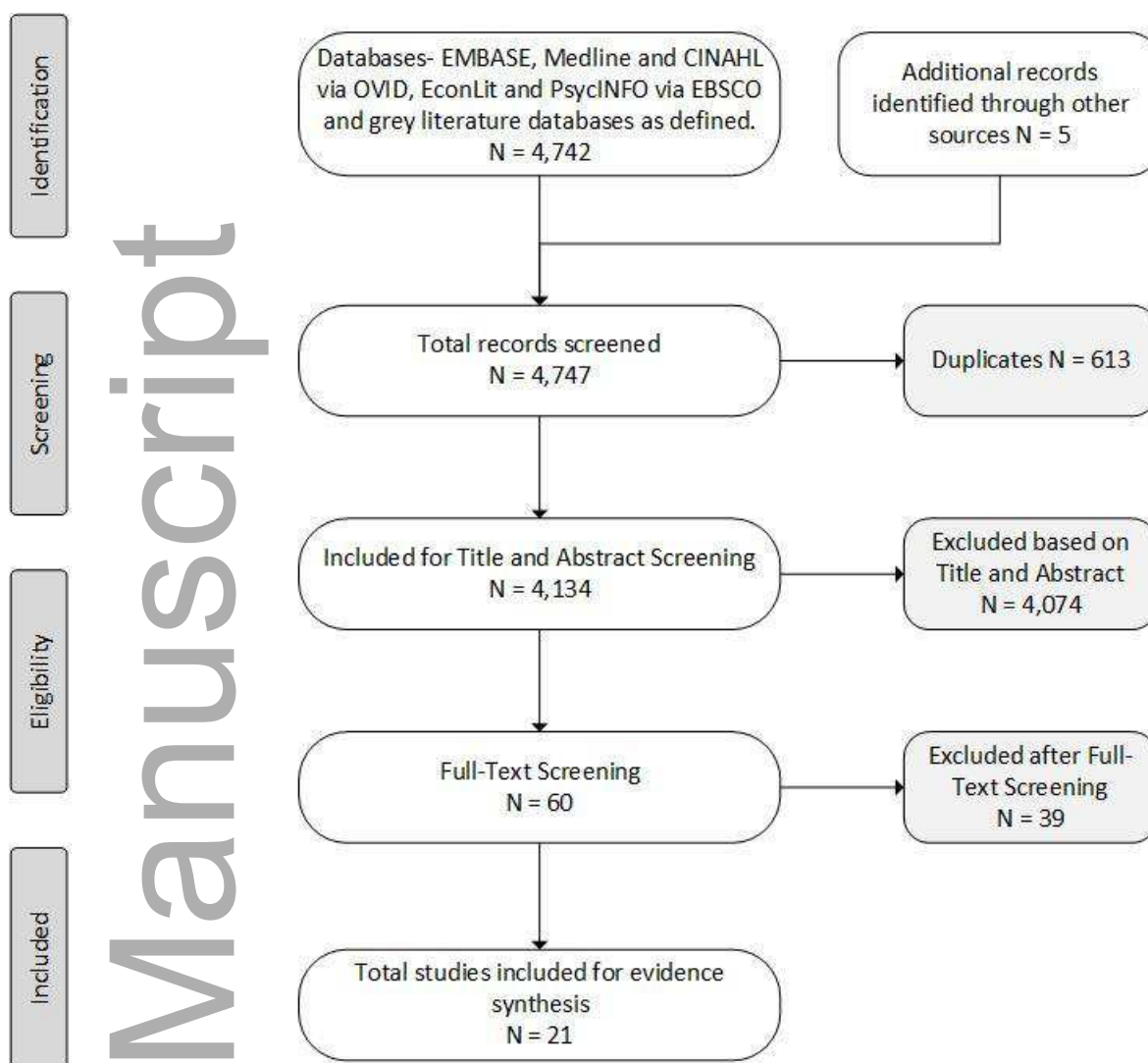


Figure 1. Preferred Reporting Items for Systematic reviews and Meta-Analyses (PRISMA) diagram for the study selection process

Table 1

Data extraction from studies including aim, population, outcomes, intervention, comparator perspective and study design

Author/ year/ country	Aim	Populatio n	Outcomes	Intervention	Compar ator	Perspect ive	Study design
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Ammerman et al., 2017, USA	To investigate the cost-effectiveness of a home visiting program for low-income mothers	Mothers with diagnosed with major depressive disorder with low-income	Quality Adjusted Life Years (QALY)	In-Home Cognitive Behavioral Therapy (CBT)	Home visiting without CBT therapy.	Public payer perspective	RCT
Aracena et al., 2009, Chile	To assess the cost-effectiveness of a home visiting program for adolescent mothers	Adolescent mothers	Child's physical health Child's psychomotor skills Indicators of child abuse Nutritional state of mother Mental health of mother	Home visits during trimester 3 until child one year of age. 12, one-hour visits on average Intervention aimed to address: - woman's identity - development of life plans - parenting skills - basic healthcare practices - relationship building	Standard prenatal and well-baby care.	Not stated	RCT
Au et al., 2006, Canada	To report the costs of a supplementary prenatal care program and the healthcare costs	All pregnant women attending one of three large family-physician clinics with low	Cost comparison between groups	Three trial arms: 1) Standard care 2) Standard care plus nurse visits 3) Standard care plus nurse visits plus home visits	Standard care.	Health service	RCT

	relating to the program	medical risk					
Barger et al., 2017, Bolivia	To isolate additional costs of integrating maternal and neonatal home visits into current program To evaluate the sustainability and feasibility of program at scale	Pre- and post-natal women living in areas of high neonatal mortality	Increased facility activity (number of antenatal, delivery and postnatal attendance in health centres and district hospitals)	Ante- and post-natal home visits of a newborn health package	Usual care without home visits in other districts	Ministry of Health in Bolivia.	Cluster RCT
Barlow et al., 2007, UK	Evaluation of effectiveness and cost effectiveness of intensive home visiting program	Vulnerable pregnant women	Dyad interaction Maternal psychological health attitudes and behaviour Infant functioning / development Risk of neglect or abuse.	Weekly home visits from antenatal 6 months to 12 month postpartum.	Standard care	Societal	RCT
Barlow et al., 2019, UK	To evaluate the effectiveness and cost-effectiveness of the intervention	Parents receiving treatment for drug or alcohol problems, and	Drug use (hair test) Severity of Dependence Scale HITS scale to measure	Parents under pressure program delivered in family homes to enhance parental	Treatment as usual was not standardised, to represent real	Public health and personal social services	RCT

		primary carer of child under 2.5 years	involvement in domestic violence Risk Abuse Scale from the Brief Child Abuse Potential Inventory was primary assessment to measure risk of child abuse Various other scales QALY data collected using the EQ-5D-5l health-related quality of life questionnaire for parents	emotional regulation (treatment was 6 months)	world services, and included family support, counselling and group parenting programs	perspectives	
Bell et al., 2019, UK	To document relevant health and non-health related resource use and associated costs	Young, first time mothers with low socio-economic profile attending a community midwifery setting	Tobacco use in late pregnancy, birth weight, emergency attendance and hospital admission for infant, and second pregnancy within 24 months	Intensive, nurse-led home visiting program (Family Nurse Partnership programme)	Care from local maternity and health visiting services as per usual practice	Considered from the perspective of the mother	RCT
Browne et al., 2001, Canada	To assess effects and costs of	Sole-support parents and	Exits from social	Five-armed RCT. featuring provider	Self-directed approach	Societal	RCT

	adding a mix of provider-initiated interventions to the health and social services that are typically self-directed.	children receiving social assistance	assistance within 1 year Savings regarding social assistance payouts.	initiated health promotion, employment retraining, and recreation activities for children			
Burwick, 2014, USA	To measure the costs associated with supplying home visiting programs	Families with children at risk of maltreatment	Costs only	Five separate home visiting programs in a 5 year grant program from 25 implementing agencies in 13 states.	Costs were calculated from different groups and centres	Implementing agency's perspective	Cross-site
Dalziel, 2015, Australia	Provision of advice to policy makers on 'value for money' of the intervention	Methadone - Maintained Parents	Child Abuse Potential Inventory was the primary outcome measure.	Home visiting intervention (Parents under Pressure) of up to 20 weeks (mean 10.5) of 1-2 hours with an intervention therapist	Routine care provided by staff at methadone clinic.	Societal	RCT
Deidda, et al, 2018, UK	To evaluate effectiveness and cost-effectiveness of a complex intervention for parents of children entering foster care	Parents of children entering foster care aged 0-60 months	Primary outcome measure is Strengths and Difficulties Questionnaire. (SDQ) for the economic analysis. Longer term impact on mental health	Complex intervention (New Orleans model), to improve abused children's mental health by improving parent-child relationship	Case management	Societal	RCT

			and allied services				
Dijkstra et al., 2018, Netherlands	To examine short- and long term cost-effectiveness of Family Group Conferencing to improve child safety	Families in child welfare, where child safety is at stake, mostly with multiple and complex problems.	Improving child safety, empowerment and social support, empowerment of family and social support of family	Family Group Conferencing for families to make a safety plan	Care as usual, families made a safety plan without Family Group Conferencing	Societal	RCT
Kuklinski 2020a, United States	To report net costs compared to long-term benefits of the program.	Parents of toddler aged 10-24 months, subject to recent report alleging maltreatment.	Adverse outcome such as child abuse/neglect	10 session attachment and strengths-based home visiting interventions	Resource and referral	Societal (participants, taxpayers, other society members)	RCT
Kuklinski 2020b, United States	To study the costs of intervention implementation	Families at risk with target children aged between 2 and 2.9 years at recruitment.	Cost of offering the intervention over 4 years	Early Steps intervention targeting early behaviour problems	Control: "business as usual", however control group not considered in this study	Societal	RCT
Mathewos et al., 2017, Ethiopia	To determine the cost-effectiveness	Mothers, pre- and post-natal	Per death avoided and per DALY avoided	Improved implementation of the Health Extension	Improve d implementation of	Provider perspective	Cluster RCT

	of a health extension program with and without program management			Program, plus improved equipment, monitoring and supervision	the Health Extension Program,		
	To assess the affordability of the program and quantify financial implications of scale up						
McIntosh et al., 2009, UK	To evaluate the cost-effectiveness of an intensive home visiting program, antenatal and postnatal	Mothers at risk by a broad definition.	Mother-child interaction Maternal mental health Maternal sensitivity Infant cooperativeness Infant mental and emotional development Quality of home environment Infants identifies as maltreated and removed from the home.	Weekly home visiting by trained home visitor	Care as usual	Societal and Healthcare	RCT
Olds et al., 2011, USA	To determine cost-effectiveness	Pregnant women, of low income	Pregnancy outcomes	Up to 30 months of home visiting program for mothers.	Type of control group not described	Societal and governmental.	Return on investment

	of a Nurse and Paraprofessional Home Visiting program in Denver	with first pregnancies	Child health and development Maternal life-course	Information about the intervention not explicitly stated. Appears to be related to smoking cessation.	Unable to find the original paper that the data were taken from.		
Sharac et al. 2011, UK	To determine the cost-effectiveness of two interventions to help control children with difficult behaviours	Adoptive parents	SDQ and 4 further parenting scales	Two interventions: 1) cognitive behavioural approach 2) educational approach	Comparing the two interventions. All interventions done in homes.	Not stated	RCT
Sonuga-Barke et al., 2018, UK	To compare the home visiting intervention to group parenting or care as usual.	Parents of children with ADHD	Child behavioural, health etc.	Intervention 1: 12-week individual, delivered in the home to address ADHD Intervention 2: Parenting training program; 12-week group-based	Treatment as usual in the region	Combined societal/NHS perspective	RCT

Thanh 2015, Canada	To determine the cost- effectiveness and cost- benefit implications of the intervention.	Women at risk of alcohol affected pregnancie s. Women were pregnant or up to six months postpartum ,	Probability of women staying in the intervention, using alcohol, pregnant, pregnant in first trimester, live birth, reduction in alcohol use during enrolment (retrieved from data collected by the agency). Other inputs retrieved from other sources. Fetal alcohol spectrum disorder information from systematic reviews.	The intervention aims to prevent subsequent alcohol exposed births through the use of mentors to address health and wellbeing through home visiting.	Modelled from previous studies	Societal	Pre- post & previo us literatu re
Wilson, 2016, USA	To describe methodology of creation of decision support tool	At-risk adolescent s	School attendance and dropout School recidivism Healthcare insurance Attendance at religious service. Physician contacts,	Family-centred support program	Detentio n and out-of- home placemen t of juveniles.	Governm ental and Societal	Pre- post & previo us literatu re

Personal
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Table 2

Information on methodological aspects of included papers by economic evaluation type

Type of economic evaluation	Paper	Modelling	Study design	Time horizon	Main costs
Cost comparison					
	Au et al., 2006	No	RCT	12 months	Overnight stay was the highest unit cost, and group costs were highly dependent on hospital use
	Burwick et al., 2014	No	Cross-site	12 months	Three top main expense were salary expense (57%), fringe benefits (15%), and indirect (overhead) costs (8%).
	Barger et al., 2017	Modelling of scale up	Cluster RCT	One year	Calculates costs of intervention, but not reduced healthcare costs
	Kuklinski et al 2020	No	RCT, but only intervention group considered here	Four years	Labour, Spplies, travel, overheads, training, intervention delivery, ongoing support/technical assistance
Cost-consequence					
	Browne et al., 2001	No	RCT	Two years	Cost breakdown not supplied
	Deidda et al., 2018	Long-term economic model	RCT	2.5 years for long term model	Protocol paper only, no results or costs collected
	Bell et al., 2019	No	RCT	24 months	Health resource use, non-health resource use (education, other supportive services, foster care, temporary accommodation)

Within trial cost-effectiveness

Aracena et al., 2009	No	RCT	15 months	Cost breakdown not supplied
Barlow et al., 2007	No	RCT	18 months	Cost breakdown not supplied
Barlow et al., 2019	No	RCT	12 months	Cost breakdown not supplied
Dalziel et al., 2015	No	RCT	6 months	Highest group delivery costs were for therapists
Dijkstra et al., 2018	No	RCT	6 and 12 months	Highest unit costs were related to hospital visits
Mathewos et al., 2017	No	Cluster RCT	12 months	For set up of the intervention, training accounted for most of the costs. For implementation, Meetings for supervision, salaries of supervisors and transport supervision were the highest costs
McIntosh et al., 2009	No	RCT	12 months	The highest cost were associate with home visitor home visits
Sharac et al., 2011	No	RCT	6 months	Highest costs were associate with employment of the social worker and classroom assistant, plus after school club, and then hospital.
Sonuga-Barke et al., 2018	No	RCT	6 months	Highest costs associate with health services, treatment delivery, therapist travel, administration, and preparation,
Cost utility				
Ammerman et al., 2017	Markov model	RCT	3 years	Only cost parameters presented, without a cost breakdown. Hospitalisation for depression was by far the highest unit cost.
Cost benefit (or Benefit-cost)				
Thanh et al., 2015	Decision tree	Pre-post & previous literature	3 years	Costs of cases of Foetal alcohol spectrum disorder avoided
Wilson et al., 2016	Decision support tool	Pre-post & previous literature	12 months	Cost breakdown not supplied
Kulinski et al., 2020a	Washington State Institute for Public	RCT	15 months	Intervention, support, costs

Policy
(WSIPP)
model

Return on investment

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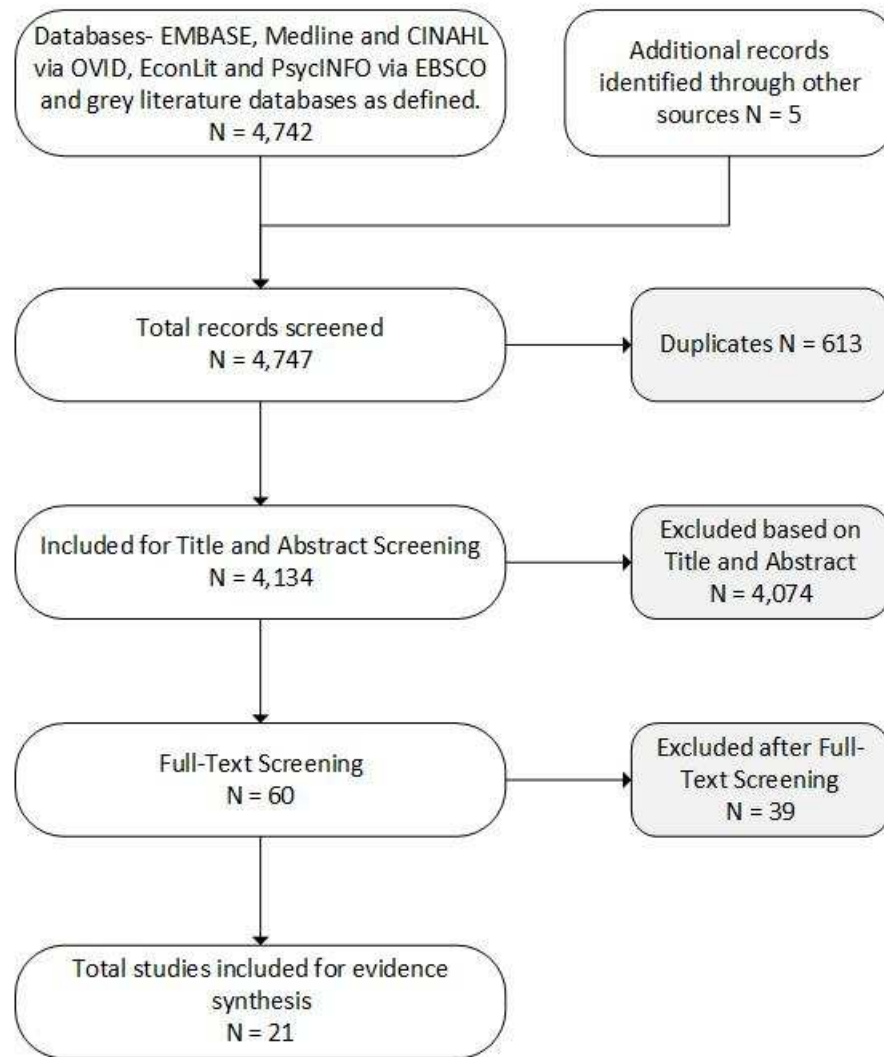
Identification

Screening

Eligibility

Included

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