Economic analysis for public policy decisions

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Abstract

Economics is the study of optimally allocating society's limited resources. Public policy, what governments choose to do or not to do, involves distributing resources across sectors. Economic analysis encompasses all analytical approaches used to explore economics, including the costs and outcomes of policies and interventions. It therefore plays a crucial role in public policy decisions, supporting the optimal allocation of resources.

Five publications are presented in this thesis, each applying economic analysis to a different policy issue. Collectively, the papers demonstrate how economic analysis can support decision-making in various sectors. A critical analysis is presented alongside the publications, exploring the development of the studies and their contribution to the field, individually and collectively.

Chapter 1 introduces the critical analysis by providing context to the publications and the central theme that runs through them. Chapter 2 builds on this to develop an analytical framework used to critique each of the papers in turn, considering the background of the research, the methods and the contribution to the field. The papers are discussed collectively in Chapter 3, pulling together the insights from Chapter 2, including the influence of knowledge mobilisation and methods on the impact of the research, and how circumstances influence the approach taken.

Co-design with end-users appeared to improve research quality and reach, although the research with most impact provided decision-makers with information on value for money. The overarching economic analysis (e.g., cost-effectiveness analysis vs. cost-utility analysis) and the within-method approach (e.g., cost perspective) both influenced the results of the research, and potentially the policy decision.

The critical analysis demonstrated that the selection of economic analysis depends on practical constraints such as the existing evidence base, research question, timing of the analysis, data, time and resource. Synthesising insights from the critical analysis, a questionnaire was developed to provide researchers and decision-makers with advice on the appropriate considerations to be made and potential analytical approaches given their context.

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List of tables and figures

Abbreviations

A&E	Accident and emergency
ABS	Antimicrobial stewardship
AMR	Antibiotic resistance
CAIS-P	Child Anxiety Impact Scale-parent report
CBA	Cost-benefit analysis
CBT	Cognitive behavioural therapy
CCA	Cost-consequence analysis
CEA	Cost-effectiveness analysis
CHEERS	Consolidated Health Economic Evaluation Reporting Standards
CHU9D	Child Health Utility-9 Dimensions
CRUK	Cancer Research UK
CUA	Cost-utility analysis
DALY	Disability-adjusted life year
DHSC	Department for Health and Social Care
ESR	Electronic Staff Record
HCAI	Healthcare-associated infection
HEE	Health Education England
HRQoL	Health-related quality of life
ICER	Incremental cost-effectiveness ratio
IPC	Infection prevention and control
L&D	Liaison and Diversion
MDS	Microbiology and diagnostic stewardship
NHS	National Health Service
NICE	National Institute of Health and Care Excellence
NIHR	National Institute for Health Research
OSI	Online Support and Intervention
QALY	Quality-adjusted life year
RCT	Randomised controlled trial
REVERSE	pREVention and management tools for rEducing antibiotic Resistance in high prevalence Settings
UK	United Kingdom
UKRI	UK Research and Innovation

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The portfolio of publications included in this thesis is the product of collaborations with many researchers. The research underpinning these publications was conducted while working at RAND Europe, the University of Oxford, and the UK Health Security Agency. I am grateful to my colleagues, past and present, for their expertise, experience, and collaboration throughout this work. I would also like to thank the external collaborators involved in each publication, without whom this research would not have been possible.

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Author declaration

I declare that I have not previously submitted any of the material offered in this thesis for a degree in this or any other university. The work contained in this critical analysis is mine alone. Co-authorship of the included publications is explicitly acknowledged, with full details of my contribution outlined in the 'Publications and contributions' section below. I was the lead analyst of the economic analysis undertaken in each of the publications. Co-author declarations confirming my contribution are provided in Appendix F.

The critical analysis contains 15,851 words, including footnotes, endnotes, the abstract and the reference list.

Publications and contributions

1. George, J., Gkousis, E., Feast, A., Morris, S., Pollard, J., & Vohra, J. (2020). **Estimating the cost of growing the NHS cancer workforce in England by 2029**. London: Cancer Research UK. URL: https://www.rand.org/pubs/external_publications/EP68310.html

Referred to as Paper 1. A full copy of the publication is available in Appendix A. Research took place at RAND Europe from 2019-2020.

Contribution of the candidate: Contributed to the development of the research idea and led the methodological design of the research, including leading the writing of the research proposal. Led the acquisition of data from stakeholders and contributed to the collection of additional data sources through desk research. Led the development of the economic model, undertook the stakeholder interviews, and contributed to the validation workshop. Conducted the economic analysis and data visualisation. Led the writing of the first draft of the publication. All with the support and guidance of the principal investigator (George, J.).

2. Guthrie, S., Pollard, J., Parkinson, S., Altenhofer, M., Leach, B., & Lichten, C. A. (2020). **Pregnancy research review: Data and methods report**. Santa Monica, CA: RAND Corporation. URL: https://www.rand.org/pubs/research_reports/RR4340.html

Referred to as Paper 2. A full copy of the publication is available in Appendix B. Research took place at RAND Europe from 2018-2020.

Contribution of the candidate: Contributed to the development of the research idea and methodological design of the research, including contributing to the writing of the research proposal.

Led the identification and collection of data on the health needs and healthcare expenditure associated with pregnancy. Led the evidence summary of the costs associated with non-standard pregnancies and the creation of a bespoke tool to compare pregnancy research spend with health needs and healthcare expenditure. Conducted the economic analysis and data visualisation. Led the writing of the 'Contextualisation of research spend' sections of the publication. All with the support and guidance of the principal investigator (Guthrie, S.).

3. Disley, E., Gkousis, E., Hulme, S., Morley, K. I., Pollard, J., Saunders, C., Sussex, J. & Sutherland, A. (2021). **Outcome Evaluation of the National Model for Liaison and Diversion**. Santa Monica, CA: RAND Corporation. URL: https://www.rand.org/pubs/research_reports/RRA1271-1.html

Referred to as Paper 3. A full copy of the publication is available in Appendix C. Research took place at RAND Europe from 2017-2020.

Contribution of the candidate: Contributed to the methodological design of the difference-in-differences analysis and led the methodological design of the economic evaluation. Led the data curation of the accident and emergency (A&E), and mental health services datasets, the collection of service costs associated with the intervention, and the identification of unit cost data in the literature. Conducted the difference-in-differences analysis of A&E and mental health service utilisation, and the economic evaluation. Led the writing of the economic analysis sections of the publication and contributed to the writing of the A&E and mental health service utilisation sections. All with the support and guidance of the principal investigator of the statistical analysis (Saunders, C.).

4. Creswell, C., Taylor, L., Giles, S., Howitt, S., Radley, L., Whitaker, E., ..., Pollard, J., Violato, M., … & Yu, L. M. (2024). **Digitally augmented, parent-led CBT versus treatment as usual for child anxiety problems in child mental health services in England and Northern Ireland: a pragmatic, non-inferiority, clinical effectiveness and cost-effectiveness randomised controlled trial**. *The Lancet Psychiatry*, 11(3). URL: https://doi.org/10.1016/S2215-0366(23)00429-7

Referred to as Paper 4. A full copy of the publication is available in Appendix D. Research took place at University of Oxford from 2020-2024.

Contribution of the candidate: Contributed to the methodological design of the economic evaluation (e.g., cost-effectiveness and multiple imputation statistical models). Led the curation of the economic data (e.g., service utilisation, health related quality of life) collected in the trial and the identification of unit cost data in the literature, with the support of co-authors. Conducted the economic analysis and

data visualisation, with the support of co-authors. Led the writing of the first draft of the economic component of the publication, which was then amalgamated with the clinical effectiveness component by co-authors for final publication. All with the support and guidance of the principal investigator of the economic evaluation (Violato, M.).

5. Pollard, J., Agnew, E., Pearce-Smith, N., Pouwels, K. B., Salant, N., Robotham, J. V., & REVERSE Consortium. (2025). **Umbrella review of economic evaluations of interventions for the prevention and management of healthcare-associated infections in adult hospital patients**. *Journal of Hospital Infection*, 158, 47-60. URL: https://doi.org/10.1016/j.jhin.2025.01.006

Referred to as Paper 5. A full copy of the publication is available in Appendix E. Research took place at UK Health Security Agency from 2024-2025.

Contribution of the candidate: Led the development of the research idea and led the methodological design of the research. Led the creation of the eligibility criteria, identification of studies, data extraction and quality assessment, all with the support of co-authors. Conducted the data synthesis and data visualisation. Led the writing of the publication. All with the support and guidance of the principal investigator (Robotham, J. V.).

Chapter 1. Introduction

This thesis presents a critical analysis of a portfolio of publications representing a coherent programme of research; the application of economic analysis to pertinent policy issues to inform decision-making. It reflects on a study estimating the cost of growing the NHS cancer workforce (Paper 1 [1]), and another estimating UK pregnancy research expenditure and contextualising it with relevant healthcare expenditure, health outcomes and other conditions (Paper 2 [2]). It also reflects on an economic evaluation of a criminal justice intervention to support vulnerable service users (Paper 3 [3]), an economic evaluation of a novel treatment for child anxiety problems (Paper 4 [4]), and a review of economic evaluations of interventions addressing healthcare-associated infections (HCAIs) in hospitals (Paper 5 [5]).

The structure of the critical analysis is presented in Figure 1.

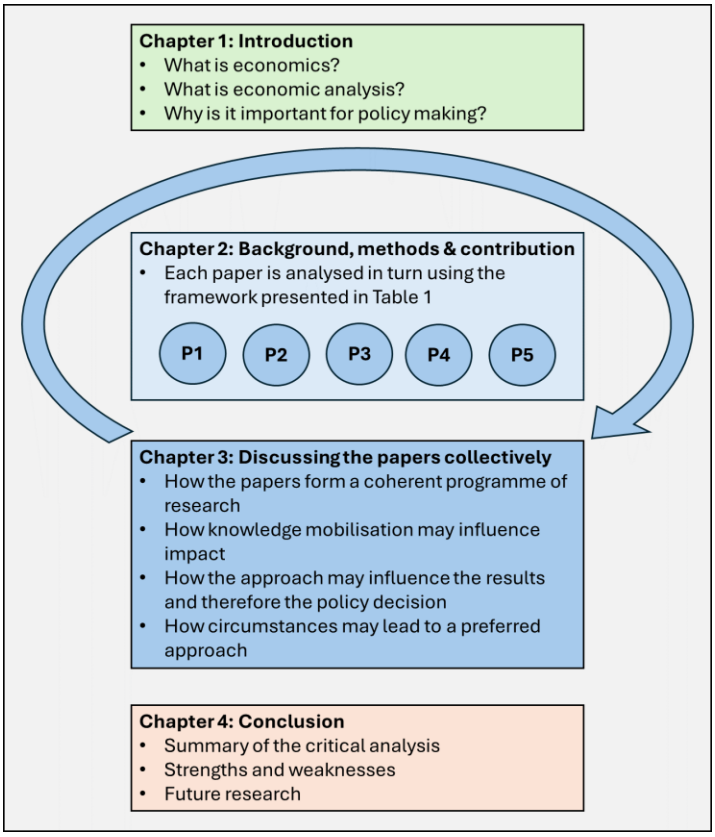

Figure 1. Structure of the critical analysis

1.1 Background

Economics is about the use of resources to meet society's needs and desires at various levels, including individual, household, organisation and government [6]. Resources are the inputs needed to provide goods and services, including labour, raw materials, land and energy [6]. Formally, economics can be defined as the study of optimally allocating limited resources to produce benefit to society [7].

The optimal allocation of resources is particularly important for 'efficient' economies, where it is not possible to make an individual better off without making at least one other worse off [8]. Such economies have no spare capacity in the system, meaning trade-offs are necessary when new policies are introduced. If resources are already maximised, then some level of existing inputs must be reallocated, with the outputs associated with its original use lost [6]. This loss is known as opportunity cost. Given the scarcity of resources, judgements about the costs and benefits associated with their use enable their optimal allocation [6].

Economic analysis is a broad term encompassing all analytical approaches used to explore economic phenomena, including the supply, demand or allocation of society's limited resources [6]. Economics can be divided into two concepts: microeconomics and macroeconomics. Microeconomics is the study of how individuals and organisations make decisions and interact in markets to allocate scarce resources, to determine relative prices, wages and rents [9]. Macroeconomics is the study of the aggregate outcomes of economic behaviour, concerned with employment, output and inflation [10].

The portfolio of publications presented here focuses on economic analysis in the context of microeconomics, and specifically that of sectoral policies, which relate to the allocation of limited resources for publicly funded programmes within and across sectors [11].

In this context, economic analysis is concerned with one or more of three main elements: costs, outcomes, and/or the comparison of costs and outcomes [12]. Studies which focus on costs alone (e.g., costing studies), or the comparison of costs and outcomes (e.g., full economic evaluations), are examples of economic analysis. However, for outcome-only studies to be considered economic analysis, the outcome must be economic-related.

Economic-related outcomes are those that capture the value placed on the consequences of an intervention, programme or topic. For example, in health economics, preference-based valuation of health-related quality of life (HRQoL) is used to estimate quality-adjusted life years (QALYs), using methods such as time trade-off and standard gamble [13]. In environmental economics, stated-

preference methods, such as discrete choice experiments, estimate willingness-to-pay for environmental policies [14], while in transport economics, revealed-preference methods, such as value of time analyses, estimate the monetary value of transport interventions [15].Costs refer to the inputs associated with a given intervention, overarching programme of activity, or specific topic (e.g., a specific health condition). These include direct costs, such as those associated with implementing (e.g., staff time) and/or receiving (e.g., out-of-pocket costs) the intervention, and indirect costs, which are those affecting the wider economy (e.g., productivity) [16]. Outcomes refer to the consequences associated with an intervention, programme or topic and can be economic-related (e.g., QALYs) or not (e.g., clinical). Outcomes can be directly (e.g., reduced mortality due to vaccination) or indirectly associated with an intervention (e.g., herd immunity due to vaccination). Outcomes can also be tangible, i.e., easily quantifiable (e.g., life years gained) or intangible, i.e., not easily quantifiable (e.g., value of improved health) [16].

Public policy has been defined as 'whatever governments choose to do or not to do' [17]. In practice, governments collect resources and redistribute them to pursue their objectives. Decisions about how these resources are allocated, often through sectoral policies, constitute public policy decisions. This thesis focuses on such decisions. For example, governments seek to provide health through healthcare, education through schooling and security through policing, among many other activities across sectors. Economic analysis is therefore crucial for public policy decisions. In an editorial on what makes research useful for policy, Whitty agrees, noting that policymakers are fundamentally making economic decisions [18].

Standard economic theory states that individuals are rational and self-interested, seeking to maximise their own 'utility', or pleasure, given their resources [10]. This is expected utility theory. The theory states that free markets, where individuals redistribute their resources through mutually beneficial trade, naturally reach general equilibrium, where prices 'clear' markets, i.e., supply is equal to demand [10]. If markets are perfectly efficient and supply equals demand throughout the economy then there is no need for the government intervention [11].

However, empirical evidence suggests that the assumptions of expected utility theory are often violated [19]. In many instances free markets do not lead to general equilibrium, and resources are therefore not allocated efficiently [20]. This is market failure, and can occur for various reasons [11]. Markets only meet the demands of those with sufficient purchasing power, will not optimally allocate resources where externalities occur, will not produce efficient outcomes where individuals lack full information, fail when

there is asymmetry of information between the principal (e.g., salesperson) and the agent (e.g., consumer), and underproduce public goods.

There is clear rationale for governments to intervene and correct such market failures through sectoral policies [11]. It is important that these provide good value for money [21]. This desire has led to a public value framework being developed in the UK, which provides a practical tool for departments across government to maximise the value they deliver from the public purse [22]. The framework assesses whether the inputs (e.g., spend), processes (e.g., service) and/or outcomes (e.g., population health) of a specific policy delivers public value. For example, it asks whether resource use is regularly reviewed, whether the relative effectiveness of different interventions has been established, and whether the existing allocation of resources remains the most productive.

There have recently been calls to broaden the framework used to determine the value for money of public policy. Frayman et al. (2024) [23] argue that the objective of government should be to improve wellbeing. They therefore argue decisions should be made based on the trade-off between the cost of a policy and its impact on wellbeing, measured in monetary terms. Their analysis suggests such an approach would lead to a major rethink of policy priorities in the UK.

Whichever framework is used, economic analysis is crucial. The three aforementioned elements of economic analysis can be utilised individually, or in combination, to provide information to policymakers about the costs, economic-related outcomes and/or cost-effectiveness of a given intervention, programme of activity, or topic. This supports policymakers to efficiently allocate society's limited resources, i.e., maximise outcomes for citizens from public spending, based on the best available evidence. This is best achieved through independent and rigorous evaluation that incorporates economic analysis [11].

Economic analysis can be grouped into one of six categories based on whether they consider costs and outcomes, and whether a comparison of two or more alternatives is made (Figure 2) [16]. Full economic evaluation is a form of economic analysis that compares the costs and outcomes of two or more alternatives (4). These can fully inform sectoral policy decisions. The five other types of economic analysis are partial evaluations. These provide crucial information for decision-makers, as will be explored in this thesis, but in isolation cannot fully inform choices between alternative policies.

Where no comparison of two or more alternatives is made, analysis of economic-related outcomes alone (i.e., outcomes that capture the value placed on relevant consequences) is an economic-related outcome

description (1A), and analysis of costs alone is a cost description (1B). The analysis of costs and outcomes without the comparison to an alternative is a cost-outcome description (2). The comparison of costs of two or more alternatives is a cost analysis (3B). The comparison of economic-related outcomes of two or more alternatives is an economic-related effectiveness evaluation (3A). Outcome descriptions and effectiveness evaluations that do not consider economic-related outcomes are not economic analysis.

Comparison of two or more alternatives?	Both costs and outcomes examined?		
No	**No**		**Yes**
	Economic-related outcomes only	**Costs only**	
	1A Economic-related outcome description	1B Cost description	2 Cost-outcome description
Yes	3A Economic-related effectiveness evaluation	3B Cost analysis	4 Full economic evaluation

Figure 2. Characteristics of economic analysis. Source: adapted from Drummond et al. (2015) [16]

Various considerations need to be made when undertaking economic analysis. There is a gap between theoretical economics and the reality that economists are faced with, with the assumptions underpinning expected utility theory often violated [19]. The rigid application of economic analysis that fails to adjust for reality is inappropriate [21], and it is important for economists to acknowledge the theoretical limitations and account for the reality of the situation in their analyses [24].

This means the approach must be relevant (i.e., represent and account for the policy issue) and practical (i.e., utilise methods that are useable) [25], account for behaviours in practice not just theory [26], and present results in a way that is easily understood [24]. All while dealing with the challenges faced by limited data, and the time and resource constraints associated with producing timely results [24]. These factors will be considered in more depth in this thesis.

Economic analysis has been conducted within and across numerous policy areas, with a variety of well-established methodologies available. Paper 1 presents a cost analysis (Figure 2, compartment 3B), estimating the cost of growing the NHS cancer workforce by 45% compared to the alternative of status quo workforce growth. Paper 2 presents a cost-outcome description (Figure 2, compartment 2), examining the healthcare cost and research expenditure of pregnancy, alongside the associated health outcomes, without comparison to an alternative. Both are partial economic evaluations [16].

Multiple full economic evaluation methodologies are also available (Figure 2, compartment 4). Paper 3 presents a cost-consequence analysis (CCA) comparing the costs and outcomes of two or more alternatives, the national Liaison and Diversion model compared to no national model, in a disaggregated manner [27]. Cost-effectiveness analyses (CEAs) capture outcomes in a single natural unit (e.g., life years) and (assuming dominance does not occur) present results in a single measure of efficiency, the incremental cost-effectiveness ratio (ICER). This is the ratio of the difference in costs to the difference in outcomes between two interventions [16]. Cost-utility analyses (CUAs) combine quantity and quality of life into a single metric, the QALY. Paper 4 presents a CEA, with anxiety symptoms the outcome, and a CUA of a novel child anxiety intervention compared to treatment-as-usual. Finally, cost-benefit analyses (CBAs) measure costs and outcomes in monetary terms [16].

Accurate synthesis of existing evidence, including well-conducted systematic reviews, has been argued to be the most valuable form of research for guiding public policy decisions [18]. Systematic reviews also play an important role in economic analysis. Broadly speaking, systematic reviews can be used to identify evidence to be incorporated into economic analysis, synthesise the results of economic analysis to inform policy, or both [6]. In the context of the former, systematic reviews can identify data on the types, quantities and values of resources and outcomes associated with an intervention, programme, or topic, which can then be used to undertake economic analysis [28]. In the context of synthesising economic analysis results, systematic review can be used to identify the quantity and quality of evidence related to a specific cost, economic-related outcome and/or cost-effectiveness question, as well as results directly relevant to decision-makers specific policy issue, and information on the context and conditions that promote the effectiveness and efficiency of different interventions [29]. Paper 5 presents a systematic review undertaken to synthesise evidence from full economic evaluations (Figure 2, compartment 4) and identify data to be incorporated into future economic analysis.

In summary, there are various methodological approaches available to conduct economic analysis to inform public policy decisions. The identification of potential approaches for the given context depends on many of the issues discussed in this chapter, including the research question, the target audience, the practicality of the method given the real-world context, the available data, and the time and resource constraints. The approach itself may also influence the results of the analysis and therefore, potentially, the policy decision [25]. These factors will be considered in detail in the remainder of this thesis.

1.2 Selection of the published works

The included papers were published between 2020 and 2025, with research commencing in 2017. They have been chosen because of the methods applied in each and their contribution to the field. Each presents economic analysis or the systematic synthesis of economic analysis to inform public policy decisions, with a variety of approaches taken. I made a significant contribution to each of the papers, as described in the 'Publications and contributions' section, with co-author declarations in Appendix F providing substantiation.

1.3 Aims

Five publications demonstrating a coherent programme of research and significant contribution to understanding are submitted alongside this critical analysis (Appendix A, B, C, D and E). The aim of the critical analysis is to reflect on this portfolio of publications to consider the application of economic analysis to inform public policy decisions.

Chapter 2. Background, methods and contribution

The considerations to be made when conducting economic analysis, identified in Chapter 1, were developed into an analytical framework (Table 1) to explore the background, methods and contribution of each publication. Additionally, the CHEERS checklist was completed for Paper 1 to Paper 4 to guide the critical analysis (Appendix H) [30], and the JBI Checklist for Systematic Reviews and Research Synthesis was completed for Paper 5 (Appendix I) [31]. The critical analysis of each publication extends beyond the aforementioned checklists, and the strengths and limitations discussed within the papers, by exploring the context of the research, the justification for the approaches taken, and the contribution of the research to the field. Table 2 summarises the critical analysis of each publication.

Table 1. Analytical framework for the critical analysis of each publication

Analytical theme	Purpose of analysis	Key considerations
Background	To reflect on the context of the research, what was known from previous research, and what research questions remained unanswered.	Policy context
		Previous research
		Research question
Methods	To reflect on the justification of the methodological approach, the strengths and weaknesses of the approach given the context of the research, and to draw on any lessons that can be learned.	Methodological approach
		Target audience
		Practicality (i.e., utilise methods that are appropriately useable)
		Available data
		Time and resource constraints
		Interpretability of the results
Contribution	To reflect on how the research advanced the knowledge base, and how it was utilised to influence policy and future research.	Research findings
		Forward citation analysis (see Appendix G for details)

2.1 Paper 1. Estimating the cost of growing the NHS cancer workforce in England by 2029

2.1.1 Background

Cancer causes over 450 deaths daily in the UK [32]. In 2019 the NHS Long Term Plan set targets to improve cancer outcomes in England over the next ten years [33]. In 2017 Health Education England (HEE), responsible for workforce planning across NHS England, published a cancer workforce plan highlighting seven key professions of concern [34]. In 2018, HEE reported that the cancer workforce would need to grow by 45% over the next ten years to deliver world class services [35]. Considering the Long Term Plan alongside HEE's cancer workforce plan suggested a mismatch between planned services and the workforce available to deliver them. Despite this, no long-term funding had been committed to HEE.

Previous research estimated the additional staff needed across the NHS to maintain the *status quo* [36, 37], and a scenario where waiting list targets are met [36]. However, neither study focussed on the cancer workforce, or the staff training costs. HEE projected the size of several cancer workforces from 2017 to 2022 [35] and Cancer Research UK (CRUK) estimated the demand for numerous cancer professions by 2027 [38]. However, neither estimated the associated training costs. It therefore remained unclear how many individuals (in absolute terms) would need to be recruited to deliver world class cancer services, and how much this would cost.

CRUK commissioned Paper 1 to estimate how much additional funding HEE would need to achieve 45% growth in seven key cancer professions from 2019 to 2029.

2.1.2 Methods

A cost analysis (Figure 2, compartment 3B) was undertaken to directly address the research question commissioned by CRUK. Although not a full economic evaluation, it provided information on the additional budget HEE would require, at a time when government spending was under review and the NHS long term workforce plan was being formulated [39].

A demographic stock-flow model was developed for each of the professions to estimate future staff numbers based on *status quo* inflows and outflows over the previous three years. To estimate total HEE costs, the total tariff paid to train a specialist was multiplied by the number of staff required to achieve 45% growth for each profession. See Appendix A. Paper 1 for detailed methods.

The model enabled use of the best available data as it could be developed at different levels of aggregation. This allowed the sex and age categorisation of the NHS Electronic Staff Record (ESR) data to be used, i.e., the NHS payroll system. Model inputs and outputs could also be aggregated to various levels, enabling validation with profession-specific workforce censuses.

The approach was practical, as the use of well-established methods meant the literature could be used for guidance. This enabled the analysis to be completed in a timely manner prior to the upcoming spending review, which is crucial for research to influence policy [18], while remaining within relatively limited resource constraints. The approach was also reproducible, with the same model structure used for all seven professions. The model and cost analysis also produced easy-to-interpret results, providing estimates of the staff shortfall for each profession and the total cost to train the necessary staff.

Although responsible for workforce planning, HEE did not bear all costs associated with the various workforce inflows into the NHS (e.g., international recruitment). Therefore, the analysis did not

incorporate the total public sector costs associated with achieving the growth targets. However, given the limited data available on the cost of workforce inflows, and the perspective defined by CRUK, a HEE focus was necessary.

The model took a simple approach to projecting labour supply. The mean inflows and outflows of the past three years were assumed to remain constant over the next ten. This is unlikely to be the case, with various factors influencing labour supply. A deterministic sensitivity analysis explored the impact of changes in workforce inflows and outflows, with each increased and decreased by an arbitrary 10%. Given this, and the cost perspective, the cost associated with growing the cancer workforce may be underestimated and lead to policy that results in greater expenditure than expected.

Cost analyses fail to consider whether the necessary investment provides good value for money. Previous research found 45% growth was necessary to provide world class services [35]. Paper 1 implicitly assumes this is desirable, and it is merely the cost that needs to be estimated. However, the opportunity cost of the intervention needs to be considered to determine whether it represents good value for money.

2.1.3 Contribution

Paper 1 found six of the seven cancer workforces were expected to grow by less than the 45% target. HEE require an additional £260m (2019/20 prices) to increase the cancer workforce through the training of new staff alone, or £142m if the workforce was increased through a mixture of inflows. See Appendix A for detailed results.

Eight articles were included in the forward citation analysis (Appendix G, Figure G1).

Articles cited the cost analysis as justification for additional workforce investment. A research report cited it as evidence more investment was required, particularly after the COVID-19 pandemic [40]. A news article in the BMJ also highlighted the argument for greater investment [41], while a journal article argued Paper 1 demonstrated the need for greater investment to achieve cancer survival targets [42]. The cost analysis in Paper 1 was highlighted by CRUK when arguing for more investment [43], as well as professional bodies the Royal College of Pathologists [44] and the Society of Radiographers [45]. CRUK also cited the cost analysis in written evidence to the *Cancer services* parliamentary inquiry [46], as did the Royal College of Pathologists when submitting evidence to the *Clearing the backlog caused by the pandemic* inquiry [47].

2.2 Paper 2. Pregnancy research review

2.2.1 Background

Prior to Paper 2 there was a drive to improve maternity care across the UK. In 2011, the Welsh government outlined their strategic vision for maternity services in the NHS [48]. The following year Northern Ireland's strategy for maternity care was published [49], as was Scotland's in 2016 [50]. While England's maternity review was being conducted [51], the Chief Medical Officer recommended a review of pregnancy research needs and expenditure across the UK [52]. Separately, a UK government review recommended assessing the impact of diseases to help determine public health and, in turn, research priorities [53].

The level of maternal and perinatal research expenditure in the UK had previously been estimated, but only considered funding up to 2007 [54]. Previous studies had contextualised research expenditure alongside the impact of the condition, although none had focussed on pregnancy or related reproductive, antenatal and postnatal care [55, 56]. The current level of spend on pregnancy research in the UK was therefore unclear, as was how this compared to the health and economic impact of pregnancy.

Paper 2, commissioned by the National Institute for Health Research (NIHR) and the Wellcome Trust on behalf of the UK Clinical Research Collaboration, sought to estimate the level of expenditure on pregnancy research in the UK, and contextualise this with the health and economic consequence of pregnancy. Economic analysis was used for the latter and will be the focus of the critical analysis.

2.2.2 Methods

A cost-outcome description (Figure 2, compartment 2) was undertaken to estimate the economic and health impact of pregnancy in the UK. A top-down estimate of NHS spend on pregnancy was calculated from the latest available condition-specific budgeting data, considering 'Maternity and reproductive' and 'Neonates' cost categories, accounting for the broad definition of pregnancy used in Paper 2, covering conception, contraception, antenatal and postnatal care. The prevalence of maternities and the associated disability-adjusted life years (DALYs) [57] were used to measure health impact. Pregnancy research funding, calculated separately, was contextualised by comparing it to the economic and health impact. See Appendix B. Paper 2 for detailed methods.

The costing and subsequent contextualisation was practical, adopting usable methods from Luengo-Fernandez et al. (2012) [55]. It also enabled the use of multiple datasets to provide a holistic overview of

the economic and health impact of pregnancy. The approach also allowed us to compare the contextualisation of pregnancy research spending with the equivalent for cancer, heart disease, dementia and stroke [56].

The costing was, however, relatively simple. Direct NHS healthcare costs were estimated from NHS programme budgeting data, which incorporates the whole care pathway, including primary and secondary care [58]. For a comprehensive estimate of the economic impact of pregnancy, a societal perspective is necessary [59]. Additionally, a prevalence-based approach was taken, where costs are considered for a given period. An incident-based approach could have been adopted to estimate the associated lifetime costs [59].

Pregnancy is not a disease. Healthy pregnancies are desirable for society. However, there are various associated complications (e.g., preterm birth) and inefficiencies that would preferably be eradicated. Despite this, the cost analysis reported the overall healthcare cost. Ideally, the undesirable cost (associated with complications and healthcare inefficiencies) would have been separated from the desirable cost (associated with efficiently treating healthy pregnancies), to estimate the avoidable cost in a best-case scenario, where all pregnancies are healthy and there are no healthcare inefficiencies.

An evidence summary was conducted to explore the desirable and undesirable costs associated with pregnancy. However, it was not possible to separate these with the available NHS budgeting data, and a considerably more intensive approach would have been required to quantify these separately. The costing approach was deemed most appropriate given the time, resource and data constraints.

Cost-outcome descriptions do not provide information on value for money. Methods are available to estimate health research return-on-investment, to determine whether the research outputs and outcomes justify the inputs [60, 61]. However, the contextualisation of pregnancy research funding was part of a wider project, which also sought to estimate the spend, by type and topic of research, and future priorities. Given the time and resource allocated to the contextualisation, it was not feasible to estimate return-on-investment.

The approach addressed the policy issue, accounting for guidance that the impact of conditions be considered when determining research priorities [53], through easy-to-interpret, high-level results that enabled the funding to be contextualised and compared with various conditions.

2.2.3 Contribution

On average, the NHS spent £5.8 billion per year on pregnancy from 2013-2017. Approximately £0.01 was spent on pregnancy research for every £1 spent on healthcare, considerably less than other conditions. Approximately £67 and £143 was spent on research for each maternity and DALY, respectively. See Appendix B for detailed results.

Eleven articles were included in the forward citation analysis (Appendix G, Figure G2).

Two articles cited Paper 2 to highlight research into pregnancy research needs and priorities. The Department for Health and Social Care (DHSC) referenced it in the Women's Health Strategy for England [62], and the National Childbirth Trust highlighted it on their website [63].

Most of the articles cited the economic analysis as evidence of underfunding. A policy report calling for improvements in pregnancy-related care [64] and an article published by an advocacy group [65] argued that pregnancy research is underfunded compared to the cost of the condition. A House of Lords paper, published as part of the *Preterm birth* inquiry, highlighted the comparison of pregnancy research funding to the cost of the condition and levels of research funding of other conditions [66].

Evidence submitted by the Academy of Medical Sciences to the *Women's reproductive health conditions* inquiry [67], a journal article exploring the impact of weight loss medication on fertility [68], and a report on pregnancy loss by two charities [69] all also argued that pregnancy research is underfunded compared to NHS spend, and research in other conditions. Two PhD theses argued the same [70, 71]. Finally, a journal article identifying research priorities in diabetes and pregnancy cited Paper 2 as evidence of the potential cost savings that could be achieved by improving pregnancy outcomes [72].

2.3 Paper 3. Outcome Evaluation of the National Model for Liaison and Diversion

2.3.1 Background

Policymakers have long grappled with the high prevalence of vulnerabilities, including mental health problems, among criminal justice system users. Liaison and Diversion (L&D) aims to identify vulnerable criminal justice system users and refer them to appropriate support, such as mental health services. A patchwork of local services had been operating for over 25 years, varying in nature and quality, before a 2009 UK government review recommended a national L&D model [73]. This was part of a drive to divert individuals from the criminal justice system, as it is known to exacerbate vulnerabilities [73]. A national model was developed and rolled out across ten trial sites in 2014 [74], followed by 13 more sites in 2015 and a commitment to achieve 75% coverage of England by 2018 [75].

A process evaluation of the L&D national model assessed the implementation of the service in the ten geographically separate trial sites, with an economic evaluation also conducted [74]. However, only the four most developed sites could be included and there was no data on service utilisation post-referral. The before-and-after approach meant differences could not be attributed to the intervention. The impact of the L&D national model on healthcare and criminal justice outcomes, and the associated economic impact, was therefore unknown.

DHSC commissioned an outcome evaluation of the L&D national model, to assess the impact on healthcare and criminal justice outcomes and, if any impact was found, what the associated economic effect was. Economic analysis was used for the latter and will be the focus of the critical analysis.

2.3.2 Methods

Data were collated from four healthcare and two criminal justice datasets to assess the impact of L&D on healthcare referral and utilisation, and offending and court processes, using difference-in-differences analysis. A CCA, where costs and outcomes are presented in a disaggregated manner, was undertaken to assess the economic impact of L&D. A societal perspective was taken with a one-year time horizon, using a top-down approach to estimate costs, combining the results of the difference-in-differences analyses with resource use and unit cost data to estimate consequences. See Appendix C. Paper 3 for detailed methods.

A CCA was most practical. It is a well-established method that utilised the difference-in-differences analyses that had already been undertaken. It also directly addressed the research question, to estimate the associated economic effect of any impact the intervention had on healthcare and criminal justice outcomes.

One reason a CCA was undertaken, rather than a more comprehensive approach that produced a single measure of efficiency, was that the economic analysis was somewhat a secondary consideration. This is typical of academic research, despite economic evidence being crucial for policymakers [18]. Applying for and accessing the various datasets took a considerable amount of time and resource, as did preparing the data and conducting the difference-in-differences. Relatively little time was therefore dedicated to the economic evaluation. The economic evaluation was also not embedded in the rollout of the L&D national model, as is often the case in randomised controlled trials (RCTs), which would have made a more comprehensive approach more feasible.

The CCA relied heavily on the statistical significance of the difference-in-differences results. This involved quantifying the economic impact of L&D where the difference-in-differences analysis reported a statistically significant impact. For example, the difference-in-differences analysis found L&D significantly reduced the probability of a custodial sentence. This was combined with the average length of a sentence and the unit cost of a prison place to estimate the economic impact. Outcomes were excluded from the CCA if there was no statistically significant difference.

There are clear limitations with this approach. Claxton (1999) [76] argues that it is not appropriate to apply the rules of classical statistical inference to economic evaluations of interventions. Interventions should be selected based on their mean net benefit, regardless of statistical significance. This is because one of the mutually exclusive alternatives must be chosen, and the fact that history dictates which option is current practice is irrelevant to the decision.

Appendix C, Table H29 outlines the longlist of potential costs and consequences of L&D considered in the economic evaluation. Data were only available for 12 of the 29 listed. Data were not available on the health of participants, meaning we were unable to undertake a CEA or CUA. A CCA was therefore most appropriate, as numerous outcomes of interest could not be included, meaning a single measure of efficiency (e.g., as provided in a CBA) may not have been reliable. Despite this, the L&D outcome evaluation created and analysed a large-scale linked dataset combining various sources for the first time, without which an economic evaluation would not have been possible.

The CCA produced easy-to-interpret results, with a total value presented for each of the costs and consequences. Given the results were not combined into a single measure of efficiency, the overall value for money of L&D was unknown. Therefore, the results may be of limited use for the DHSC. This is because DHSC are tasked with maximising health gain for their budget, and a single measure of efficiency (e.g., incremental cost per QALY) enables the comparison of various interventions across a variety of policy domains. However, the disaggregated nature of the results may be beneficial to others, such as those working in specific domains (e.g., criminal justice), as the results clearly demonstrate the domain-specific economic impact of L&D.

2.3.3 Contribution

The L&D national model cost £29.31m. Reduced custodial sentences were associated with savings of £12.03m to £38.14m for the criminal justice system and productivity gains of £1.06m to £3.37m. See Appendix C for detailed results.

Eleven articles were included in the forward citation analysis (Appendix G, Figure G3).

Most cited Paper 3 as evidence of the potential savings L&D can provide to the criminal justice system and wider society, focussing on the upper end of the saving estimates. A commentary on L&D services over the past 30 years highlighted the cost of the national model alongside the savings [77]. Two PhD theses highlighted the cost savings of L&D to the criminal justice system [78, 79]. A report considering vulnerable criminal justice system users combined the disaggregated savings to present an overall estimate [80]. A paper reviewing sentencing practices of those with mental health problems [81], and a book chapter exploring the relationship between mental health and crime [82], combined the disaggregated CCA costs and savings to argue that L&D was associated with an overall saving.

Two blogs disseminating the findings of the L&D national model outcome evaluation also highlighted the economic analysis [83, 84]. A journal article cited the economic evaluation while arguing for greater L&D provision in magistrates' courts [85], as did a report calling for a national L&D model in Scotland, which also combined the disaggregated CCA results [86]. The final article was a research brief produced to disseminate the findings [87].

2.4 Paper 4. Economic evaluation of a novel intervention for child anxiety problems

2.4.1 Background

Over a quarter of people will experience an anxiety disorder [88], with many first experienced in childhood [89]. Annually, anxiety disorders lead to 5.7m years lived with disability among young people globally [90] and societal costs can be up to 21 times higher compared to children in the general population [91]. Child anxiety problems also persist into later life [92]. Despite the prevalence, burden and persistence of child anxiety problems, only a minority receive evidence-based treatment, such as cognitive behavioural therapy (CBT) [93].

Brief forms of CBT, such as therapist-supported, parent-led approaches, can be effective at treating child anxiety disorders [94]. Digital augmentation may further improve efficiency and accessibility [95]. Given their potential, the National Institute of Health and Care Excellence (NICE) recommended guided self-help digital CBT for child anxiety problems for early use in the NHS [96], while calling for more evidence [97]. Online Support and Intervention (OSI) for child anxiety was one of four interventions recommended by NICE. OSI is an online platform used as part of a therapist-supported, parent-led CBT approach. Early evidence had found OSI to be feasible, acceptable and associated with improved outcomes [98]. However, its clinical and cost-effectiveness remained unclear.

An economic evaluation was funded by a joint DHSC-UK Research and Innovation (UKRI) and NIHR research grant to assess the cost-effectiveness of OSI plus therapist support, compared to treatment-as-usual, for child anxiety problems in routine practice.

2.4.2 Methods

A within-trial CUA and CEA was undertaken with a six-month time horizon, the follow-up period of the non-inferiority RCT. Outcomes were measured using quality-adjusted life years (QALYs) for the CUA, derived from the Child Health Utility-9 Dimensions (CHU9D) instrument, and the Child Anxiety Impact Scale-parent report (CAIS-P) for the CEA. Costs were considered from a healthcare perspective in the base-case analysis, and a societal perspective in sensitivity analysis. See Appendix D. Paper 4 for detailed methods.

The six-month time horizon was relatively short. The cost-effectiveness of an intervention within its trial period can differ considerably from what would have been observed had follow-up continued [99]. Economic models can be used to extrapolate results beyond the end of the trial to capture the longer-term impacts. However, the development of such models is resource intensive, dependent on the existence of sufficient data and not always necessary [100]. Given the time and resource allocated to the economic evaluation, and the limited long-term data in the literature [92], such economic modelling was not feasible.

The approach was practical. Methods for economic evaluation alongside clinical trials are well-established and have been applied in various contexts, with guidelines available [99]. The economic evaluation was embedded in the RCT, meaning all necessary data were collected to conduct a CEA and CUA.

Most economic evaluations of this type are undertaken alongside superiority trials, that determine whether one intervention is more effective than another. However, the RCT in Paper 4 was a non-inferiority trial, that determine whether one intervention is no worse than another. Advice exists for economic evaluations of the latter, although it is relatively limited. Bosman et al. (2008) [101] argue intention-to-treat and per-protocol results should be presented with equal importance, as Paper 4 does. Xie et al. (2019) [102] present a framework that simultaneously considers non-inferiority and willingness-to-pay for a QALY, but this is applied to model-based economic evaluations, and further work is needed to extend the approach to economic evaluations alongside non-inferiority trials.

There are no well-established guidelines for conducting economic evaluations alongside non-inferiority trials. Unlike superiority trials, non-inferiority trials are not powered to detect clinical differences between interventions, meaning differences in economic outcomes and costs may also be harder to detect. More guidance is therefore needed.

CEAs measure outcomes in a natural unit. It can be challenging to make policy decisions based on the results. If the CEA of a child anxiety intervention measures outcomes using the CAIS-P, and the CEA of a cancer treatment in life-years, it is not possible to directly compare the cost-effectiveness of the two. Moreover, the willingness-to-pay for natural units is unknown, making it challenging to determine whether an intervention is cost-effective.

This may not be an issue for decision-makers at the condition-specific level, as they seek to maximise health gain for their condition-specific budget. Compared to treatment-as-usual, OSI was associated with clinical improvements and lower healthcare costs, i.e., it dominated. Service providers could therefore conclude that OSI is a more efficient use of their fixed resources and recommend it as routine practice.

For decisions to be made at the sectoral level (e.g., healthcare), outcomes need to be captured using a generic measure, e.g., QALYs. A willingness-to-pay per QALY gained is also necessary to determine whether an intervention is cost-effective. NICE has a willingness-to-pay threshold of £20,000-£30,000 [103]. CUA therefore enables decisions about interventions across the healthcare sector.

Importantly, the CUA followed NICE's methodological guidelines for economic evaluations of health technologies, which is necessary for new treatments to be recommended for routine use [103]. Despite this, the results of the CUA were somewhat inconclusive. Unlike the CEA, OSI did not dominate treatment-as-usual in the CUA.

Holding all else constant, the cost-effectiveness of OSI depended on the value set used for the CHU9D instrument in the intention-to-treat analysis. With the UK adult value set, the probability of OSI being cost-effective was 35% at £20,000 willingness-to-pay per QALY, with the mean ICER exceeding the willingness-to-pay threshold. This increased to 60% with the Australia adolescent value set, with the mean ICER below the willingness-to-pay threshold. No guidance exists on the preferred value set, so a value judgement is needed to decide which is most appropriate and, ultimately, whether the intervention is cost-effective or not.

This phenomenon may be a product of the economic evaluation being undertaken alongside a non-inferiority trial, as the mean difference in QALYs approximated zero. Minor changes in incremental QALYs,

such as from the use of different value sets, can therefore change the cost-effectiveness results, as the willingness-to-pay threshold passes through the origin of the cost-effectiveness plane. This reiterates the need for guidance on non-superiority trials and the preferred CHU9D value set.

2.4.3 Contribution

QALY differences between OSI and treatment-as-usual approximated to zero. OSI costs were lower, driven by reduced therapist time. In the CUA the cost-effectiveness depended on the value set used for the CHU-9D instrument, while OSI dominated treatment-as-usual in the CEA. See Appendix D for detailed results.

Twelve articles were included in the forward citation analysis (Appendix G, Figure G4).

A commentary cited Paper 4 as an exemplar RCT and called for more real-world trials, highlighting the economic findings when reflecting on the results of the paper [104]. A systematic review of parental involvement in digital CBT for child anxiety cited the paper as evidence of a cost-effective intervention [105]. A preprint cited Paper 4 as evidence of cost-effective digital mental health interventions with human centred design [106], while another argued it demonstrated parent-led digitally augmented CBT improves efficiency [107].

Economic results were disseminated through a University of Oxford press release [108], a *The Lancet Psychiatry* editorial [109] and a NIHR plain English summary [110]. Three news articles presented the cost-effectiveness results [111-113], as did a blog [114]. Finally, NHS Education for Scotland cited the paper when recommending computerised CBT with practitioner support be delivered to parents for children with anxiety [115].

2.5 Paper 5. Umbrella review of economic evaluations of interventions for HCAIs in hospitals

2.5.1 Background

HCAIs are increasing in Europe, with around five million occurring annually [116]. Many HCAIs are resistant to antibiotics, with almost two-thirds of all resistant infections in Europe associated with healthcare [117]. The emergence of antibiotic resistance (AMR) threatens to undermine the effectiveness of antibiotics, with AMR already causing around 36,000 deaths annually in Europe [118]. The European Union funded the REVERSE (pREVention and management tools for rEducing antibiotic Resistance in high prevalence Settings) project, which aims to develop interventions for the prevention and management of multi-drug resistant HCAIs. The interventions are split into three categories: antimicrobial stewardship (ABS), infection prevention and control (IPC), and microbiology and diagnostic

stewardship (MDS). The project will assess the cost-effectiveness of these interventions using an economic model.

Systematic reviews have previously synthesised the cost-effectiveness evidence for interventions addressing HCAIs and AMR. However, they have either focussed solely on HCAIs [119] or AMR [120], or on specific types of interventions, such as whole genome sequencing [121] or point-of-care diagnostics [122]. No formal synthesis of the overarching cost-effectiveness evidence of interventions addressing HCAIs and AMR in hospitals had been undertaken.

A review was conducted as part of the REVERSE project to summarise the cost-effectiveness evidence of ABS, IPC and MDS for the prevention and management of HCAIs in hospital, with a focus on resistant infections.

2.5.2 Methods

In summary, an umbrella review (i.e., a systematic review of systematic reviews) was undertaken to summarise the cost-effectiveness evidence of ABS, IPC and MDS for the prevention and management of HCAIs in hospital, with a focus on resistant infections. The objective was twofold. Firstly, to identify which interventions are cost-effective and where more research is required. Secondly, to identify data to parameterise the economic model being developed to assess the cost-effectiveness of the REVERSE interventions. See Appendix E. Paper 5 for detailed methods.

Multiple recent systematic reviews synthesised the relevant cost-effectiveness evidence. An umbrella review was therefore deemed most appropriate for synthesising the evidence base and addressing the research question [123]. The approach also used well-established, reproducible methods.

Results were also easy to interpret. Appendix E, Table 1 provided a summary of the synthesised results for each of the interventions, with a narrative description of the cost-effectiveness evidence, and the quantity and quality of the evidence. The quantity of the evidence base (Appendix E, Figure 2) and the cost-effectiveness results (Appendix E, Figure 3) were summarised in heatmaps.

Cost-effectiveness evidence is ultimately aimed at decision-makers, as it seeks to inform the efficient allocation of resources. Such evidence is often of limited generalisability, due to setting-specific unit cost, resource use and outcome data [124]. The context of an intervention influences such data and therefore the cost-effectiveness results. This includes the incidence of infection, prevalence of AMR, healthcare systems and willingness-to-pay for health gains.

Although the accurate synthesis of existing evidence has been suggested as the most valuable form of research for policy [18], the results of the umbrella review may be of limited use for decision-makers. As an umbrella review was conducted, analysis of the context of the underlying studies was out of scope. Even traditional systematic reviews of economic evaluations can be of limited use, as the synthesis of cost-effectiveness evidence from various settings is often inadequate for those making decisions in specific contexts [29].

However, researchers on the REVERSE project were the immediate target audience, as they sought to develop an economic model. In this context the purpose of the review is to identify data for model development, including data on the main event pathways (e.g., length of stay), the probabilities or rates associated with these, and the subsequent resource use and outcomes [28]. The umbrella review enabled this, with researchers able to review the underlying economic evaluations to identify the necessary data to parameterise the model.

As Paper 5 was part of a wider project assessing the cost-effectiveness of the REVERSE interventions, relatively limited time and resource was allocated to the evidence synthesis. As a result, and because the review aimed to identify model parameters, an umbrella review was undertaken given the breadth of evidence they can identify in a relatively short timeframe. The review took six months from conceptualisation to journal submission. Traditional systematic reviews can take years. An umbrella review was determined to be just as effective at identifying data for the model while using considerably less resource. However, additional work was required to extract the relevant data from the underlying economic evaluations.

2.5.3 Contribution

Twenty-four systematic reviews were included. Most evidence focussed on screening, with selective screening cost-effective in most situations. IPC bundles were mostly cost-effective but highly heterogeneous. The evidence base was sparse for the remaining interventions. See Appendix E for detailed results.

Zero articles were included after forward citation searches and screening (Appendix G, Figure G5), given how recently the paper was published.

Table 2. Summary of the critical analysis of each publication

Paper	Background: Context of the research, what was known, and what remained unanswered	Methods: Justification of the approach, strengths and weaknesses, and lessons learned	Contribution: How the research advanced the knowledge base, and how it was utilised
Paper 1: Estimating the cost of growing the NHS cancer workforce in England by 2029	• HEE's cancer workforce plan highlighted the need to grow seven key cancer workforces. • There was a mismatch between planned cancer services and the workforce to deliver them. • No long-term funding committed to HEE, responsible for workforce planning, with a government spending review due soon. • Workforce growth of 45% was required by 2029 to deliver world class services, but the associated cost was unknown.	• A cost analysis was the most appropriate approach given the background and research question. • Utilisation of a demographic stock-flow model enabled use of the best available data. • The model was practical and reproducible, producing easy-to-interpret results in a timely manner, within the projects resource constraints. • Costs were considered from a HEE perspective, and therefore did not consider all costs to the system. • The model took a simple approach to projecting labour supply. • Cost analyses do not consider whether the investment provides good value for money.	• Six of the seven workforces were expected to grow by less than the 45% target. • HEE would require an additional £142m to £260m to achieve the necessary workforce increases. • Cited to argue for additional funding for the cancer workforce, through research reports, articles from charities and professional bodies, and written evidence submitted to parliamentary inquiries.
Paper 2: Pregnancy research review	• There was a drive to improve maternity care across the UK, with each nation producing a plan to achieve this. • A government review recommended assessing the impact of health conditions to help determine research priorities. • The Chief Medical Officer recommended a review of pregnancy research needs and expenditure. • Estimates of pregnancy research expenditure were outdated, and it was	• A cost-outcome description was the most practical approach, enabling the use of various data, and comparison across conditions. • The cost analysis was relatively simple. A return-on-investment study would have provided information on value for money. • However, the analysis was part of a wider project and was all that was feasible given the time and resource available. • The cost analysis did not account for the fact pregnancy is not a disease, due to the	• The NHS spent £5.8b per year on pregnancy. • Approximately £0.01 was spent on pregnancy research for every £1 spent on healthcare, considerably less than cancer, heart disease, dementia and stroke. • Cited to highlight research that is being conducted into pregnancy research spending. • Cited as evidence of underfunding in pregnancy research, compared to healthcare expenditure and other

Paper	Background: Context of the research, what was known, and what remained unanswered	Methods: Justification of the approach, strengths and weaknesses, and lessons learned	Contribution: How the research advanced the knowledge base, and how it was utilised
	unknown how it compared to the health and economic impact.	available data and the time and resource available. • The approach directly addressed the policy question, producing easy-to-interpret results for the target audience.	conditions, as well as potential cost savings of future research.
Paper 3: Outcome Evaluation of the National Model for Liaison and Diversion	• Liaison and Diversion (L&D) aims to identify vulnerable criminal justice system users and refer them to appropriate support. • A government review recommended fragmented local L&D services be replaced by a national model. • The national model was rolled out across 10 trial sites, followed by 13 more sites in 2015 and a commitment to achieve 75% nationwide coverage by 2018. • A process evaluation of the 10 trial sites was published in 2016, although the impact of the L&D national model on healthcare and criminal justice outcomes, and the associated economic impact, was unknown.	• A cost-consequence analysis (CCA) was the most practical approach, drawing on the difference-in-differences analysis. • It also directly addressed the research question. • The economic evaluation was a secondary consideration, meaning time and resource was limited, and the economic evaluation was not embedded in the rollout of L&D. • The CCA relied heavily on the statistical significance of the results in the difference-in-difference analysis. This is a limitation of the research, as interventions should be evaluated based on their mean net benefit, rather than statistical significance. • The CCA made best use of the limited data that were available. • The approach produced easy-to-interpret results, although they were not combined into a single measure of efficiency, meaning they may be of limited use.	• The L&D national model cost £29.31m. • Reduced custodial sentences were associated with savings of £12.03m-£38.14m in the criminal justice system, and productivity gains of £1.06m-£3.37m. There was no impact on healthcare outcomes. • Cited as evidence of the potential savings L&D can provide to the criminal justice system and wider society, and as justification for greater L&D provision across the UK. • Several of the articles citing the paper combined the disaggregated results of the CCA to estimate overall savings.
Paper 4: Economic evaluation of a novel intervention for child anxiety problems	• Despite the prevalence, burden and persistence of child anxiety problems, few receive evidence-based treatment. • Brief CBT can be effective and improve efficiency and access, with digital	• A CEA and CUA of OSI plus therapist support compared to treatment-as-usual was the most practical approach given the available data, time and resource constraints.	• The difference in QALYs approximated to zero. • Costs were lower in the OSI arm, driven by less therapist time delivering treatment.

Paper	Background: Context of the research, what was known, and what remained unanswered	Methods: Justification of the approach, strengths and weaknesses, and lessons learned	Contribution: How the research advanced the knowledge base, and how it was utilised
	augmentation furthering such improvements. • NICE recommended OSI, a digitally augmented brief CBT, for early use in the NHS based on initial evidence. • The clinical and cost-effectiveness remained unclear, with NICE calling for further evidence.	• CEA does not enable policy decisions at the sectoral (e.g., health) level, although can inform condition-specific decision-makers. • CUA enables decisions at the sectoral level, as outcomes are measured in QALYs. However, they can provide conflicting results. • The approach followed NICE's methodological guidelines. • More guidance is needed for economic evaluations alongside non-superiority trials, and for the most appropriate value set for the CHU9D health-related quality of life instrument in UK evaluations.	• The CEA found OSI dominated treatment-as-usual, while the CUA results were mixed. • Cited as an exemplar real-world trial, and as evidence of a cost-effective digitally augmented CBT for child anxiety. • Citing papers highlighted the cost savings driven by less therapist time on OSI. • NHS Scotland cited the paper when recommending therapist-supported computerised CBT be delivered to parents for child anxiety.
Paper 5: Umbrella review of economic evaluations of interventions for the prevention and management of healthcare-associated infections in adult hospital patients	• Millions of healthcare-associated infections (HCAIs) occur annually, many of which are resistant to antibiotics. • The REVERSE (pREVention and management tools for rEducing antibiotic Resistance in high prevalence Settings) project aims to develop cost-effective interventions for the prevention and management of multi-drug resistant HCAIs. • Multiple recent systematic reviews have synthesised the cost-effectiveness evidence for interventions addressing HCAI and AMR. However, they have focussed on HCAIs or AMR separately, or on specific interventions.	• An umbrella review was therefore undertaken to synthesise the overarching cost-effectiveness evidence of interventions addressing HCAIs and AMR in hospitals. • The approach was appropriate for the research question and objective, and produced easy-to-interpret high-level results. • The synthesis of cost-effectiveness evidence from various contexts is of limited use for policymakers, given the cost-effectiveness of an intervention is context specific. • A main objective of the evidence synthesis was to identify data to parameterise the REVERSE economic	• Twenty-four systematic reviews were included, with evidence identified on 10 intervention types and 14 infection/organism categories. • Most evidence focussed on screening, with selective screening cost-effective in most situations. • IPC bundles were mostly cost-effective but highly heterogeneous. • The evidence base was sparse for the remaining interventions. • Zero articles were identified from the forward citation analysis, given how recently the paper was published.

Paper	Background: Context of the research, what was known, and what remained unanswered	Methods: Justification of the approach, strengths and weaknesses, and lessons learned	Contribution: How the research advanced the knowledge base, and how it was utilised
		model, which the umbrella review enabled. • An umbrella review was also most practical given the limited time and resource available, particularly when compared to traditional systematic reviews.	

Chapter 3. Discussing the publications collectively

Chapter 3 reflects on the publications as a coherent programme of research and how they collectively advance the knowledge base. Contributions of each paper are compared, with consideration of how knowledge mobilisation approaches may have influenced impact. Reflections are made about how methodological choice affected research results and therefore potentially the policy decision, and the practical circumstances that leads to an approach being preferred.

3.1 Contribution and knowledge mobilisation

The five papers collectively demonstrate the application of economic analysis to address pertinent policy-related issues to inform decision-making across a variety of topic areas.

Various methods were applied across the five publications, with the evidence generated from the studies used for different purposes. The outputs from the economic analyses were cited as justification for additional investment, for example in the NHS cancer workforce (Paper 1) and pregnancy research (Paper 2). It was also presented as evidence of underfunding in the example of pregnancy research and used to inform research priority setting (Paper 2). The papers were cited as evidence of cost savings associated with interventions, such as L&D (Paper 3) and OSI (Paper 4), and the cost-effectiveness of the latter. The two papers were also cited in arguments for the wider adoption of the two interventions, including in other countries. Finally, the evidence was cited to demonstrate desirable methodology, alongside a request for more research using the desired approach (Paper 4).

3.1.1 Engaging with end-users

Research that engages with end-users throughout the project appears more likely to be cited by influential organisations seeking direct policy impact. Although Paper 1 has fewer citations than the other primary analyses (i.e., Papers 2-4), it was cited by professional bodies [44, 45] and CRUK, the charity that commissioned the work [43], including in evidence submitted to parliamentary inquiries [46, 47], and a policy thinktank [40].

This is likely because Paper 1 engaged with the target audience throughout, including charities and professional bodies seeking to influence policy, and decision-makers. This stakeholder engagement is a form of co-design, i.e., meaningful end-user involvement in the research [125]. It benefitted the research, as the relationship developed with HEE resulted in them sharing the best available data, without which we would have had to rely on various publicly available data sources, impacting the external validity. The research was regularly reviewed by experts interviewed to validate the model

inputs and assumptions. Preliminary results were presented at a workshop to ensure the output was useable and to disseminate early findings. This suggests an analyst working in isolation may be less successful than one engaging in co-design. Previous studies have drawn similar conclusions, although this has rarely been tested empirically [125].

Paper 2 engaged with a steering group throughout the project, including the research commissioners, professional bodies, charities, healthcare providers and decision-makers. However, the involvement of end-users was considerably less than that in Paper 1. They were not interviewed to inform the research, for example. Despite this, Paper 2 was also cited by influential organisations looking to impact policy. For example, it was cited as part of multiple parliamentary inquiries [66, 67], by the DHSC [62], and national charities [69]. This suggests that even with less intensive end-user engagement results can still be disseminated to influential end-users if appropriate stakeholders are included in a project steering group that is active throughout the research.

Paper 3 preliminary results were presented to the commissioners of the research, DHSC. However, end-users were not engaged with the research throughout the project as in Paper 1 and, to a lesser extent, Paper 2. This might explain why the citing sources were less influential. For example, Paper 3 was not cited in evidence which was submitted to parliamentary inquires. Key end-users likely to influence policy may have been unaware of the research or may have had less confidence in the results as they were not involved in co-designing the study, and therefore less likely to utilise them.

Ongoing engagement with policy-focussed end users supports dissemination but is not necessarily essential. There was no clear evidence that Paper 1, 2 or 3 directly influenced policy change. Paper 4, however, was cited by NHS Education for Scotland when recommending computerised CBT with practitioner support delivered to parents for children with anxiety [115]. Such parents, a key end-user, co-designed certain aspects of the research, such as participant questionnaires. Policy-focussed end-users were not engaged in co-design throughout the project, although preliminary results were presented to a steering group towards the end of the economic analysis. Despite this, Paper 4 appeared to have most policy impact of all the analyses.

3.1.2 What could have been done differently?

This suggests a different approach could have been taken for some of the economic analyses, and corresponding knowledge mobilisation, to provide end-users with the information they desired.

The disaggregated results of the CCA were combined by many of the sources citing Paper 3 to estimate the overall economic impact of L&D, despite the paper stating many relevant costs and outcomes were not included. An economic evaluation providing results in a single measure of efficiency appears to have been desired by end-users. Paper 4 provided such results and appeared to have most policy impact. Had the research engaged in co-design, the desire for a single measure of efficiency may have become clear earlier. Knowledge mobilisation should have emphasised the importance of considering the results in a disaggregated manner.

While Paper 1 and 2 were cited as evidence for greater funding, their argument may have been stronger had they presented evidence on the value for money of such investments. Paper 4 did analyse value for money and, again, appeared to have most policy impact.

Paper 1 could have taken a broader perspective for the costs of increasing the cancer workforce, and included outcomes, such as waitlist reductions and improved patient outcomes. This could then be compared to the *status quo* to determine value for money.

Paper 2 could have gone beyond the contextualisation of pregnancy research spending and explored value for money by comparing the research outputs and outcomes to the inputs (i.e., funding), to determine the expected return-on-investment [61].

3.1.3 Economic analysis is only part of the picture

The included publications demonstrate how economic analysis can support decision-making, particularly when value for money is analysed. However, economics is only part of the picture. Politics and ideology often outweigh economics when policy decisions are made, even when economists are aligned in their views [126]. One reason for this is that we live in a society and not an economy. Efficiency is therefore not the only consideration when spending taxpayer money. There are many other influences at play, including justice, national security and compassion [21]. Even if economic analysis provides positive evidence on the value for money of an intervention, there is no guarantee it will influence policy.

Decision-makers are increasingly concerned with the winners and losers of policies and interventions [127]. Traditional economic analysis considers the efficient allocation of resources but does not consider equity. Distributional cost-effectiveness analysis addresses this issue by examining how costs and outcomes are distributed across different groups (e.g., age, sex, socioeconomic status) and incorporating the trade-offs between improving total health (i.e., efficiency) and equity (e.g., reducing health

inequalities) [128]. In practice, however, the application of distributional CEA has been relatively limited [129]. None of the publications included in this thesis explicitly considered equity.

The conclusions drawn by reflecting on the contribution and knowledge mobilisation of the publications extend beyond the suggestions made by Whitty in his editorial on what makes research useful for policy [18]. Specifically, among the included publications, engaging in co-design appeared to improve the quality and reach of the research, and, in the context of economic analysis, the publications that provided information on value for money appeared to have most policy influence.

3.2 Influence of the approach on the results and policy decision

The forward citation analysis showed the publications were often cited by sources seeking to impact policy. The approach of an economic analysis can influence the results, and therefore policy decisions. This can be driven by the overarching approach, such as whether a costing study or full economic evaluation was conducted, or within-method approaches, such as the cost perspective, time horizon or value set.

3.2.1 Influence of the overarching approach

The CEA in Paper 4 found that OSI dominated treatment-as-usual. However, the CUA found that OSI led to a small reduction in QALYs, meaning it lowered costs but slightly worsened outcomes. This appears to be because the CEA and CUA were picking up different aspects in their outcome measures. The CEA mean ICER fell in the south-east quadrant of the cost-effectiveness plane (less costly, more effective), while the CUA mean ICER fell in the south-west (less costly, less effective). Despite, or perhaps because of, the difference in results, most of the sources cited the paper as evidence of the cost-effectiveness of OSI, largely focussing on the cost-savings associated with the intervention. This may be appropriate for decision-makers seeking to maximise child mental healthcare budgets but may not lead to the most efficient allocation of resources across the healthcare sector, as explained in section 2.4.2.

End-users often combined the disaggregated results of the CCA in Paper 3. However, a CCA was undertaken as only 12 of the 29 relevant cost and outcome categories were incorporated in the analysis. It would therefore be inappropriate to draw conclusions from the combined results. For example, L&D may have had a negative impact on QALYs which, if known, would have impacted the results. A CCA purposely presents disaggregated results to allow decision-makers to consider the individual costs and outcomes in their own context, as well as other factors, when making decisions [130].

Systematic reviews of cost-effectiveness evidence may also lead to inappropriate policy decisions. Paper 5 drew conclusions about the cost-effectiveness of HCAI interventions by synthesising international literature. However, a cost-effective intervention in one setting may not be in another [124]. Inappropriate interventions may therefore be implemented if policy decisions are solely informed by systematic reviews of economic evaluations across different settings.

3.2.2 Influence of the cost perspective

Paper 1 took a HEE-only cost perspective, meaning the full cost to the taxpayer was unknown. Decision-makers are interested in the total cost associated with an intervention, making it challenging to implement policy based on such a narrow cost perspective.

Paper 2 took a healthcare perspective to estimate the total cost of pregnancy to the NHS. However, pregnancy is not a disease and healthy pregnancies are desirable. Arguably, only undesirable pregnancy costs (i.e., those associated with pregnancy complications and healthcare inefficiencies) should have been considered, rather than total pregnancy costs, as this is the cost that pregnancy research is seeking to eradicate. The cost would have likely varied considerably, and the results of the contextualisation differed, had the analysis been able to tease apart desirable and undesirable pregnancy healthcare costs.

Taking a societal perspective, rather than a healthcare perspective, can lead to different results. Paper 4 presented the cost-effectiveness results from both perspectives. OSI was associated with parents missing more work time compared to treatment-as-usual, likely because they were more involved in the treatment. OSI was therefore associated with lower cost savings from a societal perspective. If the preferred cost perspective was societal, as it is in the Netherlands for example [131], OSI would be less likely to be recommended.

The economic evaluation in Paper 3 relied on the results of the difference-in-differences analysis to determine which costs and outcomes were incorporated in the CCA. This approach inappropriately excluded potential impacts of L&D based on classical statistical inference [76]. For example, L&D was associated with a small statistically insignificant increase in A&E attendance and decrease in mental healthcare attendance, which if included may have impacted the results.

3.2.3 Influence of the costing approach

Paper 4 took a bottom-up micro-costing approach, where each resource used (e.g., A&E attendance) was identified, measured and multiplied by a unit cost [16]. The mean difference in costs associated with OSI could be disaggregated and the driver of the savings (i.e., reduced therapist time) identified. This may

explain why Paper 4 was cited by NHS Scotland when recommending such interventions as standard practice, due to the efficiencies it offered mental health services, even though the CUA results were inconclusive.

Paper 2 and 3 took a top-down approach to estimating costs, where overall expenditure is used to estimate costs [132]. Paper 2 drew on data aggregated at the condition-specific level to estimate the total healthcare cost of pregnancy. Disaggregated data differentiating between standard pregnancy costs and those associated with undesirable complications or inefficiencies would have been more useful for policymaking, but this would have required a more nuanced, bottom-up approach.

Paper 3 estimated the total cost of L&D by analysing the cost of commissioning the programme for each site. This is unlikely to reflect the true cost of the intervention. For example, it does not capture in-kind costs, such as the opportunity cost of existing facilities being made available to L&D staff, and the time dedicated by non-L&D staff. In practice, decision-makers are interested in the whole cost of an intervention, including opportunity costs. The top-down approach taken means the full cost of L&D is unclear. Time, resource and data precluded more detailed costing in Paper 2 and 3.

3.2.4 Influence of the time horizon

Most of the papers adopted a relatively short time horizon. Paper 3 considered the costs and outcomes associated with L&D over a one-year period, whereas Paper 4 only considered the six-month follow-up period of the trial. The cost-effectiveness observed in a trial can differ from what would have been observed had follow-up continued, particularly when an intervention is likely to have a long-term impact [99]. Given that L&D targets vulnerable criminal justice users, where contact with such services has shown to exacerbate long-term vulnerabilities [73], and OSI treats child anxiety problems, which have been shown to impact outcomes into adulthood [92], the relatively short time horizons may not appropriately capture the costs and outcomes associated with the interventions [133].

Paper 2 considered a longer 5-year time horizon. A prevalence-based approach was taken, where costs are only considered for the period of interest. However, as pregnancy complications, such as preterm birth, are associated with poorer outcomes later in life [134] an incidence-based approach may have been more appropriate for estimating the total healthcare costs associated with pregnancy, as it adopts a lifetime horizon [59].

Estimating the long-term costs and outcomes of an intervention typically involves economic modelling to extrapolate the results beyond the time horizon of the trial. However, reliable modelling depends on the

availability of good quality long-term data on the effectiveness and treatment adherence of such interventions, which is often not available [100]. Additionally, it is argued that long-term modelling is only necessary if it is likely to result in a different cost-effectiveness conclusion being drawn to that of the within-trial analysis [100]. Otherwise, a within-trial cost-effectiveness is likely to be sufficient [100].

3.3 Preference for an approach

The strengths and weaknesses of different approaches to economic analysis have been discussed in depth elsewhere [16, 135]. This critique will focus on the context of the five publications and the practicalities that led to the approaches that were taken, providing empirical evidence to compliment these textbook discussions.

3.3.1 Preferred approach

Ideally, a full economic evaluation would be undertaken with the results captured in a single generic measure of efficiency, such as a cost per QALY gained ICER, so that they can be compared to established willingness-to-pay thresholds and equivalent analyses of different interventions. This is in line with NICE guidelines, which seeks to ensure common methods are used for the evaluations of health technologies [103].

The forward citation analysis demonstrated that end-users largely sought such evidence as they attempted to influence policy. The time horizon should be long enough to incorporate all the potential costs and outcomes associated with the intervention, taking both a healthcare and societal perspective to account for different decision-making preferences. In practice, this is often not feasible for a variety of reasons and alternative approaches, even if they do not provide information on value for money, may be adopted.

3.3.2 Importance of the research question

The research question may largely be predetermined by the research commissioner. This was true of Paper 1. As HEE were responsible for workforce planning, CRUK commissioned the research to estimate the additional funding the organisation would need to grow the cancer workforce. A cost analysis was therefore sufficient, as was the narrow cost perspective, despite the associated limitations. Ultimately, the approach needs to address the research question at hand. If this is driven by stakeholders external to the researcher, then overarching approach selection may be limited.

3.3.3 Economic analysis as a secondary consideration

Economic analysis can be a secondary consideration, particularly when part of a larger project, as highlighted by Whitty [18]. This was the case with Paper 3, where data access and the difference-in-differences analysis consumed most of the time and resource dedicated to the project. This meant that, in practice, researchers were tasked with undertaking an economic evaluation of L&D after the data had been collected and with relatively limited time and resource.

Ideally, this would be avoided. Co-design, via end-user engagement, can help identify the desire for economic analysis at an early stage and the research can be designed appropriately. For example, the economic analysis can be embedded in the wider project, as it was with Paper 4. This allowed for a more comprehensive economic evaluation to be conducted, enhancing translation to policy impact.

If a researcher is faced with conducting an economic analysis as a secondary consideration, the scope and design of the research will be determined by existing available data. A first step would therefore be to determine which costs and outcomes could be incorporated into an analysis, assuming there is not sufficient resource to undertake additional primary data collection. With this information, the most appropriate approach can be identified, given the resources that are available. For example, no HRQoL data were available for Paper 3, which explains why a CEA and CUA were not undertaken. Less than half of the relevant costs and outcomes were included in the analysis, precluding a CBA as the cost and outcome perspective was too narrow. A CCA was therefore deemed most appropriate.

3.3.4 Data availability

Even if the economic analysis is not a secondary consideration, the method is often determined by the available data. This was true of Paper 1, when deciding which approach to use for the cost analysis. Given that detailed workforce data were available, demographic stock-flow models were developed to maximise use of the data's granularity, by modelling the workforce by sex and year of age. The methodological choice in Paper 2 was also largely a product of the available data. The total healthcare cost of pregnancy was estimated and incorporated into the contextualisation of research spending, as the data were not granular enough to distinguish between desirable and undesirable pregnancy costs.

Ultimately, the overarching approach of Paper 1 and 2 was largely driven by the research question. However, the specific method used to undertake the cost analyses was dependent on the available data. Even if the research question requires a full economic evaluation with a single measure of efficiency, data may not be available to undertake such a comprehensive analysis within the available time and

resource, as was the case with Paper 3. Economic modelling can be undertaken when the necessary data are lacking. For example, cost-effectiveness can be explored across plausible ranges or informed by expert opinion. However, as previously stated, developing such models is resource intensive.

3.3.5 Time and resource

Even where the economic analysis is integrated into the research project and the necessary data is available, the available time and resource are crucial in determining the approach. The modelling underpinning the cost analysis in Paper 1 was highly reproducible, with a single model structure developed and reproduced for the seven cancer professions, using the relevant data for each. This enabled the economic analysis to address the research question within the relatively limited resource available. It also ensured timely results, before the next spending review, which is crucial for research to influence policy [18].

The RCT undertaken in Paper 4 was designed to ensure all appropriate data were collected to undertake a CEA and CUA, from a healthcare and societal perspective. However, resource was not available to estimate the long-term costs and outcomes associated with OSI, which would have required the development of a *de novo* economic model. The cost-effectiveness of OSI was therefore only considered within the 6-month trial period.

The cost analysis and contextualisation of pregnancy research in Paper 2 was part of a wider project. It was therefore not possible to undertake a more comprehensive return-on-investment study given the resource allocated to the economic analysis. Similarly, the outcome evaluation in Paper 3 utilised most of the time and resource allocated to the project, which largely explains why the economic evaluation of L&D was a secondary consideration and a relatively low resource intensive CCA was undertaken.

3.3.6 Evidence synthesis

The assumption so far has been that the necessary economic analysis has not already been conducted. If multiple studies have already assessed the cost-effectiveness of the intervention of interest, in the context that it will be implemented, then a synthesis of the evidence base would be most appropriate, given the evidence is of sufficient quality to inform policy. In fact, Whitty argues the accurate synthesis of existing evidence is the most valuable form of research for policy making [18].

Systematic reviews use well-established, transparent and reproducible methods that allow the relevant evidence to be identified and synthesised to determine cost-effectiveness, as per Paper 5. However, it is

important that the context of the underlying analyses is considered before decision-makers implement policy based on the results of the evidence synthesis.

3.3.7 Summary

A full economic evaluation with a single generic measure of efficiency, an appropriate time horizon, and a healthcare and societal perspective is the gold standard for decision-making. However, this is often not appropriate or feasible. Table 3 summarises the discussion by presenting various queries decision-makers and researchers often face when undertaking economic analysis to inform public policy. It is not a comprehensive guide accounting for all eventualities. Instead, the questionnaire summarises the lessons learned from the critical analysis, providing advice on the considerations to be made and guidance on the potential approaches to be taken for the economic analysis, given the query of interest. In practice, researchers and decision-makers may face some, none, all, or more queries than those presented in Table 3 when deciding what type of economic analysis to undertake.

Table 3. Questionnaire to guide potential economic analysis approaches

Query	Considerations	Potential approaches
Are there one or more studies evaluating cost-effectiveness in your specific context?	If yes, is the evidence strong enough to inform policy? If no, are there reasons it hasn't been done yet? Is it feasible? Is it appropriate?	If strong evidence exists, consider using it to inform policy. If multiple studies exist, consider synthesising the evidence base. If no or weak evidence exists, consider undertaking a full economic evaluation, if feasible and appropriate.
Has a research question been predetermined that does not require a full economic evaluation?	If yes, do end-users want information on value for money? If no, is a full economic evaluation desired? Is it feasible? Is it appropriate?	If a full economic evaluation is not desired, consider undertaking a partial economic evaluation that addresses the research question. Consider engaging research commissioner and end-users in co-design to help ensure the desired output is produced.
Is the economic analysis a secondary consideration in a wider research project?	If yes, what is the best you can do with the available data, time and resource? If no, what output does the commissioner and end-user desire?	If the economic analysis is a secondary consideration, consider listing all potential costs and consequences and map them to the available data to decide on the approach, given time and resource constraints. If it is not a secondary consideration, consider co-designing the research with end-users and embedding the economic analysis within the wider project, if applicable.
Are data available to undertake a full economic evaluation?	If yes, is a full economic evaluation desired? Is it feasible? Is it appropriate? If no, what is the best you can do with the available data?	If data are available, consider undertaking a full economic evaluation, if feasible and appropriate. If data are not available, consider undertaking a partial economic evaluation that maximises the available data, or consider economic modelling if time and resource allow.
If a full economic evaluation is feasible and appropriate, is there time, resource and data for an appropriate time horizon, and a healthcare and societal perspective?	If yes, can you comply with established guidelines? If no, what is the best you can do with the available data? Can you comply with established guidelines?	If there is time, resource and data, consider undertaking a full economic evaluation with an appropriate time horizon, and a healthcare and societal perspective. If there is not time, resource and data, consider undertaking a full economic evaluation prioritising decision-makers preferred perspective, with a time horizon as appropriate as possible.

Chapter 4. Conclusion

4.1 Summary

A critical analysis of five publications conducting economic analyses has been presented in this thesis.

The introduction of the critical analysis provided context to the publications and the central theme that runs through them. Economic analysis is crucial for public policy as it provides decision-makers with the information needed to efficiently allocate society's limited resources.

Chapter 2 drew on the introduction's contextualisation of economic analysis to develop an analytical framework (Table 1) that was applied to critically analyse each of the papers in turn. The background, methods and contribution of the research was considered, all of which were largely driven by the context of each research project.

The publications were then discussed collectively in Chapter 3, pulling together the insights gained from Chapter 2. Meaningful end-user involvement appeared to improve the research and its reach. Ultimately, though, the research that appeared to have most policy impact was that which provided decision-makers with information on value for money. However, none of the publications explicitly considered equity, i.e., how costs and outcomes are distributed across different groups.

The influence of the methodology on the results and therefore the policy decision was also explored. This found that the overarching approach of the economic analysis (e.g., CEA vs. CUA) and the within-method approaches (e.g., healthcare vs. societal perspective) can lead to different results.

The thesis then reflected on how the circumstances of the research can lead to the preference for a particular approach to the economic analysis. In the context of the included publications, the approach was guided by the current evidence base, the research question, the stage of the research within the wider project, and the available data, time and resource.

A questionnaire was developed based on the learnings from the critical analysis, providing advice on the considerations to be made and the potential economic analysis approaches to be taken when faced with various pertinent queries (Table 3). This may be of use for researchers looking to conduct economic analysis and decision-makers looking to use it to inform their decisions.

4.2 Strengths and weaknesses of the critical analysis

The critical analysis should be considered in light of several strengths and limitations.

A key strength of the critical analysis was the systematic approach to the critique, through the development of an analytical framework (Table 1) facilitating robust assessment against pre-defined criteria. The framework enabled the consideration of factors beyond those included in typical reporting standard checklists (e.g., the CHEERS checklist [30]) to explore the circumstances that led to the economic analysis approach, and what could have been done differently. However, the framework was developed based on the contextualisation of economic analysis presented in the introduction and is therefore not necessarily comprehensive, potentially failing to capture aspects relevant for other economic analyses.

Formal forward citation analysis was undertaken for each of the papers to inform the discussion of the contribution of the publications individually and collectively. Several of the publications included in this thesis are policy reports and therefore not well suited to being identified through searches of traditional databases (e.g., PubMed). To account for this, the literature search was highly comprehensive, undertaking extensive searches of Google and Google Scholar to ensure as much evidence was identified as possible.

A tangible output of the critical analysis is the questionnaire that was developed to provide advice to researchers and decision-makers on the considerations to be made, and the potential economic analysis approaches to be taken, given typical queries they may be faced with (Table 3). This aims to support the undertaking of more useful and translational research, in turn supporting evidence-based policy and rational decision-making. However, the questionnaire is not a comprehensive guideline and only considers factors that arose from the critical analysis of the five publications. In practice, there are likely to be many other factors to be considered that are not included.

4.3 Future research

The critical analysis has reflected on ways in which future research can develop on the economic analyses presented in each of the publications. For example, the cost analysis in Paper 1 could have taken a broader perspective to estimate the total cost of increasing the NHS cancer workforce. Paper 2 could have built on the cost analysis and contextualisation of pregnancy research to determine the return-on-investment of the research funding. Paper 3 could have undertaken an economic evaluation of the L&D programme based on the mean net benefit of the intervention, rather than the statistical significance of the difference-in-differences analysis. Paper 4 could have considered a longer time horizon to capture the longer-term costs and outcomes associated with OSI. The umbrella review in Paper 5 could have considered the context of the underlying economic evaluations to better enable

policy decisions based on the results. The publications could also have considered the distribution of costs and outcomes across different groups, rather than focussing solely on efficiency. The practicalities faced by researchers precluded such analyses at the time, as explained in the critical analysis. However, future research could address these issues.

Future critical analysis of such work could seek to develop a more comprehensive analytical framework (Table 1) to aid the critique of specific economic analyses. As mentioned, the framework was developed based on the general discussion of economic analysis in the introduction. Future research could systematically identify all the key considerations that should be explored in a comprehensive analytical framework that moves beyond typical reporting standard checklists.

Future research could also attempt to develop a comprehensive guideline regarding which economic analysis approach should be taken given the context faced by researchers, building on the questionnaire presented in Table 3. For example, a flow diagram could be developed which provides clear guidance on approach based on responses to the different situations a researcher might face.

4.4 Conclusion

In conclusion, this thesis has demonstrated that economic analysis can provide evidence to support funding allocation, research priorities and intervention choice across sectors and countries, even in the face of various practical constraints (e.g., data, time, resource). It has also provided a novel, systematic analytical framework (Table 1) and questionnaire (Table 3) for decision-makers and researchers to aid the consideration of methodological choice for economic analysis.

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Appendix

Appendix A. Paper 1

George, J., Gkousis, E., Feast, A., Morris, S., Pollard, J., & Vohra, J. (2020). **Estimating the cost of growing the NHS cancer workforce in England by 2029**. London: Cancer Research UK. URL: https://www.rand.org/pubs/external_publications/EP68310.html

Estimating the cost of growing the NHS cancer workforce in England by 2029

October 2020

Together we will beat cancer

CANCER RESEARCH UK

Reference

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Cancer Research UK

Cancer Research UK is the world's largest independent cancer charity dedicated to saving lives through research. We support research into all aspects of cancer through the work of over 4,000 scientists, doctors and nurses. In 2019/2020, we spent £455 million on research institutes, hospitals and universities across the UK. We receive no funding from Government for our research.

This research was funded by the Policy Development team and conceived in collaboration with the Cancer Policy Research Centre, Cancer Research UK.

Cancer Research UK is a registered charity in England and Wales (1089464), Scotland (SC041666) and the Isle of Man (1103).

http://www.cancerresearchuk.org/

RAND Europe

RAND Europe is a not-for-profit research organisation that helps to improve policy and decision-making through research and analysis. RAND's publications do not necessarily reflect the opinions of its research clients and sponsors.

This research was carried out through a collaboration between researchers at the University of Cambridge and at RAND Europe, The Cambridge Centre for Health Services Research (CCHSR). Co-led by Steve Morris, RAND Professor of Health Services Research at University of Cambridge, and Jon Sussex, Chief Economist at RAND Europe, CCHSR's aim is to inform policy through evidence-based research on health services.

http://www.randeurope.org

List of acronyms

AfC	Agenda for Change (NHS pay scales)
ALBs	Arm's-Length Bodies
CPD	Continuing Professional Development
DHSC	Department of Health and Social Care
ESR	Electronic Staff Record
FTE	Full-Time Equivalent
GDPR	General Data Protection Regulation (EU)
HEE	Health Education England
LTP	(NHS) Long Term Plan
MFF	Market Forces Factor
NHS	National Health Service
NQT	Newly Qualified Trainee

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Foreword

The UK public has always held doctors, nurses and health professionals in high regard – this is nothing new. What is new is what we saw during the height of the coronavirus (COVID-19) pandemic – millions of us uniting to show our appreciation for our NHS staff; week in, week out.

NHS staff do heroic things, but many would tell you that they are not themselves heroes. They are highly trained, highly skilled professionals, doing the job they are paid to do. Recruiting and training these professionals requires investment.

Even before the coronavirus pandemic began, NHS staff were working harder than ever to support a system that was too often running overwhelmed, with not enough capacity to meet the needs of all patients. In large part, this is due to a persistent failure from successive governments to properly invest in NHS staff.

When we stood on our doorsteps and balconies to clap for the NHS what we acknowledged, more than anything, was the immense pressure that NHS staff are under and our pride for the work they do. This pressure did not begin with COVID-19 and will not end after the pandemic – unless the Government takes significant action to provide adequate funding and prioritises the workforce in future NHS policy.

This has been all too apparent in cancer services. Early diagnosis is critical for good cancer outcomes. Growing pressure in diagnostic services due to staff shortages has led to a significant decline in performance against key cancer waiting times standards, and to staff delivering vital cancer treatments reporting that shortages are harming patient care.

Without action, this situation is only going to get worse. The number of people being diagnosed with cancer is growing all the time – by 2035, nearly 438,000 people are expected to be diagnosed with cancer in England every year, an increase of more than 40 per cent on 2015 levels, with 130,000 more cases projected. While the NHS Long Term Plan has set welcome ambitions for cancer services, meeting these ambitions will increase demands on staff even further.

We do not yet know what the full impact of the COVID-19 pandemic will be on the NHS' need for staff in the future and its ability to recruit them – but it is likely that it will be significant and require Government action. The alarming figures recently seen on the backlog of people awaiting cancer tests or treatment will clearly impact on staff capacity to cope and may further shape the future landscape.

But what we do know – and what this report shows – is that, notwithstanding the impact of COVID-19, significant growth is needed almost across the board to equip the NHS workforce to diagnose and treat cancer patients in the future. This report makes clear that the Government must make significant, targeted investments now to make sure the NHS is ready for what lies ahead.

This important report sets out the investment needed in the recruitment and training of staff to support the delivery of excellent healthcare. Investing in the health care workforce is expensive, and too often Governments' have baulked when faced with such a large cost. But less visible is the cost of not investing – staff shortages, lack of capacity and a reliance of temporary staffing options. Investing in staffing now is critical to delivering the high value health service we all want and need.

Anita Charlesworth

Director of Research and Economics
The Health Foundation

Executive summary

Over the last 50 years, the UK has made significant progress in improving survival outcomes for people diagnosed with cancer. In the 1970s, only 1 in 4 cancer patients would survive their disease for ten years or more. By 2010, this had risen to 2 in 4, and survival outcomes continue to improve.[1] There are several drivers for this, from the introduction of screening programmes and innovative treatments to ongoing improvements to health care pathways – all of which has been underpinned by the continued and tireless efforts of staff from across the NHS.

However, there is still much to be done. Cancer remains one of the leading causes of death in England. Every day around 1,000 people in England are diagnosed with cancer, and around 450 die from the disease.[1] Demand for cancer services is growing: more cases each year are diagnosed, and people are on average living for longer with cancer. An ageing and growing population means the cancer incidence continues to rise at an alarming rate, with many patients having increasingly complex needs. By 2035, the number of people estimated to be diagnosed with cancer in England is expected to reach over half a million per year, an increase of 40 per cent since 2015.[2] We also see lower survival in the UK than in comparable countries around the world, and significant variation in outcomes across the UK too.[3,4]

Critical to addressing these challenges is our ability to diagnose cancer at an early stage. Patients diagnosed early, at stages 1 and 2, have the best chance of curative treatment and long-term survival.[5] But in the UK, we currently diagnose just over half of patients at an early stage.[6] The importance of this challenge is recognised by the UK Government and NHS England, in their commitment to diagnose 75 per cent of cancers at an early stage by 2028.[7]

But the diagnosis, treatment and support of people living with cancer relies on a range of skilled NHS staff conducting specialist tasks such as performing and reporting on diagnostic tests and providing different forms of treatment and support. Ensuring that the NHS has enough skilled staff, now and in the future, is therefore a vital part of fulfilling the ambitions of the LTP and improving outcomes for cancer patients.

Despite attempts to increase size of the cancer workforce, key cancer-related professions have remained under pressure, with vacant posts and staff shortages. Cancer Research UK found that nearly three in four staff surveyed in non-surgical oncology services see staff shortages as a barrier to providing excellent patient experience.[8] Capacity constraints, particularly due to staff shortages in diagnostic services, are associated with poor performance against NHS waiting times standards.[9] Even before the COVID-19 pandemic, the NHS had been reporting worsening performance against both the two-week wait for urgent suspected cancer referrals and the 62-day Cancer Waiting Times treatment standards.[10]

In June 2018, the government announced a long-term funding settlement for the NHS,[11] whereby NHS England's annual budget would rise by an extra £20.5 billion by 2023–24. No

announcement has been made beyond 2020–21 for other health bodies such as Health Education England (HEE), even though HEE is responsible for planning for and training the staff that will be required to deliver the government's commitments.

Understanding mechanisms for workforce growth and their associated costs

In 2017 the NHS, led by NHS England and HEE, published a Cancer Workforce Plan for England.[12] This assessed the cancer workforce against the increasing demand for diagnostic capacity and for cancer care and raised particular concerns about whether the NHS had sufficient staff in seven key specialisms most closely related to cancer diagnosis and treatment: clinical and medical oncology, gastroenterology, histopathology, clinical radiology, diagnostic radiography, therapeutic radiography and specialist cancer nursing. In 2018 HEE set out that, to provide a world-class service for NHS cancer patients, the workforce in these seven priority professions would likely need to increase by 45 per cent by 2029.[13]

This study aimed to understand the increase in HEE's future budget, beyond existing trends based on current levels of investment, required to recruit and train more staff to increase the NHS cancer workforce by 45 per cent by 2029 in England. These findings can inform the Government's spending review by providing an estimate of the additional budget settlement required to promote workforce growth.

This report identifies several different ways of increasing staff numbers: by increasing training places, international recruitment and encouraging higher retention levels. It is important to note that the costs identified in this report do not represent the full cost to the health sector of increasing staffing in key professions, only those that HEE is responsible for. Many of the costs of international recruitment, for example, are borne by NHS Trusts, which are funded by the NHS England funding settlement.

Workforce and cost data have been identified for each of the priority professions in order to build a demographic stock-flow model, with workforce data based on number of staff in post rather than the number of funded positions – this means the modelling does not consider vacancies in funded posts. The model allows us to build up an idea of how many people we would expect to be in post in each profession each year, if current trends of entering and leaving that particular workforce continue. Using this model, we identified how many more people, over and above existing trends, HEE or other parts of the health system would need to recruit to meet an increase of 45 per cent in staffing by 2029. We tested the logic and validity of our findings by consulting a stakeholder advisory panel and holding a validation workshop to obtain feedback on the data and emerging messages.

Key findings

Aside from gastroenterology, all professions would need additional government investment to meet a growth of 45 per cent by 2029

Workforce numbers for six of the seven professions are already on an increasing trajectory, assuming a continuation of current trends. For one profession, gastroenterology, our model estimated that the workforce would increase by more than 45 per cent if its existing trajectory continues. Therefore, no *additional* measures to recruit and train staff would be required. For five of the other six professions, we estimated that the size of the workforce would increase to differing degrees over the period, but that there would be a need for additional intervention to achieve the needed growth rate of 45 per cent.

Table 1 sets out the estimated growth of each profession on the basis of current trends, and the estimated *additional* FTE required to achieve 45 per cent growth.

Table 1: Summary of the estimated growth of cancer specialists and additional number required to reach 45 per cent growth

Cancer profession	Estimated number of FTE staff in 2019	Expected growth by 2029 (based on current trends)		Estimated additional growth required to meet 45 per cent growth (above expected growth)		Total (expected and additional) increase in FTE staff needed to reach 45 per cent growth
		Percentage growth	Estimated FTE staff increase	Additional percentage growth	Additional FTE staff required	
Clinical and medical oncology	1,185	40%	477	5%	57	534
Gastroenterology	1,290	48%	623	0%	0	623
Histopathology	1,228	-2%	-27	47%	580	553
Clinical radiology	3,087	33%	1,004	12%	384	1,388
Diagnostic radiography	14,997	28%	4,158	17%	2,591	6,749
Therapeutic radiography	2,844	24%	672	21%	609	1,281
Specialist cancer nurses	4,135	28%	1,149	17%	710	1,859
Total	**28,766**	**-**	**8,056**	**-**	**4,931**	**41,753**

Source: RAND Europe modelling using NHS ESR data and other data.

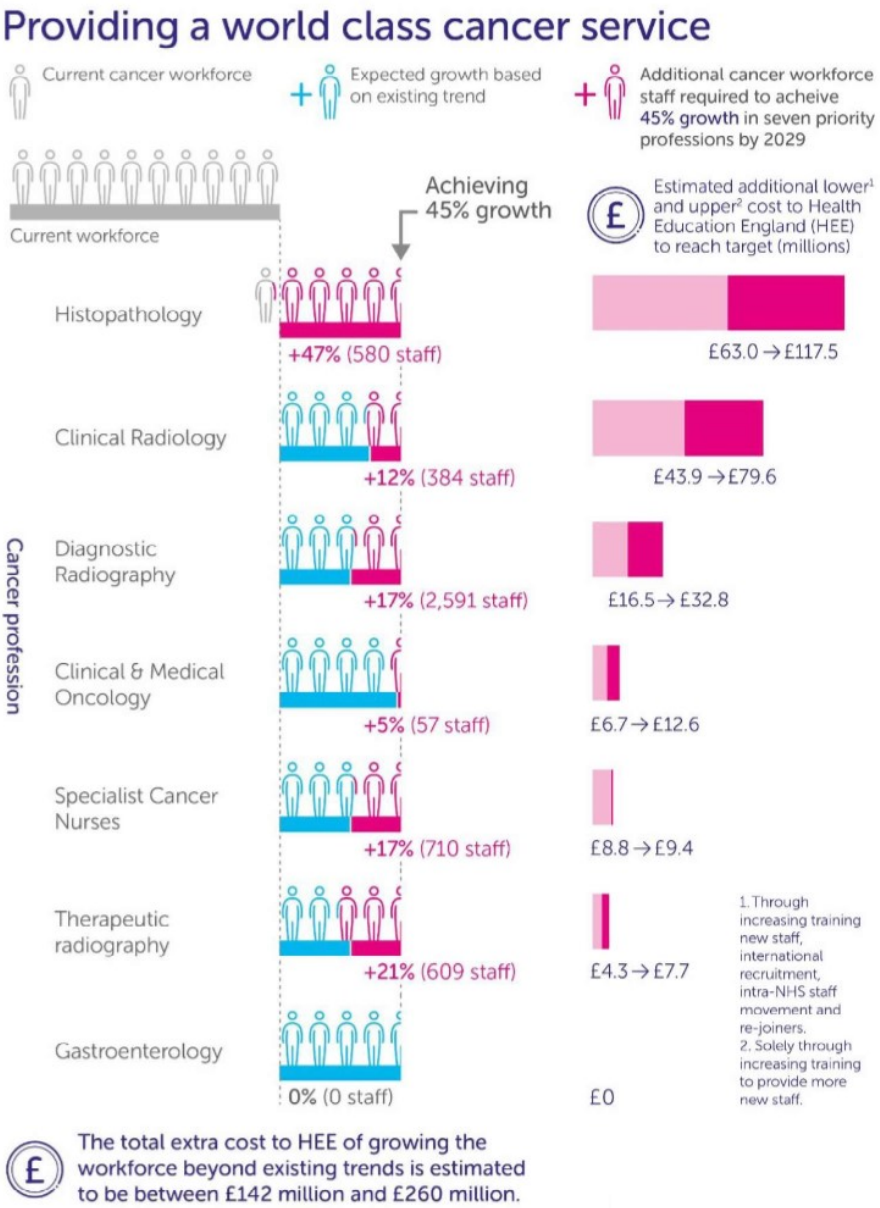
The histopathology workforce is forecast to decline

The results of the economic modelling show that achieving a growth of 45 per cent may be more difficult in some professions than others. For example, in clinical and medical oncology, if current trends continue this workforce would only need to grow by a further net addition of 57 staff to meet the 45 per cent growth scenario. However, in diagnostic radiography, an

estimated additional 2,591 staff would be needed in addition to the existing trend.

The modelling shows that, without any new intervention, the number of histopathologists is expected to reduce by 2 per cent by 2029. Therefore, to meet a 45 per cent growth, the profession would require 580 more staff at a potential maximum cost to HEE of £118 million.

Figure 1: Summary of the total additional number of cancer specialists required over ten years to 2029 and the total cost to HEE of different scenarios



On the number of staff required, we recommend:

1. NHS England and Improvement should ensure that their next People Plan adequately reflects the NHS's ambitions for cancer care in England by clearly articulating details of how many staff will be needed to deliver quality services to a growing number of patients in the long-term.
2. NHS England and Improvement, working with HEE, should review how it can influence its pipeline of staff to the professions where they are most needed, with a particular focus on histopathology. The forecast of histopathology workforce numbers is particularly concerning, as this is the only profession with current trends suggesting a decline in staff numbers by 2029.

The total extra cost to HEE is estimated to be between £142 million and £260 million

Estimates for each profession explored three different scenarios that vary according to the extent to which the additional staff would be coming through specialist training or via other routes.

We estimate that the total additional cost to HEE to achieve a 45 per cent growth in the key cancer workforce professions would be between £142 - £260 million (Figure 1). (NB. The higher estimate of £260 million is approximately 6 per cent of HEE's spending for one year). The actual cost will depend on the route taken to increase the inflows over the next ten years to 2029. This funding would need to be front-loaded due to the time required to train staff. Of the total estimated cost to HEE, almost half would be required to train additional histopathologists due to the forecast decline in numbers in that specialism by 2029.

Since the analysis was completed, the NHS – led by NHS England, NHS Improvement and HEE – published an NHS People Plan for 2020/21, which takes some steps to increasing staff numbers in some of the key cancer professions. Whilst this is welcome, they do not address the long term needs outlined in this report.

On the cost of increasing the cancer workforce, we recommend that:

3. The UK Government should provide long-term additional funding to HEE, aligned to the NHS Long Term Plan, to secure a sufficient pipeline of future NHS cancer staff. Initial estimates suggest that, to achieve a 45% growth in the seven key cancer professions, *additional* funding for HEE will need to be at least £142 million but possibly as much as £260 million over the next three to five years. This should be taken into account in the next Comprehensive Spending Review.

There are some considerations about the feasibility of the modelled scenarios

Although the research modelled different cost scenarios, there may be some limitations in practice to their feasibility. Scenarios that include staff increases through a mixture of routes including international recruitment and encouraging rejoiners will be affected by external circumstances such as the COVID-19 pandemic and the UK's exit from the EU. Equally, scenarios that rely heavily on increasing the number of professionals trained may be limited by the

number of work placements available.

Two additional salient points are:

- The importance of extending foundation-level training, to provide sufficient doctors so that different parts of the NHS are not competing for the same limited pool of clinicians.
- The need to understand a nuanced picture of required future staffing, including the possibility of different roles and multidisciplinary working. For example, there may be a greater demand in future for some of these professions than for others.

On the scenarios for increasing the workforce we recommend that:

4. NHS England and Improvement, along with HEE, should assess the feasibility of the different scenarios for increasing inflows. This is particularly important in light of the likely restrictions arising from the COVID-19 pandemic and the UK's exit from the EU, which may reduce international recruitment.
5. Building on the NHS People Plan 2020/21, NHS England and Improvement, in collaboration with HEE, should ensure that consideration of likely future demand and planned future modelling of the workforce is translated into clear long-term ambitions for the growth of different cancer professions.
6. This research project identified several important areas for future research. NHS England and Improvement, the National Institute for Health Research and other funders should commission work to fill these research gaps, to inform its approach going forwards with a fuller understanding of the needs of the cancer workforce and the constraints to its growth.

Conclusion

This research is important in starting to understand the likely requirements for the NHS, and particularly for HEE, of increasing the number of staff in seven priority cancer professions by 2029. Whilst all seven professions will require continued investment to maintain current growth trends, this modelling indicates that this will not be enough and that *additional* investment will be required.

The modelling also contributes important information about which workforces will require the most significant interventions by HEE if they are to grow by 45 per cent, as well as an estimate of the associated specialist training costs of such interventions. In addition to HEE's important role in securing staffing, the report also explains that other parts of the health sector, particularly NHS trusts, also play a role.

While three different scenarios for increasing the workforce are set out, external factors such as the impact of COVID-19 will likely have significant implications for the NHS's ability to attract more staff through certain routes. This study will allow policymakers to consider the relative costs and feasibility of alternative options.

Contents

Introduction

Background and rationale

Cancer Research UK commissioned RAND Europe to carry out a study to inform understanding of the future budget that Health Education England (HEE) would require to recruit and train more staff, if the NHS in England is to make sufficient progress on its key workforce requirements for cancer by 2029.

Cancer is one of the leading causes of death in the UK. Every day over 800 people in England are diagnosed with cancer, and nearly 400 die from the disease.[14] Demand for cancer services is growing: more cases each year are diagnosed, and people are on average living for longer with cancer. An ageing and growing population is likely to mean that nearly 438,000 people per year are expected to be diagnosed with cancer in England by 2035, an increase of 40 per cent since 2015.[15]

The NHS in England has placed a strong emphasis on cancer: the NHS Cancer Transformation Programme has been created, and the NHS Long Term Plan (LTP) aims to improve early diagnosis of cancer as well as patient outcomes.[16] The diagnosis, treatment and support of people living with cancer relies on a range of skilled NHS staff performing specialist tasks such as performing and reporting on diagnostic tests and providing different forms of treatment and support. Ensuring that there are sufficient skilled staff in place is therefore a vital part of improving cancer services and their outcomes for patients.

In 2017 the NHS published a Cancer Workforce Plan for England, led by NHS England and HEE.[17] This assessed the cancer workforce against the increasing demand for diagnostic capacity and for cancer care, and raised concerns about whether the NHS had sufficient staff in seven key NHS specialisms that were most closely related to cancer diagnosis and treatment: clinical and medical oncology, gastroenterology, histopathology, clinical radiology, diagnostic radiography, therapeutic radiography and specialist cancer nursing. The plan contained actions to grow the cancer workforce by a total of 4,126 staff across six of the seven priority professions by 2021, a planned increase of 18.5 per cent.

Although the Cancer Workforce Plan focused on seven priority professions where there were particular concerns about capacity, there are a huge number of other health professionals who routinely deal with patients living with cancer, including allied health professionals, medical physicists, palliative care staff, surgeons, clinical endoscopists, advanced clinical practitioners and a wider range of support staff. It is also important to note that some of the seven professions, such as oncologists, work exclusively in cancer, whereas others, such as gastroenterologists and histopathologists, also care for patients with a range of other diseases, and not solely cancer. Nevertheless, in this report we focus on the seven professions identified as a priority by the 2017 Cancer Workforce Plan.

Cancer workforce under pressure and the impact on patients

Despite attempts to increase the cancer workforce, key cancer-related professions have remained under pressure. For example, the Society of Radiographers reported in its 2018 survey that almost one in ten diagnostic radiography posts were vacant, meaning services have been struggling to meet demand.[18] Cancer Research UK found that nearly three in four staff surveyed in non-surgical oncology services see staff shortages as a barrier to providing efficient cancer treatments and excellent patient experience.[19] One third of cancer patients believe that there were sometimes or rarely sufficient nurses on duty to care for them in hospital.[20] Capacity constraints, particularly due to staff shortages in diagnostic services and increased pressure on emergency services, are associated with poorer performance against NHS waiting times standards.[21] Even prior to the COVID-19 pandemic, the NHS had been reporting worsening performance against both the two-week wait for urgent suspected cancer referrals and 62-day Cancer Waiting Times treatment standards.[22] In September 2019, 77 per cent of patients were seen within the 62-day standard.[23]

There is a large and increasing amount of evidence for a relationship between low staffing levels and adverse patient outcomes, including higher mortality rates.[24] For example, the Francis Report identified inadequate staffing levels as one of the key factors that led to poor quality care at Mid Staffordshire NHS Trust.[25] The *Review into the Quality of Care & Treatment provided by 14 Hospital Trusts in England*, led by Sir Bruce Keogh, similarly pointed to the impact of workforce issues on hospital mortality.[26] More specifically there is also evidence on staffing levels and patient outcomes for patients living with cancer.[27]

Ambitions for the future cancer workforce

The emphasis in the NHS Cancer Transformation Programme on early diagnosis and treatment of cancer was renewed in the LTP, published in January 2019.[28] This plan outlined an ambition that by 2028 for every four patients diagnosed with cancer, three will be diagnosed at an early stage. It stated that currently only around half of patients with cancer are diagnosed while the disease is at an early stage. The LTP set out its aims to improve cancer outcomes by improving national screening programmes, giving people faster access to diagnostic tests, and investing in new technologies so that more patients can benefit from highly personalised treatments. Initiatives in the LTP should help to improve early diagnosis rates but will also require the NHS to diagnose and treat more people. However, it will be difficult to achieve these ambitions and improve cancer outcomes without significant increases in staff numbers.

In June 2019, the NHS published the Interim NHS People Plan,29 which focused on the need to transform the workforce through new roles and multidisciplinary work, setting out immediate actions for 2019/2020. Then, in July 2020, the NHS published "We are the NHS: People Plan 2020/21 – action for us all",30 which set out some further steps to expand the cancer workforce. Commitments included training 450 reporting radiographers, providing training grants for 350 nurses to become cancer nurse specialists and chemotherapy nurses, and

training 400 clinical endoscopists, amongst others. It also set out short-term actions to improve workforce capacity by promoting international recruitment and encouraging former staff to return to practice.

Whilst these are positive steps, further detail is required that extend beyond the short-term. The NHS has said that more details on expanding the workforce in the longer term will be published after the next government spending review, scheduled to take place in Autumn 2020.

The role of HEE and NHS funding

Historically, HEE has played a significant role in workforce development, with responsibilities for providing system-wide leadership and oversight for workforce planning, education and training across England, working alongside system partners including other NHS arm's-length bodies (ALBs), NHS Providers and Royal Colleges. Despite governance changes in the health system, HEE remains responsible for workforce planning, education and training. As part of its strategic workforce planning relating to key cancer professions, HEE's internal modelling estimated in 2018 that to deliver world-class cancer services by 2029, the NHS in England would require an aggregate growth of 45 per cent in its cancer workforce.[31]

In June 2018, the government announced a long-term funding settlement for the NHS, whereby NHS England's annual budget would rise by an extra £20.5 billion by 2023–24.[32] This funding increase allowed NHS England a longer planning horizon than most other central government bodies. However, no announcement was made for other health bodies including HEE, despite the latter's vital role in workforce planning and training. In September 2019[33] and March 2020,[34] the NHS budget was increased further, and similar increases were also announced for capital investment, public health and staff education and training. But unlike the core NHS funding increases, funding for these areas has not been determined beyond 2020/21.

This project therefore seeks to inform understanding of the increase in the future budget, beyond existing trends, that HEE would require to recruit and train more staff in order to increase the NHS cancer workforce by 45 per cent by 2029. It looks at the costs for three scenarios (see next section) for how such an increase in NHS staff might be achieved. Its findings will be useful in informing the next government spending review by outlining the budget settlement required to promote workforce growth. Although there are interlinkages between HEE and other national and local organisations with respect to workforce planning, training and recruitment, this project only seeks to identify the costs relating to HEE. Note, that the analysis in this report does not take into account commitments made to expand parts of the cancer workforce in the NHS People Plan 2020/21 as these commitments were announced after the analysis was conducted.

Research aims and objectives

The aim of this research was to identify how much additional funding Health Education England (HEE) would need in order to achieve three different growth scenarios in seven key cancer professions by 2029. These seven professions are:

- clinical and medical oncology
- gastroenterology
- histopathology
- clinical radiology
- diagnostic radiography
- therapeutic radiography
- specialist cancer nursing.

The seven professions were identified by HEE in its 2017 Cancer Workforce Plan as being of most significant concern at that time in terms of capacity.[35]

The key research objectives for the study were to:

- Establish how many NHS staff in England currently work in each of the seven key cancer professions.
- Model how many additional staff would be required in each profession's workforce to achieve 45 per cent growth.
- Understand how much of its budget HEE currently spends on each of the seven cancer professions, including differing routes of entry to the professions such as domestic specialist training places or those rejoining the workforce.
- Estimate how much HEE would have to spend in order to increase the cancer workforce by 45 per cent in 2029 under three different scenarios.

We focused on three different scenarios that alter the inflows to each profession, through specialist training, international recruitment, intra-NHS transfers between professions[36] and rejoiners. The scenarios focus on inflows as these are of more direct relevance to HEE than retention programmes, which have been primarily led by NHS Improvement. The three scenarios modelled an increase in staff:

1) Solely through increasing training to provide more new staff.
2) Through increasing training of new staff and international recruitment only.
3) Through increasing training of new staff, international recruitment, intra-NHS staff movement and rejoiners.

Methods

This section provides a summary of the four main methods used in this research by the RAND Europe and University of Cambridge team. We provide fuller details of our methods in Section 1 of the supplementary information document,[37] available online).

In summary, we identified the last available data on the seven key cancer workforces as well as HEE's associated budgeting and spending data. Alongside this we undertook expert consultation with key stakeholders to discuss any data identified up to that point and the existence of any additional data, and to validate the overarching assumptions in the economic modelling. We then collated the information gathered in these two stages and used it to develop a demographic stock-flow economic model, where inflows (e.g. through training) appreciate and outflows (e.g. through retirement) depreciate a given workforce stock over time. The purpose of the demographic stock-flow economic model was to project the stock of the workforce up to 2029. Finally, a workshop was undertaken with key stakeholders to validate the data input into the model, the assumptions made and the preliminary output. This was all undertaken with oversight from a small stakeholder advisory group throughout (see below).

Data collection

We collected data on workforce numbers and on the cost to HEE of growing the cancer workforce. In accordance with good ethics practice, any interviewees were fully informed about the study and had the opportunity to consent or to decline to take part. The research team collected and stored data for research activities securely and complied with EU General Data Protection Regulation (GDPR) requirements throughout the study.

Workforce data

We identified the latest available data on workforce numbers for each of the seven key professions. We were interested in both the current number of Full-Time Equivalent (FTE) staff in each profession (stock), as well as the number of FTE staff who join or leave the profession per year (inflows and outflows). The data on the latest number of NHS staff in each profession provided us with baseline stock of staff, which enabled us to establish the baseline for an economic model to estimate how expected trends in inflows and outflows would impact on the overall number by 2029. We identified the necessary data through two methods:

- Desk research using targeted searches to identify additional publicly available data that may be relevant for the economic model.
- Approaching key stakeholders to request relevant data. HEE was able to give us selected workforce data taken from the NHS Electronic Staff Record (ESR).[38] In addition, some data were available from other stakeholders such as census information carried out by Macmillan Cancer Support, the Royal College of Radiologists or by the Royal College of Pathologists.

We compared the different data sources to triangulate the data (see Section 2 in the supplementary information pack) and provide us with assurance on the likely accuracy of our data. We then used the data to populate a demographic stock-flow economic model.

Budgeting and spending data

As with the workforce data, we identified the necessary financial data through two methods:

- Desk research using targeted searches to identify additional publicly available data that may be relevant for the economic model.
- Approaching key stakeholders to request relevant data.

We identified the latest publicly available data on HEE's budget and spending data for each of the seven key professions. We used extensive desk research to identify as much data as possible on HEE's budgets and spending from published and grey literature. These data included HEE's published financial reports, business plans and board minutes, as well as wider online material about particular spending streams and information from other sources such as the Department of Health and Social Care (DHSC) and organisations training specialist students.

We applied the cost information gained from these methods to the demographic stock-flow economic model on workforce numbers. This approach allowed us to estimate how much additional investment HEE would require in three different scenarios to achieve a 45 per cent growth in the cancer workforce by 2029. The funding would need to be front-loaded in order to train sufficient specialist staff by 2029, as the average specialist training for each of the professions varies between three and five years. The model only includes the cost to HEE of specialist training, but not core and foundation training because these stages were outside the scope of the modelling.

The full details of the cost calculation are available in Section 1 of our supplementary information pack. In summary, to estimate the training cost for each newly qualified specialist, we used the tariff prices for training set by DHSC and similar marginal costs of training each person. We adjusted our calculation to reflect the number of individual students who must be trained in order to reach the desired final FTEs of additional staff, factoring in drop-out rates and those working part-time.

We also investigated the cost to HEE of the other types of inflow to the professions: international recruitment, intra-NHS moves and rejoiners. For most of the seven professions HEE does not routinely incur costs associated with these types of inflow, with much of the cost falling to the NHS Trust or organisation employing the individual. The exception was that HEE does have some costs associated with some nurses being recruited internationally or rejoining the NHS. We estimated the costs of nurses recruited through this method where we could; however, these routes are not specific to specialist cancer nurses.

Expert consultation

We undertook targeted semi-structured interviews with specialist representatives from seven organisations relevant to the seven cancer workforce professions within the scope of our review. All the stakeholders that we approached agreed to engage with us, including HEE and various Royal Colleges and Societies. We conducted the interviews by telephone and used our conversations to:

- Discuss the accuracy, robustness and reliability of the data we identified from public sources.
- Identify any further sources of data that would help to inform our economic modelling.
- Validate our proposed assumptions, such as retirement age, for the economic model and discuss adjustments where necessary.

Economic modelling

We developed a demographic stock-flow economic model, building on the stock-flow approach taken by the majority of health workforce planning models, for each of the seven targeted cancer professions.[39]

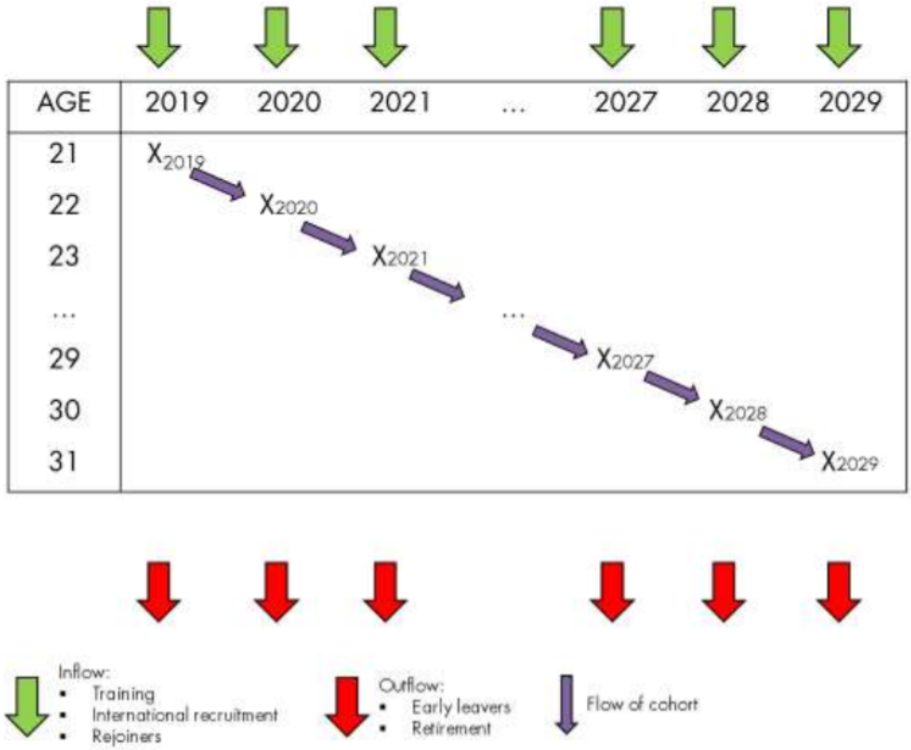
Rather than assuming that the number of FTE staff would remain constant from the baseline year to 2029 without additional investment, we have assumed that the 'status quo' baseline is a continuation each year of the average annual inflows and outflows observed over the most recent three years in each profession. We considered it more realistic to assume a similar growth than to model based on no 'status quo' growth given previous trends, but adopted an increase based on the flow numbers rather than percentage increases as this is more cautious. There are of course limitations in making any status quo growth assumptions, as particular initiatives to recruit more staff and particular circumstances (e.g. the UK's exit from the EU) may have a bearing on projections of future growth.

We considered it was better to use the most recent data for each of the two groups of professions (medical and non-medical) even though they do not cover exactly the same time period. For the medical workforces (clinical and medical oncology, gastroenterology, histopathology and clinical radiology) this was three years prior to 31 March 2018, and for the non-medical workforces (diagnostic radiography, therapeutic radiography and specialist cancer nursing) this was three years prior to 31 March 2019. Figure provides a more detailed visual representation of the demographic stock-flow model. Within the model each cohort of professionals (defined by age – in the example in Figure , cohort 'X' is people whose 21st birthday falls in 2019, 22nd birthday in 2020, and so on) is followed from the baseline year (the year with the latest available data) until 2029. A proportion of the cohort exits the workforce at an earlier point in their working life than retirement or due to retirement (outflows), while a proportion of the workforce joins the cohort through training, international recruitment and returning to practice (inflows).

We only include specialist training levels in our model. This is because the total time to train to be a specialist consultant is often about ten years, and thus a new trainee starting medical

training today may not qualify by 2029, the end date for this model. Instead we model based on the time it takes for a doctor to train from being newly qualified to a consultant, which is often approximately five or six years for a medical consultant, and sometimes less for some of the non-medically qualified professions covered in this study. The modelling also accounts for changes in the average number of FTE worked per individual (changes in FTE). Additionally, each cohort exits the model at the assumed latest age of retirement for each key cancer profession, given as 69 years.

Figure 2: Demographic stock-flow economic model



Source: RAND Europe.

The model uses a number of key assumptions. For example, we have assumed that all people over the age of 55 who leave the NHS ESR are retiring and so will not re-enter the workforce at a later date. Equally we model that all people entering the ESR from outside the NHS under the age of 30 are newly qualified staff. We set out all of the assumptions in more detail in Section 1 of the supplementary information pack.

Starting from the growth estimated in the 'status quo' approach described above, we have modelled three different ways of achieving 45 per cent growth by 2029. In all three scenarios we do this by increasing the inflows as this is where HEE is more closely involved. The three scenarios are:

- The necessary additional workforce growth is achieved entirely through increasing newly qualified specialist staff (i.e. training).
- The necessary additional workforce growth is achieved through increasing training and international recruitment, in the same proportion relative to each other as was observed over the past three years.
- The necessary additional workforce growth is achieved through training, increasing international recruitment, intra-NHS moves and people rejoining the NHS, so that each element makes up the same proportion of inflows that were observed over the past three years.

The three scenarios focus on inflows to the cancer workforce (the green arrow in Figure) as the research was focused on HEE, whose work directly affects these. Outflows from the cancer workforce may be affected by staff retention programmes, which have been primarily led by NHS Improvement. Outflows are assumed to stay at the same rate as seen over the last three years, with retention programmes and other initiatives staying at the same relative level.

After modelling how many FTE of each key cancer profession a net increase of 45 per cent by 2029 would represent, and the means through which they flow into the workforce, we combined this information with the available data on the cost to HEE of training each of the seven types of cancer professionals. This allowed us to estimate how much HEE would have to spend on specialist training places to increase the stock of each cancer workforce in the three different growth scenarios by 2029.

Validation workshop

We invited a range of participants to a half-day workshop facilitated by the RAND Europe team. The purpose of the workshop was to sense check the emerging workforce and finance data, seek any additional data and to test whether the modelling assumptions were reasonable. A total of 13 individuals attended from governmental organisations, relevant Royal Colleges and Societies, and cancer charities, as well as research team members from RAND Europe and the University of Cambridge. Invitees who were not able to attend were asked instead to provide written comments on the information contained in the validation workshop pre-read material and on the notes of the discussion, and several did so. We used the validation workshop outputs and additional comments to refine and complete the workforce modelling and cost scenarios.

Project oversight

In addition to the work already described, we created a stakeholder group comprising Cancer Research UK, Macmillan Cancer Support, HEE, DHSC and NHS England and NHS Improvement. This group provided oversight of the project, as well as a readily accessible group with whom the research team could discuss queries as they arose to reach appropriate solutions. The stakeholder group contributed to an inception meeting and also validated our findings through participation in the wider validation workshop and through commenting on the factual accuracy of an early draft of this report.

Results

In this section we set out:

- Baseline workforce numbers as at 2019 in the seven cancer professions included in this research.
- An overview of HEE's spending and costs.
- Demographic stock-flow modelling of each of the seven professions, covering:
    - Clinical and medical oncology (combined)
    - Gastroenterology
    - Histopathology
    - Clinical radiology
    - Diagnostic radiography
    - Therapeutic radiography
    - Specialist cancer nursing.
- An overall compilation of the modelling results and costs for HEE.

Baseline workforce numbers in the seven cancer professions

Table 2 shows the baseline data identified for each of the seven professions and the year of that baseline. To promote consistency across the economic modelling of the seven professions we used NHS Electronic Staff Record (ESR) data in our model where possible, and we validated the reasonableness of the data through desk research and expert consultation. ESR data capture individual-level information on 99 per cent of NHS staff working in organisations that directly employ NHS staff in the Hospital and Community Health Services (HCHS) sector in England (as well as staff from ALBs, Clinical Commissioning Groups and some from the independent sector).[40] This means that the modelling used the number of staff, rather than the number of funded places for each profession. The ESR contains information on profession, employing organisation, contracted FTE and some demographic information. We received two separate workforce datasets from the ESR: one for the non-medical workforces (diagnostic radiographers, therapeutic radiographers and adult nurses); and the other for the four medically qualified professions (clinical radiologists, gastroenterologists, histopathologists and clinical and medical oncologists). For a detailed discussion of the ESR data, including its strengths and limitations, please refer to the supplementary information pack.

Table 2: Baseline FTE numbers of NHS staff in each of the seven priority professions

Cancer-related profession	Year of baseline data	FTE of NHS staff in England
Clinical and medical oncology	2018	1,137
Gastroenterology	2018	1,222
Histopathology	2018	1,220
Clinical radiology	2018	2,965
Diagnostic radiography	2019	14,997
Therapeutic radiography	2019	2,844
Specialist cancer nursing (adult)	2017	3,851

Sources: RAND Europe analysis of NHS Electronic Staff Record (all professions except specialist cancer nursing) and Macmillan 2017 Cancer Workforce in England census (for specialist cancer nursing). Note: For all professions except specialist cancer nurses, these baseline data were recorded as at 31 March in the year listed. The baseline for specialist cancer nurses is taken from the Macmillan cancer workforce census which described the workforce on 9 October 2017.

Overview of HEE's spending

HEE's publicly available information includes annual data on spending on the future workforce, as well as projections. These data are broken down by region and by category: non-medical, undergraduate, postgraduate and other. However, they are not broken down by profession, so we were not able obtain data specific to the seven cancer professions in this way. Nevertheless HEE's Annual Reports and Accounts provide an overview of HEE's spending and trends (see Table 3).[41]

Table 3: Breakdown of HEE's spending, 2016–17 to 2018–19

Type of expenditure	2016–17 (£ m)	2017–18 (£ m)	2018–19 (£ m)
HEE staff costs	**151**	**158**	**143**
Education and training	**4,527**	**4,404**	**4,023**
Of which:			
Undergraduate medical and dental training	892	883	894
Postgraduate medical and dental training	1,846	1868	1,896
Non-medical staff training(1)	1,788	1,654	1,233
Other operating expenditure	**390**	**352**	**404**
TOTAL	**5,069**	**4,914**	**4,570**

Source: Health Education England Annual Report and Accounts 2017–18 and 2018–19.

Note (1): HEE support for students commencing nursing programmes ceased in September 2017. The reduction in HEE expenditure in this area between 2017–18 and 2018–19 can be seen under the 'non-medical' heading.

This table shows that HEE's budget decreased slightly overall between 2016–17 and 2018–19. The decrease can be explained by the loss of HEE support for students commencing nursing programmes via a nursing bursary in September 2017. In other lines of future medical workforce spending, there have been modest increases of approximately 1 per cent to the postgraduate medical training budgets each year.

Newly qualified training tariffs

The DHSC publishes tariffs each year for the amount to be paid by HEE to training providers for education and training placements for specialist training. A Market Forces Factor (MFF) is then applied to these tariff payments. The MFF is a multiplier for the basic tariff payment which varies based on geographical location.[42] This approach allows funding to take into account an estimate of unavoidable cost differences between healthcare providers. The latest guidance for the 2019–20 tariff shows:

- A non-medical placement costing £3,270 + MFF
- A medical undergraduate placement costing £33,286 + MFF
- A medical postgraduate costing £11,418 + MFF
- A study leave budget of £734 per student and a contribution to basic salary costs.[43]

We then adjusted these costs to allow for drop-out rates and people who work part-time (i.e. the need to train more than one person to achieve one FTE). The above information is sufficient to model the cost of increasing the workforce for each of the seven professions by training of new staff (see next section for detailed results).

HEE costs of other inflows to cancer professions

We found that attracting rejoiners, recruiting from overseas or improving retention rates seldom incurs a cost to HEE. This is because these costs fall to others within the health sector, often to the NHS Trust who is or will be employing the professional in question. For example, in international recruitment, the National Audit Office reported in 2020 that the estimated average cost to recruit a nurse from overseas is £12,000,[44] but it is NHS providers who are responsible for the recruitment, with HEE providing support for the overseas recruitment strategy and planning through its regional teams.

However, for nursing, we found some costs are met by HEE. For rejoiners, HEE pays about £2,600 per rejoiner, although many of the nurses recruited through this scheme will not be specialist cancer nurses. This comprises:

- a £500 stipendiary (one-off payment)
- a £1000 recruitment fee per rejoiner
- a £1,100 contribution to placement costs.

HEE also contributes to international recruitment of nurses who join under the Global Learners Programme, by paying the fixed central administrative and educational costs. However, we were unable to obtain a figure for these costs, and the unit costs of recruitment are borne by the recruiting trust. This year HEE also planned to launch a partnership with other organisations to bring overseas clinical radiologists to the UK, but we do not know the cost to HEE of this programme.

Demographic stock-flow modelling by workforce to 2029, including cost estimates

The results of the economic modelling are presented in the sections below, for each of the seven professions in turn. These data show what the growth of the workforce might be, if projected forward from trends in the three previous years using the demographic stock-flow economic model – i.e. no change other than the continuation of existing trends. The existing trend was identified using the average of three years' of data for each inflow and outflow. The growth of the workforce is compared to the reference year 2019, highlighted by the vertical line in the figures and green cells in the tables. All costs are expressed in 2019/20 prices and assume that the real cost of training per trainee does not change over time.

Economic modelling by profession

Clinical and medical oncology

Context

Clinical oncologists use radiotherapy and systemic therapies, such as chemotherapy, to treat and manage patients with cancer.[45] Medical oncologists diagnose, assess, treat and manage patients with cancer, and conduct research that has practical applications.[46] Clinical and medical oncology are complementary disciplines, with some overlap in their respective roles.[47] Issues with occupational coding mean it is not possible to distinguish between clinical and medical oncologists in the ESR data, meaning they are modelled simultaneously.[48]

Once medical students have completed their two-year foundation training and a further two years of core medical training, they can specialise in clinical oncology, which usually takes a further five years,[49] or more for part-time students. Specialising in medical oncology takes a minimum of four years[50]; however, most trainee medical oncologists will also undertake some form of applicable research alongside their training and therefore take longer than the minimum four years.[51] Our modelling begins at specialty trainee level 3 (ST3), where both specialty training routes begin.[52] The first round fill rate for clinical oncologists was 100 per cent in 2019.[53] The latest data we have on fill rates for medical oncologists is from 2016, when 82 per cent of places were filled in round 1.[54]

Historic workforce stock and flows

On 31 March 2018 there were 1,137 FTE consultant clinical and medical oncologists working in the NHS in England. Over the three years prior, the following average **annual** FTE flows are observed:

- Inflows:
    - Newly qualified from specialist training: 61 (53 per cent of inflows)
    - Intra-NHS[55]: 19 (16 per cent of inflows)
    - Rejoiners and international recruitment: 36 (31 per cent of inflows)
- Outflows:
    - Early leaver: -42 (70 per cent of outflows)
    - Retirement: -18 (30 per cent of outflows)

Overall, there is an average annual total of 116 FTE inflows to the workforce and -60 FTE outflows from the workforce, with a net average annual inflow of 56 FTE.

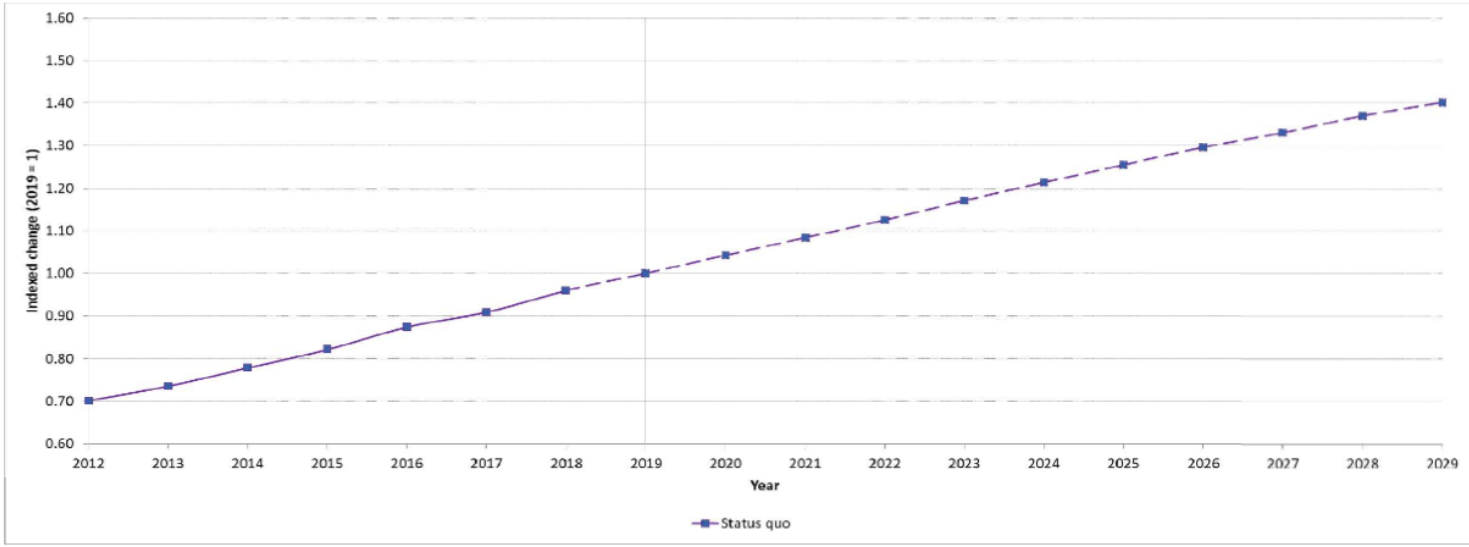
The validation workshop highlighted the following key points:

- Many countries internationally do not have an equivalent clinical oncologist role, meaning that international recruitment is not a major inflow for the profession.[56] This suggests that most of the international recruitment for this profession is related to medical oncology.
- Approximately 5 per cent of the workforce are locums.
- For clinical and medical oncology, the number of rejoiners is relatively low.

Projected status quo change

Figure 3 shows the projected status quo growth of clinical and medical oncologists from 2018 to 2029 (dashed line), assuming a continuation of inflow and outflow trends over the past three years (i.e. in the results achieved from HEE's current activity level) and applying those 'flow' trends to the known 'stock', including age and FTE characteristics, of the current population of oncologists at that time. This approach is not the same as a simple linear extrapolation of increases since the model builds in stock characteristics such as, for example, if a high proportion of the population are nearing retirement. Using 2019 as the reference year, the demographic stock-flow economic model projects a 40 per cent increase in FTE, a growth of 477 FTE from 1,185 in 2019 to 1,662 in 2029, as outlined in Table 4. This is in line with the historic growth in the workforce, as shown by the solid line in Figure 3.

Figure 3: Historic and projected status quo changes in the clinical and medical oncology workforce, indexed to 2019



Source: RAND Europe. Note: For the medically qualified workforces (clinical radiologists, gastroenterologists, histopathologists and clinical and medical oncologists), values up to and including 2018 are historic (solid line) and values from 2019 onwards are outputs of the modelling (dashed line). This reflects the latest available data at the time of modelling.

Table 4: Historic and status quo changes in the clinical and medical oncology workforce

	2012	2013	2014	2015	2016	2017	2018	2019	2020	2021	2022	2023	2024	2025	2026	2027	2028	2029
Total FTE	832	872	923	974	1037	1078	1137	1185	1236	1284	1334	1388	1439	1488	1537	1577	1625	1662
Indexed change	0.70	0.74	0.78	0.82	0.87	0.91	0.96	1.00	1.04	1.08	1.13	1.17	1.21	1.26	1.30	1.33	1.37	1.40

Source: RAND Europe. Note: Indexed change from reference year.

Scenario growth and costing

We calculate that to achieve 45 per cent growth in the clinical and medical oncology workforce by 2029, an additional 57 FTE are required, as the status quo increase is projected to be already at 40 per cent.

Table 5 outlines the required FTE increase to achieve the 45 per cent growth target in each of the three scenarios. The required inflow and cost to HEE of each scenario is as follows:

- Scenario 1: an additional 57 Newly Qualified Trainee (NQT) FTEs are required to achieve the necessary growth. An NQT is a doctor who has just completed specialist training. As clinical and medical oncologists work at an average of 0.92 FTE per individual,[57] and assuming an average training drop-out rate of 10 per cent, this means an additional 68 individuals need to be trained by 2029, at a total additional cost of £12.6m to HEE over the ten years.
- Scenario 2: an additional 36 NQT and 21 rejoiner and international FTE are required. Applying the average FTE per individual of 0.92, and assuming an average training drop-out rate of 10 per cent, this means an additional 43 individuals need to be trained, at an NQT cost to HEE of £8.0m over the ten years.
- Scenario 3: an additional 30 NQT, 9 intra-NHS and 18 rejoiner and international FTE are required. Applying the average FTE per individual of 0.92, and assuming an average training drop-out rate of 10 per cent, this means an additional 36 individuals need to be trained, at an NQT cost to HEE of £6.7m over the ten years.

As stated in the methods section above, since HEE pays costs associated with specialist training we can estimate the cost to HEE of training each additional person by using the standard tariff rates set by DHSC and then adjusted for other factors such as the market forces factor, the average FTE worked per individual, and the need to train more people than positions required to allow for drop-out rates.

Scenarios 2 and 3 include other entry routes which create costs elsewhere in the health system. Such costs tend mainly to fall to the Trust or NHS body employing the individual as Continuing Professional Development (CPD) and international recruitment costs are their responsibility. Our understanding is that no costs arise to HEE from these entry routes, so this does not affect the total estimated cost to HEE. However, when interpreting the overall costs of different scenarios, it must be remembered that this represents the cost to HEE only, not the total cost to the health sector.

Table 5: Scenario growth of the clinical and medical oncology workforce

	Total required FTE increase (numbers)			Total estimated cost to HEE
Scenario	**Newly qualified (specialist) training (NQT)**	**Intra-NHS**	**Rejoiners and international recruitment**	
Scenario 1	57	-	-	**£12,638,325**
Scenario 2	36	-	21	**£7,991,882**
Scenario 3	30	9	18	**£6,690,878**

Source: RAND Europe.

Gastroenterology

Context

Gastroenterologists investigate, diagnose, treat and work to prevent all gastrointestinal (intestines and stomach) and hepatological (biliary tree, gallbladder, liver and pancreas) diseases.[58] Once medical students have completed their two-year foundation training and a further two years of core medical training they can specialise in gastroenterology, which begins at specialty trainee level 3 (ST3) and takes at least five years to complete.[59] Our modelling begins at this level. Specialist gastroenterology training places have a fill rate of 100 per cent nationally.[60]

Historic workforce stock and flows

On 31 March 2018 there were 1,222 FTE consultant gastroenterologists working in the NHS in England. Over the three years prior, the following average **annual** FTE flows are observed:

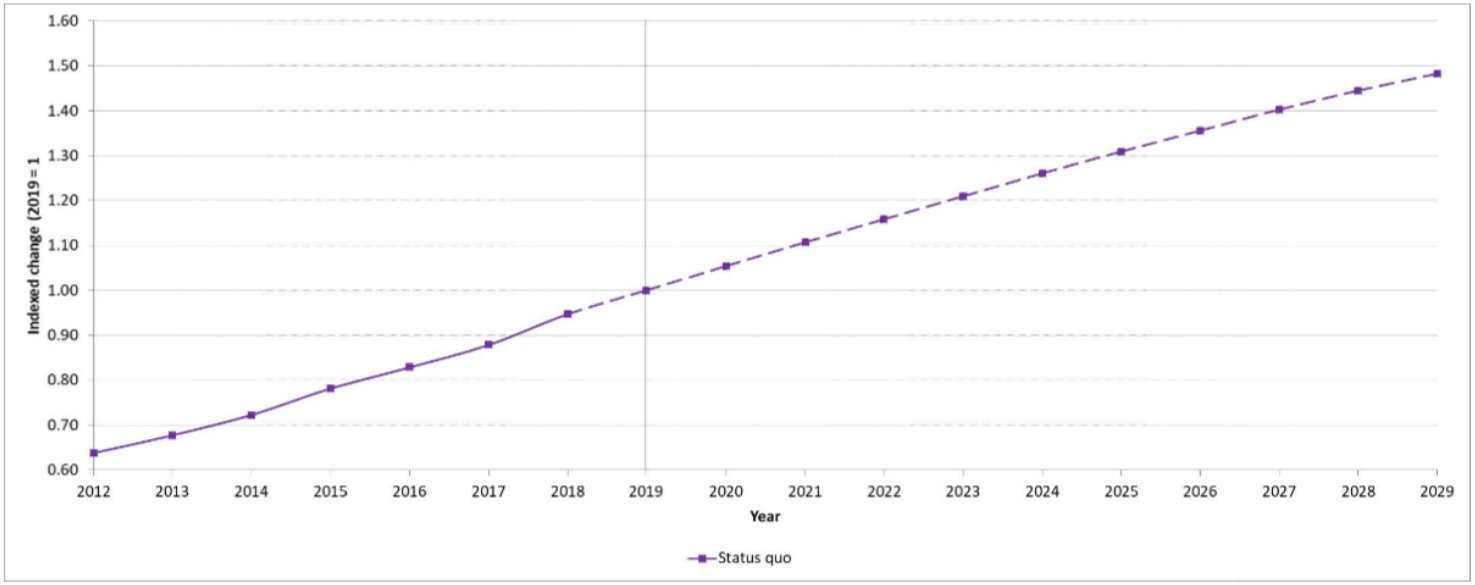
- Inflows:
  - Newly qualified from specialist training: 72 (66 per cent of inflows)
  - Intra-NHS: 5 (4 per cent of inflows)
  - Rejoiners and international recruitment: 32 (29 per cent of inflows)
- Outflows:
  - Early leaver: -24 (61 per cent of outflows)
  - Retirement: -15 (39 per cent of outflows)

Overall, there is an average annual total of 109 FTE inflows to the workforce and -39 FTE outflows from the workforce, with a net average annual inflow of 70 FTE.

Projected status quo increase

Figure 2 shows the projected status quo growth of gastroenterologists from 2018 to 2029 (dashed line), assuming a continuation of inflow and outflow trends over the past three years (i.e. without any change by HEE in its current activity level). Using 2019 as the reference year, the demographic stock-flow economic model projects a 48 per cent increase in FTE, a growth of 623 FTE from 1,290 in 2019 to 1,913 in 2029, as outlined in Table 6. This is in line with the historic growth in the workforce, as shown by the solid line in Figure 2 .

Figure 2 : Historic and status quo changes in the gastroenterology workforce, indexed to 2019



Source: RAND Europe. Note: For the medically qualified workforces (clinical radiologists, gastroenterologists, histopathologists and clinical and medical oncologists), values up to and including 2018 are historic (solid line) and values from 2019 onwards are outputs of the modelling (dashed line). This reflects the latest available data at the time of modelling.

Table 6: Historic and status quo changes in the gastroenterology workforce

	2012	2013	2014	2015	2016	2017	2018	2019	2020	2021	2022	2023	2024	2025	2026	2027	2028	2029
Total FTE	823	874	932	1008	1069	1133	1222	1290	1359	1428	1494	1560	1626	1689	1748	1809	1863	1913
Indexed change	0.64	0.68	0.72	0.78	0.83	0.88	0.95	1.00	1.05	1.11	1.16	1.21	1.26	1.31	1.36	1.40	1.44	1.48

Source: RAND Europe. Note: Indexed change from reference year.

Scenario growth and costing

The status quo increase of the gastroenterology workforce is 48 per cent from 2019 to 2029, which is greater than the 45 per cent growth scenario target, meaning that growth scenarios were not undertaken for this workforce, although of course to achieve status quo growth, HEE will still need to invest money in continuing its training of this workforce.

Histopathology

Context

Histopathologists are responsible for diagnosing and studying disease in tissues and organs through the examination of samples of tissue.[61] Histopathologists determine the cause of death by undertaking autopsies and are integral to the management of patients with cancer, through the staging and grading of tumours.[62] There are important areas of growth in histopathology – one is the increasing use of digital pathology, which includes the acquisition, management, sharing and interpretation of pathology information in a digital format. Artificial intelligence (AI) also plays an increasing role, carrying out repetitive tasks and sifting through data to find common features and to make conclusions based on statistical probability although it is anticipated to augment the role of a histopathologist rather than replace it. Both of these changes may affect the size and nature of the histopathologist workforce in the longer term

Training in histopathology is separated into four stages and takes at least five years to complete when in training full time.[63] In the validation workshop we heard that there may often be seven years between starting and completing training. Training for histopathology is a run-through course[64] which begins as speciality trainee level 1 (ST1).[65] In 2019 there was a training place fill rate of 100 per cent in the first round of recruitment for that year; however, in 2018 this was only 47 per cent in the first round.[66]

Historic workforce stock and flows

On 31 March 2018 there were 1,220 FTE histopathologists working in the NHS in England. Over the three years prior, the following average **annual** FTE flows are observed:

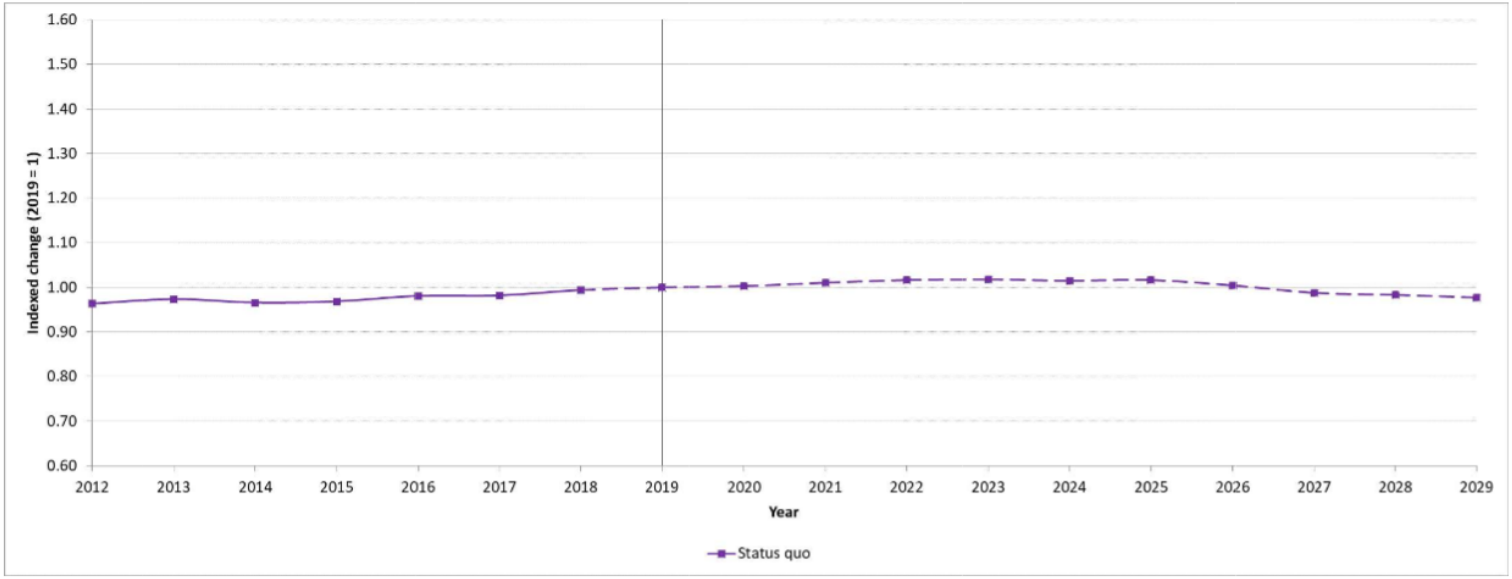
- Inflows:
    - Newly qualified training: 46 (54 per cent of inflows)
    - Intra-NHS: 9 (11 per cent of inflows)
    - Rejoiners and international recruitment: 31 (36 per cent of inflows)
- Outflows:
    - Early leaver: -34 (49 per cent of outflows)
    - Retirement: -35 (51 per cent of outflows)

Overall, there is an average annual total of 86 FTE inflows to the workforce and -69 FTE outflows from the workforce, with a net average annual inflow of 17 FTE.

Projected status quo change in workforce

Figure 3 shows the projected status quo growth of histopathologists from 2018 to 2029 (dashed line), assuming a continuation of inflow and outflow trends over the past three years (i.e. in the results achieved from HEE's current activity level) and applying those 'flow' trends to the known 'stock', including age and FTE characteristics of histopathologists. Using 2019 as the reference year, the demographic stock-flow economic model projects a 2 per cent decrease in FTE, a decline of 27 FTE from 1,228 in 2019 to 1,201 in 2029, as outlined in Table 7. This is initially in line with the historic growth in the workforce, as shown by the solid line in Figure 3, until 2026 when the stock of the workforce begins to consistently decline as a result of an ageing workforce and outflows becoming greater than inflows.

Figure 3: Historic and status quo changes in the histopathology workforce, indexed to 2019



Source: RAND Europe. NOTE: For the medically qualified workforces (clinical radiologists, gastroenterologists, histopathologists and clinical and medical oncologists), values up to and including 2018 are historic (solid line) and values from 2019 onwards are outputs of the modelling (dashed line). This reflects the latest available data at the time of modelling.

Table 7: Historic and status quo changes in the histopathology workforce

	2012	2013	2014	2015	2016	2017	2018	2019	2020	2021	2022	2023	2024	2025	2026	2027	2028	2029
Total FTE	1182	1195	1185	1189	1204	1205	1220	1228	1232	1241	1247	1249	1245	1248	1233	1213	1207	1201
Indexed change	0.96	0.97	0.97	0.97	0.98	0.98	0.99	1.00	1.00	1.01	1.02	1.02	1.01	1.02	1.00	0.99	0.98	0.98

Source: RAND Europe. Note: Indexed change from reference year.

Scenario growth and costing

To achieve 45 per cent growth in the histopathology workforce by 2029 an additional 580 FTE are required, as the status quo change projects a 2 per cent decrease in the size of the workforce.

Table 8 outlines the required FTE increase to achieve the 45 per cent growth target in each of the three scenarios. The required inflows and cost of each scenario to HEE is as follows:

- Scenario 1: an additional 580 NQT FTEs are required to achieve the necessary growth. Histopathology-specific data on average FTE could not be identified, however NHS consultants work at an overall average of 0.94 FTE per individual in 2018 (the year the baseline data corresponds to), which was used as a proxy for histopathologists.[67] Assuming a 10 per cent training drop-out rate, this means an additional 677 individuals need to be trained by 2029, at a total additional cost of £117.5m to HEE over the ten years.
- Scenario 2: an additional 349 NQT and 231 rejoiner and international FTE are required. Applying the average FTE per individual of 0.94, and assuming an average training drop-out rate of 10 per cent, this means an additional 407 individuals need to be trained, at an NQT cost to HEE of £70.6m over the ten years.
- Scenario 3: an additional 310 NQT, 63 intra-NHS and 206 rejoiner and international FTE are required. Applying the average FTE per individual of 0.94, and assuming an average training drop-out rate of 10 per cent, this means an additional 363 individuals need to be trained, at an NQT cost to HEE of £63m over the ten years.

As stated in the methods section above, since HEE pays costs associated with specialist training we can estimate the cost to HEE of training each additional person by using the standard tariff rates set by DHSC and then adjusted for other factors such as the market forces factor, the average FTE worked per individual, and the need to train more people than positions required to allow for drop-out rates.

Scenarios 2 and 3 include other entry routes which create costs elsewhere in the health system. Such costs tend mainly to fall to the Trust or NHS body employing the individual as CPD and international recruitment costs are their responsibility. Our understanding is that no costs arise to HEE from these entry routes so this does not affect the total estimated cost to HEE. However, when interpreting the overall costs of different scenarios, it must be remembered that this represents the cost to HEE only, not to the total cost to the health sector.

Table 8: Scenario growth of the histopathology workforce

	Total FTE increase			Total cost to HEE
Scenario	**Newly qualified (specialist) training (NQT)**	**Intra-NHS**	**Rejoiners and international recruitment**	
Scenario 1	580	-	-	**£117,453,896**
Scenario 2	349	-	231	**£70,611,131**
Scenario 3	310	63	207	**£62,977,495**

Source: RAND Europe.

Clinical radiology

Context

Clinical radiologists use images to diagnose, treat and manage medical conditions and diseases. Clinical radiologists work closely with radiographers and collaborate with a wide range of other health professionals. They are responsible for reporting most imaging procedures used in diagnosing medical conditions and will perform many interventional procedures, such as minimally invasive surgery.[68]

Training in clinical radiology takes around five years to complete when in full-time training and, like histopathology, is a run through course which begins at ST1.[69] Our modelling begins at this stage. When trainees studying part-time are included, training can take about six years to complete. In 2019 there was a training place fill rate of over 99 per cent in the first round of recruitment for that year, and in 2018 this was 100 per cent.[70] The final fill rate has been 100 per cent for several years.

There is currently a Global Radiologists Programme for clinical radiologists, offered by a partnership of four organisations, including HEE and the Royal College of Radiologists. The purpose is to attract and recruit experienced clinical radiologists from other countries to come and work for three years in a suitable NHS trust in England on an earn, learn, return basis. We do not have further information on the numbers recruited through this scheme or the costs to HEE of taking part.[71]

Historic workforce stock and flows

On 31 March 2018 there were 2,965 FTE consultant clinical radiologists working in the NHS in England. Over the three years prior, the following average **annual** FTE flows are observed:

- Inflows:
  - Newly qualified from specialist training: 133 (54 per cent of inflows)
  - Intra-NHS: 2 (1 per cent of inflows)
  - Rejoiners and international recruitment: 112 (45 per cent of inflows)
- Outflows:
  - Early leaver: -57 (53 per cent of outflows)
  - Retirement: -51 (47 per cent of outflows)

Overall, there is an average annual total of 247 FTE inflows to the workforce and -108 FTE outflows from the workforce, with a net average annual inflow of 139 FTE. Just under half (45 per cent) of inflows come from those rejoining the workforce or entering through international recruitment, of which a considerable number come from South Africa, India and Pakistan.[72]

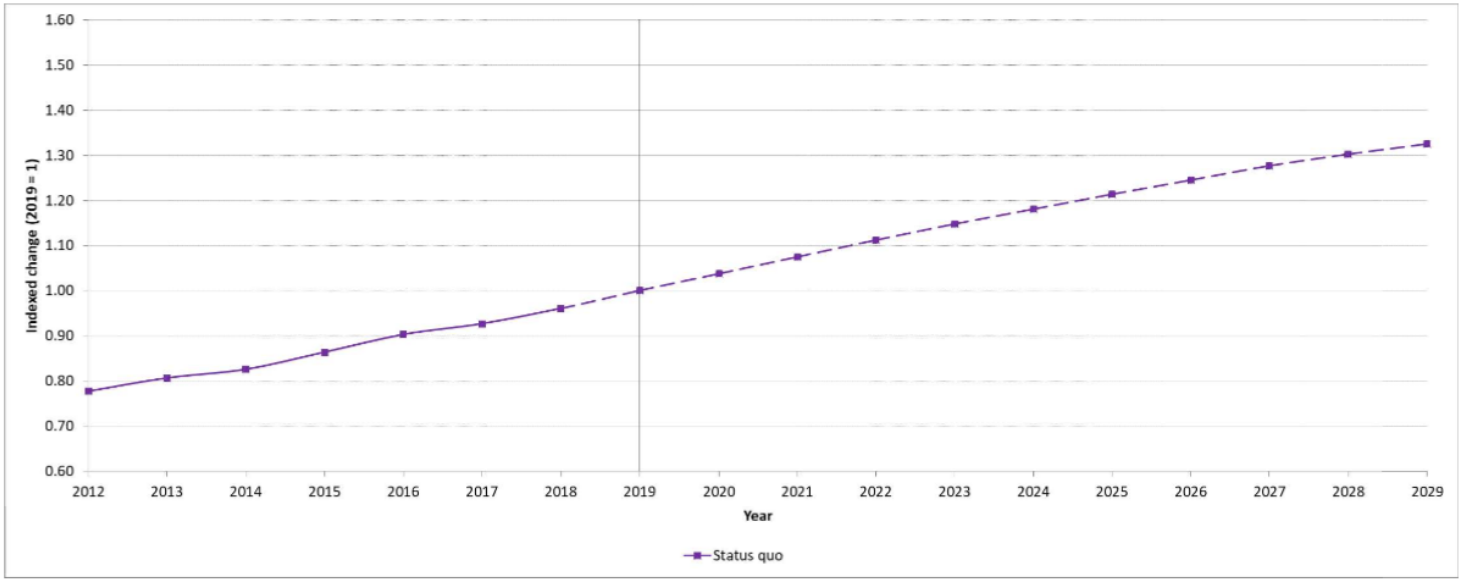
Insights from the validation workshop

- For clinical radiology, approximately 6 per cent of the workforce are locums.
- For radiologists, recruiting abroad is common compared with some of the other professions in this study.
- The number of rejoiners is relatively low for clinical radiologists.

Projected status quo change

Figure 4 shows the projected status quo growth of clinical radiologists from 2018 to 2029 (dashed line), assuming a continuation of inflow and outflow trends over the past three years (i.e. in the results achieved from HEE's current activity level) and applying those 'flow' trends to the known 'stock' of clinical radiologists, including age and FTE characteristics. Using 2019 as the reference year, the demographic stock-flow economic model projects a 33 per cent increase in FTE, a growth of 1,004 FTE from 3,087 in 2019 to 4,091 in 2029, as outlined in Table 9. This is in line with the historic growth in the workforce, as shown by the solid line in Figure 4.

Figure 4: Historic and status quo changes in the clinical radiology workforce, indexed to 2019



Source: RAND Europe. NOTE: For the medically qualified workforces (clinical radiologists, gastroenterologists, histopathologists and clinical and medical oncologists), values up to and including 2018 are historic (solid line) and values from 2019 onwards are outputs of the modelling (dashed line). This reflects the latest available data at the time of modelling.

Table 9: Historic and status quo changes in the clinical radiology workforce

	2012	2013	2014	2015	2016	2017	2018	2019	2020	2021	2022	2023	2024	2025	2026	2027	2028	2029
Total FTE	2398	2489	2548	2666	2787	2860	2965	3087	3205	3319	3433	3543	3644	3747	3842	3940	4019	4091
Indexed change	0.78	0.81	0.83	0.86	0.90	0.93	0.96	1.00	1.04	1.08	1.11	1.15	1.18	1.21	1.24	1.28	1.30	1.33

Source: RAND Europe. Note: Indexed change from reference year.

Scenario growth and costing

We calculate that to achieve 45 per cent growth in the clinical radiology workforce by 2029 an additional 384 FTE are required, as the status quo increase is projected to be a 33 per cent increase in the size of the workforce. Table 10 outlines the required FTE increase to achieve the 45 per cent growth target in each of the three scenarios. The required inflow and NQT cost of each scenario is as follows:

- Scenario 1: an additional 384 newly qualified trainee (NQT) FTEs are required to achieve the necessary growth. As clinical radiologists work at an average of 0.92 FTE per individual,[73] and assuming an average training drop-out rate of 10 per cent, this means an additional 459 individuals need to be trained by 2029, at a total additional cost of £79.6m to HEE over the ten years.
- Scenario 2: an additional 208 NQT and 176 rejoiner and international FTE are required. Applying the average FTE per individual of 0.92, and assuming an average training drop-out rate of 10 per cent, this means an additional 249 individuals need to be trained, at an NQT cost to HEE of £43.2m over the ten years.
- Scenario 3: an additional 207 NQT, 3 intra-NHS and 174 rejoiner and international FTE are required. Applying the average FTE per individual of 0.92, and assuming an average training drop-out rate of 10 per cent, this means an additional 247 individuals need to be trained, at an NQT cost to HEE of £42.9m over the ten years.

Table 10: Scenario growth of the clinical radiology workforce

	Total FTE increase			Total known cost to HEE
Scenario	**Newly qualified (specialist) training**	**Intra-NHS**	**Rejoiners and international recruitment**	
Scenario 1	384	-	-	**£79,632,700**
Scenario 2	208	-	176	**£43,199,439**
Scenario 3	207	3	174	**£42,852,455**

Source: RAND Europe. Note: Total known cost to HEE does not include any financial contribution made by HEE to the Global Radiologists Programme as we do not have any information on this.

As stated in the methods section of the report, since HEE pays costs associated with specialist training, we can estimate the cost to HEE of training each additional person by using the standard tariff rates set by DHSC and then adjusted for other factors such as the market forces factor, the average FTE worked per individual, and the need to train more people than positions required to allow for drop-out rates.

Scenarios 2 and 3 include other entry routes which create costs elsewhere in the health system. Such costs tend mainly to fall to the Trust or NHS body employing the individual as CPD and international recruitment costs are their responsibility. We believe in this case there may also be some additional cost to HEE of participating in the Global Radiologists Programme but we do not have this information. When interpreting the overall costs of different scenarios, it must be remembered that the cost stated above represents the cost to HEE of specialist training only, not to the total cost to the health sector.

Diagnostic Radiography

Context

Diagnostic radiographers use the latest technology in clinical imaging to screen, diagnose and undertake ongoing surveillance of patients, such as cancer patients after treatment.[74] They use a range of imaging technology and techniques such as x-ray, fluoroscopy and ultrasound.[75]

Traditionally, training in diagnostic radiography is provided in a three-year full-time undergraduate degree, which can also be completed part-time in around six years.[76] Those with a relevant undergraduate degree can undertake a postgraduate programme in diagnostic radiography, although it can be hard for individuals to acquire funding for such courses and, as a result, not many newly qualified trainees take this route.[77]

Historic workforce stock and flows

On 31 March 2019 there were 14,997 FTE diagnostic radiographers working in the NHS in England. Over the three years prior, the following average **annual** FTE flows are observed:

- Inflows:
    - Newly qualified training: 880 (50 per cent of inflows)
    - Intra-NHS: 318 (18 per cent of inflows)
    - Rejoiners: 375 (21 per cent of inflows)
    - International recruitment: 181 (10 per cent of inflows)
- Outflows:
    - Early leaver: -919 (82 per cent of outflows)
    - Retirement: -208 (18 per cent of outflows)

Overall, there is an average annual total of 1,754 FTE inflows to the workforce and -1,127 FTE outflows from the workforce, with a net average annual inflow of 627 FTE.

Other relevant information

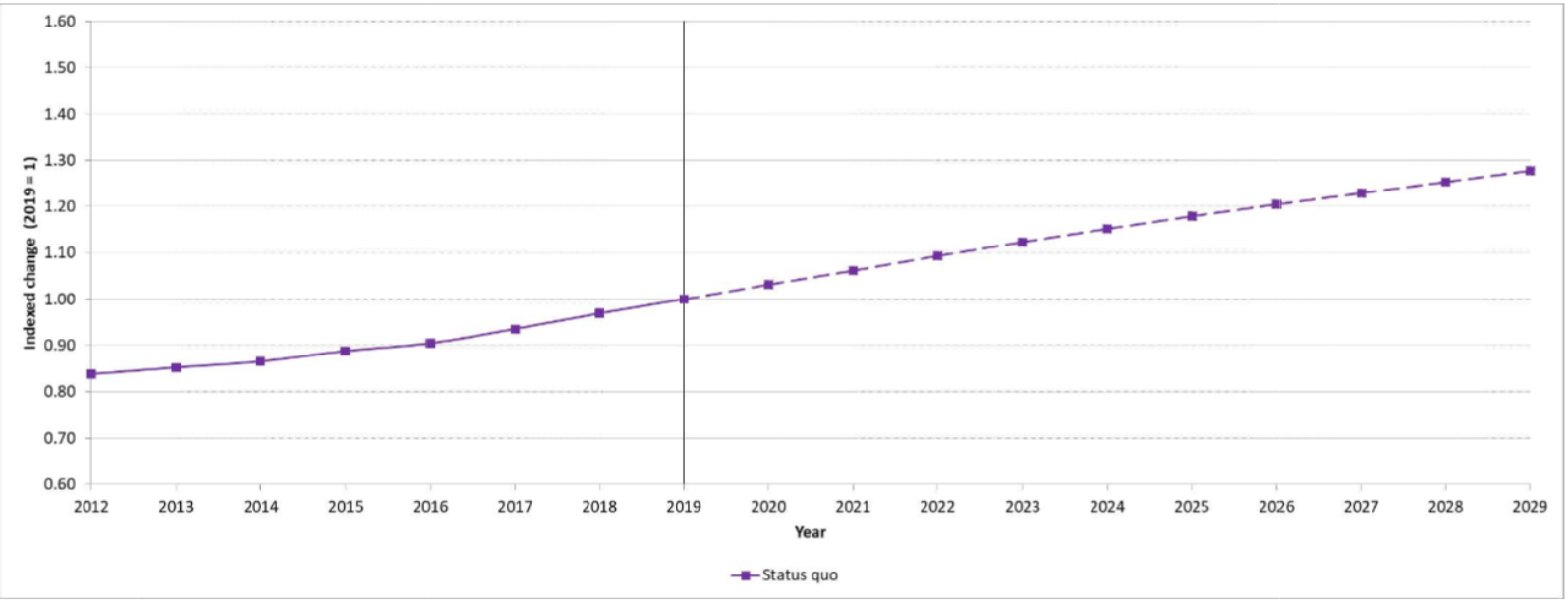
The Interim NHS People Plan, published in June 2019,[78] stated that over the following five years, more clinical degree-level apprenticeships would be introduced, including for therapeutic and diagnostic radiographers. Furthermore, in July 2020, the NHS published the NHS People Plan 2020/2021,[79] which made a commitment to training 450 reporting radiographers.

We were not able to build these commitments into our model for diagnostic radiographers because the data were not available at the time of conducting the modelling.

Projected status quo change

Figure 5 shows the projected status quo increase in diagnostic radiographers from 2019 to 2029 (dashed line), assuming a continuation of inflow and outflow trends over the past three years (i.e. in the results achieved from HEE's current activity level) and applying those 'flow' trends to the known 'stock' of diagnostic radiographers. Using 2019 as the reference year, the demographic stock-flow economic model projects a 28 per cent increase in FTE, a growth of 4,158 FTE from 14,997 in 2019 to 19,155 in 2029, as outlined in Table 11. This is in line with the historic growth in the workforce, as shown by the solid line in Figure 5.

Figure 5: Historic and status quo changes in the diagnostic radiography workforce, indexed to 2019



Source: RAND Europe. Note: For diagnostic radiographers and therapeutic radiographers, values up to and including 2019 are historic (solid line) on the graph, and values from 2020 onwards are estimates from the modelling (dashed line).

Table 11: Historic and status quo changes in the diagnostic radiography workforce

	2012	2013	2014	2015	2016	2017	2018	2019	2020	2021	2022	2023	2024	2025	2026	2027	2028	2029
Total FTE	12570	12783	12979	13317	13570	14032	14535	14997	15463	15930	16393	16843	17276	17682	18064	18436	18801	19155
Indexed change	0.84	0.85	0.87	0.89	0.90	0.94	0.97	1.00	1.03	1.06	1.09	1.12	1.15	1.18	1.20	1.23	1.25	1.28

Source: RAND Europe. Note: Indexed change from reference year.

Scenario growth and costing

We estimate that to achieve 45 per cent growth in the diagnostic radiography workforce by 2029 an additional 2591 FTE are required, as the status quo projects a 28 per cent increase in the size of the workforce. Table 12 outlines the required FTE increase to achieve the 45 per cent growth target in each of the three scenarios. The required inflow cost to HEE of each scenario is as follows:

- Scenario 1: an additional 2,591 NQT FTEs are required to achieve the necessary growth. Diagnostic radiography-specific data on average FTE could not be identified, however therapeutic radiographers work on average 0.92 FTE per individual, which was used as a proxy for diagnostic radiographers.[80] Assuming an average drop-out rate of 10 per cent, this means an additional 3,103 individuals need to be trained by 2029, at a total additional cost of £32.8m to HEE over the ten years.
- Scenario 2: an additional 2,149 NQT and 442 international recruitment FTE are required. Applying the average FTE per individual of 0.92, and assuming an average drop-out rate of 10 per cent, this means an additional 2,574 individuals need to be trained, at an NQT cost to HEE of £27.2m over the ten years.
- Scenario 3: an additional 1,300 NQT, 470 intra-NHS, 554 rejoiner and 267 international FTE are required. Applying the average FTE per individual of 0.92, and assuming an average drop-out rate of 10 per cent, this means an additional 1,557 individuals need to be trained, at an NQT cost to HEE of £16.5m over the ten years.

Table 12: Scenario growth of the diagnostic radiography workforce

	Total FTE increase				Total known cost to HEE
Scenario	**Newly qualified (specialist) training**	**Intra-NHS**	**Rejoiner**	**International recruitment**	
Scenario 1	2591	-	-	-	**£32,822,089**
Scenario 2	2149	-	-	442	**£27,226,574**
Scenario 3	1300	470	554	267	**£16,469,221**

Source: RAND Europe.

As stated in the methods section of the report, since HEE pays costs associated with specialist training, we can estimate the cost to HEE of training each additional person by using the standard tariff rates set by DHSC and then adjusted for other factors such as the market forces factor, the average FTE worked per individual, and the need to train more people than positions required to allow for drop-out rates.

Scenarios 2 and 3 include other entry routes which create costs elsewhere in the health system. Such costs tend mainly to fall to the Trust or NHS body employing the individual as CPD and international recruitment costs are their responsibility. Our understanding is that no costs arise to HEE from these entry routes, so this does not affect the total estimated cost to HEE. However, when interpreting the overall costs of different scenarios, it must be remembered that this represents the cost to HEE only, not to the total cost to the health sector.

Therapeutic Radiography

Context

Therapeutic radiographers deliver radiotherapy treatments, such as x-rays and other ionising radiation, to treat patients with medical conditions, largely cancer and tumours.[81] They care for patients along the whole care pathway, from referral to post-treatment follow up, working within multidisciplinary teams.[82]

Similarly to diagnostic radiography, training in therapeutic radiography is traditionally provided in a three-year full-time undergraduate degree, which can also be completed part-time in around six years.[83] Those with a relevant undergraduate degree can undertake a postgraduate programme in therapeutic radiography, although it can be hard for individuals to acquire funding for such courses and, as a result, not many newly qualified trainees take this route.[84] Furthermore, some training spaces have recently been left unfilled, largely because student bursaries have recently been removed for therapeutic radiography.[85]

Historic workforce stock and flows

On 31 March 2019 there were 2,844 FTE therapeutic radiographers working in the NHS in England. Over the three years prior, the following average **annual** FTE flows are observed:

- Inflows:
    - Newly qualified training: 219 (56 per cent of inflows)
    - Intra-NHS: 111 (28 per cent of inflows)
    - Rejoiners: 49 (13 per cent of inflows)
    - International recruitment: 11 (3 per cent of inflows)
- Outflows:
    - Early leaver: -260 (90 per cent of outflows)
    - Retirement: -30 (10 per cent of outflows)

Overall, there is an average annual total of 390 FTE inflows to the workforce and -290 FTE outflows from the workforce, with a net average annual inflow of 100 FTE.

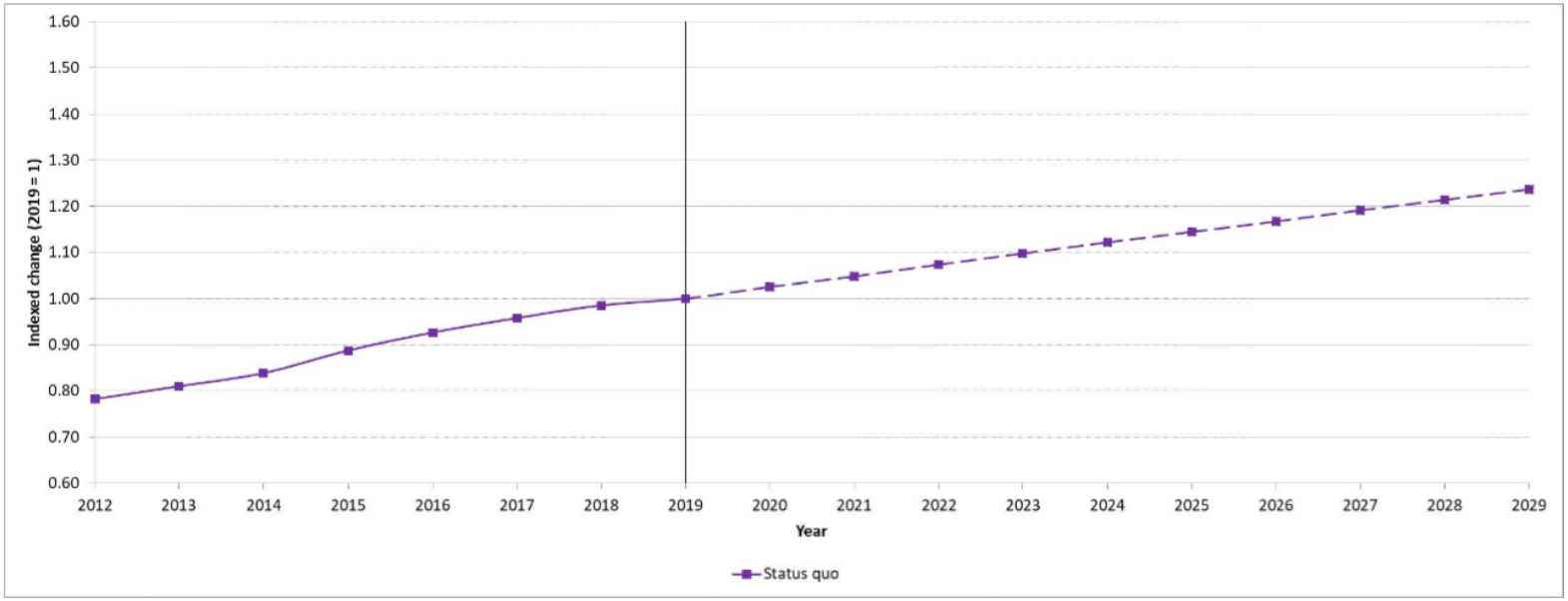
Other relevant information

The Interim NHS People Plan, published in June 2019,[86] stated that over the following five years, more clinical degree-level apprenticeships would be introduced, including for therapeutic and diagnostic radiographers. It also said that in 2019–20, there would be a focus on increasing applications to undergraduate education, particularly in professions such as therapeutic radiography where there is a shortage. Furthermore, the NHS People Plan 2020/2021,[87] published in July 2020, also reinforced the commitments to addressing workforce shortages within radiography. We were not able to build these plans into our model for therapeutic radiographers because the data were not available at the time the analysis was conducted.

Projected status quo growth

Figure 6 shows the projected status quo growth of therapeutic radiographers from 2019 to 2029 (dashed line), assuming a continuation of inflow and outflow trends over the past three years (i.e. in the results achieved from HEE's current activity level) and applying those 'flow' trends to the known 'stock' of therapeutic radiographers. Using 2019 as the reference year, the demographic stock-flow economic model projects a 24 per cent increase in FTE, a growth of 672 FTE from 2,844 in 2019 to 3,516 in 2029, as outlined in Table 13. This is in line with the historic growth in the workforce, as shown by the solid line in Figure 6.

Figure 6: Historic and status quo changes in the therapeutic radiography workforce, indexed to 2019



Source: RAND Europe. Note: For diagnostic radiographers and therapeutic radiographers, values up to and including 2019 are historic (solid line) on the graph, and values from 2020 onwards are estimates from the modelling (dashed line).

Table 13: Historic and status quo changes in the therapeutic radiography workforce

	2012	2013	2014	2015	2016	2017	2018	2019	2020	2021	2022	2023	2024	2025	2026	2027	2028	2029
Total FTE	2225	2303	2385	2524	2634	2724	2802	2844	2915	2983	3054	3123	3190	3255	3320	3387	3454	3516
Indexed change	0.78	0.81	0.84	0.89	0.93	0.96	0.99	1.00	1.02	1.05	1.07	1.10	1.12	1.14	1.17	1.19	1.21	1.24

Source: RAND Europe. Note: Indexed change from reference year.

Scenarios and costing

We estimate that to achieve 45 per cent growth in the therapeutic radiography workforce by 2029 an additional 609 FTE are required, as the status quo projects a 24 per cent increase in the size of the workforce. Table 14 outlines the required FTE increase to achieve the 45 per cent growth target in each of the three scenarios. The required inflow and cost to HEE is as follows:

- Scenario 1: an additional 609 NQT FTEs are required to achieve the necessary growth. As therapeutic radiographers work on average 0.92 FTE per individual,[88] and assuming an average drop-out rate of 10 per cent, this means an additional 729 individuals need to be trained by 2029, at a total additional cost of £7.7m to HEE over the ten years.
- Scenario 2: an additional 578 NQT and 30 international recruitment FTE are required. Applying the average FTE per individual of 0.92, and assuming an average drop-out rate of 10 per cent, this means an additional 693 individuals need to be trained, at an NQT cost to HEE of £7.3m over the ten years.
- Scenario 3: an additional 341 NQT, 173 intra-NHS, 77 rejoiner and 18 international FTE are required. Applying the average FTE per individual of 0.92, and assuming an average drop-out rate of 10 per cent, this means an additional 409 individuals need to be trained, at an NQT cost to HEE of £4.3m over the ten years .

Table 14: Scenario growth of the therapeutic radiography workforce

	Total FTE increase				**Total cost to HEE**
Scenario	**Newly qualified (specialist) training**	**Intra-NHS**	**Rejoiners**	**International recruitment**	
Scenario 1	609	-	-	-	**£7,711,023**
Scenario 3	578	-	-	31	**£7,330,231**
Scenario 2	341	173	77	18	**£4,326,212**

Source: RAND Europe.

As stated in the methods section of the report, since HEE pays costs associated with specialist training, we can estimate the cost to HEE of training each additional person by using the standard tariff rates set by DHSC and then adjusted for other factors such as the market forces factor, the average FTE worked per individual, and the need to train more people than positions required to allow for drop-out rates.

Scenarios 2 and 3 include other entry routes which create costs elsewhere in the health system. Such costs tend mainly to fall to the Trust or NHS body employing the individual as CPD and international recruitment costs are their responsibility. Our understanding is that no costs arise to HEE from these entry routes, so this does not affect the total estimated cost to HEE. However, when interpreting the overall costs of different scenarios, it must be remembered that this represents the cost to HEE only, not to the total cost to the health sector.

Specialist cancer nursing

Context

Cancer nurses play a critical part in the delivery of cancer services across the pathway, ensuring personalised care and support for people living with and beyond cancer. The HEE workforce plan focused on specialist cancer nurses who have high levels of specialised technical skills as a priority profession. However, nursing for cancer patients is much wider than these specialist nurses – nurses support people with a wide variety of health conditions in both the community and acute settings.

However, the way in which nurses are currently recorded by the NHS means we cannot identify the wider cancer nursing workforce, so instead this analysis focuses on just the specialist cancer nurses who were included in the Macmillan Cancer Workforce in England census.[89] This includes nurses on Agenda for Change Bands 5 to 9 who spend more than half of their time directly supporting adults living with cancer and have a documented training record declaring them to be specialists in cancer care. It includes secondary and tertiary care in both hospitals and the community.

Individuals cannot become a specialist cancer nurse directly from newly qualified training routes as experience is required as an adult nurse before specialisation in cancer is possible. This means that inflows largely come from individuals in the wider nursing pool who undertake ad hoc training to become a specialist cancer nurse.[90] Generally, specialist cancer nurses require at least five years' post-registration clinical experience, including two in cancer or a similar area, as well as a specialist qualification in cancer, palliative care or similar. As a result, all individuals under 30 years of age entering the workforce from outside the NHS (i.e. a private organisation) are classed as inflows from continued professional development, rather than newly qualified training.

Historic workforce stock and flows

On 9 October 2017 there were 3,851 FTE specialist cancer nurses working in England. Data on

the flows of specialist cancer nurses were not available, meaning that data on inflows and outflows of Agenda for Change[91] Band 6, Band 7 and Band 8a nurses (which cover 99 per cent of specialist cancer nurses) were used as a proxy for flows into and out of the specialist cancer nurse workforce. This assumption is the best available but there are likely to be important differences. For example, 9 per cent of specialist cancer nurses working in 2017 were from outside the EU and a further 5 per cent were from the EU excluding the UK,[92]which may not be the same proportion as general nursing. Over the three years leading up to 2019, the following average **annual** FTE flows are observed:

- Inflows:
    - Joining the NHS: 5 (1 per cent of inflows)
    - Intra-NHS: 713 (91 per cent of inflows)
    - Rejoiners: 56 (8 per cent of inflows)
    - International recruitment: 7 (1 per cent of inflows)
- Outflows:
    - Early leaver: -544 (88 per cent of outflows)
    - Retirement: -73 (12 per cent of outflows)

Overall, there is an average annual total of 781 FTE inflows to the workforce and -617 FTE outflows from the workforce, with a net average annual inflow of 101 FTE.

Other relevant information

- The proportion of specialist cancer nurses aged 50 or over has increased from 33 per cent in 2014 to 37 per cent in 2017, which could result in a peak of retirements in the next ten years
- Brexit may have had an impact and the loss of bursaries has reduced the numbers of nurses in training.
- There has also been a rapid increase of specialist cancer nurses between 2014 and 2017, in part caused by the establishment of acute oncology services. Some of these were funded by Macmillan and this growth is unlikely to be replicated.
- There is a Global Learners Programme which supports nurses who wish to migrate to the UK to work in the NHS. It includes an online educational package to support nurses in meeting the regulatory requirements to register in the UK. Currently, HEE runs a programme team and bears the fixed central administrative costs. However, these figures can not be included in our costings as we do not know the average amounts. The unit costs of recruitment via the Global Learner Programme are borne by NHS Trusts. This programme is aimed at all nurses; not just specialist cancer nurses.
- Very few specialist cancer curses are recruited internationally because specialist cancer nursing is not well-developed in other parts of the world.
- HEE also has a rejoining scheme for nurses. For each nurse, it pays approximately £2,500 in course fees, contribution to placement costs and stipendiary. However, the COVID-

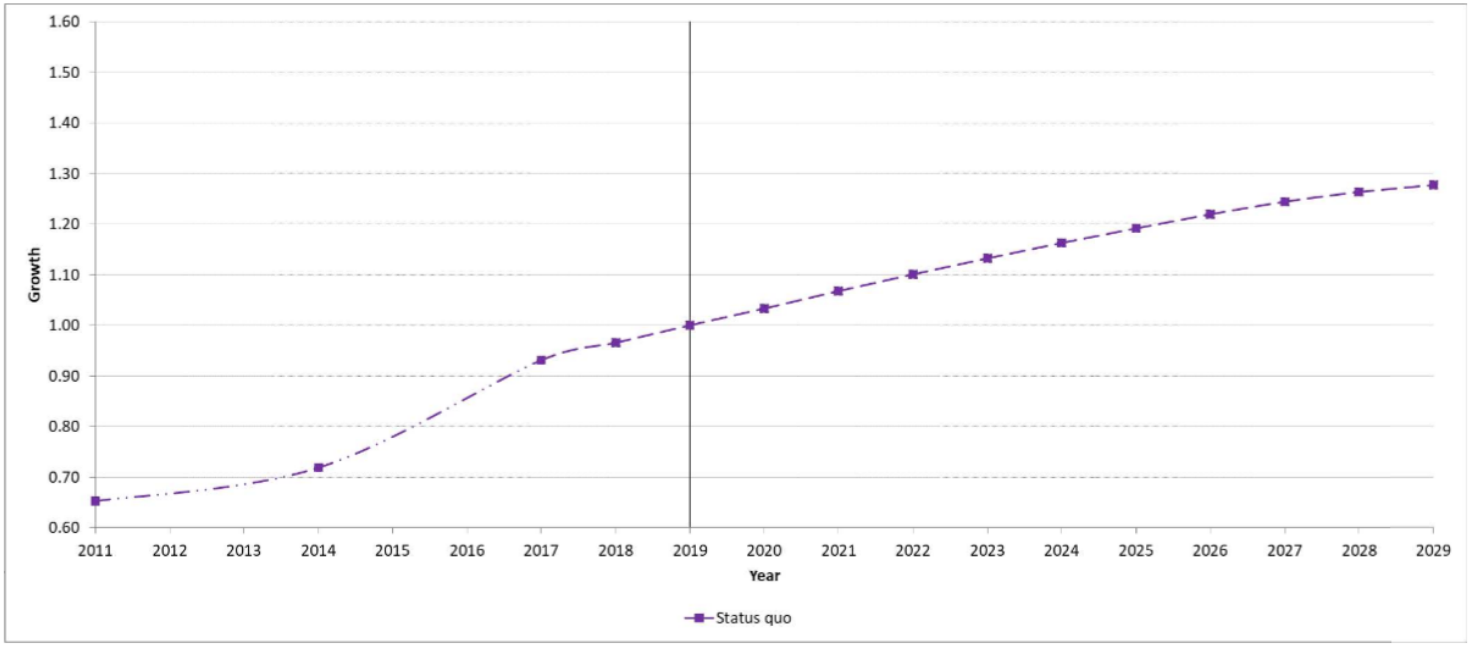
19 response may have severely limited the pool of possible rejoiners over the medium term.

- Subsequent to conducting this analysis, the NHS published the NHS People Plan 2020/2021,[93] which set out commitments to offer training grants for specialist cancer nurses.

Projected status quo change

Figure 7 shows the projected status quo growth of specialist cancer nurses from 2017 to 2029 (dashed line), assuming a continuation of inflow and outflow trends over the past three years (i.e. in the results achieved from HEE's current activity level). Using 2019 as the reference year, the demographic stock-flow economic model projects a 28 per cent increase in FTE, a growth of 1,149 FTE from 4,135 in 2019 to 5,284 in 2029, as outlined in Table 15. This represents a decrease in the growth rate of the workforce compared to historic trends, as shown by the dotted and dashed line in Figure 7. This is perhaps unsurprising given the rapid increase in specialist cancer nurses when setting up acute oncology teams after 2014. However, the historic trend line is dotted and dashed, rather than solid, because different methodologies were applied when undertaking the census in 2011,[94] 2014[95] and 2017. This means differences in the stock of specialist cancer nurses over time may be a genuine increase in the size of the workforce, or a result of the different methodologies applied in the censuses over time, or a combination of the two – which is most likely. However, for the purposes of calculating the historic growth rate from 2011 to 2017 we have assumed that all of the differences in the stock of specialist cancer nurses over time are a result of genuine increases in the size of the workforce.

Figure 7: Historic and status quo changes in the specialist cancer nursing workforce



Source: RAND Europe.

Table 15: Historic and status quo changes in the specialist cancer nursing workforce

	2011	2012	2013	2014	2015	2016	2017	2018	2019	2020	2021	2022	2023	2024	2025	2026	2027	2028	2029
Total FTE	2702	-	-	2974	-	-	3851	3993	4135	4274	4413	4550	4684	4811	4930	5043	5147	5223	5284
Indexed change	0.65	-	-	0.72	-	-	0.93	0.97	1.00	1.03	1.07	1.10	1.13	1.16	1.19	1.22	1.25	1.26	1.28

Source: RAND Europe. Note: Indexed change in reference year; - = data not available.

Scenario growth and costing

There is no single qualification that enables an individual to become a specialist cancer nurse, with nurses entering the workforce through CPD rather than NQT, and no national information is available on the cost of the ad hoc training that adult nurses undertake to become a specialist in caring for patients with cancer.[96] As a result we used the necessary training to become an adult nurse as a proxy for training to become an specialist cancer nurse. This was both for staff joining the NHS and transferring within the NHS. Training to become an adult nurse is traditionally a three-year full-time undergraduate degree.[97] However, it is a big assumption that cancer nurses will behave in the same way in terms of inflows and outflows as the remainder of nurses.

To achieve 45 per cent growth in the specialist cancer nurse workforce by 2029 an additional 710 FTE are required, as the status quo projects a 28 per cent increase in the size of the workforce. Table 16 outlines the required FTE increase to achieve the 45 per cent growth target in each of the three scenarios. The required inflow and NQT cost of each scenario is as follows:

- Scenario 1: an additional 710 CPD FTEs are required to achieve the necessary growth. As specialist cancer nurses work on average 0.88 FTE per individual,[98] and assuming an average drop-out rate of 10 per cent, this means an additional 892 individuals need to be trained by 2029, at a total additional cost of £9.4m to HEE over the ten years.
- Scenario 2: an additional 703 nurses transferring intra-NHS or joining the NHS and 7 international recruitment FTE are required. Applying the average FTE per individual of 0.88, and assuming an average drop-out rate of 10 per cent, this means an additional 887 individuals need to be trained, at an NQT cost to HEE of £9.3m over the ten years.
- Scenario 3: an additional 651 intra-NHS, 53 rejoiner and 6 international FTE are required. Applying the average FTE per individual of 0.88, and assuming an average drop-out rate of 10 per cent, this means an additional 822 individuals need to be trained, at a CPD cost to HEE of £865,000 over the ten years, plus 53 rejoiners at a cost of £2,500 per rejoiner = £132,500.

Table 16: Scenario growth of the specialist cancer nursing workforce

	Total FTE increase			Total known cost to HEE
Scenario	**Joining the NHS / Intra NHS**	**Rejoiners**	**International recruitment**	
Scenario 1	710	-	-	**£9,435,161**
Scenario 2	703	-	7	**£9,342,138**
Scenario 3	651	53	6	**£8,783,612**

As stated above we can estimate the cost to HEE of training each additional person using the proxy of basic three-year training. In addition, for Scenario 3, we can estimate HEE's share of the cost for a rejoiner. However, much of the cost of the rejoiner is still met by the trust, so this estimation only represents the cost to HEE only, not the total cost to the health sector.

Summary of results

Workforce numbers for six of the seven professions were already on an increasing trajectory, assuming a continuation of current trends (Table 17). For one profession, gastroenterology, our model estimated that the workforce would meet a 45 per cent growth in the number of staff if it continued its existing trajectory, meaning that no additional measures to recruit and train more staff beyond a continuation of the same growth trend would be required. For five of the other six professions, we estimated that the size of the workforce would increase to differing degrees over the period in question, but that there would be a need for additional intervention to achieve a growth rate of 45 per cent. For the histopathology workforce, the modelling shows that without any new intervention, the number of histopathologists would actually be forecast to reduce by 2 per cent by 2029. Therefore, to meet a growth of 45 per cent, the profession would require 580 more staff at a potential maximum cost to HEE of £118 million.

Table 17: Summary of the estimated growth of cancer specialists and additional number required to reach 45 per cent growth

Cancer profession	Estimated number of FTE staff in 2019	Expected growth by 2029 (based on current trends)		Estimated additional growth required to meet 45 per cent growth (above expected growth)		Total (expected and additional) increase in FTE staff needed to reach 45 per cent growth
		Percentage growth	Estimated FTE staff increase	Additional percentage growth	Additional FTE staff required	
Clinical and medical oncology	1,185	40%	477	5%	57	534
Gastroenterology	1,290	48%	623	0%	0	623
Histopathology	1,228	-2%	-27	47%	580	553
Clinical radiology	3,087	33%	1,004	12%	384	1,388
Diagnostic radiography	14,997	28%	4,158	17%	2,591	6,749
Therapeutic radiography	2,844	24%	672	21%	609	1,281
Specialist cancer nurses	4,135	28%	1,149	17%	710	1,859
Total	**28,766**	**-**	**8,056**	**-**	**4,931**	**41,753**

Source: RAND Europe modelling using NHS ESR data and other data.

Our model estimated the *additional* cost to HEE consequent on meeting the 45 per cent growth target across all seven professions, beyond what we would expect to see if current growth trends continue. We provided estimates for a range of scenarios that vary according to the extent to which the additional staff would be coming through specialist training. We therefore

estimated the total extra cost to HEE of increasing all workforces as being between £142.2 million and £259.7 million, depending on the route taken to increase the inflows over the ten years to 2029.

We wanted to understand the extent to which our model was sensitive to our assumptions about inflows being inaccurate. We therefore applied deterministic sensitivity analysis at the 10 per cent range to the results of our modelling to identify the extent to which the additional funding required is sensitive to the changes in the inflows and outflows to each of the cancer professions. We looked at how many more (or less) FTE staff would be required through training if the other assumptions changed. This enabled us to see which are the most important inflow and outflows. For most of the workforces, the model was most sensitive to changes in the early leaver assumption, as this represents a large proportion of the outflows from the profession. Clinical and medical oncology and clinical radiology appear to be the most sensitive to almost all of the inflows and outflows, whereas the histopathology model is not sensitive to any of the assumptions. The full results of the sensitivity analysis are provided in section 4 of the supplementary information pack.

Discussion

This study firstly aimed to assess how many more staff would be required in seven key cancer professions in order to achieve a 45 per cent growth in those workforces by 2029. Secondly, it aimed to estimate the additional funding, above and beyond that which would be expected if current trends continue, that HEE would require if it were to meet those increases. Our research used the best available NHS data to estimate the baseline stock position and then build a demographic stock-flow economic model that projected the 'status quo' workforce growth out to 2029 for each of the seven key cancer professions, assuming the previous three-year average inflows and outflows remained constant (i.e. the results achieved from HEE's current activity level). The model then estimated how many more staff in each profession would be required to achieve an overall growth of 45 per cent by 2029. Finally, it estimated the training cost to HEE of that increase using available data. We found that if all of the additional growth in the cancer workforce was through training, then the total cost over ten years would amount to about 6 per cent of HEE's annual budget.

The study's methodological approach – that there would be a 'status quo' change in each workforce even without additional HEE intervention – is similar to that used in many economic models. As we did not identify any specific one-off workforce initiatives that might mean the data we collected is not representative, and as there are many specialist trainees already in training, it is reasonable to assume that trends from the past three years may continue to be valid for flows into and out of the workforce in the near future. The relationship between HEE's budget and that planned growth is less clear. Our analysis of HEE's accounts for the past three years indicates an overall decline in its budget (largely because student nurses are no longer entitled to bursaries). However, even looking at post-graduate medical and dental courses only, HEE's spending has only increased by approximately 1 per cent in cash terms from one year to the next (see Table 2). This is a modest change alongside the increases in staffing that we have already seen in each of our professions (bar histopathology). The small increase in spending may indicate that increasing staff numbers in each workforce may not be closely dependent on large HEE budget increases, although time lags between training costs and the subsequent inflow of qualified staff are also an important caveat when considering the link between the HEE's spending and the number of trained staff.

The results of our economic modelling show that the target of 45 per cent growth is more stretching in some professions than others. This may be in terms of the health sector's ability to scale up its activity, or about the financial cost of increasing staffing. As Figure 3 shows, if current trends continue in clinical and medical oncology, that profession's numbers would increase by 40 per cent by 2029 without additional intervention; the workforce only needs to grow by an additional 57 staff to meet the 45 per cent growth scenario. However, the cost per professional trained is high. In diagnostic radiography, however, the modelling estimated the need for an additional 2,591 staff to meet a 45 per cent increase, although the cost of training

each person is significantly less than for a clinical or medical oncologist. As might be expected, we noted that the costs of training per person are substantially less for those workforces that are not medical doctors (radiographers and nurses), even though the numbers required are larger. The costs presented only include specialist training rather than foundation-level training, because students starting their medical training in 2020 may not be consultants by 2029.

There are also some issues regarding the feasibility of different scenarios modelled in this study. For some professions, particularly clinical oncology, there is often no international equivalent role and therefore international recruitment is difficult. External circumstances such as the UK's exit from the EU and the COVID-19 pandemic will further affect the feasibility of recruiting internationally. Equally, scenarios that rely heavily on increasing the number of professionals trained may find constraints in the number of work placement places to be a limiting factor. Relying heavily on increased training would also create a need to front-load recruitment of any additional staff towards the start of the ten-year period, due to the time taken to complete specialist training. Therefore, this will also require a front-loaded budget for HEE.

The model looks at the cost of specialist training but increases in all of the cancer professions highlighted in this study may mean that several different professions seeking staff are all searching primarily in the same pool of foundation-qualified doctors. It will be important to understand the extent to which there is any tension between different professions using the same pool of foundation-level qualified doctors to attract specialty recruits, or whether the NHS is able to increase its number of foundation and core training places, to accommodate such increases. If it cannot, then any increase to doctors specialising in cancer may be at the expense of other parts of the NHS. For this reason, it will be important for HEE to understand how many undergraduate medical placements will be required and the potential impact in other NHS sectors.

The true picture of required future staffing is likely to be more nuanced than a simple 45 per cent increase across all seven professions. For example, there may be a greater demand in future for some of these professions than for others. Histopathology is a profession of particular concern, given the almost static size of its workforce under the status quo assumptions, unlike the other six professions. Histopathology is likely to remain a crucial cancer workforce, given that it provides the definitive diagnostic for most cancers. Technological advances and new, more flexible, job roles can also be expected to affect the growth scenarios, such as the increasing use of non-medical staff or even artificial intelligence to report on some of the imaging tests undertaken, a role historically carried out solely by radiologists.[99]

Study strengths and limitations

We are not aware of other previous studies that have attempted to estimate the associated costs to HEE of increases to the seven cancer professions identified in the 2017 Cancer

Workforce Plan as being priorities for growth. Such estimates are important as HEE has a pivotal role in ensuring an adequate future workforce for the NHS, but it is yet to receive a long-term funding commitment. This study will therefore hopefully assist the government and HM Treasury in understanding the implications for funding of workforce increases in a vital area. Other key strengths of the work are:

- The study is based on the most complete source of NHS workforce data.
- The study builds forward from historic growth trends rather than modelling scenario increases from a static baseline, which would be less realistic.
- We have used a collaborative approach throughout and engaged many relevant stakeholders to test and validate data sources and assumptions. There has been a good deal of consensus over the results of this work.

There are, however, some limitations and caveats with the economic modelling. We give a summary of these here, and describe them in more detail in the Section 3 of the supplementary information pack.

- The ESR workforce data we have used has some minor flaws. In particular, it does not give us complete information about the inflows and outflows of staff, the coding of staff to particular professions is not always 100 per cent accurate, and it cannot give us detailed data on specialist cancer nurses.
- The model assumes that previous growth rates will continue, but in reality a wide range of factors may influence this. For example, it would be reasonable to assume that there is a finite pool from which international staff and rejoiners will come, and therefore the health sector may find it increasingly difficult to recruit from these fields once they have picked the 'low-hanging fruit'.
- There are significant costs in increasing the seven workforces that do not fall to HEE, so the cost information described cannot be used in itself to make comparisons between the total costs of different scenarios for increasing the workforce. In modelling different cost scenarios for achieving a 45 per cent growth, we do not consider the contribution or costs of other parts of the NHS.
- We did not look at the changing nature and productivity of the different workforces; for example, advances in imaging technology may allow the same number of staff to meet the needs of more patients.
- We could not fully consider the feasibility of the modelled scenarios for increasing the workforces. There may be particular limitations to the feasibility of different scenarios, not least that the current COVID-19 pandemic and the UK's exit from the EU may impact the recruitment of international nurses and doctors to the NHS.

Conclusion

Although there are several caveats and limitations associated with our analysis (see above), this research aims to understand the requirements for the NHS, and particularly for HEE, of increasing the seven priority cancer workforces by 45 per cent by 2029. The modelling identifies the workforces that will likely require the most significant additional interventions by HEE if they are to grow by 45 per cent and offers an estimate of the associated specialist training costs of such interventions. In addition to HEE's important role in securing staffing, the report also explains that other parts of the health sector, particularly NHS trusts, also play a role. While three different scenarios for increasing the workforce have been set out, external factors such as the impact of COVID-19 and the UK leaving the EU will likely have significant implications for the NHS's ability to attract more staff through some routes and this study will allow policymakers to think about the relative costs and feasibility of alternative options. It should also be noted that the analysis does not take into account commitments made to expand parts of the cancer workforce in the NHS People Plan 2020/21 as these commitments were announced after the analysis was conducted.

Future Research

Some of the limitations and uncertainties described above might be addressed through future research studies. In our view, the most important of these are:

- Considering the growth in the cancer workforce in the context of updated forecasts of increasing demand. Understanding both modelled demand and capacity will allow a clearer understanding of what investment may be needed in order to maintain or improve the quality and timeliness of cancer services in line with the NHS's ambitions. Modelling future demand would need to take into account of not only the changing UK population demographics, cancer incidence, survival and the needs of people living with cancer, but also the likely impact of NHS policies to improve early diagnosis, and to improve treatment personalisation.
- Linked to the question of future demand, it would be useful to explore the implications of new ways of working for the mix of skills and professions required to diagnose, treat and support people living with cancer. The research reported here assumed that the same percentage increase will be required for all seven cancer professions by 2029, but it might be possible to refine this approach by looking at where the biggest capacity constraints and delays are in a cancer patient's journey, as this would inform understanding of how to prioritise workforce growth among the different specialisms.
- The present research focused on the implications and costs for HEE, in line with the research brief. Future research might map the responsibilities for workforce training, recruitment, retention and deployment, and use these to obtain fuller costings across all organisations of the different options of increasing the size of key cancer professions by 45 per cent by 2029. Although this report focuses primarily on inflows, it would also be important to consider the possible role of staff retention in increasing numbers given

the large proportion of staff who leave the workforce pre-retirement.

- Collecting more granular data on how international staff are recruited to the NHS, including greater understanding of the numbers attracted through recruitment programmes versus those who join the NHS individually due to family or other circumstances. The impact of the UK's exit from the EU and the COVID-19 pandemic may also significantly affect the number and nationality of staff recruited from overseas. It would also be useful to understand the barriers to joining the NHS and to assess the cost of recruiting an international doctor to specialist posts, including any hidden costs such as greater turnover of international staff. This might give a better view of the extent to which the status quo assumptions for international staff are likely to be feasible in practice.
- Seeking better information about the impact of increased workforce capacity on patient experience and outcomes. This might include modelling how more staff affect a patient's flow through the system or their waiting time for cancer care, assessing geographical variation in services provided, and even comparing England to international data on the link between staffing and outcomes for cancer diagnosis, treatment and support.

Policy recommendations

The research team makes the following recommendations for health bodies if they are to secure sufficient staff to meet the ambitions of earlier diagnosis and improved outcomes as set out in the NHS Long Term Plan:

1. The government must provide long-term funding, aligned to the NHS Long Term Plan, to secure a sufficient pipeline of future NHS cancer staff. Initial estimates suggest that, to achieve a 45 per cent growth in the seven key cancer professions, this will need to be at least £142 million but closer to £260 million over the next three to five years. This should be taking into account in the next Comprehensive Spending Review.
2. NHS England and Improvement should ensure that their next People Plan adequately reflects the NHS's ambitions for cancer by clearly articulating how many staff will be needed to deliver quality services to a growing number of patients in the long term.
3. NHS England and Improvement, working with HEE, should review how it can influence its pipeline of staff to the professions where they are most needed, with a particular focus on histopathology. The forecast histopathologist numbers are concerning, as this is the only profession with current trends suggesting a decline in the number of staff by 2029.
4. Building on the NHS People Plan 2020/21, NHS England and Improvement, in collaboration with HEE, should ensure that consideration of likely future demand and planned future modelling of the workforce is translated into clear long-term ambitions for the growth of different cancer professions.
5. This research project identified a number of important areas for future research. NHS England and Improvement, the National Institute for Health Research and other funders should commission work to fill these research gaps, to inform its approach going forwards with a fuller understanding of the needs of the cancer workforce and the constraints to its growth.
6. NHS England and Improvement, along with HEE, should assess the feasibility of the different scenarios for increasing inflows. This is particularly important in the light of the likely restrictions arising from the COVID-19 pandemic, which may make international recruitment more difficult.

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Appendix B. Paper 2

Guthrie, S., Pollard, J., Parkinson, S., Altenhofer, M., Leach, B., & Lichten, C. A. (2020). **Pregnancy research review: Data and methods report**. Santa Monica, CA: RAND Corporation. URL: https://www.rand.org/pubs/research_reports/RR4340.html

An extract of the publication relevant to the economic analysis is presented here. Specifically:

- 1. Introduction
- 2. Methods
    - Overview of study approach
    - 2.2 Task B: Contextualisation of research spend
- 4. Task B results: Contextualisation of research spend
- References

EUROPE

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SARAH PARKINSON, MARLENE ALTENHOFER

Pregnancy research review

Data and methods report

For more information on this publication, visit www.rand.org/t/RR4340

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Preface

This report sets out the results of a study – commissioned by the UK Clinical Research Collaboration – that aimed to characterise the pregnancy-research landscape in the UK, assess the level of funding for pregnancy research in the UK, and understand the extent to which funded research addresses the research priorities identified by stakeholders. This data and methods report sets out the methods used for the study, and presents in detail the results of the study and the data it produced. It is accompanied by a policy report that focuses on presenting the key policy-relevant messages emerging from the work. This report is likely to be of interest to research funders and those working in research related to pregnancy.

For more information about this report, or RAND Europe, please contact:

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Abbreviations

ABPI	Association of the British Pharmaceutical Industry
AHRC	Arts and Humanities Research Council
AMRC	Association of Medical Research Charities
ART	Assisted Reproductive Therapy
BBSRC	Biotechnology and Biological Sciences Research Council
CCG	Clinical Commissioning Group
CORDIS	Community Research and Development Information Service
CRNCC	Clinical Research Network Coordinating Centre
CS	Caesarean Section
CSO	Chief Scientist Office
DALY	Disability-Adjusted Life Year
EPSRC	Engineering and Physical Sciences Research Council
ESRC	Economic and Social Research Council
GBD	Global Burden of Disease
GDP	Gross Domestic Product
HSC R&D	Health and Social Care Research and Development
ICD	International Classification of Diseases
ISRCTN	International Standard Randomised Controlled Trial Number
ITT	Invitation to Tender
JLA	James Lind Alliance
LCRN	Local Clinical Research Network
MBRRACE-UK	Mothers and Babies: Reducing Risk through Audits and Confidential Enquiries across the UK
MRC	Medical Research Council
NC3Rs	National Centre for the Replacement, Refinement and Reduction of Animals in

	Research
NCT	National Childbirth Trust
NERC	Natural Environment Research Council
NICE	National Institute for Health and Care Excellence
NICU	Neonatal Intensive Care Unit
NIHR	National Institute for Health Research
PMC	PubMed Central
PPIE	Patient and Public Involvement and Engagement
R&D	Research and Development
SIGN	Scottish Intercollegiate Guidelines Network
STFC	Science and Technology Facilities Council
UKCRC	UK Clinical Research Collaboration
UKRI	UK Research and Innovation
US	United States
WHO	World Health Organisation

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This study was made possible thanks to the support and engagement of many people and organisations, to whom we are very grateful. We are grateful to our expert advisory-group members Prof. Angela Harden, Prof. Jane Norman, Dr Oliver Rivero Arias and Rachael Leonard for their guidance and helpful support throughout the study. We would like to thank the UKCRC project steering group, and in particular Leanne Dew and Natalie Owen, for their continued engagement throughout the study. We are very grateful to all the funders who took time to share their funding data, provide other valuable input and respond to our queries. Thanks also to Jocelyn LeBlanc at the Association of Medical Research Charities for assisting us with obtaining data from AMRC members. We are thankful to the Association of the British Pharmaceutical Industry (ABPI) for helpful insights about industry spend on pregnancy research, and in particular to Sheuli Porkess who assisted with circulating our request about funding information and the link to our survey to ABPI members.

We would also like to thank Dr Jim Carter from the Medical Research Council (MRC) who helped us with Health Research Authority (HRA) data.

Our sincere thanks also go to Prof. Sophie Staniszewska from Warwick Research in Nursing Warwick Medical School, who advised us on the work at the request of NIHR INVOLVE.

We would like to express our gratitude to all the (anonymous) individuals who contributed to our crowdsourcing exercise and the priority-setting survey, and to those who shared the link to the survey with their networks.

This study would also not have been possible without the members of the public who participated in our workshop: we are immensely grateful to the women and their partners who shared what really matters to them when it comes to pregnancy research.

Our sincere thanks also go to Kerry Smith from the National Childbirth Trust (NCT) who helped organise this workshop, and to the South Bretton Family & Community Centre in Peterborough, who kindly allowed us to hold this workshop in their premises.

We would also like to thank the participants in our emerging-findings workshop for their engaging discussions and helpful input.

Finally, we are grateful to our quality assurance reviewers, Dr Sarah Parks, Dr Katherine Morley and Dr Advait Deshpande from RAND Europe, for their constructive and useful guidance and reviews.

1. Introduction

1.1. Context and aims

It is essential to base decisions about research investment on the best available evidence, particularly when funding comes from a limited public purse. However, assessing priorities in health research can be challenging given the need to balance multiple factors, including societal needs, research capacity and potential return on investment. Understanding societal needs ideally involves gathering views from stakeholders with varying interests, experiences and expertise. Meanwhile, research outcomes and impacts can be unpredictable, creating uncertainty about the level of, and time frame for, return on research investment. Despite these challenges, clear, systematic processes for priority-setting can help ensure that funded health research has potential for impact, meets needs, and that resources are used fairly and efficiently.

Regarding pregnancy-related health, there is mounting evidence that more research is needed to improve outcomes for women and babies. Recent policies have emphasised the need to improve pregnancy care, while noting a lack of research in key areas, including pre-conception interventions, screening tests, pregnancy treatments and models for perinatal care.[1] MBRRACE-UK (Mothers and Babies: Reducing Risk through Audits and Confidential Enquiries across the UK) has highlighted slow progress in reducing rates of extended perinatal mortality with more research needed. The issue is not new; a 2009 review concluded that UK maternal and perinatal health research is underfunded compared to other conditions, with the UK devoting a lower proportion of funding than other English-speaking countries.[2]

In 2014, the Chief Medical Officer recommended a review of research needs and expenditure in pregnancy in the UK.[3] The objective of this study was to deliver that review, and generate a sound evidence base on UK pregnancy research needs and priorities, and how that compares to the current funding landscape.

---

[1] Policies such as NHS England's *National Maternity Review* (2016), the *Strategic Vision for Maternity Services in Wales* (Welsh Government 2011), the Scottish five-year plan *The Best Start* (Scottish Government 2017), Northern Ireland's *Strategy for Maternity Care* (Department of Health, Social Services and Public Safety 2012) and NIHR's *Better Beginnings* (NIHR 2017).

[2] Fisk and Atun (2009).

[3] Chief Medical Officer (2015).

The main research questions of this study were:

1. What is the current level of spend on pregnancy research in the UK?
2. What is currently funded, in terms of type of research and topic?
3. How does the current pregnancy research spend in the UK compare to other health research areas?
4. What are the main priorities for future pregnancy research in the UK?

1.2. Structure and purpose of this report

This report sets out in detail the methods used to address the four research questions outlined above, and the results and data produced through these research activities. A separate accompanying report sets out the policy implications of this research and analysis. The remainder of this report is structured as follows:

Chapter 2: Methods – sets out our methods in detail, including limitations and caveats.

Chapter 3: Task A results: Funding analysis – details results from the analysis of data on funded projects in pregnancy research.

Chapter 4: Task B results: Contextualisation of research spend – sets out results from our analysis of spend on research compared to costs of pregnancy, and other comparators.

Chapter 5: Task C results: Prioritisation exercise – includes results from the prioritisation exercise.

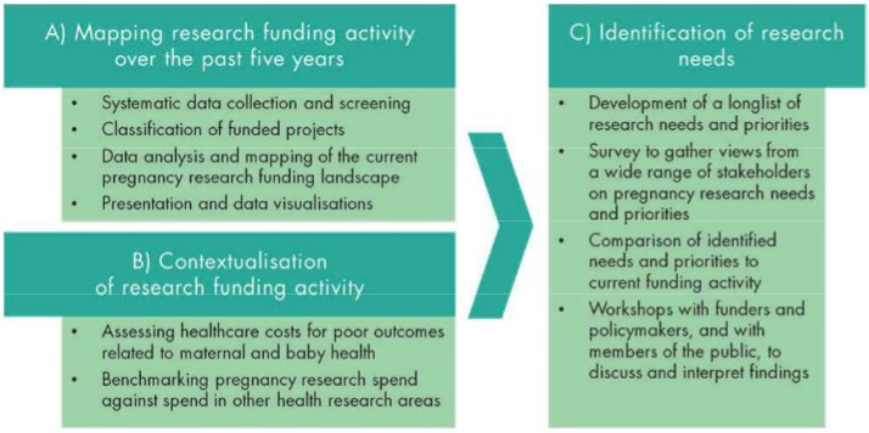
In addition, the full content of the research tools used – for example the survey questions and agendas for the workshops – are provided as annexes. An extensive set of data tables covering elements of the prioritisation exercise are also provided in the annexes.

2. Methods

Our study approach consisted of three tasks – A, B and C – as illustrated in Figure 1.

For the purposes of all the tasks in this study, we took a broad definition of pregnancy to encompass not just the period of being pregnant and giving birth, but also spanning conception, contraception, the antenatal period and postnatal outcomes and experiences linked to the process of being pregnant and giving birth, for both women and their families. We also took a broad scope in terms of what is classified as pregnancy research, covering all topics, disciplines and research approaches in which the ultimate aim of the research is to improve outcomes for pregnant women or women trying to become pregnant, and their families.

Figure 1: Overview of study approach



Task A was focused on mapping funded research related to pregnancy in the UK. We identified pregnancy-related research by searching existing databases and requesting data from funders. We then screened these data, looking at award titles and abstracts, to remove irrelevant funding awards and classify awards that were relevant based on their topic, funder, research approach, funding type, and the stage of pregnancy they address.

Task B aimed to contextualise the amount of funding allocated to pregnancy research, both against the costs and burden of pregnancy as a health condition, and against the amount invested in other areas of research compared to their costs.

Task C was a prioritisation exercise spanning the whole of pregnancy research, and was intended to provide a picture of the key topics of importance to different stakeholders, including researchers, healthcare professionals, charities, funders and members of the public.

The remainder of this chapter sets out the methods used for Tasks A, B and C in detail, followed by discussion of caveats and limitations of the analysis.

2.1. Task A: Funding analysis

The aim of Task A was to produce an overall picture of the current UK pregnancy research funding landscape by mapping research funding activity over a period of five years (2013 to 2017).[4] This enabled us to understand the current funding picture and thus facilitate decision-making on future investment relative to research priorities. The main research questions relating to this task were:

- What is the current level of spend on pregnancy research in the UK?
- What is this being spent on, in terms of type of research and topic?

Task A was conducted in a format similar to a systematic review – databases were systematically searched to generate a list of research projects funded by UK funders. This list was then manually screened, classified and analysed.

2.1.1. Data collection

Identification of relevant datasets and funders

Relevant datasets and a list of relevant funders were identified based on information from the study's initial project specification and inputs from the study team, the UKCRC steering group and the study's expert advisory group. The initial list of funders was developed using an approach that aimed to be as inclusive as possible. We sought to include funders identified in at least one of the following ways:

- Funders covered in the UK Clinical Research Collaboration (UKCRC) Health Research Analysis database (2014) (UKCRC 2014).
- Funders in the UK Research and Innovation (UKRI) 'Gateway to Research' database of UK public funders (UKRI 2019).
- Other UK public research funders identified in consultation with the UKCRC steering group.
- Relevant UKCRC partners and related organisations.[5]
- Relevant charity funders recommended by the Association of Medical Research Charities.
- Funders with relevant research projects identified in the 360Giving GrantNav database (Grantnav homepage 2019).

[4] The data collection started in early 2019. At this time, data for 2018 were incomplete and therefore not included.

[5] For example, The Academy of Medical Sciences is a partner; we have also included analogous organisations in other disciplines, including the Royal Society, the Royal Academy of Engineering and the British Academy.

2.2. Task B: Contextualisation of research spend

The aim of Task B was to gain a better understanding of the landscape that pregnancy and pregnancy research sits in, in the context of the UK from 2013 to 2017. The main research question relating to this task was:

- How does the current pregnancy research spend in the UK compare to other health research areas?

In order to answer this research question, the following were considered:

- Health needs surrounding pregnancy:
  - The number of maternities,[32] births and disability-adjusted life years (DALYs)[33] resulting from pregnancy and pregnancy-related conditions.
- Health spend surrounding pregnancy:
  - NHS spend on pregnancy and pregnancy-related conditions.
- Comparison of:
  - The health needs and health spend in pregnancy compared to the health needs and health spend of the UK as a whole;
  - The funding for pregnancy-related research compared to the health needs and health spend on pregnancy across the UK;
  - The funding for pregnancy-related research compared to research funding for other health conditions treated by the NHS across the UK.

In order to undertake the above contextualisation of pregnancy-related research in the UK, data were collected from multiple sources, analysed and compared with a paper in the literature contextualising research spend in other health areas.[34] Further details on data sources and the approach to analysis are provided in the following sections. All data for this study were collated and analysed in Microsoft Excel 2010.

2.2.1. Literature summary

Firstly, in order to gain a better understanding of the costs associated with non-standard pregnancies, a brief search and summary of relevant literature was undertaken. This was particularly important because the data on the health needs and health spend associated with pregnancy and pregnancy-related conditions do not distinguish between needs and spend related to standard pregnancies and those related

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[32] Maternities are defined by the Office for National Statistics (ONS) as: 'Women having babies (including stillbirths). A maternity is a pregnancy resulting in the birth of 1 or more children, therefore, these figures are not the same as the number of babies born.'

[33] The World Health Organisation defines DALYs as: 'One DALY can be thought of as one lost year of "healthy" life. The sum of these DALYs across the population, or the burden of disease, can be thought of as a measurement of the gap between current health status and an ideal health situation where the entire population lives to an advanced age, free of disease and disability' (WHO 2019).

[34] Luengo-Fernandez et al. (2015).

to complications surrounding pregnancies. Google and Google Scholar were searched using terms such as 'pregnancy outcomes economic cost UK', 'pregnancy smoking outcomes economic costs UK' and 'high risk pregnancy costs' and literature relevant to complications (e.g. preterm birth, low birth-weight, stillbirth and multiple pregnancies) as well as lifestyle issues (e.g. smoking and obesity) during pregnancy was identified, reviewed and summarised.

To find additional literature on the costs associated with pregnancy complications, the following search was run in PubMed: *economic AND (costs OR impacts OR consequences) AND (birth OR pregnancy OR preterm OR stillbirth)*. The titles of the first 100 hits (sorted by 'Best Match') were screened for relevance, limiting the results to the past 10 years. This resulted in 39 references being selected for potential inclusion. They were exported to an EndNote library. Reviews were identified and read in full. Information was extracted from all relevant reviews (10 in total) relating to the costs associated with pregnancy care and pregnancy complications wherever available (including intangible costs that were covered in the reviews). There was a focus on data from the UK, where available. This information was then summarised in the report. The results of the literature summary are provided in Section 4.2 below.

2.2.2. Data collection

Collection of health-needs data

In order to quantify pregnancy-related health needs in the UK, data were collected on the incidence of births and maternities annually across England, Wales, Scotland and Northern Ireland, as well as DALYs caused by pregnancy and pregnancy-related conditions. Table 5 provides a breakdown of the data sources used to collect the necessary health-needs data.

Table 5: Data sources for pregnancy-related health needs

Data description	Data source	Available data
Statistics on live births, stillbirths, total births and maternities for England and Wales.	Office for National Statistics (ONS). Births in England and Wales: summary tables.[35]	2013, 2014, 2015, 2016, 2017
Statistics on live births, stillbirths, total births and maternities for Scotland.	National Records of Scotland (NRS). Births Time Series Data.[36]	2013, 2014, 2015, 2016, 2017
Statistics on live births, stillbirths, total births and maternities for Northern Ireland.	Northern Ireland Statistics and Research Agency. Registrar General Annual Report 2013–17.[37]	2013, 2014, 2015, 2016, 2017
Statistics on DALYs as a result of pregnancy and pregnancy-related	Global Burden of Disease (GBD) Collaborative Network. Global	2013, 2014, 2015, 2016, 2017

[35] Office for National Statistics (2018).

[36] National Records of Scotland (2018).

[37] NISRA (2019).

conditions.	Burden of Disease Study 2017 (GBD 2017) Results.[38]

An inclusive approach was taken when collecting data on DALYs caused by pregnancy and pregnancy-related conditions. All conditions categorised as 'Maternal and neonatal disorders' by the GBD Collaborative network were included, as outlined in Table 6. As a result, the DALYs considered in this study reflect both the mother ('Maternal disorders') and the newborn child ('Neonatal disorders'). For each of the conditions included in the GBD 2017 Results, details of the International Statistical Classification of Diseases (ICD)-10 codes, diagnosis, symptom and procedural codes – used by healthcare professionals globally – can be found on the GBD website. It is important to note that stillbirths are not included in the 'Maternal and neonatal disorders' DALYs provided in the GBD.

Table 6: GBD 2017 conditions categorised as 'Maternal and neonatal disorders'

Maternal disorders	Neonatal disorders
Maternal haemorrhage	Neonatal preterm birth
Maternal sepsis and other maternal infections	Neonatal encephalopathy due to birth asphyxia and trauma
Maternal hypertensive disorders	Neonatal sepsis and other neonatal infections
Maternal obstructed labour and uterine rupture	Hemolytic disease and other neonatal jaundice
Maternal abortion and miscarriage	Other neonatal disorders
Ectopic pregnancy	
Indirect maternal deaths	
Late maternal deaths	
Maternal deaths aggravated by HIV/AIDS	
Other maternal disorders	

Collection of health-spend data

In order to quantify NHS spend on pregnancy and pregnancy-related conditions across the UK, programme budgeting data were collected from numerous sources.[39] An inclusive approach was taken when collecting health-spend data, with costs categorised as 'Maternity and reproductive' and 'Neonates' included to capture pregnancy and pregnancy-related conditions. Table 7 provides a breakdown of the data sources from which the necessary health-spend data was collected. Unfortunately, the NHS spend data categories are not broken down any further beyond 'Maternity and reproductive' and 'Neonates',

[38] GHFx (2019).

[39] As explained by NHS Networks: 'Programme budgeting provides a framework for estimating NHS expenditure across healthcare conditions, also known as 'programmes categories', across the whole care pathway' (NHS Networks 2019).

however, detailed information on the health services that make up these categories can be found on the NHS Networks website.[40]

Table 7: Data sources for pregnancy-related health spend

Data description	Data source	Available data
Statistics on NHS spend in England related to 'Maternity and reproductive' and 'Neonates' health services	NHS Networks. 2012/13 Programme Budgeting data is now available[41].	2012–13
Statistics on NHS spend in Wales related to 'Maternity and reproductive' and 'Neonates' health services	Stats Wales. NHS Expenditure by budget category and year.[42]	2013–14, 2014–15, 201–16, 2016–17
Statistics on NHS spend in Scotland related to 'Maternity and reproductive' and 'Neonates' health services	The Scottish Government. Programme Budgeting in NHS Scotland.[43]	2011–12

No programme budgeting information could be identified for Northern Ireland meaning that the data had to be imputed using the values from England, Wales and Scotland. In addition, data were not available for all years of interest in England, Wales and Scotland, meaning that data had to be extrapolated beyond the years for which it was available. Details of this imputation and extrapolation are outlined in Section 2.2.3 below.

Collection of data on comparator conditions

In order to compare the funding for pregnancy-related research to that of other health conditions treated by the NHS across the UK, data were collected from a study undertaken by Luengo-Fernandez et al. (2015) considering levels of research funding for different health conditions in the UK. The authors collected data on research spend, health needs and health spend for cancer, heart disease, dementia and stroke in 2012 to undertake a similar analysis to that outlined below. For the purposes of this study, information on research spend, health needs and health spend for cancer, heart disease, dementia and stroke were taken from the paper, rather than collected from primary sources, due to resource limitations.

Luengo-Fernandez et al. (2015) took a similar approach to estimating the health needs and health spend of cancer, heart disease, dementia and stroke as that taken in this study. The authors adopted a 'top down' approach, using aggregate data to estimate the health spend in the NHS across the UK (as well as predicting the wider societal costs, which was deemed out of scope for this project), as described in detail in Luengo-Fernandez et al. (2012). Furthermore, GBD data were used to estimate the health need,

[40] NHS Networks (2019).

[41] NHS Networks (2019).

[42] Stats Wales (2018).

[43] Scottish Government (2015).

specifically DALYs, associated with the conditions of interest. Finally, with respect to analysis of research funding, the authors included research infrastructure expenditure, whereas this study has not accounted for all potentially relevant research infrastructure expenditure. The authors excluded research funding provided by industry, the same approach taken in this study.

Furthermore, data on the total level of (non-industry) health research funding in the UK in 2014 were obtained from the UK Clinical Research Collaboration analysis of UK health research. It was then adjusted for the remaining years of interest (2013, 2015, 2016 and 2017) using UK-wide Gross Domestic Product (GDP) deflators obtained from HM Treasury.[44] The UKCRC analysis of UK health research funding distinguishes 'direct awards', which directly fund research, and 'indirect' expenditure, which is mainly infrastructure spending. As the direct funding more closely matches the data set gathered in this study, we used those figures. It should also be noted that although these data capture a significant proportion of UK health research funding, they do not provide information on the total level of funding, as data were not captured from organisations that did not participate. Finally, the data set only provides information on research awards that were active (i.e. taking place) in the calendar year 2014, meaning it does not necessarily provide an accurate estimate of what was spent on health research in 2014. This was not accounted for in our analysis.

2.2.3. Analysis

Extrapolation and imputation of health-spend data

Firstly, in countries where health-spend data were identified, such data had to be extrapolated from the years that were available to all years of interest (2013 to 2017). In England the latest available national-level programme budgeting data set was provided in the financial year 2012–13; in Wales it was 2016–17 and in Scotland it was 2011–12. In order to extrapolate these data to the remaining years of interest the available data were scaled up using UK wide GDP deflators obtained from HM Treasury.[45] This is likely to underestimate the true health spend on pregnancy and pregnancy-related conditions (as well as all other conditions) because NHS inflation rates are typically higher than GDP deflator values, however data on NHS inflation rates for the time period in question were not identified.

Secondly, because no programme budgeting information could be identified for Northern Ireland, it had to be imputed using the values from England, Wales and Scotland. To do this the number of births, both live and still, in Northern Ireland was compared to the number of births in the rest of the UK to determine how many births occur in Northern Ireland as a proportion of births in the rest of the UK. This proportion of the health spend on pregnancy and pregnancy-related conditions in England, Wales and Scotland was then assigned to Northern Ireland. In other words, if the proportion of births in Northern Ireland was 5 per cent of that of the rest of the UK, 5 per cent of the health spend on pregnancy and pregnancy-related conditions in England, Wales and Scotland was assigned to Northern Ireland.

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[44] HM Treasury (2019).

[45] HM Treasury (2019).

In order to estimate 'All other' health expenditure – i.e. total NHS spend minus NHS spend on pregnancy and pregnancy-related conditions – the same extrapolation and imputation approach outlined above were adopted for each year of interest across all four countries of the UK. Briefly, for England, Wales and Scotland, where total health-spend data were available, UK wide GDP deflators were used to extrapolate the data to the years it was missing. For Northern Ireland, where no total health-spend data were available, a proportion of total health spend in England, Wales and Scotland was assigned to Northern Ireland based on the proportion of births occurring in Northern Ireland compared to the rest of the UK.

Analysis of the health-needs data

In order to contextualise pregnancy and pregnancy-related research funding and health needs across the UK, several approaches were taken (see Section 4.4.1 for results):

- The average number of maternities annually over the five years from 2013 to 2017 compared to the prevalence of cancer, heart disease, dementia and stroke in 2012;
- Proportion of DALYs accounted for by pregnancy and pregnancy-related conditions; and
- The average number of annual DALYs due to pregnancy and pregnancy-related conditions over the five years from 2013 to 2017 compared to the number of DALYs due to cancer, heart disease, dementia and stroke in 2012.

Analysis of health-spend data

In order to contextualise pregnancy and pregnancy-related research funding and health spend across the NHS in the UK, several approaches were taken (see Section 4.4.2 for results; note that all values are given in 2018–19 prices):

- Proportion of NHS expenditure accounted for by pregnancy and pregnancy-related conditions; and
- The average annual NHS expenditure on pregnancy and pregnancy-related conditions over the five years from 2013 to 2017 compared to the NHS expenditure on cancer, heart disease, dementia and stroke in 2012.

Analysis of research funding

In order to contextualise pregnancy and pregnancy-related research funding on other health conditions across the UK several approaches were taken (see Section 4.4.3 for results):

- Proportion of all health-relevant research funding that is accounted for by research funding for pregnancy and pregnancy-related conditions;
- The average annual level of research funding for pregnancy and pregnancy-related conditions over the five years from 2013 to 2017 compared to the research funding for cancer, heart disease, dementia and stroke in 2012;
- The average annual level of research funding per maternity over the five years from 2013 to 2017 compared to the research funding per case for cancer, heart disease, dementia and stroke in 2012;

- The average annual level of research funding per DALY attributable to pregnancy and pregnancy-related conditions over the five years from 2013 to 2017, compared to the research funding per DALY attributable to cancer, heart disease, dementia and stroke in 2012; and
- The average annual level of research funding per £1m spent on pregnancy and pregnancy-related conditions in the NHS over the five years from 2013 to 2017, compared to the research funding per £1m spent on cancer, heart disease, dementia and stroke in the NHS in 2012.

2.2.4. Caveats and limitations of the work

Firstly, it is important to recognise that pregnancy is not a disease, whereas cancer, heart disease, dementia and stroke are. As a result, the optimal health needs and health spend of pregnancy and pregnancy-related conditions will always be greater than zero, as the birth of children is desirable for society. On the other hand, cancer, heart disease, dementia and stroke are diseases that would ideally be eradicated, resulting in optimal health needs and health spend of zero. However, data on the health needs and health spend associated with pregnancy and pregnancy-related conditions does not distinguish between needs and spend related to standard pregnancies (i.e. health needs and spend we do not want to eradicate) and complications surrounding pregnancies (i.e. health needs and spend we do want to eradicate). We did explore potential approaches to separate health needs and health spend related to standard pregnancies from those related to complications surrounding pregnancies, however this was not deemed feasible given the available data and scope of the study. As a result we have considered overall health needs and health spend, although it would be preferable if we were able to consider only health needs and health spend associated with complications surrounding pregnancy. If we were able to distinguish between desirable and undesirable health needs and health spend associated with pregnancy, firmer conclusions could be drawn about how pregnancy research funding in the UK compares to other health research areas.

Secondly, data on the health needs, health spend and level of research funding across the multiple health conditions have been collated from different sources that used different approaches to gather the data. Specifically, Luengo-Fernandez et al. (2015) and UKCRC (2014) have applied their own methods, and therefore caution should be taken when comparing the information on pregnancy to that on the other conditions and the total health-relevant research spend. For example, Luengo-Fernandez et al. (2015) included research infrastructure expenditure in their analysis of research funding, whereas this study has not accounted for all potentially relevant research infrastructure expenditure. We have, however, attempted to align our methods with those applied in both studies in order to make comparisons as meaningful as possible. Specifically, in their analysis of research funding Luengo-Fernandez et al. (2015) excluded research funding provided by industry, with the same approach taken in this study. Moreover, we compared the research spend on pregnancy research to the 'direct awards' identified in the UKCRC (2014) analysis, as this approach aligns most closely with the method adopted in this study. Furthermore, although Luengo-Fernandez et al. (2015) took a more detailed approach to estimating the health spend associated with cancer, heart disease, dementia and stroke by analysing a greater number of sources (e.g. Hospital Episode Statistics and the Labour Force Survey), as well as estimating the wider societal costs, we took the same 'top down' approach in this study, using aggregate data. Finally, Luengo-Fernandez et al. (2015) also utilised GBD data to capture the DALYs associated with their conditions of interest, with the same approach applied in this study.

4. Task B results: Contextualisation of research spend

4.1. Introduction

Pregnancy research funding figures can be hard to contextualise on their own. Supplementing such information with data on the health needs and health spend of pregnancy and other conditions, as well as the research spend on such conditions, allows more meaningful conclusions to be drawn. Furthermore, a 2006 UK governmental review into the allocation of medical research funding across public bodies recommended that the impact of conditions on both the population and the economy should be considered when determining society's health priorities, as well as research priorities (Cooksey 2006).

In order to contextualise the funding of pregnancy research in the UK we attempted to gain a better understanding of the health impacts of pregnancy, as well as the landscape that pregnancy and pregnancy-related research sits in, particularly in the context of the UK from 2013 to 2017. This was done by undertaking a brief literature summary of the costs associated with poor pregnancy outcomes – presented in Section 4.2 below – and analysing numerous sources of data to better understand the health needs, health spend and research funding associated with pregnancy, and how these relate to other conditions (also presented in Section 4.2 below).

Note that this chapter sets out the results of the work only – analysis and comparison with findings from the other tasks is provided in the accompanying policy report.

4.2. Costs associated with poor pregnancy outcomes

As outlined in Section 2.2.4 (which discussed the limitations of the contextualisation work), data on the health needs and health spend associated with pregnancy and pregnancy-related conditions do not distinguish between needs and spend related to standard pregnancies and those related to complications surrounding pregnancies. In order to gain a better understanding of the costs associated with non-standard pregnancies, a brief search and summary of relevant literature was undertaken. In summary, poor pregnancy outcomes related to complications (e.g. preterm birth and low birthweight), or risky behaviours during pregnancy (e.g. smoking), are associated with considerably higher economic costs.

4.2.1. Pregnancy complications

In the literature considering the costs associated with poor pregnancy outcomes related to complications, several papers were identified and reviewed considering complications such as preterm birth, low birth-weight, stillbirth, and high-risk and multiple pregnancies (i.e. women giving birth to more than one

child). Overall, women who suffer from such complications during pregnancy incur significantly greater healthcare costs during pregnancy and for some time afterwards.

Preterm birth

Preterm birth is a factor that can increase the costs associated with pregnancy, both in terms of birth costs and in costs throughout life. Infants that are born prematurely have increased direct medical costs – due to heightened risk of neonatal ICU admission and of hospital admission beyond the neonatal period – as well as increased indirect costs from social and education services, lost productivity, home adaptations, informal carers and other non-medical costs (Petrou 2019). After pregnancy and birth, there are long-term costs associated with prematurity, including costs associated with cerebral palsy, mental retardation, vision impairment and hearing loss, as well as informal care provision for preterm infants, loss of productivity for parents of preterm infants and psychological distress, anxiety and depression of parents and family members (Frey and Klebanoff 2016).

Gestational age tends to be inversely related with pregnancy costs, despite differences in follow-up times and costs included between studies that have looked at the costs of prematurity. The costs associated with prematurity are around $100,000 for early prematurity, $10,000–30,000 for moderate prematurity and less than $4,500 for late prematurity (Soilly et al. 2014). One estimate suggests that the costs of preterm birth to the public sector in the UK are £2.946 billion over the long term for each annual cohort (Mangham et al. 2009). In a review of the literature on the economic consequences of preterm birth and low birthweight, Petrou (2003) found that direct hospital costs for newborns weighing less than 1kg were on average 75 per cent higher than for babies born weighing between 1–1.49kg, and were on average more than four times higher than direct hospital costs for newborns weighing at least 1.5kg. Furthermore, in the first year of life, babies born weighing less than 1.5kg incurred significantly higher medical costs than those weighing 1.5kg or more. Similarly, newborns weighing less than 1kg at birth were observed to have significantly higher health services utilisation in the first 8–9 years of their lives.

Multiple pregnancies

Multiple pregnancies can also result in additional costs in provision of care. In an observational study of four district hospitals in southeast England over 15 months, Mistry et al. (2007) compared the healthcare costs for multiple pregnancies (i.e. women giving birth to more than one child) to the costs for high-risk and low-risk singleton pregnancies. Multiple pregnancies were found to be substantially more costly than low-risk single-child pregnancies, with antenatal costs nearly double the cost for low-risk singleton pregnancies, and the obstetric costs – due to 63 per cent of births being by caesarean section – £1,000 higher at £3,393 on average. However, women having high-risk singleton pregnancies as a result of pregestational diabetes were found to have slightly greater costs than those having multiple pregnancies on average.

Stillbirths

Stillbirths are a pregnancy complication associated with significant costs, both in terms of tangible and intangible costs. The direct costs of stillbirth have been estimated at $1,450–8,067, and it is estimated that costs are approximately 10–70 per cent greater for stillbirths compared to live births (Heazell et al.

2016). There are also additional costs from stillbirths associated with increased fetal monitoring in subsequent pregnancies, which cost approximately £558–1,735 more than pregnancies for women whose previous birth was an uncomplicated birth, as well as indirect costs from funerals and cremations and productivity losses.

Along with these costs, there are also significant costs to the NHS from post-mortem examinations and investigations that occur after stillbirth. Costs of post-mortem examinations and NHS investigations ranged from £1,242–1,804 per stillbirth, and the total costs of litigation and damages paid by the NHS from 2003–2007 – derived from 62 cases – were more than £1.76m (Mistry et al. 2013). In this time, the cost of care for subsequent pregnancies for women with previous stillbirths was over £15.1m (Mistry et al. 2013).

In a systematic review undertaken to better understand the economic consequences of stillbirths globally, Heazell et al. (2016) found that direct financial costs (including investigation into cause of death) varied from $1,450 to $8,067, and care costs associated with stillbirths were 10–70 per cent higher than care costs associated with livebirths. Moreover, the productivity of bereaved families was found to be significantly impacted, through absenteeism and presenteeism: 10 per cent of parents stayed off work for 6 months afterwards and productivity was greatly reduced when they did return, with as little as 26 per cent of normal productivity after 30 days, rising to 63 per cent after 6 months. Finally, families were also found to incur significant funeral and burial or cremation costs.

Stillbirth is also associated with intangible psychological and social costs such as grief, anxiety and fear, relationship breakdown, decreased care for surviving siblings, social isolation and presenteeism, and psychological effects on healthcare professionals (Heazell et al. 2016; Murphy and Cacciatore 2017; Ogwulu et al. 2015).

Perinatal mental health

Perinatal depression and anxiety are also a significant sources of cost in terms of pregnancy complications. The total lifetime costs of perinatal depression per patient are estimated to be £75,728, and the total lifetime costs of perinatal anxiety are £34,811, including costs such as health and social care costs, productivity losses, quality-of-life-related costs and behavioural and health costs for children. The majority of these costs are associated with negative impacts on children, with the minority of costs being associated with negative impacts on women. This translates to a cost of £8,500 for every woman giving birth in the UK, and an aggregated cost of £6.6 billion per year (Bauer et al. 2014).

4.2.2. Lifestyle and risk behaviours

With respect to the literature considering the costs associated with poor pregnancy outcomes related to lifestyle issues, two papers were identified on smoking and obesity. These papers find that women who smoke or are overweight or obese incur substantially higher healthcare costs during pregnancy. In their analysis of neonatal healthcare costs associated with smoking during pregnancy using secondary data from the US, Adams et al. (2002) found that maternal smoking increased the relative risk of newborns being admitted to a neonatal intensive care unit (NICU) by nearly 20 per cent. Maternal smoking was found to

add over $700 in neonatal costs, which amounts to a cost of almost $367m across the US (in 1996 US dollars).

Morgan et al. (2014) undertook a retrospective prevalence-based study to estimate the direct healthcare costs associated with being obese or overweight during pregnancy in Wales. After adjusting for maternal age, ethnicity and comorbidities, women who were overweight or obese had mean total costs respectively 23 per cent and 37 per cent higher than those not overweight. Adjusting for smoking and alcohol consumption did not significantly impact the results. The paper found that women of a healthy weight had a mean direct healthcare cost estimate of £3,546, whereas overweight and obese women had a mean cost estimate of £4,244 and £4,717 respectively.

In conclusion, the contextualisation work undertaken in the remainder of this chapter does not distinguish between health needs and health spend associated with poor pregnancy outcomes and those associated with standard pregnancy outcomes. The literature on costs associated with poor pregnancy outcomes highlights the types of poor pregnancy outcomes that exist, from low birth-weight to stillbirth, and demonstrates that there are significant economic costs associated with these outcomes.

4.3. Estimates of the costs to the NHS of pregnancy complications

Although the NHS does not separate costs associated with regular pregnancy care from costs associated with avoidable pregnancy complications, there is some literature available on the cost of pregnancy complications. We extracted information from relevant reviews, including those discussing the costs of pregnancy complications including perinatal depression, perinatal anxiety, stillbirth, preterm birth and the cost of different delivery modes and the cost of birth in different settings.

Although there is no widespread classification system that would allow for a direct comparison of costs and outcomes across different modes of delivery, there is some evidence that can provide cost estimates, which suggests that instrumental vaginal delivery and planned caesarean sections (CS) are more costly than uncomplicated vaginal deliveries (Fahy et al. 2013). For example, a 2001 study found that the costs of an uncomplicated vaginal delivery in the UK ranged from £629–1,298, compared to £1,512–4,337 for a planned caesarean section (Henderson et al. 2001: reviewed in Fahy et al. 2013). This difference in cost between delivery modes continues after the initial delivery. Also, a 2002 study in Scotland found significant differences between total costs of delivery modes for the first two months post-partum: £1,698 for spontaneous vaginal delivery, £2,262 for instrumental vaginal delivery and £3,909 for a planned CS (Petrou and Glazener 2002: reviewed in Fahy et al. 2013). There are also additional costs associated with certain pregnancy complications. The additional cost for an external cephalic version (compared to a normal spontaneous vaginal birth) was estimated to be £187 in a 2001 study, and the additional cost for an assisted breech delivery were estimated to be £425, although both of these were less costly than an elective CS for uncomplicated breech pregnancies (James et al. 2001: reviewed in Fahy et al. 2013). Table 10 provides an overview of some of the complications and conditions covered in the literature and the costs and impacts associated with them.

Table 10: Summary of costs and outcomes associated with complications of pregnancy based on a review of the literature.

Condition or outcome	Estimated costs	Other impacts not quantified	Review reference
Perinatal depression	Up to **£75,728** over lifetime of mother and child for costs associated with health and social care, suicide, health-related quality-of-life losses and productivity losses (for women), and preterm birth, infant death, emotional, intellectual and conduct problems (for children)		Bauer et al. 2014
Perinatal anxiety	Up to **£34,811** over lifetime of mother and child based on same costs as above.		Bauer et al., 2014
Stillbirth	**$1,450–8,067** (direct costs) **£558–1,735** (cost of increased fetal monitoring for subsequent pregnancy as compared to woman with previously uncomplicated birth) **£1,242–1,804** (post-mortem examinations and NHS investigations) **£1.76m** (62 cases of litigation and damages paid by the NHS 2003–2007) **£15.1m** (subsequent pregnancy care for women with previous stillbirth)	Indirect costs: funerals and cremations, productivity losses, presenteeism Psychological and social costs such as grief, anxiety. depression, social isolation and fear, relationship breakdown, decreased care for surviving siblings and psychological effects on healthcare professionals	Heazell et al. 2016 Mistry et al. 2013 Murphy and Cacciatore 2017 Ogwulu et al. 2015
Preterm birth	**$100,000** for early prematurity, **$10,000–30,000** for moderate prematurity **Less than $4,500** for late prematurity	Increased indirect costs from social and education services, lost productivity, home adaptations, informal carers and other non-medical costs Cerebral palsy, mental retardation, vision impairment, hearing loss Informal care provision, lost productivity for parents, psychological distress, anxiety and depression of parents and family members	Soilly et al. 2014 Petrou 2019 Frey and Klebanoff 2016
Different modes of delivery	Review found no internationally accepted childbirth cost/outcome classification for comparison across modes of delivery. Estimates from reviewed studies in the UK: Costs of an uncomplicated vaginal delivery in the UK ranged from **£629–1,298**, compared to **£1,512–4,337** for a planned caesarean section (CS) Costs of first 2 months post-partum: **£1,698** for spontaneous vaginal delivery, **£2,262** for instrumental vaginal delivery and **£3,909** for a planned CS Additional cost compared to normal spontaneous vaginal delivery: **£187** for external cephalic version and **£425** for assisted breech delivery		Fahy et al., 2013

Source: RAND Europe analysis of multiple sources as specified.

The NHS also accrues costs associated with litigation when pregnancy outcomes are poor. As described by Dickson et al. (2016), litigation costs associated with pregnancy are amongst the highest in terms of costs reported to the NHS Litigation Authority. It is highlighted that 'between 2000 and 2009, over 5,000 claims were made totalling £3.1 billion' (NHS Litigation Authority 2012, p.4). More recently, the litigation claims related to pregnancy were around fifty times the current pregnancy research spend, estimated at £2.5bn in 2018–19 (NHS Resolution 2019).

4.4. Contextualisation of pregnancy research funding, health needs and health spend

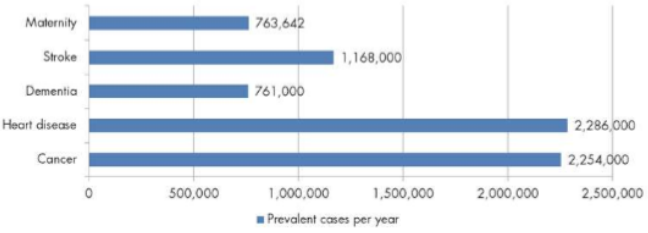
4.4.1. Health needs across the UK

In order to quantify pregnancy-related health needs in the UK, data were collected on the incidence of births and maternities annually across England, Wales, Scotland and Northern Ireland, as well as DALYs caused by pregnancy and pregnancy-related conditions.[60] Further information on the data collected and methods used to analyse them can be found in Section 2.2.

Condition prevalence

From 2013–2017 a total of 3.9 million births occurred, resulting from 3.8 million maternities across the UK, which translates to an average of 775,890 births per year resulting from 763,642 maternities per year. Figure 70 shows the average number of maternities annually over the five years from 2013 to 2017 compared to the prevalence of cancer, heart disease, dementia and stroke in 2012. The number of individuals in the population who suffer with dementia is similar to the number of women having maternities, while nearly three times the number of people have heart disease and cancer compared to maternities.

Figure 70: Prevalent cases of different health conditions, in millions.



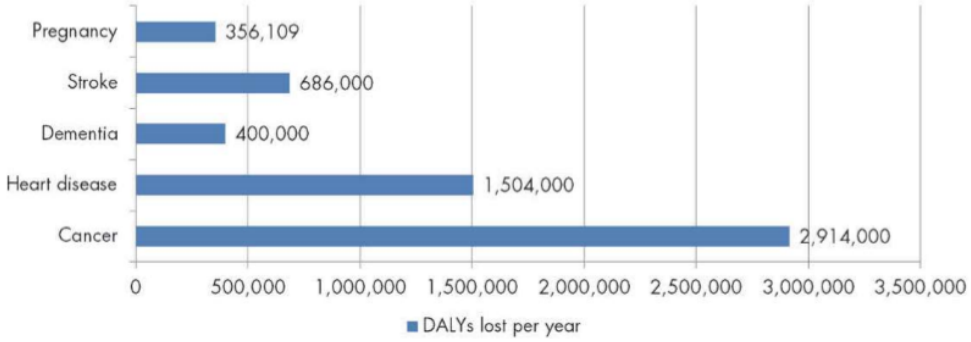
Source: RAND Europe analysis, building on work of Luengo-Fernandez et al. (2015).

[60] The World Health Organisation defines DALYs as: 'One DALY can be thought of as one lost year of "healthy" life. The sum of these DALYs across the population, or the burden of disease, can be thought of as a measurement of the gap between current health status and an ideal health situation where the entire population lives to an advanced age, free of disease and disability' (WHO 2019).

DALYs

A total of 1.8 million DALYs in the UK from 2013–2017 were attributable to pregnancy and pregnancy-related conditions (which includes both the mother and newborn child), the equivalent to 356,109 DALYs annually. DALYs attributable to pregnancy and pregnancy-related conditions accounted for 1.97 per cent of all DALYs during this period. Figure 71 highlights the average number of annual DALYs attributable to pregnancy and pregnancy-related conditions over the five years from 2013 to 2017 compared to the number of DALYs attributable to cancer, heart disease, dementia and stroke in 2012. A similar number of DALYs are attributable to both pregnancy and dementia each year, but considerably more are attributable to heart disease, and significantly more to cancer. It is worth noting that there are a number of limitations to the use of DALYs in this context. Firstly, many pregnancies have positive outcomes and/or are not subject to complications, so this metric does not capture or account for those cases. Secondly, the implications and costs (societal, and to the NHS) from poor pregnancy outcomes can be long-term and these longer term implications – on child health or long-term maternal health – will not be captured by this analysis.

Figure 71: DALYs attributable to different health conditions.



Source: RAND Europe analysis, building on work of Luengo-Fernandez et al. (2015).

4.4.2. Health spend across the UK

In order to quantify NHS spend on pregnancy and pregnancy-related conditions across the UK, programme budgeting data – which provide a framework for estimating NHS expenditure across different health conditions over the whole care pathway – were collected from numerous sources.[61] Further information on the data collected and methods used to analyse them can be found in Section 2.2.

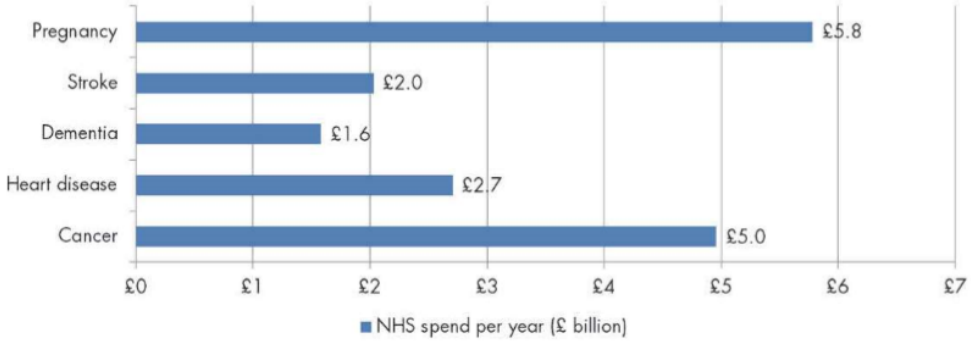
NHS expenditure

The NHS across the UK spent a total of £28.9bn on 'maternity and reproductive health' and 'neonates' from 2013–2017. On average this accounted for £5.8bn expenditure per annum, making up 4.62 per

[61] NHS Networks (2019).

cent of expenditure in the NHS across the whole of the UK. Figure 72 displays the average annual NHS expenditure on pregnancy and pregnancy-related conditions over the five years from 2013 to 2017 compared to the NHS expenditure on cancer, heart disease, dementia and stroke in 2012. The NHS spends more on pregnancy than any other condition considered, and considerably more than stroke, dementia and heart disease.

Figure 72: NHS spend on different health conditions, in £ billions.



Source: RAND Europe analysis, building on work of Luengo-Fernandez et al. (2015).

4.4.3. Research funding across the UK

In order to compare the funding for pregnancy-related research to research funding for other health conditions treated by the NHS across the UK, data were collected from a study considering different health conditions and levels of research funding for those conditions in the UK.[62] Furthermore, data on the total level of health research funding in the UK in 2014 were obtained from the UK Clinical Research Collaboration analysis of UK health research.[63] Further information on the data collected and methods used to analyse them can be found in Section 2.2.

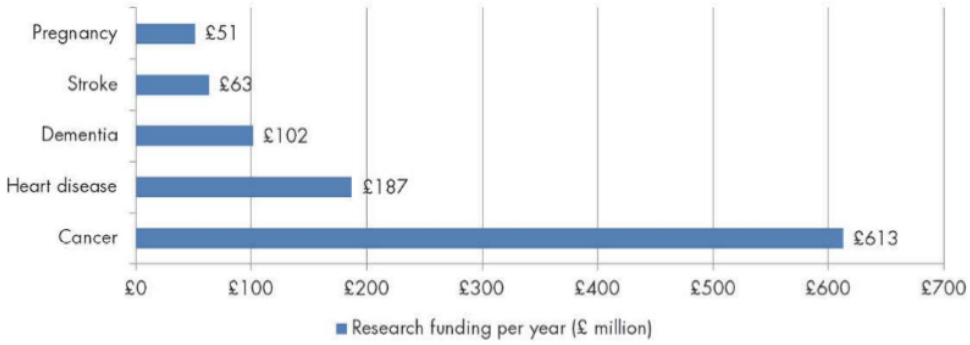
Overall research funding

A total of £255.1m was spent on pregnancy research from 2013–2017 in the UK, meaning an annual average spend during that period of £51m. Given that, according to HRA analysis, overall UK health research expenditure (direct, non-industry) in 2014 was £2.03bn, on average, funding for pregnancy research accounted for 2.4 per cent of funding for all health research annually.[64] Figure 73 shows the average annual level of research funding for pregnancy and pregnancy-related conditions over the five years from 2013 to 2017 compared to the research funding for cancer, heart disease, dementia and stroke in 2012. It is worth noting that research funding for pregnancy and pregnancy-related conditions does

[62] Luengo-Fernandez et al. (2015).
[63] UKCRC (2014).
[64] UKCRC (2014).

not include all infrastructure spending, whereas the other conditions do, as explained in Section 2.2. Pregnancy and stroke research receive a similar level of research funding, which is lower than dementia and heart disease and considerably lower than cancer research.

Figure 73: Research funding for different health conditions, in £ millions.

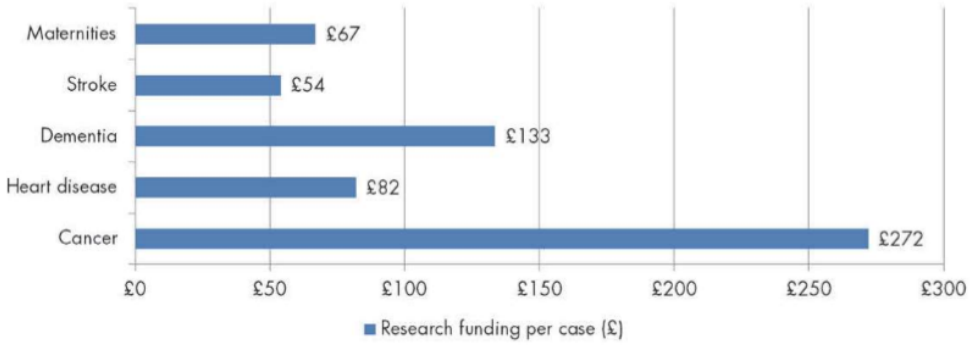


Source: RAND Europe analysis, building on work of Luengo-Fernandez et al. (2015).

Research funding and health need

Figure 74 highlights the average annual level of research funding per maternity over the five years from 2013 to 2017, compared to the research funding per case for cancer, heart disease, dementia and stroke in 2012. Pregnancy research receives less funding per case than dementia and heart disease research, and considerably less than cancer research, although more than stroke research per case.

Figure 74: Research funding per case for different health conditions, in £.

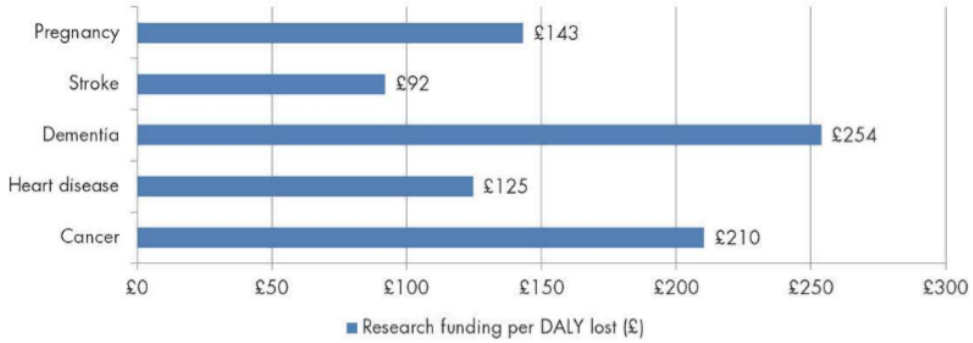


Source: RAND Europe analysis, building on work of Luengo-Fernandez et al. (2015).

The average annual level of research funding per DALY attributable to pregnancy and pregnancy-related conditions over the five years from 2013 to 2017, compared to the research funding per DALY attributable to cancer, heart disease, dementia and stroke in 2012 is shown in Figure 75. Unlike research funding per case, pregnancy research funding per DALY is relatively high, receiving a similar amount to

heart disease research, somewhat less than dementia and cancer research, and considerably more than stroke research per DALY.

Figure 75: Research funding per DALY for different health conditions, in £.

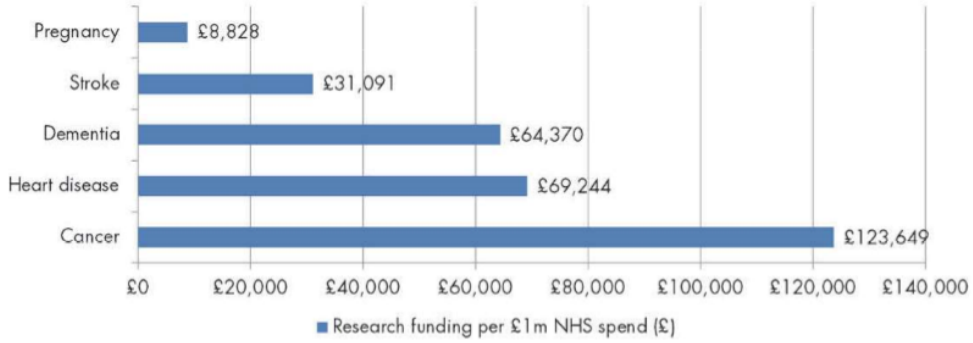


Source: RAND Europe analysis, building on work of Luengo-Fernandez et al. (2015).

Research funding and health spend

Figure 76 outlines the average annual level of research funding per £1m spent on pregnancy and pregnancy-related conditions in the NHS over the five years from 2013 to 2017, compared to the research funding per £1m spent on cancer, heart disease, dementia and stroke in the NHS in 2012. Pregnancy research receives considerably less funding per £1m spent by the NHS across the UK than stroke, dementia, heart disease and cancer funding per £1m spent.

Figure 76: Research funding for every £1m spent in the NHS for different health conditions, in £.



Source: RAND Europe analysis, building on work of Luengo-Fernandez et al. (2015).

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Appendix C. Paper 3

Disley, E., Gkousis, E., Hulme, S., Morley, K. I., Pollard, J., Saunders, C., Sussex, J. & Sutherland, A. (2021). **Outcome Evaluation of the National Model for Liaison and Diversion**. Santa Monica, CA: RAND Corporation. URL: https://www.rand.org/pubs/research_reports/RRA1271-1.html

An extract of the publication relevant to the economic analysis is presented here. Specifically:

- Summary and key findings
- 11. Summary of economic evaluation of the National Model for L&D
- 12. References
- Annex H. Complete economic analysis

EUROPE

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Outcome Evaluation of the National Model for Liaison and Diversion

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Preface

This document is the final report of an independent evaluation of the National Model for Liaison and Diversion (L&D). L&D schemes operate primarily in police custody suites and courts. They aim to identify and assess people with vulnerabilities as they pass through the criminal justice system, to ensure their health and other needs are known about and that they are referred to appropriate services for treatment or support.

The Liaison and Diversion Programme was created in 2010 to facilitate development of a model for commissioning L&D services, and to support existing L&D service providers. From 2011 to 2013, the Programme developed a national model for L&D services, with an initial focus on the diversion of people who are brought into police custody. In 2014 the *Liaison and Diversion Operating Model* was published by the Programme and NHS England launched a new National Model for L&D services.

This study of the National Model is funded by the National Institute for Health Research (NIHR) Policy Research Programme (PR-ST-0715-10013). The views expressed are those of the author(s) and not necessarily those of the NIHR or the Department of Health and Social Care. The research was undertaken by RAND Europe.

This report has been prepared for the Department of Health and Social Care and NHS England as the final output for the evaluation, but will also be of interest to policy makers and practitioners responsible for designing and implementing liaison and diversion services, as well as those commissioning such schemes. It is also of relevance to anyone designing future research or evaluations relating to L&D services.

RAND Europe is an independent, not-for-profit research institution. For further information about RAND Europe or this evaluation please contact:

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Summary and key findings

Background to Liaison and Diversion services

Liaison and Diversion (L&D) services aim to identify those in the criminal justice system (CJS) who have mental health needs and other vulnerabilities, refer them to appropriate support services, and ensure that information about those needs is available to decision-makers in the CJS.

L&D services have operated in some form for over 25 years, but their nature and quality varied between locations and there are many areas of the country where no L&D services were available. Between 2011 and 2013, the Department of Health was supported by an external partner, the Offender Health Collaborative, to develop a national L&D model. In 2014, NHS England launched the national *Liaison and Diversion Operating Model* for L&D services (NHS England Liaison and Diversion Programme 2014).

Following the launch of the National Model in 2014, it was implemented at ten trial sites in England, followed by a further 13 sites in 2015. In July 2016, funding was announced to expand implementation of the National Model further, with the goal of providing services to 75% of the country by April 2018.[1]

The National Model for L&D provides 24-hour, seven days per week services for people of all ages in the adult and youth justice pathways, covering a range of health issues and 'vulnerabilities'[2] including mental health, physical health and learning disabilities.[3] The intended outcomes of L&D services are fourfold:

- Improve access to healthcare and support services for vulnerable individuals and a reduction in health inequalities.
- Reduce reoffending or escalation of offending behaviours.
- Divert individuals, where appropriate, out of the youth and criminal justice systems into health, social care or other supportive services.
- Deliver efficiencies within the youth and criminal justice systems.

[1] https://www.england.nhs.uk/commissioning/health-just/liaison-and-diversion/news/programme-updates/ [accessed March 2020].

[2] 'Vulnerabilities' is the term used in the operating model and service specification.

[3] The National Model for L&D is contained in two documents: an operating model (NHS England Liaison and Diversion Programme 2014) and a service specification (Liaison and Diversion Programme 2014). The service specification is based on the operating model. The service specification has since been updated, most recently in 2019.

Evaluation of the National Model for L&D

Prior evaluations

In 2015, RAND Europe was commissioned by the then Department of Health to evaluate the impact of implementation of the National Model for L&D at the ten initial sites where it was established in 2014 (Disley et al. 2016). The evaluation involved consultation with key stakeholders, including L&D practitioners and team leaders, and members of the judiciary. Stakeholders were overwhelmingly positive about the National Model, particularly regarding its impact on identifying people in need of support (Disley et al. 2016).

The current evaluation focuses on the impact of the National Model for L&D on health services utilisation and criminal justice outcomes

RAND Europe was commissioned by the Department of Health and Social Care (DHSC) in 2016 to conduct an independent outcome evaluation of the National Model for L&D as implemented at 27 sites in England. The primary aim of the evaluation was to investigate the impact of implementation of the National Model for L&D on healthcare and criminal justice outcomes, addressing five research questions:

1. What impact does the National Model have on **health service utilisation** (in particular, use of A&E, mental health services and drug and alcohol treatment services)?
2. What impact does the National Model have on **reconviction**?
3. What impact does the National Model have on **diversion from the criminal justice system** (in particular, being diverted from prosecution, from court or from a custodial sentence)?
4. What impact does the National Model have on the **timeliness of court processes** (in particular, the number of adjournments, number of hearings per case, duration of a case and time from arrest to sentence)?
5. If any impacts on offending and health care utilisation are found, what is the **economic effect** of those?

A secondary aim was to understand the implementation of the National Model for L&D (as at the time when the impact evaluation was conducted), and how this may affect the outcomes achieved.

The current evaluation uses a novel administrative data linkage approach to overcome the challenges presented in evaluating the National Model for L&D

For this evaluation, we created a large-scale linked data set combining information from four separate healthcare sources (Hospital Episode Statistics Accident & Emergency, Mental Health Services Datasets, Improving Access to Psychological Therapies database, National Drug Treatment Monitoring System) and two separate criminal justice sources (Police National Computer, Her Majesty's Courts and Tribunals Service). To our knowledge this is the first example of such an extensive, cross-sector data linkage study. It enabled us to overcome the key challenges of evaluating the implementation of the National Model for L&D services, namely:

- The National Model was implemented before the evaluation was commissioned, so it was not possible to directly gather long-term baseline data from sites before the intervention was deployed. The linked data cohort provides extensive information on contact with health and criminal justice services both before and after referral to L&D services.
- During the evaluation period, the National Model was being rolled out across England, so there was no contemporaneous control group (i.e. without L&D services) and a randomised controlled design could not be used to assess causation. We used the longitudinal linked administrative data to create a 'historic' control cohort that enabled us to attribute causation, using data from an earlier time period before the implementation of the National Model of L&D.

To provide broader insight into the results from the linked data cohort we analysed two additional sources of information:

1. The internal programme monitoring data set (the 'Minimum Data Set', MDS), which provides insights into those reached by L&D services across the evaluation sites, and helps to understand overall implementation of the model.
2. An update of our previous process evaluation of the National Model via telephone interviews of staff from evaluation sites to aid interpretation of the quantitative data. This does not amount to a process evaluation, but it provides some insight into implementation of the National Model.

Multiple outcomes across the health and criminal justice domains were included in the evaluation

The linked administrative dataset was used to assess the impact of implementation of the National Model of L&D on a range of outcomes including:

- Accident & Emergency attendances
- Referrals and attendances at specialist mental health services
- Referrals and attendances at Improving Access to Psychological Therapies services
- Referrals and attendances at drug and alcohol treatment services, and data on quality of life and other wellbeing measures
- Offending, including offence type and outcome
- Court processes, including duration and number of hearings.

Data on these outcomes were available for an evaluation cohort of 8,729 service users referred to L&D services between January and September 2017 and followed-up for at least 12 months. The evaluation cohort was broadly representative of the wider population referred to L&D services during this period, although people from ethnic minority groups may be under-represented in this cohort. We also used these linked data to create an historic control group. To do this, we identified an historic offence for which 1,699 members of the evaluation cohort were charged during 2013/4, before the National Model of L&D was implemented, and used the data before and after this offence.

Strengths and limitations of the evaluation

As discussed above, the major **strength** of this evaluation is the creation of the novel, large-scale linked data set combining information from four healthcare sources and two criminal justice sources. This data set provides longitudinal information on the evaluation outcome measures that could not have been reliably obtained by other methods, and allows the creation of an historic cohort that provides a comparator for the evaluation cohort and enables attribution of causation.

There are four main **limitations** of our evaluation approach:

1. The sites included in the evaluation were not randomly selected and may not be representative of all current L&D services in England.
2. The use of an historic comparator group is not as robust as conducting a randomised controlled trial. As mentioned, the implementation approach for the National Model of L&D precluded the use of a randomised study design.
3. The quality of the administrative data available for research, and the processes used for linking these data, are not perfect.
4. The healthcare outcomes we evaluated were all related to referrals for treatment and subsequent attendance; we did not examine changes in the healthcare conditions that the treatment was intended to address.

Summary of key findings from the evaluation

L&D services following the National Model have succeeded in engaging with people who have a broad range of vulnerabilities

Overall, 88% of people referred to Liaison & Diversion services had at least one vulnerability identified. Almost three-quarters (71%) of those referred had a mental health need, and half (52%) experienced drug or alcohol misuse. Other vulnerabilities identified included risk of suicide or self-harm, unstable accommodation, being an abuse victim, financial needs, and needs relating to physical health, communication difficulties, or learning disabilities. Some service users had more than one vulnerability; for example, almost 20% were recorded as having more than one mental health need. This may present a challenge for L&D staff if multiple vulnerabilities are present, but none of these meet the criteria for referral. A quarter (26%), however, of people referred to L&D services had no prior service contacts recorded, indicating L&D services were potentially providing their first contact with health and social care services. This demonstrates that L&D services have accomplished the aim of engaging people with multiple vulnerabilities to facilitate access to support services.

L&D services appear to intervene at a point of crisis

Using the novel cross-sector linked data set we identified increased use of multiple healthcare services in the months leading up to the arrest that precipitated referral to an L&D service. The 6-12-month prior period to L&D referral is often characterised by a steep increase in contact with Accident & Emergency (A&E) services, specialist mental health services, and declining self-reported health in those attending drug

treatment services. However, L&D service users with and without previous criminal justice system contact were as likely to go to A&E after involvement with the L&D service. This suggests that for L&D service users it is acute health vulnerabilities that lead to contacts with the CJS, rather than the other way around. We also observed an increase in detentions under the Mental Health Act (MHA) (1983, amended 2007) in the six months prior to referral to an L&D service, suggesting that referral to L&D was not their first crisis-related contact with police for some service users. We did not observe such a marked increase in offending behaviour, except in the one to two months immediately prior to the arrest leading to L&D referral. This pattern of repeated health service contacts over an extended period suggests there may be a window of opportunity for intervention prior to L&D referral.

The interventions offered by L&D services vary by individual and by L&D site

Interventions provided by L&D services, and their uptake by service users, varies widely. Reported interventions spanned a wide spectrum, from advice and brief interventions, to primary care referral, to detention under the MHA for psychiatric assessment. Referral to health care services varied significantly between sites, particularly for the national Improving Access to Psychological Therapies (IAPT) service. Variability between sites is a planned aspect of the National Model as it is intended that referrals and other interventions are tailored to individual needs. Yet, insofar as this suggests variation in the *delivery* of the National Model, this represents an unplanned aspect of its implementation. Intervention uptake also varied, with people with substance use vulnerabilities in particular being more likely to decline L&D referral and interventions overall, despite being more likely to have multiple vulnerabilities. Results from the qualitative interviews indicated that timeliness of accessing onward referral services may contribute to variation in both provision and uptake; lengthy delays decrease the likelihood that service users engage with services, but interviewees reported that in some instances waiting times could be up to six months.

Following L&D referral, there is a short-term increase in referral to mental health services

Referral to L&D services appears to be followed by an increase in referral to IAPT services and non-L&D specialist mental health services. We did not, however, find evidence that these referrals translated into a substantial increase in face-to-face attendances in the post-L&D referral period. For IAPT services, there was no evidence for an increase in attendances. There may have been some increase in attendances at specialist mental health services, but due to how information was recorded in the data source, we could not differentiate between specialist mental health services, so this may just represent contact with L&D services.

Drug and alcohol treatment referral *and* attendance may increase following L&D referral

After L&D referral, drug and alcohol treatment service referrals appear to increase. A substantial minority of the drug and alcohol treatment service referrals were for individuals without previous recorded contact with these services. Additionally, referrals to drug and alcohol treatment services appear to translate into increased attendance at appointments, although this increase was not statistically significant.

Referral to L&D services does not appear to reduce offending

Overall, we did not find any evidence for an impact of referral to L&D services on offending behaviour in the post-L&D referral period. This lack of effect was at odds with the perceptions of L&D service staff

regarding the impact of the National Model. Our interview participants consistently identified that L&D services aimed to reduce offending by addressing the unmet needs of offenders through appropriate assessment and referral. This discrepancy between the information from the interviews and the overall data linkage analyses may partially be due to heterogeneity in the population targeted by L&D services, and the wide variation in interventions offered both within and between services. We also found, however, that (after accounting for other vulnerabilities) contact with the criminal justice system is not a predictor of healthcare utilisation after L&D referral, which **suggests that offending behaviour in this cohort is a symptom of a wider health and/or social problem that a single contact with a L&D service may not be sufficient to address.**

L&D services appear to increase diversion from custodial sentences

L&D services may reduce the proportion of offences resulting in custodial sentences and thus increase diversion from the criminal justice system. Considering changes in the proportion of offences resulting in a custodial sentence over time, the likelihood of service users receiving a custodial sentence after involvement with L&D services was almost half that of the historic control group. This estimate of impact of the National Model was statistically significant ($p = 0.05$). There was, however, no evidence for an impact of the National Model on the length of custodial sentences.

Court processes are not significantly affected by L&D services

The duration of court proceedings was reduced for offences committed at or just after referral to L&D services, but the available data do not provide sufficient statistical support to conclusively demonstrate an impact of the National Model on this outcome. There was no evidence for a statistically significant impact on number of hearings per court case. Following L&D referral, the time from first hearing to completion was almost 7 days shorter in the evaluation cohort compared to the historic control, but this difference was not statistically significant. The qualitative interviews, however, did provide additional support for the role L&D staff play in expediting court decisions via drafting on-the-day reports for pre-sentencing.

The L&D programme contributes to savings in the criminal justice system, but not in the healthcare system

L&D services appear to directly contribute savings of between £13.1 million and £41.5 million in the criminal justice system through diversion from custody and consequent increases in productivity. The economic analysis (see Box S1 for approach and limitations) hinges on custodial sentence length; when considering the diversion from custody based on average sentence length, L&D is associated with a total saving of £38.1 million, or £858 per L&D referral not declined. If we also consider increases in productivity due to avoidance of custody, then the savings increase to £933 per L&D referral (average sentence length) or £294 per L&D referral (median sentence length). We estimate the National Model of L&D costs £659 per L&D referral not declined. **Based on the average sentence length, the cost of L&D referral is thus more than offset by the £933 of savings due to diversion from custody**. These analyses do not incorporate costs or savings related to healthcare utilisation outcomes as the national L&D programme was

not found to have any statistically significant effect on attendance at A&E, (non-L&D) mental health specialist services, IAPT, or drug and alcohol treatments services.

Box S1 Approach and limitations of the economic evaluation

Approach

- Comparison of NHS England and NHS Improvement commission costs of the national L&D programme with the impacts on costs more widely and on benefits achieved.
- Cost-consequences analysis examining the effects on the health service and criminal justice system, as well as impacts in terms of service users' net contribution to the economy.
- Considers costs and benefits over a one-year period resulting from the national L&D scheme against the historic 'control' cohort identified from based on those charged with an offence three to four years prior to referral to L&D.

Limitations

- A cost-effectiveness analysis, where an overall measure of efficiency would be provided, was not possible given the available data.
- A lack of data meant that many potential costs and benefits could not be quantified in the economics analysis (see Chapter 11).
- As per the main evaluation analyses, these results are based on data from the first two waves of L&D implementation; findings may not generalise to all L&D services in England.

Evaluation conclusions

This evaluation used a novel, large-scale linked data set combining nationally-collected administrative data from both the healthcare sector and the criminal justice sector. This unique data set provided insights into contact with a range of services both before and after referral to a L&D service which could not have been reliably obtained in other ways.

Analyses conducted across these data sources consistently showed that **L&D services are successfully engaging with a group of service users with a broad range of vulnerabilities, often at a time of acute crisis when they are most in need to support**. There was, however, substantial variation between L&D services in the types of interventions offered, and referrals to healthcare services often did not translate into face-to-face contact with health service providers. This is likely to be due to a combination of factors including differences in service user needs at each site, variation in availability of services to which L&D staff can refer people, and length of waiting time for face-to-face appointments.

The **impact of the National Model for L&D on healthcare and criminal justice system outcomes appears to be focused on increasing referrals to mental health and drug and alcohol treatment services and diversion from custodial sentences**. It may also lead to an increase in attendances at drug and alcohol treatment services, and in the number of hearings per court case, but there was no evidence for a statistically significant impact on these outcomes in this study.

The evaluation identified only limited improvement in some outcomes after L&D referral, but there was **no evidence that outcomes became worse due to L&D referral, and we did not identify any unintended**

consequences of referral. Evaluating the programme is challenging due to heterogeneity in the implementation of the intervention and the population targeted. There are, however, some limiting factors that could be addressed to improve the overall impact of L&D services; most notably increasing capacity for onward referrals and developing approaches to support people who have multiple vulnerabilities but are not currently eligible for referral because no single vulnerability meets a required therapeutic threshold.

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Abbreviations

A&E	Accident and Emergency
BBV	Blood-borne viruses
CJS	criminal justice system
HES	Hospital Episode Statistics
HIV	Human immunodeficiency virus
HMCTS	HM Courts & Tribunal Service
IDU	Injecting drug use
L&D	Liaison and Diversion
MDS	Minimum Dataset
MHA	Mental Health Act
MHSDS	Mental Health Services Dataset
MoJ	Ministry of Justice
NDTMS	National Drug Treatment Monitoring System
NHS	National Health Service
PHE	Public Health England
PNC	Police National Computer

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11. Summary of economic evaluation of the National Model for L&D

11.1. Introduction and approach

The research team has undertaken an economic analysis of the impact of the national L&D scheme; comparing, as far as possible with the data available to the team, the costs of the national scheme with the costs, savings and other impacts it has had. Where appropriate, we have used the data for a historic cohort of individuals as a comparator. This historic cohort was identified from among the evaluation cohort, who had also been charged with an offence three to four years prior to their L&D referral.

We have adopted a societal perspective for the analysis. We have thus considered the scope for impacts on all groups of stakeholders: the individuals receiving the L&D services, their families, L&D service providers, the CJS, health services and the economy in general. Table 11.1 lists the potential costs and impacts foreseen, grouped by stakeholder type.

Limitations to the data available and consequently the analyses possible, mean that we focus on the impacts detectable in the year following L&D referral, and are able to present quantified evidence on only a subset of those categories. In essence, we conduct a cost-consequence analysis taking explicit account of the items indicated in Table 11.1. Unfortunately, no data were available to enable us to estimate the impact of L&D on the health and well-being of individuals or their families.

The analysis draws on the data described elsewhere in this report and on data from a targeted review of the literature. Full details of the economic analysis are in Annex H. The main points from it are summarised in this chapter.

Table 11.1: Potential costs and benefits associated with the L&D scheme that were considered in the economic evaluation. Costs and benefits are grouped by thematic area with an indication of whether these could be included in the analysis based on available data.

Thematic area	Costs/benefits	Included
Individual		
Health and well-being	Psychological health of individual	
	Physical health of individual	
	Quality of life of individual	
	Psychological health of individual's family	
	Quality of life of individual's family	
Service providers		
L&D service costs	Capital costs; e.g. installing NHS servers	
	Revenue costs; e.g. staffing costs	X
	In-kind costs; e.g. facilities provided by police/courts	
Criminal justice system		
	Offending rates	X
Efficiencies in the police system	Number of arrests	
	Processing of detainees in police custody	
Timeliness of court processes	Number of hearings	X
	Time from arrest to completion of case	X
	Arrest to laying of information	X
	Laying of information to first hearing	X
	First hearing to completion	X
	Number of psychiatric and psychologist reports required	
Diversion from criminal justice service	No further action	
	Diversion from prosecution	
	Diversion from custody	X
Health services		
Primary care utilisation		
Secondary care utilisation	Accident & Emergency utilisation	X
	Mental health service utilisation	
	IAPT utilisation	X
	Drug and alcohol treatment utilisation	X
	Learning disability services utilisation	
	Other NHS utilisation	
	Health service access inequality	
Wider society		
	Productivity	X

Source: RAND Europe analysis

11.2. Findings from the economic analysis

Table 11.2 brings together in one place the various monetised impacts of the national L&D scheme that we were able to estimate. All monetary values are given in 2018/19 prices. We express costs both in total and per person who received L&D services (that is, people who were referred to L&D and did not decline that referral). In our analysis, we estimate 44,469 L&D referrals taken up, over a 12-month period.[7]

We find that the national L&D scheme costs a total of £29.31 million reported to us by NHS E&I, which equates to £659 per L&D referral not declined. The national L&D scheme may also be associated with a further total cost of £21.55 million, or £485 per L&D referral not declined, due to additional court hearings. This impact was not significant at conventional levels (p=0.1), and so could be ignored. We report it here, however, as the low p-value suggests at least that the possibility of there being additional court hearings as a result of L&D could be worth further research in future if additional data become available.

Table 11.2: Summary of L&D economic impacts

Costs (shown as negative) and benefits (positive)	Total	Per L&D referral not declined
Service providers		
L&D service costs	- £29.31m	- £659
Criminal justice system		
Timeliness of court processes		
Number of hearings*	*(- £21.55m)*	*(- £485)*
Diversion from criminal justice service		
Diversion from custody, based on median/mean sentence length avoided	£12.03m/£38.14m	£270/£858
Health services		
Secondary care utilisation		
A&E utilisation	£0	£0
IAPT utilisation	£0	£0
Drug and alcohol treatment utilisation	£0	£0
Wider societal		
Productivity, based on median/mean sentence length avoided	£1.06m/£3.37m	£24/£76

Source: RAND Europe analysis

*Not statistically significant at conventional levels but p=0.1

The national L&D scheme appears to be associated with some savings to other parts of the criminal justice system. The national L&D scheme was found to have a statistically significant impact on the likelihood of receiving a custodial sentence, reducing the probability by 45% in the 12-month post-L&D period. Police National Computer (PNC) data shows that across the evaluation cohort 14.5% of offences in the 12-month pre-L&D period resulted in custodial sentences, suggesting that a total of (44,469x0.145=) 6,448

[7] We estimated the number of referrals taken up over a 12-month period as we only recruited participants over a nine-month period, so extrapolated referral and uptake rates from this data set.

individuals would have received a custodial sentence without accepting a referral to L&D. A 45% reduction in this in the 12-month post-L&D period equates to 2,902 fewer individuals receiving a custodial sentence as a result of the national L&D scheme.

Data on the lengths of custodial sentences shows that they are subject to a highly skewed distribution, with a large proportion of short sentences but a substantial 'tail' of much longer sentences. As a result, the median sentence length is much shorter than the mean sentence length. We therefore illustrate the impact not only of assuming that the custodial sentences avoided are of mean length but also the more modest impact if it is more conservatively assumed that the custodial sentences avoided would have been of median length. PNC data shows that the median length of a custodial sentence for the offence leading to L&D referral is 70 days, whereas the mean is 222 days. People, however, are generally released from prison halfway through their custodial sentence, so we have assumed that only half of this time would have been served in prison (Beard 2020).

When considering the mean diversion from custody, L&D is associated with a total saving of £38.14 million, or £858 per L&D referral not declined. When considering the median diversion from custody, L&D is associated with a total saving of £12.03 million, or £270 per L&D referral not declined.

The national L&D scheme was not found to have any statistically significant effect on the quantifiable health service utilisation outcomes: neither A&E attendances, nor IAPT utilisation, nor use of drug and alcohol treatments services.

As explained above, L&D appears to result in 2,902 fewer custodial sentences per year amongst those not declining their referral. The post-April 2016 MHSDS suggests that 16.7% of the evaluation cohort are employed, suggesting that (2,902x0.167=) 485 employed individuals avoided custody because of the national L&D scheme. On the basis of the mean length of a custodial sentence served (222 days), and assuming that on average half of a sentence is served in custody, 485 employed individuals avoiding serving on average 111 days in custody implies that 53,835 additional days are worked because of the national L&D scheme. Assuming a typical working day is eight hours long and is paid at the national minimum wage of £7.83 per hour,[8] this suggests that £3.37 million of productivity is generated through wages that otherwise would not have been earned, or £76 per L&D referral not declined. If the more conservative median length of a custodial sentence served (70 days) is assumed, then the productivity gains from the national L&D scheme are correspondingly more modest, at £1.06 million of productivity generated through wages that otherwise would not have been earned, equivalent to £24 per L&D referral not declined.

The national L&D scheme thus appears to be directly contributing to savings in the CJS, through diversion from custody, as well as enabling greater levels of productivity – again due to diversion from custody. Assuming mean levels of sentence length avoided, and omitting cost impacts that are not statistically significant, we have found that the L&D service cost of £659 per L&D referral is more than offset by the £933 savings due to diversion from custody (£857+£76). If, however, we were to assume only median levels of sentence length being avoided, the L&D service costs of £659 per L&D referral not

[8] HM Government, n.d.

declined, would exceed the £294 savings due to diversion from custody (£270+£24). If in reality L&D does lead to additional court hearings, then the overall net impact would be worsened accordingly.

In conclusion, the national L&D scheme has a significant and direct impact on certain areas of the CJS and wider society, but a lack of data meant that many of the potential costs and benefits could not be quantified in the economic analysis.

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Annex H. Complete economic analysis

H.1. Introduction

The objective of this economic analysis is to compare the NHS England and NHS Improvement (NHS E&I) commissioning costs of the national L&D scheme with the impact on costs more widely and on benefits achieved. We take a societal approach, considering the year following L&D referral.

Specifically, the economic analysis compares the costs and benefits resulting from the national L&D scheme against a historic 'control' cohort, identified from among the evaluation cohort, who were charged with an offence three to four years prior to their L&D referral. As discussed in detail in Annex C, this was the most appropriate approach to produce as robust a counterfactual as possible given the absence of a randomised controlled trial and in view of the data available to the research team.

We were able to undertake a cost-consequence analysis, where the costs and outcomes are clearly listed and compared, but not combined into a single measure of efficiency. A cost-effectiveness analysis, where an overall measure of efficiency would be provided, was not possible given the available data. In summary, the economic analysis compares the costs of introducing the national L&D scheme to NHS E&I to any benefits obtained. All values in this chapter are given in 2018/19 prices.

H.2. Method

H.2.1. Economic Framework

As recommend in the HM Treasury Green Book,[14] a long-list of potential costs and benefits associated with the national L&D scheme was drawn up, using a societal approach to account for the cross-cutting nature of the intervention. We clustered the categories of costs and benefits into thematic areas, as follows and set out in Table H.29 with further details in following paragraphs:

- Individual level
- L&D service provider level
- Criminal justice system level
- Health services level
- Wider societal level.

[14] HM Treasury (2013).

Table H.29. Potential costs and benefits associated with the L&D scheme that were considered in the economic evaluation. Costs and benefits are grouped by thematic area with an indication of whether these could be included in the analysis based on available data.

Thematic area	Costs/benefits	Included
Individual		
Health and well-being	Psychological health of individual	
	Physical health of individual	
	Quality of life of individual	
	Psychological health of individual's family	
	Quality of life of individual's family	
Service providers		
L&D service costs	Capital costs; e.g. installing NHS servers	
	Revenue costs; e.g. staffing costs	X
	In-kind costs; e.g. facilities provided by police/courts	
Criminal justice system		
	Offending rates	X
Efficiencies in the police system	Number of arrests	
	Processing of detainees in police custody	
Timeliness of court processes	Number of hearings	X
	Time from arrest to completion of case	X
	Arrest to laying of information	X
	Laying of information to first hearing	X
	First hearing to completion	X
	Number of psychiatric and psychologist reports required	
Diversion from criminal justice service	No further action	
	Diversion from prosecution	
	Diversion from custody	X
Health services		
Primary care utilisation		
Secondary care utilisation	Accident & Emergency utilisation	X
	Mental health service utilisation	
	IAPT utilisation	X
	Drug and alcohol treatment utilisation	X
	Learning disability services utilisation	
	Other NHS utilisation	
	Health service access inequality	
Wider society		
	Productivity	X

Source: RAND Europe analysis

IAPT indicates Improving Access to Psychological Therapies services

Individual

L&D is expected to have a positive impact on **individual** health and well-being outcomes. The service is expected to divert appropriate individuals away from the criminal justice system and into the health service, which should, in turn, improve their **physical and psychological health and quality of life**. As a result, L&D can also be expected to improve the **psychological health and quality of life of service user's direct family members**.

Service providers

L&D is likely to be associated with considerable **service provider** costs, which can be summarised into three categories:

1) Upfront **capital costs** to enable provision of the national L&D scheme, such as the instillation of NHS servers in numerous police stations to access health service records on site.
2) Ongoing **revenue costs**, such as the employment of additional L&D staff to deliver the services.
3) **In-kind costs** where no monetary transaction occurs but goods and services have to be provided to support the national L&D scheme, e.g. existing facilities at police stations and courts being made available to L&D staff.

As noted above, we are comparing the costs and benefits of the national L&D scheme against a historic cohort, identified from the evaluation cohort, who were charged with an offence three to four years prior to their L&D referral. As the national L&D scheme is being compared to a historic cohort with an arrest in the period three to four years prior to L&D referral, some of this historic cohort may have had *local* L&D services provided to them, although we have no information about that. This economic evaluation, however, is considering the benefits achieved by the NHS E&I-commissioned *national* L&D scheme, so the cost of any local L&D services already provided to the historic cohort are not considered.

Criminal justice system

L&D is hypothesised to impact the **CJS** in numerous ways. The diversion of appropriate individuals away from the criminal justice system and into the health service should result in reduced **offending rates** as individuals receive appropriate care, which would be expected to reduce offending. Applying this same logic, it would be expected that the **number of arrests** should reduce after individuals have been referred to L&D.

L&D is also expected to improve the **processing of detainees in police custody**, i.e. reducing time and dedicated resource, as police services benefit from having a qualified health practitioner on site. Moreover, the information collated by the L&D service and provided to courts is expected to help reduce the need for adjournments and hence the **number of hearings** per case. Such L&D service information should also result in fewer **psychiatric and psychological reports** being requested by courts before decisions can be made in the system. As a result of the information collated by the L&D services potentially reducing the number of hearings and requests for psychiatric and psychological reports, this should in turn reduce **time from arrest to completion** of cases.

The diversion of appropriate individuals away from the criminal justice system and into the health service should increase diversion from criminal justice services, as **no further action** would then be taken on individuals who are deemed to require health services rather than criminal punishment, which should also see an increase in **diversion from prosecution and custody**.

Health services

L&D is expected to have differing impacts on various parts of the health service. L&D may have a positive or a negative impact on **primary care utilisation**. On the one hand, individuals may be directly referred to GPs through the scheme while, on the other hand, individuals may be diverted away from primary care to more appropriate specialised care, e.g. drug and alcohol treatment services.

L&D is hypothesised to have a varied impact on secondary care utilisation. Firstly, it is expected to reduce **A&E utilisation** as individuals are diverted to appropriate health services and are therefore receiving treatment outside of A&E. L&D, however, may be expected to increase **mental health, IAPT, drug and alcohol, and learning disability service utilisation**, as L&D seeks to divert appropriate individuals into the health service. As a result, it is also unclear what net impact L&D might have on the **utilisation of other NHS** services.

It is anticipated that the diversion of appropriate individuals away from the criminal justice system and into the health service may reduce **health service access inequality**, as some vulnerable individuals will be accessing health services who otherwise would not have done.

Wider society

It is anticipated that L&D services will have a positive impact at the wider societal level. **Productivity** in the economy will be directly impacted if L&D leads to an increase in diversion from custody, as employed individuals may be able to maintain their job and continue to earn a wage. **Productivity** may also be impacted indirectly if L&D improves individual outcomes (e.g. physical and psychological health), as this may lead to reductions in absenteeism and presenteeism among employed individuals.

H.2.2. Quantification and monetisation

It was then considered whether each item in Table 1 was quantifiable based on the data available to us, and if so whether a monetary value could be assigned to it from our available data or the wider literature. The research team undertook a targeted search of the literature to identify as many monetary values as possible for items that were quantifiable. Where items in the economic framework could be quantified, the impact of the national L&D scheme was considered up to one year after the intervention (the follow-up data available to the research team only covered this time period).

In this way the research team identified several service provider, criminal justice system, health service and wider societal outcomes and costs that could be both quantified and monetised, as outlined below.

- Service providers:
  - National L&D scheme commissioning costs.
- Criminal justice system:
  - Reoffending rates
  - Number of hearings
  - Diversion from custody.
- Health services:
  - A&E utilisation
  - IAPT utilisation

- Drug and alcohol treatment utilisation.

- Wider society:
  - Productivity.

No quantified data were available for the health and well-being impacts on individuals or their families.

Table H.30 outlines which of the items were quantifiable and able to be assigned monetary values, with the sources for each provided in column two and column three respectively. The following paragraphs then explain in more detail

Table H.30: Data sources for quantifying and costing each item

Cost/Benefit	Quantification*	£ value**
Service providers		
L&D service costs	NHS England	NHS England[15]
Criminal justice system		
Offending rates	PNC	'The economic and social costs of crime' second edition[16]
Timeliness of court processes		
Number of hearings	HMCTS	Cost of a day in court[17]
Diversion from criminal justice service		
Diversion from custody	PNC	Cost per prison place and cost per prisoner[18]
Health services		
Secondary care utilisation		
A&E utilisation	HES A&E	NHS Reference Costs[19]
IAPT utilisation	IAPT	'Services for people with mental health problems'[20]
Drug and alcohol treatment utilisation	NDTMS	'Services for people who misuse drugs or alcohol'[21]
Wider society		
Productivity	PNC	'National Minimum Wage & Living Wage rates'[22]

Source: RAND Europe analysis

*Ability to quantify impact of national L&D scheme on item with data available to research team

**Ability to cost item with data available to research team

[15] Commissioning costs of the national L&D scheme were provided to the research team by NHS E&I

[16] Heeks et al. (2018).

[17] Law Society (2018).

[18] MoJ (2019).

[19] NHS England (2020).

[20] Curtis & Burns (2018).

[21] Curtis & Burns (2018).

[22] HM Government, n.d.

Service providers

Overall **L&D service costs** were provided to the research team by NHS E&I. More specifically, commissioning costs of the national L&D scheme were used as a proxy for service costs as this was the only data available. No data were available on the capital/revenue cost split, and no in-kind cost data were available. There are two important caveats to note regarding the national L&D scheme service provider costs, which are given by site in NHS E&I provided the research team with data on the cost of commissioning the national L&D scheme by site, as shown in Table H.32. In some instances, the service provider costs were provided by NHS E&I aggregated across more than one site, e.g. Northumbria and Sunderland combined. Some sites' costs were missing and so values had to be imputed for Oxfordshire and North East London, as explained in detail in the Methods.

Overall, the total L&D service provider costs were £29.31 million – or £659 per L&D referral not declined:

- £29,311,000 service cost ÷ 44,469 cases = £659 per L&D referral not declined
- .

First, no cost data were available for North East London. A value was estimated for that site based on the cost of the West London site and the proportion of L&D referrals not declined in North East London compared to West London. West London was deemed to be the most appropriate proxy for North East London costs as both sites are based in Greater London. The proportion of L&D referrals not declined in North East London compared to West London was used to weight the costs to account for differences in service activity across the two sites.

Second, cost data were provided by NHS E&I for Oxfordshire and Berkshire combined and we do not have referral information for Berkshire. Half of the combined cost was assigned to Oxfordshire. This was deemed the most appropriate approach after consultation with NHS E&I, given the data available.

Criminal justice system

Offending rates are quantifiable using PNC data, measured as rate of offending per person per month, and can be costed using 'The economic and social costs of crime'.[23]

No data were available to enable us to assess the impact of L&D on numbers of arrests or on the costs of processing by the police of detainees.

Timeliness of court processes is quantifiable using HMCTS data, and the **number of hearings** can be costed using 'Cost of a day in court';[24] this is assuming that a hearing takes half a day of court time on average, i.e. four hours, as outlined by the Institute for Government.[25]

Diversion from custody is quantifiable using PNC data, measured as the proportion of offences resulting in a custodial sentence, which can be costed using 'Costs per prison place and costs per prisoner'.[26] The average length of a custodial sentence among the evaluation cohort is also captured in the PNC data.

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[23] Heeks et al. (2018).

[24] Law Society (2018).

[25] Institute for Government (2019).

[26] MoJ (2019).

Health services

Data to enable estimation of the impact of L&D on use of primary care services were not available, nor was it possible with the data available to quantify the impact of L&D on reducing inequalities of access to health services.

A&E utilisation is quantifiable using NHS 'Hospital episode statistics (HES)'[27] A&E data, captured as A&E utilisation per person per month, and can be costed using 'NHS reference costs'.[28]

IAPT utilisation is quantifiable using IAPT data, captured as attended face-to-face IAPT appointments per person per month, and can be costed using Personal Social Services Research Unit (PSSRU) unit costs on 'Services for people with mental health problems'.[29]

Drug and alcohol treatment utilisation is quantifiable using NDTMS data, measured as attendances per person per month, which can be costed using PSSRU unit costs on 'Services for people who misuse drugs or alcohol'.[30]

Wider society

Productivity in the UK economy will be directly impacted if L&D leads to an increase in diversion from custody, as more individuals will be able to earn a wage, which is quantifiable assuming eight hours are worked a day and can be costed using the 'National Minimum Wage & Living Wage Rates'.[31]

H.2.3. Unit of the economic analysis

The unit of analysis for the economic evaluation is 'per L&D referral not declined' as we are interested in the impact of the national L&D scheme on those who actually received the service. The MDS captures the number of L&D referrals during our evaluation window (1 January 2017 to 30 September 2017) across the 27 sites included in our evaluation, shown in Table H.31; this excludes those without a referral date, those without a specified male or female gender, and those born in 2000 or later as we are only interested in adult services. These data most accurately represent the number of individuals receiving the intervention in our evaluation window.

Table H.31. Number (%) of individuals referred to L&D services during evaluation window

Time period	L&D referrals	L&D referrals not declined	L&D referrals declined
Nine-month evaluation window	45,323	33,352 (73.6%)	11,971 (26.4%)
12-month equivalent	60,431	44,469 (73.6%)	15,961 (26.4%)

Source: MDS & RAND Europe analysis

[27] NHS Digital (2020).

[28] NHS Improvement (2018).

[29] Curtis & Burns (2018).

[30] Curtis & Burns (2018).

[31] HM Government, n.d.

MDS referral data was scaled up from nine to 12 months, assuming a consistent rate of referrals for the final 3 months, to ensure it is comparable with the national L&D scheme commissioning costs, which were provided as annual values. Table H.31 shows that 26.4% of individuals referred to L&D during the evaluation window declined the referral, with 73.6% not declining and therefore assumed to receive L&D services.

H.3. Results

The following sections present the estimated monetised impacts of L&D. All of the estimates are brought together in a summary table at the end – Table H.33.

H.3.1. Service providers' costs

NHS E&I provided the research team with data on the cost of commissioning the national L&D scheme by site, as shown in Table H.32. In some instances, the service provider costs were provided by NHS E&I aggregated across more than one site, e.g. Northumbria and Sunderland combined. Some sites' costs were missing and so values had to be imputed for Oxfordshire and North East London, as explained in detail in the Methods.

Overall, the total L&D service provider costs were £29.31 million – or £659 per L&D referral not declined:

- £29,311,000 service cost ÷ 44,469 cases = £659 per L&D referral not declined

Table H.32. L&D service provider costs by site (£'000)*

Site name	Service cost (£'000)
Avon & Wiltshire	1,294
Barnsley	291
Black Country/Dudley & Warsaw	1,513
Cleveland	
Durham	
Middlesbrough	1,821
Cornwall & Devon	1,488
Coventry	664
Doncaster & Rotherham	504
Dorset	762
Hampshire	1,371
Lancashire	1,381
Leicestershire	711
Liverpool	1,972
NE London	*2,433*
Norfolk & Suffolk	2,172
Northamptonshire	670
Northumbria	
Sunderland	1,668
Nottingham	1,142
Oxfordshire	*1,446*

Sheffield	412
Somerset	596
South Essex	923
Sussex	1,394
Wakefield	414
West London	2,269
TOTAL	**29,311**

Source: NHS E&I

*Imputed or estimated costs presented in *italics*

H.3.2. Criminal justice system costs and savings

Offending rates

The national L&D scheme does appear to have a statistically significant impact on the probability of offending in the 12-month post-L&D period in the evaluation cohort compared to the historic cohort. This result, however, is an artefact of the study design, as members of the historic cohort must necessarily have committed future offences to be included in the evaluation cohort, meaning it is not an appropriate comparator for analyses of offending. As a result, it was not possible to include an analysis of the impact of L&D on offending rates.

Number of hearings

The national L&D scheme was found not to have a statistically significant impact on the probability of there being more than one hearing per case in the 12-month post-L&D period in the evaluation cohort compared to the historic cohort.

There may, however, be a positive association between L&D and the probability of having more than one hearing per case, with an average increase in probability of 36% in the year post-L&D referral (p=0.11). It is important to note that this effect is not significant at conventional levels.

A 36% increase in the probability of more than one hearing would suggest that 16,009 individuals have at least one additional hearing in their case:

- 0.36 higher probability of >1 hearing × 44,469 cases = 16,009 cases with an additional hearing.

Assuming everyone with an additional hearing has a total of one additional hearing, which takes on average half a day of court time,[32] this results in a total additional cost of £21.55 million, or £485 per L&D referral not declined:

- 16,009 cases with an additional hearing × 0.5 days x £2,692 cost of a court day = £21,548,114
- £21,548,114 cost of additional hearings ÷ 44,469 cases = £485 per L&D referral not declined.

Diversion from custody

The national L&D scheme was found to have a statistically significant impact on the likelihood of receiving a custodial sentence, reducing the probability by 42% (OR 0.58, 95% confidence interval 0.46-0.74) in

[32] Institute for Government (2019).

the L&D period and by 45% (OR 0.55, 95% confidence interval 0.43-0.70) in the 12-month post-L&D period. The overall impact of L&D on the likelihood of receiving a custodial sentence during or after the L&D period has a p-value of 0.05.

PNC data show that across the evaluation cohort 14.5% of offences in the 12-month pre-L&D period resulted in custodial sentences, suggesting that a total of 6,448 individuals would have received a custodial sentence without accepting a referral to L&D:

- 0.145 probability of custodial sentence × 44,469 cases = 6,448 expected custody cases.

L&D was found to significantly reduce the probability of receiving a custodial sentence by 45% in the in the 12-month post-L&D period, suggesting 2,902 fewer individuals received a custodial sentence as a result of the national L&D scheme:

- 0.45 lower probability of custody × 6,448 expected custodial sentence cases = 2,902 fewer custody cases.

Data on the lengths of custodial sentences show that they are subject to a highly skewed distribution, with a large proportion of short sentences but a substantial 'tail' of much longer sentences. As a result, the median sentence length is much shorter than the mean sentence length. We therefore illustrate the impact not only of assuming that the custodial sentences avoided are of mean length but also the more modest impact if it is more conservatively assumed that the custodial sentences avoided would have been of median length.

PNC data show that the median length of a custodial sentence for the offence leading to L&D referral (n=664) is 70 days, whereas the mean is 222 days, due to small numbers of offenders receiving long sentences. People, however, are generally automatically released from prison halfway through their custodial sentence, so it was assumed only half of this time was served in prison.[33] Note that we have excluded sentences longer than 15 years to avoid skewing the data, as this was the mean sentence length of murder/manslaughter assumed in the evaluation of the L&D trial schemes, based on correspondence with the MoJ.[34]

Considering the more conservative, 70-days, median length of a custodial sentence served; 2,902 individuals avoid serving, on average, 35 days in custody, suggesting that 101,570 fewer days are served in prison because of the national L&D scheme:

- 2,902 fewer custody cases × 35 days in custody = 101,570 fewer days served in prison.

Combining this information with the cost of a prison place, which is £43,213 per year or £118.39 per day,[35] the national L&D scheme results in a saving of £12.03 million, or £270 per L&D referral not declined.

- 101,570 fewer days served in prison × £118.39 cost per prisoner per day = £12,025,053
- £12,025,053 saving from fewer prison days ÷ 44,469 cases = £270 per L&D referral not declined.

[33] Beard (2020).

[34] Disley et al. (2016).

[35] MoJ (2019).

Considering the rather longer, 222-day, mean length of a custodial sentence served; 2,902 individuals avoid serving on average a 111-day custodial sentence, suggesting that 322,122 fewer days are served in prison because of the national L&D scheme:

- 2,902 fewer custody cases × 111 days in custody = 322,122 fewer days served in prison.

Combining this information with the cost of a prison place, which is £43,213 per year or £118.39 per day,[36] the national L&D scheme results in a saving of £38.14 million, or £858 per L&D referral not declined.

- 322,122 fewer days served in prison × £118.39 cost per prisoner per day = £38,136,597
- £38,136,597 saving from fewer prison days ÷ 44,469 cases = £858 per L&D referral not declined.

H.3.3. Health services' costs and savings

A&E utilisation

The national L&D scheme was found to have no statistically significant impact on A&E utilisation.

IAPT utilisation

The national L&D scheme was found to have no statistically significant impact on IAPT utilisation.

Drug and alcohol treatment utilisation

The national L&D scheme was found to have no statistically significant impact on drug and alcohol treatment utilisation.

H.3.4. Wider societal impacts

Productivity

As explained in the above discussion of diversion from custody, L&D appears to result in 2,902 fewer custodial sentences per year amongst those not declining their referral. The post-April 2016 MHSDS data (n=2,738) suggests that 16.7% of the evaluation cohort are employed, suggesting 485 employed individuals avoided custody because of the national L&D scheme:

- 0.167 probability of being employed × 2,902 fewer custody cases = 485 employed cases avoiding custody.

Considering the more conservative median length of a custodial sentence served; 485 employed individuals avoid serving on average 35 days in custody, suggesting that 16,975 additional days are worked because of the national L&D scheme:

- 485 employed cases avoiding custody × 35 days in custody = 16,975 additional days worked.

Assuming a typical working day is eight hours long and is paid at the national minimum wage is, which is £7.83 per hour for those aged 25 years and over,[37] this suggests £1.06 million of productivity is generated through wages which otherwise would not have been earned, or £24 per L&D referral not declined:

- 16,975 additional days worked × 8 hours × £7.83 minimum hourly wage = £1,063,314

[36] MoJ (2019).

[37] HM Government, n.d.

- £1,063,314 productivity generated ÷ 44,469 cases = £24 per L&D referral not declined.

Considering the rather higher mean length of a custodial sentence served; 485 employed individuals avoid serving on average 111 days in custody, suggesting that 53,835 additional days are worked because of the national L&D scheme:

- 485 employed cases avoiding custody × 111 days in custody = 53,835 additional days worked.

Assuming a typical working day is eight hours long and is paid at the national minimum wage, which is £7.83 per hour for those aged 25 years and over,[38] this suggests £3.37 million of productivity is generated through wages which otherwise would not have been earned – or £76 per L&D referral not declined:

- 53,835 additional days worked × 8 hours × £7.83 minimum wage = £3,372,224
- £3,372,224 productivity generated ÷ 44,469 cases = £76 per L&D referral not declined.

H.4. Summary

Table H.33 brings together in one place the various monetised impacts whose estimation was described above.

Table H.33. Summary of L&D economic impacts

Costs (shown as negative) and benefits (positive)	Total	Per L&D referral not declined
Service providers		
L&D service costs	- £29.31m	- £659
Criminal justice system		
Timeliness of court processes		
Number of hearings*	*(- £21.55m)*	*(- £485)*
Diversion from criminal justice service		
Diversion from custody, based on median/mean sentence length avoided	£12.03m/£38.14m	£270/£858
Health services		
Secondary care utilisation		
A&E utilisation	£0	£0
IAPT utilisation	£0	£0
Drug and alcohol treatment utilisation	£0	£0
Wider societal		
Productivity, based on median/mean sentence length avoided	£1.06m/£3.37m	£24/£76

Source: RAND Europe analysis

*Not statistically significant at conventional levels but p=0.11

[38] HM Government, n.d.

The national L&D scheme has considerable service costs, amounting to a total of £29.31 million or £659 per L&D referral not declined.

The national L&D scheme may also be associated with a further total cost of £21.55 million, or £485 per L&D referral not declined, due to additional hearings. This impact was not significant at conventional levels (p=0.11), and so could be ignored. We report it here, however, as the low p-value suggests at least that the possibility of there being additional court hearings as a result of L&D could be worth further research in future if additional data become available.

The national L&D scheme appears to be associated with some savings to other parts of the criminal justice system, as well as with productivity gains for wider society. When considering the median diversion from custody, L&D is associated with a total saving of £12.03 million, or £270 per L&D referral not declined, and total productivity gain of £1.06 million, or £24 per L&D referral not declined. When considering the mean diversion from custody, L&D is associated with a total saving of £38.14 million, or £858 per L&D referral not declined, and total productivity gain of £3.37 million, or £76 per L&D referral not declined.

Interestingly, the national L&D scheme was not found to have any impact on the quantifiable health service utilisation outcomes.

It is perhaps no surprise that the picture remains largely unclear with respect to many of the outcomes, particularly given the heterogeneous nature of both the individuals being referred into the L&D scheme and the services that are available locally once in contact with the L&D scheme.

Despite this, the economic evaluation has provided some interesting findings. The national L&D scheme appears to be directly contributing to savings in the criminal justice system, through diversion from custody, as well as enabling greater levels of productivity – again due to diversion from custody. Assuming mean levels of sentence length avoided, and omitting cost impacts that are not statistically significant, we have found that the L&D service cost of £659 per L&D referral is more than offset by the £933 savings due to diversion from custody (£857+£76). If, however, we were to assume only median levels of sentence length being avoided, the L&D service costs of £659 per L&D referral not declined, would exceed the £294 savings due to diversion from custody (£270+£24). If, in reality, L&D does lead to additional court hearings, then the overall net impact would be worsened accordingly.

In conclusion, the national L&D scheme has a significant and direct impact on certain areas of the criminal justice system and wider society, but a lack of data meant that many of the potential costs and benefits could not be quantified in the economic analysis.

Appendix D. Paper 4

Creswell, C., Taylor, L., Giles, S., Howitt, S., Radley, L., Whitaker, E., ..., Pollard, J., Violato, M., … & Yu, L. M. (2024). **Digitally augmented, parent-led CBT versus treatment as usual for child anxiety problems in child mental health services in England and Northern Ireland: a pragmatic, non-inferiority, clinical effectiveness and cost-effectiveness randomised controlled trial**. *The Lancet Psychiatry*, 11(3). URL: https://doi.org/10.1016/S2215-0366(23)00429-7

Digitally augmented, parent-led CBT versus treatment as usual for child anxiety problems in child mental health services in England and Northern Ireland: a pragmatic, non-inferiority, clinical effectiveness and cost-effectiveness randomised controlled trial

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Summary

Background Anxiety problems are common in children, yet few affected children access evidence-based treatment. Digitally augmented psychological therapies bring potential to increase availability of effective help for children with mental health problems. This study aimed to establish whether therapist-supported, digitally augmented, parent-led cognitive behavioural therapy (CBT) could increase the efficiency of treatment without compromising clinical effectiveness and acceptability.

Methods We conducted a pragmatic, unblinded, two-arm, multisite, randomised controlled non-inferiority trial to evaluate the clinical effectiveness and cost-effectiveness of therapist-supported, parent-led CBT using the Online Support and Intervention (OSI) for child anxiety platform compared with treatment as usual for child (aged 5–12 years) anxiety problems in 34 Child and Adolescent Mental Health Services in England and Northern Ireland. We examined acceptability of OSI plus therapist support via qualitative interviews. Participants were randomly assigned (1:1) to OSI plus therapist support or treatment as usual, minimised by child age, gender, service type, and baseline child anxiety interference. Outcomes were assessed at week 14 and week 26 after randomisation. The primary clinical outcome was parent-reported interference caused by child anxiety at week 26 assessment, using the Child Anxiety Impact Scale–parent report (CAIS-P). The primary measure of health economic effect was quality-adjusted life-years (QALYs). Outcome analyses were conducted blind in the intention-to-treat (ITT) population with a standardised non-inferiority margin of 0·33 for clinical analyses. The trial was registered with ISRCTN, 12890382.

Findings Between Dec 5, 2020, and Aug 3, 2022, 706 families (706 children and their parents or carers) were referred to the study information. 444 families were enrolled. Parents reported 255 (58%) child participants' gender to be female, 184 (41%) male, three (<1%) other, and one (<1%) preferred not to report their child's gender. 400 (90%) children were White and the mean age was 9·20 years (SD 1·79). 85% of families for whom clinicians provided information in the treatment as usual group received CBT. OSI plus therapist support was non-inferior for parent-reported anxiety interference on the CAIS-P (SMD 0·01, 95% CI –0·15 to 0·17; p<0·0001) and all secondary outcomes. The mean difference in QALYs across trial arms approximated to zero, and OSI plus therapist support was associated with lower costs than treatment as usual. OSI plus therapist support was likely to be cost effective under certain scenarios, but uncertainty was high. OSI plus therapist support acceptability was good. No serious adverse events were reported.

Interpretation Digitally augmented intervention brought promising savings without compromising outcomes and as such presents a valuable tool for increasing access to psychological therapies and meeting the demand for treatment of child anxiety problems.

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Introduction

Child mental health services are notoriously stretched around the world, with stark gaps between needed and available care.[1] Digitally augmented psychological treatments bring potential to dramatically increase capacity within clinical services;[2] however, such treatments have not been established in routine child mental health services. Nonetheless, the implementation

Research in context

Evidence before this study

A recent Cochrane review highlighted that there is now robust evidence for cognitive behavioural therapy (CBT) for child anxiety problems, however few children who might benefit from CBT are able to access it. Supporting parents to implement CBT strategies in their children's day-to-day lives (parent-led CBT) has been shown to be a clinically effective and cost-effective way to provide treatment, and digital augmentation could further increase accessibility. We searched OVID PsycINFO, PubMed, Web of Science, OVID EMBASE, CINAHL via EBSCOhost, and the Cochrane Central Register of Controlled Trials (CENTRAL) on Jan 26, 2023, using terms relating to child or adolescent or youth, parent or family, anxiety, and cognitive behavioural therapy (appendix p 193). We placed no restrictions on publication date or language. Thirteen papers were identified. Only one small randomised controlled trial reported on the use of therapist-supported, digitally augmented parent-led CBT. The trial included 52 pre-school children (age 3–6 years) and suggested the approach was feasible and well received by parents, and that there was some evidence of efficacy compared to a wait-list control. Economic evaluations of digitally augmented psychological interventions for child mental health problems are sparse, with none specifically focusing on anxiety problems in children. No trials to date have evaluated whether digitally augmented CBT generally, and parent-led CBT specifically, is non-inferior and cost-effective compared to routine treatment in child mental health services. In the UK, the National Institute of Health and Care Excellence has recently identified this as the critical information required in order to make clinical recommendations.

Added value of this study

To our knowledge, this is the first randomised controlled trial to test the clinical and cost-effectiveness of therapist-supported, digitally augmented parent-led CBT for child anxiety problems in routine child mental health settings compared to usual treatment. The digital platform used in this study (Online Support and Intervention [OSI] for child anxiety) was designed with therapists and families to help parents to help their children overcome problems with anxiety, with remote and brief therapist support. OSI plus therapist support brought substantial reductions in therapist time taken to deliver treatment without compromising treatment outcomes, when compared with (predominantly) evidence-based treatment as usual. When we considered the joint distribution of incremental mean costs and effects, OSI was likely to be cost effective under certain scenarios, but uncertainty was high. Parent and therapist feedback was positive—they found OSI easy to access and use and reported a wide range of benefits.

Implications of all the available evidence

CBT is well established as an effective treatment for child anxiety problems, yet few children who could benefit are able to access it. Parent-led CBT has been shown to provide an efficient way to deliver effective treatment yet barriers remain for both families and clinical teams. OSI plus therapist support is a promising new approach to increase access to effective treatment for child anxiety problems. Both the main study outcomes and therapist and parents' descriptions of their experiences suggest that implementation of this online therapist-supported parent-led CBT approach has potential to substantially increase capacity within busy child mental health services while bringing accessibility benefits for families.

See Online for appendix

of digitally enabled care within children and young people's mental health services is a current priority area, as reflected in England, for example, by a National Institute of Health and Care Excellence (NICE) Early Value Assessment.[3] Here we focus on anxiety problems as they affect over a quarter of the population during their lives,[4] bring substantial personal and economic costs,[5] and often start early in life, with a peak age of onset at 5 years.[6]

Cognitive behavioural therapy (CBT) is an effective treatment for childhood anxiety disorders[7] but only a minority of children with anxiety disorders access evidence-based treatment.[8–10] Families face extensive barriers, including high demands on services, limited available support, and long waiting lists,[11] highlighting the need for efficient mechanisms for treatment delivery. Brief forms of CBT for childhood anxiety can be effective.[7] For pre-adolescent children, they typically involve a therapist-guided, parent-led approach,[7] in which therapists support parents to implement CBT strategies in their children's day-to-day lives. In addition to reducing overall therapy time, this approach has potential to increase access to effective treatment by reducing the perceived stigma and disruption to children's usual activities by not requiring them to attend regular clinic appointments and enabling difficulties to be managed as a family.[11] There is now good evidence for this approach[12] and it is widely implemented, for example, in England,[13] as a first-line treatment approach.

Digital augmentation brings potential to further increase the efficiency and accessibility of psychological interventions such as parent-led CBT, by enabling parents to access and engage with core treatment content in their own time and space with personalised therapist support. One small trial has provided promising findings for pre-school children in comparison with a waitlist control.[14] Here we evaluated a novel, therapist-supported, parent-led CBT approach using Online Support and Intervention (OSI) for child anxiety—a platform that was designed in collaboration with families and NHS therapists[15] with the specific aims of enabling efficient, engaging, and accessible treatment for child anxiety disorders.

Therapist-supported OSI has so far shown promising evidence[16,17] but is yet to be systematically evaluated for clinical effectiveness and cost-effectiveness and acceptability in routine practice. Therefore, the primary objective of this trial was to determine whether this therapist-supported, digitally augmented, parent-led CBT brings cost savings in routine children's mental health services without compromising clinical outcomes, meeting the recent call from England's NICE for essential evidence to inform clinical recommendations.[18]

Methods

Study design and participants

We conducted a pragmatic, two-arm, multisite, randomised, controlled, non-inferiority trial of OSI plus therapist support and treatment as usual in Child and Adolescent Mental Health Services for child anxiety problems. We also examined the acceptability of OSI plus therapist support via qualitative interviews with parents and therapists. The trial was registered with the ISRCTN (12890382) and the study protocol was published.[19]

To participate, sites needed to provide child mental health care on behalf of the National Health Service (NHS) or local authorities in England and Northern Ireland. There were 34 participating sites: 29 NHS Trusts and five local authority or voluntary or community sector providers. These sites included 73 different recruiting Child Mental Health Teams; 42 teams were Mental Health Support Teams providing mental health support through schools.

To be eligible, children needed to be aged 5–12 years at intake, have a primary problem of anxiety (as determined by clinical teams in line with their usual practice), and be willing and able to assent. Parents were required to have sufficient English language to complete measures and access interventions, have access to the Internet, and be willing and able to provide consent. Participants were not eligible if the children had comorbid conditions that were likely to interfere with treatment delivery (established diagnosis of autism or learning disability, suicidal intent, or recurrent or potentially life-limiting self-harm); were identified by social services as having child protection concerns; or were a potential participant in another study where the child might receive the OSI intervention. Participants were also ineligible if the participating parent had a significant intellectual impairment or severe mental health problem that was likely to interfere with treatment delivery.

Of note, this study started when restrictions were in place due to the COVID-19 pandemic, a time when there were particular concerns about increases in demand for child mental health services[20] and when services had to quickly pivot to delivering services remotely.[21] In our study protocol we referred to "treatment as usual in the COVID-19 context", however as COVID-19 restrictions were not in place throughout the trial, and because services have continued to use strategies employed in the COVID-19 context, we have adopted the term "child mental health service treatment as usual" as this is a better reflection of what was delivered within this arm.

This study was approved by London-City & East Research Ethics Committee (20/HRA/4421).

Randomisation and masking

Participants were randomly assigned in a 1:1 ratio to OSI plus therapist support or child mental health service treatment as usual for child anxiety problems (referred to henceforth as treatment as usual). Minimisation by child age (≤8 years and ≥9 years), gender, service type (school based or not school based), and baseline anxiety-associated interference, including permuted block size, was used to ensure balance across arms. Participants were randomly assigned using a fully validated and secured web-based randomisation system called Sortition[22] that acted automatically after the participating parent completed the consent and baseline measures, and the child completed assent. Sortition then automatically sent an email, including the result of the allocation treatment arm, to the trial team, the clinical team, and the participant. Due to the nature of the trial, blinding to intervention was not possible for trial participants, however statistical analyses were conducted blind to treatment allocation.

Procedures

Participants were identified within clinical services following their usual assessment procedures. Eligible families were invited to take part in the trial at the point in time when the clinical team were confident that they could deliver either treatment arm within 12 weeks of randomisation. Members of the clinical team introduced the study to potential participants and registered them on a study website where they could access study information for parents and children (in written and video form), access contact details for further enquiries, and, if willing, provide consent (parents) or assent (children) via a secure online system. After consent or assent was provided, parents and children were asked to complete online baseline assessments (all questionnaires) before randomisation. After randomisation, parents were asked to complete the treatment expectations and acceptability measure.

Treatment in both arms was organised by the clinical teams, who were requested to start as soon as possible and at most within 12 weeks of randomisation. Participating parents and children in both arms were sent a link to complete further assessments (questionnaires) at week 14 and week 26 after randomisation via a secured online web-based database system.[23,24] The full schedule of enrolment, interventions, and assessments is provided in the trial protocol.[19]

Parents received a welcome phone call from the trial team and monthly parent bulletins with trial updates. A scheduled series of emails, text messages, and telephone

calls was made to families during the 1-month periods in which their 14-week questionnaire and 26-week questionnaire were due, to promote participant retention. Families received a £10 voucher as a thank you for completing their final assessment.

Qualitative interviews were conducted one-to-one by a researcher with qualitative expertise (FK), who was not involved in any other aspects of the trial. Purposively sampled participants were interviewed at a date and time convenient to the participant after their week 14 assessment. Participants received a £20 voucher for taking part.

Treatment in both arms could be delivered by any therapists in participating clinical services, who routinely provided psychological treatments for child anxiety problems. 188 therapists across 73 clinical teams delivered treatment within the trial. Therapists' professional backgrounds were provided for 167 of the therapists, as shown in the appendix (pp 4–5).

OSI for child anxiety was designed to digitally augment parent-led CBT for the treatment of anxiety problems in pre-adolescent children by providing parents with all the core treatment content that they need in accessible forms, including information (in text, audio, and video) and exercises (supported by worksheets and quizzes). The accompanying therapist case management system supports therapists to help parents to personalise the content for their child and overcome potential barriers that they might face. There is also an optional child game app that parents can use to motivate their child to engage with the intervention. The core intervention content is centred on empowering parents to help their child by developing an understanding of their child's anxious predictions, putting these predictions to the test in a manageable (gradual) way, and promoting problem solving to address issues that arise. The OSI intervention is provided across seven modules, and parents are supported to apply it by weekly 20-min telephone or video call sessions between the parent and a therapist, and a review session, 4 weeks after the final treatment session. Therapists were provided with a written manual and a brief training video (45 min). Ongoing supervision of the therapists was provided within their clinical services following usual procedures. The research team offered weekly drop-in sessions for therapists to address technical questions or challenges, but very few therapists regularly attended them (60 [76%] of 79 therapists who delivered an OSI case attended at least one drop-in session; median drop-in sessions attended was 1). OSI plus therapist support is considered to be a complete treatment so, although participants were not prevented from accessing other support, OSI plus therapist support was considered to be an alternative rather than an adjunct to treatment as usual.

For more on **game app** see https://osiresearch.org.uk/osi/

The comparator was whatever treatments the participating services were otherwise delivering to treat child anxiety problems. Therapists provided information on the therapeutic approach being followed, the format (individual or group), modality (in person or online), and who they worked with (child, parent, or both). Therapists in both arms also provided detailed information on the time taken to deliver the intervention, including preparation, administrative tasks, and supervision.

Outcomes

The primary outcome was the Child Anxiety Impact Scale–parent report (CAIS-P)[25] at week 26 after randomisation. Secondary clinical outcomes included child-reported anxiety interference (CAIS-C total and global scores)[25] and anxiety symptoms (Revised Child Anxiety and Depression Scale [RCADS-C]),[26] parent-reported child anxiety symptoms (RCADS–P),[27] CAIS-P global score,[25] overall functioning (Outcome Rating Scale),[28] and common comorbid emotional and behavioural problems (Strengths and Difficulties Questionnaire–parent report [SDQ-P])[29] measured at week 14 and week 26 after randomisation, and the CAIS-P at week 14 after randomisation. For all these scales, a higher score indicates worse functioning, with the exception of the Outcome Rating Scale, in which a higher score indicates better functioning. For brevity, other secondary outcomes are described in the appendix (pp 32–36).

To capture adverse events, therapists were requested to monitor and report any harms and adverse events during the treatment phase. Additionally, parents and children were invited to report any negative impact of participating in the study as part of their assessment at week 14 and 26 after randomisation.

Indicative topic guides were used to guide the post-treatment interviews with parents and therapists about their experiences of the trial and the OSI treatment, including what they found helpful and unhelpful, potential improvements, the involvement of others in treatment, and how things had been since treatment ended.

The primary economic outcome was child quality-adjusted life years (QALYs), derived from the validated parent report version of the Child Health Utility 9-Dimension measured at baseline, week 14, and week 26.[30] As no established guidelines exist on which value set is most appropriate for UK preadolescent children, individual responses were converted to utilities using preference weights obtained from both a sample of the UK adult general population[31] and of Australian adolescents aged 11–17 years.[32] Parent QALYs were derived from the EuroQol-5 dimensions, 5 levels (EQ-5D-5L) administered to parents at baseline, week 14, and week 16.[33] Utility values were derived using a validated mapping function from the UK EQ-5D-3L value set,[34] as recommended by NICE.[35] Child and parent QALYs were each calculated by combining the utility values at baseline, week 14, and week 26 assessment using the area under the curve approach, which assumes a linear relationship between utilities at different time points.[36] Parent–child QALYs were

obtained by additively combining individual parent and child QALYs.[37] The CAIS-P was used as a secondary economic outcome.

Data collected on patient-level resource use included treatment, additional health and personal social service use, and time off school for children and work for parents. To calculate the total cost of the intervention, therapists completed bespoke economic logs capturing treatment duration and type of contact, and time spent on preparation, clinical supervision, administration, and travel, as applicable. Child and parent resource use data were collected from parents at baseline (referring to the preceding 3 months) and at weeks 14 and 26 after randomisation using a modified (with PPI input) Client Service Receipt Inventory[38] including information on primary and secondary health and social care and medication use, school missed by the child, work missed by the parent, child and parent travel time, and direct costs for health and social care and for participating in the intervention. For each trial participant, resource use data were multiplied by the appropriate unit cost to calculate the total mean cost in each trial arm (appendix pp 8–22). The cost of the OSI technology is not included in our economic analyses as it is still unknown (but see appendix pp 188–190).

Parents were asked to complete the Credibility and Expectation of Improvement Scale[39] to assess their expectations and views regarding treatment credibility, after they had been randomly assigned and informed of the treatment arm (with higher scores reflecting more positive responses). Parents and therapists also completed an adapted form at week 14 after randomisation, to give a retrospective account of treatment credibility.

Choice of primary outcome

The CAIS-P captures the degree to which anxiety is interfering in the child and family's life. Although it has not previously been used as a primary outcome in trials, the CAIS-P was selected as the primary outcome because (1) measures of interference have been considered more relevant and valid than symptom measures by experts by experience in previous consultations;[40] (2) the CAIS-P has been found to align better with diagnostic measures of child anxiety disorders than anxiety symptom measures;[41] and (3) measures of interference, and the CAIS in particular, have recently been highlighted as crucial socially and ecologically valid markers of treatment need.[42] The total score is the sum of responses to 25 items rated on a 4-point scale (from 0 [not at all] to 3 [very much]; range 0–75) across three psychosocial domains (academic, social activities, and home and family environment). Two of the original items were not included as they were not appropriate for the pre-adolescent age group. We are not aware of translations into non-English languages. The CAIS-P is freely available from the authors.

Statistical analysis

We aimed to recruit between 418 and 560 children (209–280 per group) as this was considered to be sufficient to provide a standardised non-inferiority margin of 0·33 (ie, the upper bound of the confidence interval must be less than 0·33 when comparing OSI plus therapist support to treatment as usual to claim non-inferiority) with between 80% and 90% power (allowing for 30% attrition) at 2·5% one-side level of significance. This standardised non-inferiority margin was equivalent to a 4-point change in mean CAIS-P and standard deviation of 12, which is half of the standardised change in the primary outcome (of 0·63 by 6 months in treatment as usual for child anxiety problems, from a previous trial conducted in routine child mental health services).[43] The required sample size was calculated using PASS 2019.

For the qualitative interviews, we purposively sampled parents and therapists from the first 70 clinical cases to reach week 14 after randomisation until we reached saturation in terms of representation on a range of demographic and clinical characteristics. The sample comprised 12 parents and ten therapists (see appendix pp 2–3 for further information on both samples).

Statistical analyses of clinical outcomes were pre-specified in the Statistical Analysis Plan (appendix pp 23–50) before the end of the trial. Analyses were conducted using Stata version 16.1. Analysis of the primary outcome was performed using a generalised linear mixed effects model adjusting for minimisation factors to determine the treatment effect and two-sided 95% CI. The mixed effect models included the outcome as the response variable, time point, randomised group, and baseline score as fixed effects and a participant-specific random intercept. The model specified an unstructured variance-covariance structure for the random effects. An interaction between time and randomised group was fitted as a fixed effect to allow estimation of treatment effect at all time points. Non-inferiority would be claimed if the upper limit of the 95% CI around the standardised effect size was less than 0·33. A p value for non-inferiority was also calculated. The models did not deviate from normality assumptions of the generalised linear mixed effects model. A similar approach was used for the other secondary outcomes. Treatment credibility, acceptability, and experience scores were calculated and compared for both treatment groups, using a Mann–Whitney U test.

The primary analysis population was defined as all participants for whom data were available, analysed according to the groups to which they were randomly allocated, regardless of treatment compliance (ie, actual treatment received). For the primary analysis, they must have completed their assessment within 4 weeks of the week 14 and week 26 timepoints, but sensitivity analyses were carried out based on altering the timeframe allowed for the assessments. A secondary analysis was also carried

out based on a per-protocol population who had (1) received five or more treatment sessions, (2) received the treatment they were originally assigned to, (3) submitted their final questionnaire within 30 weeks of randomisation, and (4) started treatment within 12 weeks of being randomly assigned (appendix p 150). Characteristics that were found to be predictive of missingness (if the parent was partnered and if the parent was cohabiting) were included in the model in a pre-planned sensitivity analysis of the primary outcome. Post-hoc sensitivity analyses, such as best-case and worst-case scenarios, and multiple imputation, were also carried out. Multiple imputation was conducted using chained equations. The following were included in the multiple imputation model: random allocation; minimisation variables; child's age; child's gender; baseline anxiety associated interference; service type (school or clinic); and factors found to be predictive of the primary outcome being missing (not partnered and not cohabiting). Adverse events were summarised by treatment arm; no other analyses were conducted.

The Health Economics Analysis Plan (appendix pp 51–78) was signed off before the end of the trial and it adhered to current best practice.[44] The primary analysis (base-case analysis) was a within-trial cost-utility analysis comparing OSI plus therapist support with treatment as usual, with incremental costs (reported with their associated 95% CI) and incremental child QALYs (reported with their associated 95% CI) combined to calculate an incremental cost-effectiveness ratio from the NHS and Personal Social Services perspective as recommended by NICE.[35] Costs were expressed in pounds sterling (£) in 2020–21 prices. Due to the short timeframe of the trial and follow-up, discounting was not applied to costs or effects. Both an intention-to-treat and per-protocol approach were adopted in the base-case analysis. Missing data were imputed by use of mean imputation conditional on treatment arm for missing items, and multiple imputation for missing responses and cases under the assumption of missing at random.[45]

Differences in costs and QALYs between OSI plus therapist support and treatment as usual were estimated using linear regression, controlling for baseline costs and utility, respectively. A secondary cost-effectiveness analysis was undertaken, with outcomes measured using the difference in CAIS-P at week 26 and incremental costs from the NHS and Personal Social Services perspective (base-case analysis).

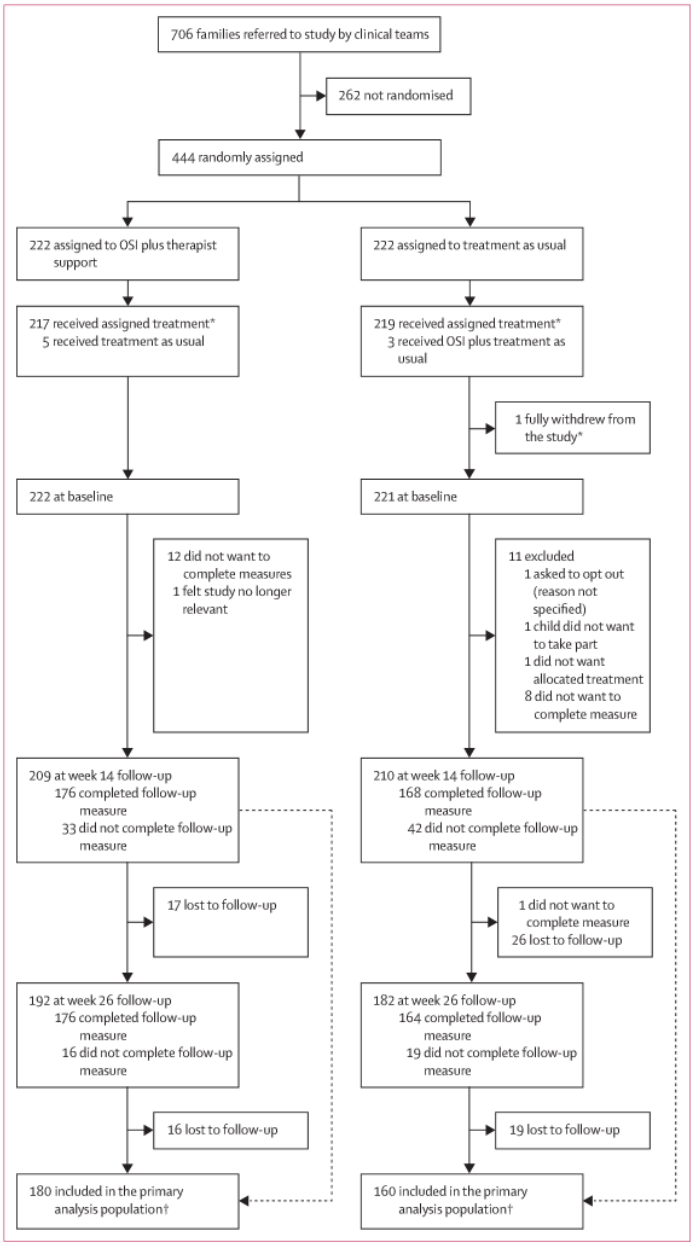

Figure 1: **Trial profile**
There was one child per family. Participants in the treatment as usual arm received treatment for child anxiety problems from child mental health services. Participants in the OSI plus therapist arm received parent-led OSI for child anxiety with therapist support. OSI=Online Support and Intervention. *One participant fully withdrew from the study and requested all data that had been collected so far to be deleted. This participant has been excluded from the analysis population. †Number of participants that completed the primary outcome of CAIS-P at week 14, or 26, or both: 21 participants in the OSI plus therapist support arm and 30 participants in the treatment as usual arm completed CAIS-P at week 14 only, and 17 participants in the OSI plus therapist support arm and 15 participants in the treatment as usual arm completed CAIS-P at week 26 only. 142 participants in the OSI plus therapist support arm and 115 participants in the treatment as usual arm completed CAIS-P at both week 14 and 26 assessments. 42 participants in the OSI plus therapist support arm and 62 participants in the treatment as usual arm were missing CAIS-P at both week 14 and 26, and are not included in the primary analysis.

	OSI plus therapist support (n=222)	Treatment as usual (n=221)	Mean/% difference
Child baseline characteristics			
Age, years	9·31 (1·83)	9·08 (1·74)	0·23
Gender			
Male	92 (41%)	92 (41%)	−0·19
Female	127 (57%)	128 (58%)	−0·71
Other	2 (1%)	1 (<1%)	0·45
Prefer not to say	1 (<1%)	0	0·45
Ethnicity			
White*	194 (87%)	206 (93%)	−5·82
Mixed†	19 (9%)	14 (6%)	2·23
Asian or Asian British‡	3 (1%)	0	1·35
Black or Black British§	1 (<1%)	1 (<1%)	0·00
Other ethnic groups¶	2 (1%)	0	0·90
Not stated	3 (1%)	0	1·35
Previous treatment for anxiety or other psychological difficulties	46 (21%)	30 (14%)	7·15
Prescribed medication for anxiety or other psychological difficulties	2 (1%)	6 (3%)	−1·81
Education			
State school	214 (96%)	209 (95%)	1·83
Independent school	4 (2%)	7 (3%)	−1·37
Special provision school	2 (1%)	2 (1%)	0·00
Home educated	2 (1%)	3 (1%)	−0·46
Special educational needs	33 (15%)	32 (14%)	0·38
Type of special educational needs‖			
Communicating and interacting	15 (45%)	11 (34%)	11·07
Cognition and learning	16 (48%)	15 (47%)	1·60
Social, emotional, and mental health difficulties	24 (73%)	20 (63%)	10·23
Sensory, or physical, or both, needs	13 (39%)	12 (38%)	1·89
CAIS-P: total score	26·87 (15·26)	25·96 (14·63)	0·91
CAIS-P: global items	6·20 (3·00)	5·86 (2·95)	0·34
CAIS-C: total score	26·13 (14·44; n=210)	25·75 (15·06; n=212)	0·38
CAIS-C: global items	5·30 (2·85; n=210)	5·17 (3·18; n=212)	0·13
RCADS-P: total anxiety score	46·35 (19·83)	45·91 (19·93)	0·44
RCADS-P: total anxiety and depression score	56·18 (23·79)	55·40 (24·17)	0·77
RCADS-C: total anxiety score	47·14 (19·68; n=204)	46·26 (19·96; n=209)	0·87
RCADS-C: total anxiety and depression score	56·98 (23·54; n=204)	55·84 (24·14; n=209)	1·13
ORS	26·25 (8·15)	27·19 (7·78)	−0·93
SDQ-P: total problems score	17·95 (7·05)	17·26 (6·53)	0·69
Parent baseline characteristics			
Age, years	39·00 (5·93)	38·28 (5·67)	0·73
Gender			
Male	9 (4%)	8 (4%)	0·43
Female	212 (96%)	213 (96%)	−0·88
Other	0	0	0·00
Prefer not to say	1 (<1%)	0	0·45
Ethnicity			
White*	203 (91%)	215 (97%)	−5·85
Mixed†	11 (5%)	2 (1%)	4·05
Asian or Asian British‡	3 (1%)	1 (<1%)	0·90
Black or Black British§	1 (<1%)	2 (1%)	−0·45
Other ethnic groups¶	2 (1%)	1 (<1%)	0·45
Not stated	2 (1%)	0	0·90

(Table 1 continues on next page)

	OSI plus therapist support (n=222)	Treatment as usual (n=221)	Mean/% difference
(Continued from previous page)			
Household circumstances			
Mortgaged or owned	137 (62%)	122 (55%)	6·51
Council rented	29 (13%)	22 (10%)	3·11
Housing association	19 (9%)	30 (14%)	−5·01
Privately rented	32 (14%)	44 (20%)	−5·50
Other	5 (2%)	3 (1%)	0·89
Is child fostered?	0	0	0·00
Is child adopted?	1 (<1%)	1 (<1%)	0·00
Education			
School completion	35 (16%)	33 (15%)	0·84
Further education	103 (46%)	101 (46%)	0·70
Higher education	39 (18%)	53 (24%)	−6·41
Postgraduate qualification	45 (20%)	34 (15%)	4·89
Partnered	177 (80%)	176 (80%)	0·09
Cohabiting (living together)**	165 (93%)	163 (93%)	0·61
Partner's education**			
School completion	50 (28%)	38 (22%)	6·66
Further education	65 (37%)	76 (43%)	−6·46
Higher education	30 (17%)	27 (15%)	1·61
Postgraduate qualification	20 (11%)	22 (13%)	−1·20
Not stated	12 (7%)	13 (7%)	−0·61
Employment			
Full time	84 (38%)	82 (37%)	0·74
Part time	87 (39%)	73 (33%)	6·16
Sheltered or supported employment	1 (<1%)	0	0·45
Unemployed	7 (3%)	20 (9%)	−5·90
Student	3 (1%)	2 (1%)	0·45
Homemaker	26 (12%)	28 (13%)	−0·96
Retired	0	0	0·00
Other	14 (6%)	16 (7%)	−0·93
Total household income, £††			
≤16 000 per year	17 (12%)	18 (13%)	−1·18
16 001–30 000 per year	27 (19%)	25 (18%)	0·77
30 001–40 000 per year	14 (10%)	18 (13%)	−3·31
40 001–50 000 per year	11 (8%)	12 (9%)	−1·02
50 001–60 000 per year	12 (9%)	17 (13%)	−3·99
60 001–70 000 per year	11 (8%)	7 (5%)	2·65
70 001–80 000 per year	8 (6%)	10 (7%)	−1·68
80 001–90 000 per year	6 (4%)	5 (4%)	0·58
90 001–120 000 per year	8 (6%)	4 (3%)	2·73
>120 000 per year	3 (2%)	6 (4%)	−2·28
Prefer not to say	24 (17%)	14 (10%)	6·73

Data are n (%) or mean (SD). Participants in the treatment as usual arm received treatment for child anxiety problems from child mental health services. Participants in the OSI plus therapist arm received parent-led OSI for child anxiety with therapist support. Percentages have been computed with the number of participants with the response available as the denominator. For all scales, a higher score indicates worse functioning, with the exception of the ORS where a higher score indicates better functioning. CAIS-C=Child Anxiety Impact Scale–child report. CAIS-P=Child Anxiety Impact Scale–parent report. ORS=Outcome Rating Scale. OSI=Online Support and Intervention. RCADS-C=Revised Child Anxiety and Depression Scale–child report. RCADS-P=Revised Child Anxiety and Depression Scale–parent report. SDQ-P=Strengths and Difficulties Questionnaire–parent report. *Including British, Irish, and any other White background. †Including White and Black Caribbean, White and Black British, White and Asian, and any other mixed background. ‡Including Indian, Pakistani, Bangladeshi, and any other Asian background. §Including African, Caribbean, and any other Black background. ¶Including Chinese, and any other ethnic group. ||Only includes those with special educational needs. **Only includes those who are partnered. ††Data were available for 141 participants in the OSI plus therapist support arm and 136 in the treatment as usual arm.

Table 1: **Baseline characteristics of participants**

A willingness to pay threshold of £20 000–30 000 per QALY gained was used to evaluate whether OSI plus therapist support was cost effective compared to treatment as usual, as per NICE guidelines,[35] representing uncertainty around the cost and effectiveness estimates, by means of acceptability curves.[46] The same approach was used in the cost-effectiveness analyses, although the maximum threshold value that the NHS or society is willing to pay for an improvement in the CAIS-P is unknown so we presented a range of possible maximum values that a decision maker might be willing to pay for a unit improvement in outcome.

Various prespecified sensitivity analyses, including a societal perspective to capture wider impacts, were undertaken to explore uncertainties around assumptions made in the base-case analyses and test the robustness of the results (appendix pp 79–81).

Qualitative data were analysed using semantic interpretative deductive and inductive thematic analysis.[47] For this paper, we used the data to deductively explore the acceptability of OSI, particularly focusing on constructs in the theoretical framework of acceptability (eg, affective attitude, effort, degree of fit with the individual's value system, understanding of the intervention, perceived effectiveness, and self-efficacy).[48]

This study is registered as an International Standard Randomised Controlled trial (ISRCTN12890382) and the protocol is publicly available.[19]

Role of the funding source

The funder of the study had no role in study design, collection, analysis and interpretation of data, in the writing of the report or in the decision to submit for publication.

Results

Participants were recruited between Dec 5, 2020, and Aug 3, 2022. 706 families (706 children and their parents or carers) were referred to the study information by the clinical teams, of whom 444 families confirmed they met inclusion criteria, provided consent, and were randomly assigned (222 to OSI plus therapist support and 222 to child mental health service treatment as usual; figure 1). All participants completed baseline assessments, however one participant subsequently requested all their data be removed. Details of the type of primary anxiety problem as determined by the clinician are provided in the appendix (p 82). Despite study procedures requiring all participants to have started treatment within 12 weeks of randomisation, only 349 (79%) of 444 participants were reported to have started their allocated treatment by the end of the trial. 181 (82%) of 222 participants allocated to the OSI plus therapist support arm and 168 (76%) of 222 allocated to the treatment as usual arm started treatment. Eight participants were incorrectly assigned to treatments by clinical teams (five in the OSI plus therapist support arm were given treatment as usual and three in the treatment as usual arm were registered to OSI). 176 (79%) participants in the OSI plus therapist support arm and 168 (76%) in the treatment as usual arm completed the week 14 assessment, and 176 (79%) in the OSI plus therapist support arm and 164 (74%) in the treatment as usual arm completed the week 26 assessment. There were more girls than boys (255 [58%] girls and 184 [41%] boys)and the majority were described as White-British (table 1). The mean age of child participants was 9·20 (SD 1·79) years.

Breakdown of the treatment approach, format, modality, and who the sessions were conducted with are provided in the appendix (pp 151–52), and therapist characteristics are also provided in the appendix (pp 4–5). 110 (85%) of 130 treatment as usual cases where information was provided on treatment approach received CBT, for 79 (72%) this was delivered through parents (with the rest [28%] being delivered through both children and parents).

As shown in the appendix (p 153), before receiving treatment, parent reports across treatment arms did not differ on how logical the treatment seemed and how confident they were in it, but scores were significantly higher for their certainty in the success of the OSI plus therapist support arm. There were no differences at the week 14 assessment. Therapist ratings also did not differ after delivering the treatment on items relating to how logical the treatment was, how prepared they felt, how successful it was, and how much they would recommend it; however, therapists reported that they felt more comfortable in delivering treatment as usual than the novel online treatment and felt they were less likely to use OSI again in the future (note: services had time limited access to OSI associated with the research trial and so were uncertain about whether they would be able to continue to use it).

The standardised mean difference between arms was less than 0·33 for the primary outcome, indicating that OSI plus therapist support was significantly non-inferior to treatment as usual, with an extremely small standardised mean difference (table 2; figure 2). The same pattern was found across all sensitivity analyses (appendix p 150). OSI plus therapist support was also significantly non-inferior to treatment as usual across all secondary analyses (figures 2 and 3; table 2). More details on the clinical results are presented in the appendix (p 150).

Descriptive data for treatment, resource use outcomes, and costs are presented in the appendix (pp 155–56); there was little difference in utility scores and QALYs between arms, after adjusting for baseline values (p 169). Cost mean differences, adjusted for baseline costs, were lower in the OSI plus therapist support arm compared with treatment as usual (appendix p 170). This was primarily accounted for by lower costs for therapist delivery; the mean therapist time delivering treatment sessions for OSI plus

	OSI plus therapist support (n=222)	Treatment as usual (n=221)	Adjusted mean difference (95% CI)*†	Standardised mean difference (95% CI)	p value for non-inferiority‡
Primary outcome					
CAIS-P					
Baseline	26·87 (15·26)	25·96 (14·63)	..	..	..
14 weeks	19·64 (16·00; n=163)	18·89 (14·52; n=145)	0·00 (−2·34 to 2·34)	0·00 (−0·16 to 0·16)	<0·0001
26 weeks§	17·99 (15·39; n=159)	18·08 (15·08; n=130)	0·14 (−2·26 to 2·53)	0·01 (−0·15 to 0·17)	<0·0001
Secondary outcome					
CAIS-P: global items					
Baseline	6·20 (3·00)	5·86 (2·95)	..	..	..
14 weeks	4·07 (3·12; n=163)	3·97 (2·88; n=145)	−0·13 (−0·63 to 0·37)	−0·04 (−0·21 to 0·12)	<0·0001
26 weeks	3·60 (3·06; n=159)	3·62 (2·84; n=130)	0·08 (−0·42 to 0·59)	0·03 (−0·14 to 0·20)	0·0003
CAIS-C: total score					
Baseline	26·13 (14·44; n=210)	25·75 (15·06; n=212)	..	..	..
14 weeks	19·27 (15·13; n=127)	20·73 (14·50; n=114)	−1·61 (−4·55 to 1·33)	−0·11 (−0·31 to 0·09)	<0·0001
26 weeks	17·03 (15·83; n=124)	19·89 (16·64; n=111)	−2·67 (−5·64 to 0·30)	−0·18 (−0·38 to 0·02)	<0·0001
CAIS-C: global items					
Baseline	5·30 (2·85; n=210)	5·17 (3·18; n=212)	..	..	..
14 weeks	3·63 (3·05; n=127)	4·03 (2·62; n=114)	−0·30 (−0·90 to 0·30)	−0·10 (−0·30 to 0·10)	<0·0001
26 weeks	3·61 (3·28; n=123)	3·40 (3·18; n=111)	0·30 (−0·31 to 0·90)	0·10 (−0·10 to 0·30)	0·012
RCADS-P: total anxiety score					
Baseline	46·35 (19·83)	45·91 (19·93)	..	..	..
14 weeks	34·09 (23·01; n=161)	34·84 (19·92; n=143)	−2·22 (−5·49 to 1·04)	−0·11 (−0·28 to 0·05)	<0·0001
26 weeks	30·57 (23·29; n=157)	32·03 (20·98; n=129)	−0·96 (−4·27 to 2·36)	−0·05 (−0·22 to 0·12)	<0·0001
RCADS-P: total anxiety and depression score					
Baseline	56·18 (23·79)	55·40 (24·17)	..	..	..
14 weeks	41·25 (28·26; n=161)	41·55 (23·89; n=143)	−2·22 (−6·16 to 1·73)	−0·09 (−0·26 to 0·07)	<0·0001
26 weeks	37·45 (28·77; n=157)	38·22 (25·39; n=129)	−0·54 (−4·54 to 3·46)	−0·02 (−0·19 to 0·14)	<0·0001
RCADS-C: total anxiety score					
Baseline	47·14 (19·68; n=204)	46·26 (19·96; n=209)	..	..	..
14 weeks	31·40 (23·18; n=127)	32·10 (21·26; n=112)	−1·29 (−5·58 to 3·00)	−0·07 (−0·28 to 0·15)	0·0002
26 weeks	29·96 (24·91; n=122)	29·53 (22·75; n=111)	1·41 (−2·89 to 5·71)	0·07 (−0·15 to 0·29)	0·0098
RCADS-C: total anxiety and depression score					
Baseline	56·98 (23·54; n=204)	55·84 (24·14; n=209)	..	..	..
14 weeks	37·91 (28·37; n=127)	38·11 (25·38; n=112)	−0·99 (−6·15 to 4·17)	−0·04 (−0·26 to 0·18)	0·0004
26 weeks	36·30 (30·86; n=122)	35·04 (27·27; n=111)	2·31 (−2·86 to 7·49)	0·10 (−0·12 to 0·31)	0·018
ORS: total score (overall functioning)					
Baseline	26·25 (8·15)	27·19 (7·78)	..	..	..
14 weeks	29·80 (7·97; n=161)	30·94 (7·00; n=143)	−0·58 (−1·90 to 0·74)	−0·07 (−0·24 to 0·09)	0·0011
26 weeks	30·68 (8·11; n=154)	31·21 (6·77; n=127)	−0·21 (−1·58 to 1·15)	−0·03 (−0·20 to 0·14)	0·0003
SDQ-P: emotional symptoms					
Baseline	6·41 (2·29)	6·21 (2·40)	..	..	..
14 weeks	4·99 (2·89; n=161)	4·62 (2·61; n=143)	0·03 (−0·45 to 0·51)	0·01 (−0·19 to 0·22)	0·0011
26 weeks	4·40 (2·76; n=154)	4·51 (2·82; n=128)	−0·24 (−0·73 to 0·25)	−0·10 (−0·31 to 0·11)	<0·0001
SDQ-P: conduct problems					
Baseline	2·84 (2·08)	2·72 (2·02)	..	..	..
14 weeks	2·48 (2·12; n=161)	2·44 (2·07; n=143)	−0·01 (−0·30 to 0·29)	0·00 (−0·15 to 0·14)	<0·0001
26 weeks	2·55 (2·16; n=154)	2·39 (2·14; n=128)	−0·05 (−0·36 to 0·25)	−0·03 (−0·17 to 0·12)	<0·0001
SDQ-P: hyperactivity or inattention					
Baseline	5·94 (2·89)	5·66 (2·75)	..	..	..
14 weeks	5·19 (3·01; n=161)	4·85 (3·06; n=143)	−0·04 (−0·46 to 0·37)	−0·02 (−0·16 to 0·13)	<0·0001
26 weeks	5·44 (3·13; n=154)	4·85 (2·74; n=128)	0·01 (−0·41 to 0·44)	0·00 (−0·15 to 0·16)	<0·0001

(Table 2 continues on next page)

	OSI plus therapist support (n=222)	Treatment as usual (n=221)	Adjusted mean difference (95% CI)*†	Standardised mean difference (95% CI)	p value for non-inferiority‡
(Continued from previous page)					
SDQ-P: peer relationship problems					
Baseline	2·77 (2·34)	2·67 (2·14)	..	..	..
14 weeks	2·57 (2·33; n=161)	2·22 (2·16; n=143)	0·19 (−0·12 to 0·49)	0·08 (−0·05 to 0·22)	0·0002
26 weeks	2·55 (2·27; n=154)	2·27 (2·03; n=128)	0·09 (−0·22 to 0·41)	0·04 (−0·10 to 0·18)	<0·0001
SDQ-P: prosocial behaviour					
Baseline	7·42 (2·33)	7·48 (2·24)	..	..	..
14 weeks	7·47 (2·31; n=161)	7·50 (2·20; n=143)	−0·03 (−0·34 to 0·29)	−0·01 (−0·15 to 0·13)	<0·0001
26 weeks	7·27 (2·35; n=154)	7·61 (2·34; n=128)	−0·15 (−0·48 to 0·17)	−0·07 (−0·21 to 0·08)	0·0002
SDQ-P: total score					
Baseline	17·95 (7·05)	17·26 (6·53)	..	..	..
14 weeks	15·24 (8·37; n=161)	14·13 (7·58; n=143)	−0·05 (−1·07 to 0·97)	−0·01 (−0·16 to 0·14)	<0·0001
26 weeks	14·93 (8·35; n=154)	14·02 (7·49; n=128)	−0·41 (−1·46 to 0·64)	−0·06 (−0·21 to 0·09)	<0·0001
Health economics outcomes					
Parent report on child CHU-9D (UK adult value set)					
Baseline	0·771 (0·132)	0·793 (0·119)	..	..	..
14 weeks	0·827 (0·133; n=173)	0·841 (0·117; n=163)	−0·001 (−0·023 to 0·020)	..	..
26 weeks	0·833 (0·141; n=172)	0·846 (0·112; n=162)	−0·002 (−0·025 to 0·021)	..	..
Parent report on child CHU-9D (Australia adolescent value set)					
Baseline	0·541 (0·256)	0·578 (0·234)	..	..	..
14 weeks	0·656 (0·265; n=173)	0·671 (0·243; n=163)	0·006 (−0·037 to 0·049)	..	..
26 weeks	0·675 (0·275; n=172)	0·686 (0·232; n=162)	0·006 (−0·040 to 0·053)	..	..
Parent self-report EQ-5D-5L (UK adult value set)					
Baseline	0·792 (0·215)	0·835 (0·175)	..	..	..
14 weeks	0·825 (0·224; n=173)	0·860 (0·159; n=164)	0·003 (−0·028 to 0·035)	..	..
26 weeks	0·847 (0·200; n=172)	0·871 (0·143; n=162)	−0·002 (−0·033 to 0·029)		

Data are mean (SD), unless otherwise indicated. Participants in the OSI plus therapist arm received parent-led OSI for child anxiety with therapist support. Participants in the treatment as usual arm received treatment for child anxiety problems from child mental health services. For all scales, a higher score indicates worse functioning, with the exception of the ORS where a higher score indicates better functioning. Generalised linear mixed effects model adjusted for randomised arm, assessment timepoint, baseline score, minimisation variables (child's age, gender, baseline anxiety associated interference, and service type), an interaction between randomised arm and assessment timepoint as fixed effects, and a random intercept for each participant. CAIS-C=Child Anxiety Impact Scale–child report. CAIS-P=Child Anxiety Impact Scale–parent report. CHU-9D=Child Health Utility 9D. EQ-5D-5L=EuroQol-5 dimensions, 5 levels. ORS=Outcome Rating Scale. OSI=Online Support and Intervention. RCADS-C=Revised Child Anxiety and Depression Scale–child report. RCADS-P=Revised Child Anxiety and Depression Scale–parent report. SDQ-P=Strengths and Difficulties Questionnaire-parent report. *OSI plus therapist support versus treatment as usual. †For health economics outcomes, the mean difference was adjusted for baseline values using an OLS model and was computed on complete observations. ‡Wald test; one-sided; level of statistical significance p=0·025. §Primary outcome.

Table 2: **Summary statistics, adjusted mean differences, standardised mean differences, and the p value for non-inferiority for the primary and secondary analyses**

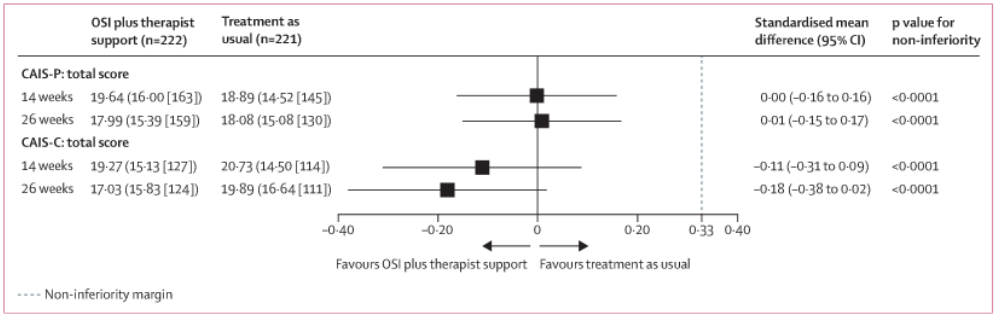

Figure 2: **Forest plot for primary and secondary outcomes: Child Anxiety Impact Scale**
Participants in the treatment as usual arm received treatment for child anxiety problems from child mental health services. Participants in the OSI plus therapist arm received parent-led OSI for child anxiety with therapist support.OSI=Online Support and Intervention. CAIS-P=Child Anxiety Impact Scale–parent report. CAIS-C=Child Anxiety Impact Scale–child report.

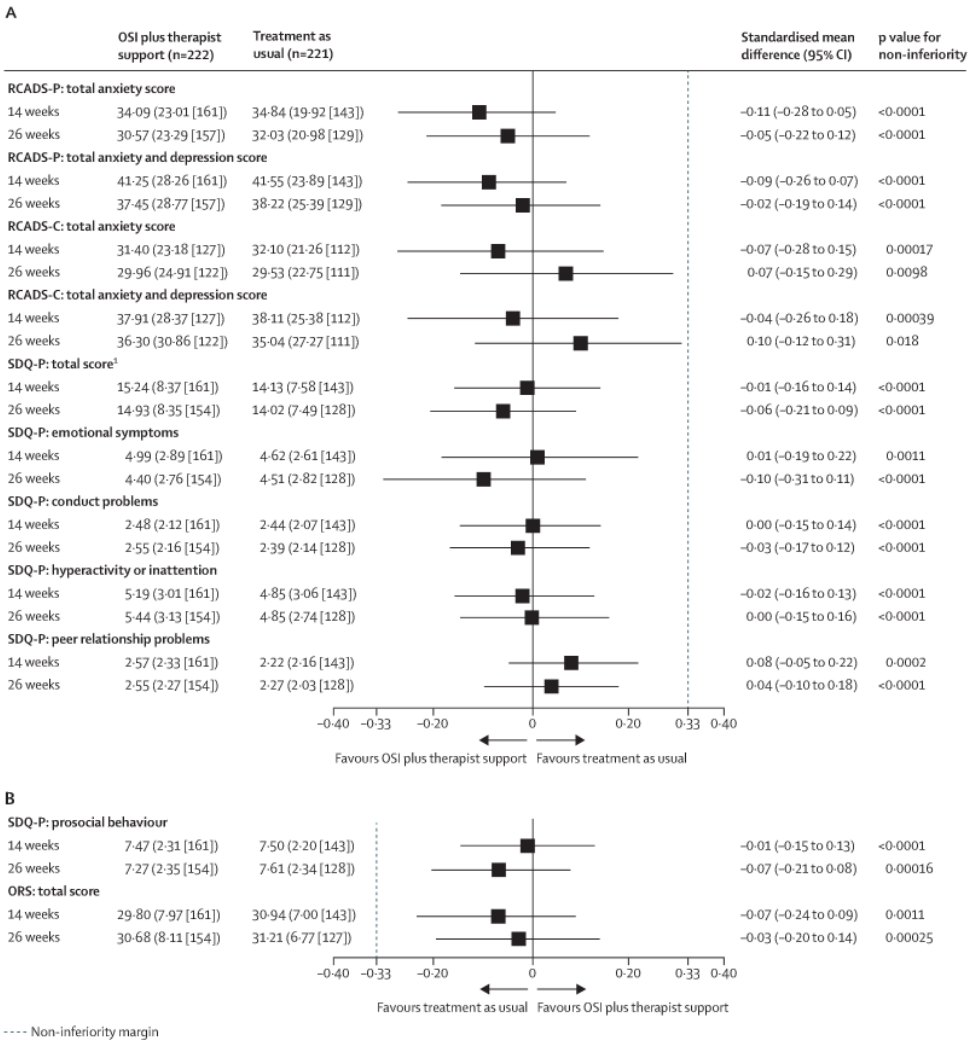

***Figure 3*: Forest plot for other secondary outcomes**

Participants in the treatment as usual arm received treatment for child anxiety problems from child mental health services. Participants in the OSI plus therapist arm received parent-led OSI for child anxiety with therapist support. Additional secondary outcomes and sensitivity analyses are described and reported in the appendix (pp 111–22). OSI=Online Support and Intervention. RCADS-P=Revised Child Anxiety and Depression Scale–parent report. RCADS-C=Revised Child Anxiety and Depression Scale–child report. SDQ-P=Strengths and Difficulties Questionnaire–parent report. ORS=Outcome Rating Scale.

therapist support (182 minutes) was 59% of the time spent delivering treatment as usual (307 minutes; appendix p 155). The various cost utility analyses across ITT and per-protocol analyses suggested that OSI plus therapist support was likely to be cost effective under certain scenarios, with the exception of the ITT analysis using the UK adult preferences dataset (appendix pp 174–80). Cost-utility analysis results were not only sensitive to the underlying values set used for deriving QALYs, but also characterised by large uncertainty surrounding the cost-effectiveness estimates. For the cost-effectiveness analysis (appendix pp 174–81), the cost-effectiveness acceptability curve, which accounts for sampling uncertainty, indicated that OSI plus therapist support is likely to be cost effective, although uncertainty remained high. However, the maximum

threshold value that society is willing to pay for a unit improvement in the CAIS-P is unknown. More details on the health economic results are presented in the appendix (pp 155–72, 174–81).

Summaries of the experiences of parents and therapists with illustrative quotes from the qualitative interviews are presented in table 3.

No serious adverse events were recorded. Ten adverse events were reported in each arm (coding by arm and details are presented in the appendix pp 123–28). The Trial Steering Committee considered all adverse events either not to relate to the treatments or trial procedures (eg, child injury after falling off bike) or to relate to routine aspects of clinical care (eg, child did not like completing standard measures).

Discussion

We found markedly similar and non-inferior treatment outcomes for the novel intervention, OSI plus therapist support, compared with the predominantly evidence-based treatment as usual. OSI plus therapist support brought substantial savings in therapist contact time and was considered to be cost-effective in several scenarios, although uncertainty in this was high. These findings highlight the potential for digitally augmented interventions like OSI plus therapist support to increase the number of children treated for anxiety problems without compromising treatment outcomes.

The novel treatment was credible to both parents and therapists, although unsurprisingly (given training was rapid and most therapists only delivered OSI plus

	Illustrative quotes	Implications
Parents' experiences of OSI plus therapist support		
Parents who initially had reservations about OSI or a parent-led approach could see the benefit once they started the programme.	"I was thinking, em, doing something online…maybe it's better to do this with speaking with somebody, like. But, actually, to be honest, actually it was much better, I found it more helpful in the end." (14P) "I didn't think it would work. You know, with like with 'me' doing it, I thought you know, she'd be better off with a professional 'cos I don't really know what I'm doing. But I mean it does help. "(19P) "I was hopeful that [my daughter] would have more involvement than she did…but if I'm honest, I got over that quite quickly 'cause the therapist was wonderful and it helped me to understand [my daughter]'s needs better. So, I saw the benefits of it once I started doing it." (04P)	It will be helpful to normalise and address any initial concerns with parents who are being offered OSI plus therapist support. Therapists would benefit from training around how to introduce this approach to parents in a way that allays any initial concerns. This could be more explicitly addressed in further developments to the OSI plus therapist support programme.
The programme was generally seen as user-friendly, well designed, and flexible, allowing parents to fit the sessions into daily life; parents appreciated being able to listen to the audio of the online content, complete sessions on a mobile phone, and download the materials.	"I'm a complete techno idiot. I'm not smart or savvy at all when it comes to tech. And if I can do it, anybody can do it." (06P) "I just thought it was really easy to navigate. It was very straightforward working through the modules." (12P) "I was washing the dishes and I was listening to it…the fact that I can come back and read the things again. I can download them. That was very good." (14P) "I could just log into it anytime that suited me. So even if it was the midnight and I was sat in bed, I could still say right, I want to get this done now." (04P)	Usability is good and for most parents there is good fit—this might provide reassurance to parents, therapists, services, and commissioners in deciding whether to receive, deliver and commission OSI plus therapist support.
Therapists were seen positively as providing support, problem-solving difficulties, and helping parents put strategies into practice.	"I had a little blockage with the step plan. She [child], she wasn't getting as far, you know she just sort of stopped at one level, and I was trying to think why that was and she [therapist] was really helpful for that." (12P) "The therapist at the time was fantastic. You know she was a massive support to me." (21P) "When I doubt myself, she picks me back up again." (09P)	Therapist support is highly valued and appears to be an essential part of treatment.
Many parents developed a sense of self-efficacy that enabled them to feel they had the tools to help their child (and other children) now and in the future.	"The course supported 'me' and empowered 'me'" (04P) "I feel confident enough that I now have the tools that if that happens with anything in life, not just the person receiving the treatment for myself and for my other child, it's changed my concept on how to deal with life as well." (09P) "I think I did learn a lot and will probably continue to evolve and learn that with her." (16P)	Parents who are being offered the programme, therapists, services, and commissioners might find it beneficial to know that the benefits appear to go beyond improvements in the child's anxiety and extend to parents' being equipped with skills to potentially manage future difficulties without the need for further professional input.
At the end of treatment, parents were generally positive about the parent-led approach and the OSI plus therapist support programme (even parents whose children were still experiencing some anxiety problems); for most parents, OSI plus therapist support led to improvements in their child's anxiety and emotional wellbeing, leading them to do things previously avoided, as well as increasing their confidence and resilience; some parents described this having a positive effect on relationships in the family.	"Where we've come from and where we are now in such a short space of time, in my eyes, is a miracle. I can't…back then I never ever dreamt that we could get to a stage where we are now. (09P) "I'd do it again in a heartbeat." (11P) "It just works, you know, so I'm, I'm really happy with it." (12P) "It hasn't particularly worked well for 'my' daughter, but I think there's a lot more going on there. But I'm so, so glad I've done it." (06P) "It's got him going back in his own bed and going to bed at a normal time without me either in there or him up all through the night." (13P) "Personally, me and my daughter are now closer. We got to a stage where we were sort of quite estranged…and we can now see a brighter future." (09P)	Providing information about the effectiveness of the treatment and parents' experiences of the programme might help parents, therapists, services, and commissioners in deciding whether to receive, deliver and commission OSI plus therapist support.
Two parents felt that they would have preferred their child to be involved in the sessions and longer face-to-face appointments where they could receive more support from the therapist; doing the programme on their own and having to manage other significant stressors in life appeared to make it difficult to engage in the programme.	"I do think there's real value of doing it face to face and involving the child." (17P) "If he'd had a bad week, I could. I mean, I can talk for England anyway, but I feel like I could have shared a lot more information…I think maybe like maybe, em, an allowance for debriefing for parents in, put into place would be good." (17P) "I think it would have been so much massively beneficial if there had been the support of the two parents… probably would have been a bit more beneficial if we were all singing off the same hymn sheet." (21P) "I'll most certainly use the techniques and I still do to a degree. But it's just so much other stuff going on to be honest with you." (21P)	It will be important to establish factors that are associated with poorer fit and outcomes for OSI plus therapist support and identify whether OSI plus therapist support could be further adapted to improve acceptability and outcomes for these families.

(Table 3 continues on next page)

	Illustrative quotes	Implications
(Continued from previous page)		
Therapists' experiences		
Generally, therapists were enthusiastic about the training and the programme in relation to its ease of use and effectiveness; therapists suggested some minor improvements and requested further training on routine outcome measures and videos illustrating the approach.	"So yeah, I felt like it was sort of good training to begin with." (02C) "I think it was really it was a great parent-led treatment definitely, and it works. And so you know, I've really enjoyed, really enjoyed, delivering it to be quite honest." (03C) "I think, generally on the whole, just kind of like, the way it was kind of easy to follow and it was really structured, the modules." (29C) "I think it's a really great way of working and I think it breaks down lots of barriers for families struggling to access treatments." (05C) "I thought would have been helpful was if the therapists, like myself, could have access to the parent website, like a test account sort of thing, so that we could actually see what they're seeing." (10C) "For those people who are kind of novice practitioners just a little 10-minute video on what each kind of ROM [routine outcome measure] means." (20C) "While it was well guided, in the instructions and the manuals, seeing in practice before would have been very helpful, I think." (02C)	In general, OSI plus therapist support is perceived as having the necessary characteristics to be implemented in services (ie, good usability and observable improvements in child anxiety). There are some minor improvements that could be made around usability, such as providing the therapist with the parent view of the programme; further training on routine outcome measures and more videos demonstrating the approach would be valued by less experienced therapists.
Therapists felt that having the questionnaires and content delivered online and being able to monitor engagement within the programme reduced burden and time for therapists; short phone calls with parents appeared to be broadly acceptable to therapists and parents.	"I can book more cases in and I can be much more flexible with them, so that's been really helpful." (05C) "I feel like a lot of the responsibility is being lifted from my shoulders because I know the information that the parent has read is good quality, accessible and I can check that they understood it." (01C) "That's what I love about the Co-CAT—so I can go on, so I might go on the day before to have a quick look." (08C) "It's easy to like track the progress with the questionnaires that they filled in and see how the scores are changing each week, so that was good. (10C) "You can build that rapport, the same way that we would anyway…and actually having check-ins with parents, 20 minutes is still enough to catch up and check in." (05C)	OSI plus therapist support is perceived to have a relative advantage over other approaches in terms of therapists' time and resources.
Some therapists were champions for the approach, voluntarily taking a particular interest in OSI plus therapist support and its adoption in the local service.	"I really, really really hope that OSI sticks around and that as a Trust we do sign up to it and that we can use it because it is like being given almost the key to the magical Kingdom that you are shown this whole other world…like Willy Wonka's Chocolate Factory and everything is brilliant and marvellous, and then go back to the way it was before…I think I would be a bit gutted to be fair." (01C) "I'm like guys it's great. You know, it's brilliant, we have to sign up for this. We need it. We need it to do well and to be rolled out across the country." (05C)	The active recruitment of champions could help spread knowledge about OSI plus therapist support and enthusiasm for the approach, and facilitate embedding it into services.
Based on small numbers of cases (therapists had only delivered OSI plus therapist support to between one and four parents), therapists expressed some ideas around who OSI plus therapist support might or might not work for; in their experience, it appeared to be acceptable to parents from multiethnic communities and those who might experience difficulties in reading or with the English language; they felt OSI plus therapist support might be less successful if children were older, had high levels of anxiety and avoidance, or were unable to articulate their worries; they also felt there were some instances where parents did not have capacity to engage in the programme and required more support.	"Something they brought up consistently is just how easy it was to understand in terms of the language. And it was easy to sort of digest information and also they had the option to have someone to narrate the text, and that was a useful function for them." (02C) "Yeah, I think for the younger ones, yes, I think it's definitely got its place for the younger ones." (23C) "As much as Mum can try and ask those questions she would say like sometimes, he just won't answer the questions, or he'll shut down when she tries to talk to him." (10C) "My sense is that I'm not sure, it works for children who aren't at school…I'm not sure if it works if the child isn't in situations where they're experiencing all their anxiety." (20C) "One of her comments was when we had the assessment was that she didn't want to be her child's therapist…then mum was a bit reluctant to start with anyway, then a few weeks in she said, oh it's too parent led. I'm gonna try something else." (23C) "I think it would really depend on the parent, so I think some parents are more suited to it than others, and some parents need that hour if you know what I mean. It can be like a therapy session for them. And some parents are that busy they just don't need, the time, just don't need you taking up the time, just need the skills…I would definitely just kinda wait and decide after I met the parents." (29C)	Therapists' initial impressions are that OSI plus therapist support appears to be acceptable to parents from multiethnic communities and those who may experience difficulties in reading or with the English language. There are initial impressions that OSI plus therapist support may be less suited to some families. Further evidence is required to determine to whom might or might not benefit from OSI plus therapist support and therefore who it should be offered. Once evidence becomes available, this should be communicated to therapists in services so that decisions are made based on the evidence.
Although they recognised the positives in using OSI plus therapist support, some therapists described preferences for, or perceived benefits in, delivering sessions in-person rather than via OSI plus therapist support. This appeared to be particularly the case for therapists who had not had professional training or were within the first year of being qualified.	"I don't mind doing online interventions, but face to face is still definitely my preference 'cause I think it's just so much easier to build that rapport and engage with someone and see how they're presenting like in front of you." (10C) "With the treatment as usual [delivering parent-led CBT in a group], it's—I probably get more out of them ones as well, I probably learn more myself, as a, as a practitioner." (29C) "I'd like to give them all the information and then get them to read the additional materials, for just, to aid more understanding. So, I'd probably talk through the anxious thoughts, the physical changes, the anxious behaviours, and then just give that [OSI plus therapist support] as additional knowledge." (08C)	Within services, there is likely to be variability in therapists' interest, motivation, values or beliefs, learning opportunities, skills and knowledge, and access to support or supervision to deliver OSI plus therapist support. If OSI plus therapist support is to be delivered by a range of therapists within services, these factors will need to be assessed and addressed via a range of strategies.

Co-CAT=Child Anxiety Treatment in the context of COVID-19. OSI=Online Support and Intervention. Parent participants are identified as P and clinician participants are labelled as C.

***Table 3*: Parents' (n=11) and therapists' (n=10) experiences regarding the acceptability of OSI plus therapist support from the qualitative interviews**

therapist support once), therapists were somewhat more comfortable delivering their usual treatment. It was also not surprising, given that OSI was provided as part of a time-limited research trial, that therapists were somewhat uncertain that they would use it again in the future; however, overall satisfaction was high. There were no serious adverse events or adverse events that were considered to relate directly to the intervention. The qualitative interviews indicated high levels of acceptability for OSI in terms of usability (for parents and therapists), effort, time, and perceived outcomes.

Therapist training in OSI plus therapist support was highly pragmatic, given the COVID-19 pandemic context, which brings both advantages and disadvantages. On the one hand, the results are particularly encouraging given the minimal training and support that therapists were given in this new online intervention. On the other hand, even better outcomes might be achieved with more substantial initial training and ongoing support and supervision. Future studies are warranted to explore the level of training required to optimise treatment outcomes. This trial included children with a broad range of anxiety problems; future studies should also explore whether outcomes differed by anxiety subtype.

We had minimal exclusion criteria for the trial and we did not require a formal diagnosis of an anxiety disorder, as this is rarely done in clinical settings.[41] An established autism diagnosis was an exclusion criteria, but many children in this age group will have not yet had a formal assessment and it is possible some participants might have received a diagnosis had this been assessed. Indeed, 15% of the children in our sample had a recognised special educational need, 32% of whom had difficulties with communicating and interacting, and 32% had difficulties with sensory or physical needs.

This study had various limitations, including that the researchers who collected data were not blind to treatment arm due to some differences in the therapist-reported data that was collected between arms. We used participant-reported outcomes, as is typical in routine practice, and we prioritised parent report as some of the outcome measures have not been validated with children as young as 5 years and in line with recent guidance.[40] This choice of reported outcome brings risk of bias from the parent, but we were encouraged that the same pattern of results was found across parent and child report measures and most of our child self-report measures somewhat favoured OSI plus therapist support, despite children not having direct therapist contact. This evaluation was conducted in a large number of routine child mental health services by a large and varied group of therapists as part of their routine caseloads. This is a strength in terms of learning about real-world implementation and no doubt contributed to this trial having a relatively diverse sample in terms of family income and parent education, but it also brings challenges associated with the demands on busy clinical teams. Given that all participating clinicians were taking part in this trial as part of their routine clinical work, there was a limit to how much information we could collect on the nature of the interventions delivered and the integrity of delivery of particular treatment models. It is possible that the COVID-19 pandemic context might have compromised delivery of treatment as usual, with the majority of contacts occurring remotely, however clinicians reported high satisfaction with their 'as usual' approach and were highly likely to continue to work in the same way beyond the trial. Clinical teams were responsible for identifying potential families for the study and we were therefore unable to obtain detailed information about families who did not participate. As such, we do not know whether the relatively low ethnic diversity among participants and therapists reflects the broader characteristics of participating clinical teams or reflects a bias in who was willing or able to participate in the trial. We relied on clinical teams to deliver the treatments in a timely manner and to report on the treatment provided, but other pressures, such as high staff turnover, presented considerable challenges; only 79% of families enrolled into the trial started treatment within 12 weeks of randomisation, and 13% did not receive any treatment within 26 weeks. However, a range of sensitivity analyses provided consistent results, which adds confidence in the robustness of the results. More participants started treatment within 12 weeks of randomisation in the OSI plus therapist support arm than the treatment as usual arm, and the families receiving OSI plus therapist support attended more sessions. This better engagement in treatment might partly explain the better overall trial retention in the OSI plus therapist support arm than in the treatment as usual arm, which leads to the need for caution in interpreting results, particularly in the per-protocol analyses. Encouragingly, most baseline demographic and clinical variables were not associated with trial attrition. The exception was that fewer participants with missing data were partnered or cohabiting, probably reflecting the fact that participating in the trial might have been particularly burdensome for single parents, as supported by the qualitative interviews. Finally, although overall the primary economic analyses results (cost-utility analyses, which are those more likely to inform policy making) indicated that OSI plus therapist support is likely to be cost effective under several scenarios, these analyses need to be considered with caution, due to their sensitivity to the underlying values sets used for deriving QALYs, and the large uncertainty surrounding the cost-effectiveness estimates (see appendix pp 184–87 for a full discussion of the health economics results).

This trial presents compelling clinical evidence and promising cost-effectiveness evidence that digitally augmented psychological therapies with therapist support can increase efficiencies in and access to child mental health services without compromising patient outcomes. Efforts are now needed to take full advantage of the opportunity that digitally augmented psychological treatments can bring to drive a step change in children's mental health services, learning from successful examples of digital implementation elsewhere in health services.[49]

Contributors

CC took overall responsibility for all aspects of the study. PW led on the qualitative aspects, LMY on statistical analyses, and MV on health economics. CC, LT, PW, MV, VR, CH, and LMY contributed to conceptualisation, funding acquisition, and methodology. LT, SG, SH,

LR, EW, EB, and FK contributed to methodology, investigation, data collection, and project administration. JvS contributed to data curation. LMY, NW, SM, VH, FK, PW, MV, JP, and SY contributed to formal analysis and visualisation. All authors contributed to writing, reviewing, and editing. JvS, NW, SM, VH, MV, SY, JP, and LM directly accessed and verified the underlying data reported in the manuscript. All authors have full access to all the data in the study and accept final responsibility to submit for publication.

Declaration of interests

CC is the author of a book for parents that is used in many of the participating clinical teams to augment treatment as usual for child anxiety problems and receives royalties from sales. CC and CH are developers of the OSI platform. They do not receive any personal financial benefits from the use of OSI. All other authors declare no competing interests.

Data sharing

Deidentified individual participant data, a data dictionary, and the analysis code will be made available on an open access data repository accompanied by the study protocol and the statistical analysis plan as soon as possible after publication; for more information contact the corresponding author.

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Appendix E. Paper 5

Pollard, J., Agnew, E., Pearce-Smith, N., Pouwels, K. B., Salant, N., Robotham, J. V., & REVERSE Consortium. (2025). **Umbrella review of economic evaluations of interventions for the prevention and management of healthcare-associated infections in adult hospital patients**. Journal of Hospital Infection. URL: https://doi.org/10.1016/j.jhin.2025.01.006

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Review

Umbrella review of economic evaluations of interventions for the prevention and management of healthcare-associated infections in adult hospital patients

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SUMMARY

Background: Healthcare-associated infections (HCAIs) result in worse outcomes for patients and greater financial burden. An estimated 4.8 million HCAIs occurred in hospitals across Europe in 2022–23. Sixty-four percent of antibiotic-resistant infections in Europe are associated with healthcare. It is therefore vital to identify cost-effective interventions.

Aim: To summarize the cost-effectiveness evidence of interventions addressing HCAIs in hospitals.

Methods: An umbrella review was conducted to identify evidence on the cost-effectiveness of antimicrobial stewardship, infection prevention and control, and microbiology and diagnostic stewardship interventions for the prevention and clinical management of HCAIs in adult hospital patients. Medline, Embase, and EconLit databases were searched. A qualitative synthesis was undertaken.

Findings: Twenty-four systematic reviews met the inclusion criteria, with 101 separate analyses extracted and grouped into 10 intervention and 14 infection/organism categories, across various countries and settings. Most evidence focused on screening followed by contact precautions, isolation and/or decolonization, with selective screening most cost-effective. Most infection prevention and control bundles were cost-effective, although interventions were heterogeneous. The evidence base was sparse for the remaining intervention categories, with more research required. The limited evidence suggests that standalone environmental cleaning, hand hygiene, diagnostics, surveillance, antimicrobial stewardship, and decolonization interventions were mostly cost-effective. The cost-effectiveness of standalone personal protective equipment, and education and training interventions was mixed. Most interventions focused on meticillin-resistant *Staphylococcus aureus* and other Gram-positive infections, with more research needed on Gram-negative infections. The comparator was unclear in many extracted analyses.

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Conclusions: Cost-effective interventions to address HCAIs in hospitals exist, although more evidence is needed for most interventions.

Introduction

Healthcare-associated infections (HCAIs) are defined as 'an infection occurring in a patient during the process of care in a hospital or other health care facility, which was not present or incubating at the time of admission. This includes infections acquired in the hospital, but appearing after discharge, and occupational infections among staff of the facility' [1]. Data collected by the European Centre for Disease Prevention and Control estimated that 4.8 million HCAIs occurred annually in acute care hospitals across Europe in 2022–23, which was up from 4.5 million in 2016–17 and 3.2 million in 2011–12 [2–4]. HCAIs lead to worse outcomes for patients, including poorer health, longer hospital stays and even death, resulting in greater financial burden for healthcare payers. Furthermore, HCAIs are often resistant to antibiotics, with 64% of all infections with antibiotic-resistant bacteria across Europe associated with healthcare [5]. Antibiotic-resistant infections resulted in almost 36,000 attributable deaths across Europe in 2020 [6].

There is therefore a clear need for cost-effectiveness evidence to support decision-making on the most appropriate interventions for the prevention and clinical management of HCAIs, and infections resistant to antibiotics, in the context of limited healthcare budgets. Capturing intervention outcomes with a generic measure, such as quality-adjusted life-years (QALYs) or disability-adjusted life-years (DALYs), as well as condition-specific measures, such as infections avoided, is vital for decision-makers to rationally allocate resources. It is also important to consider the context of the intervention, as an intervention that is cost-effective in one setting is not necessarily cost-effective in another and may depend on factors such as the incidence of infections, prevalence of antibiotic resistance, and organization of healthcare systems.

Multiple systematic reviews have recently been published examining the cost-effectiveness of different interventions tackling HCAIs and antimicrobial resistance (AMR) [7–12]. An umbrella review (i.e. a systematic review of systematic reviews) was therefore appropriate to meet the aims of this review [13]. Specifically, the objective was to summarize the cost-effectiveness evidence of interventions addressing HCAIs and AMR in hospitals. To achieve this objective, we sought to answer the following research question: What is the cost-effectiveness of antimicrobial stewardship (ABS), infection prevention and control (IPC), and microbiology and diagnostic stewardship (MDS) for the prevention and clinical management of HCAIs in adult hospital patients, with a particular focus on infections resistant to antibiotics?

Methods

The umbrella review was registered on PROSPERO (CRD42024544387) [14]. A rapid methodological approach was applied according to best practice [15]. The review was reported in line with the latest Preferred Reporting Items for Systematic reviews and Meta-Analyses (PRISMA) guidelines (Supplementary Appendix S2) [16].

Eligibility criteria

The PICO (Participants, Interventions, Comparisons, Outcomes) framework was used to establish the eligibility criteria. Retrieved records were included if they met the following criteria: peer-reviewed systematic review; adult hospital patients; ABS, IPC and MDS interventions for the prevention and clinical management of HCAIs, including infections resistant to antibiotics, compared to an alternative intervention option (e.g. standard practice, no intervention); reporting cost-effectiveness evidence. Reviews that only considered low- and/or middle-income countries were excluded.

ABS is a coherent set of integrated actions with the aim to promote the responsible and appropriate use of antimicrobials, such as education, auditing, broad-spectrum antibiotic restrictions, and shortening of treatment duration. IPC consists of interventions such as guidance and standards, education and training, hand hygiene, personal protective equipment (PPE), environmental cleaning and governance. MDS consists of interventions such as microbiological testing and rapid diagnostic workflows to enable targeted therapy of appropriate antimicrobials.

The PROSPERO protocol reports the inclusion/exclusion criteria in detail [14].

Identification of reviews

Four key concepts were used to create the search strategy: (1) HCAI, (2) AMR, (3) economic evaluation and (4) systematic review. Search strategies were created by an information scientist, N. Pearce-Smith, and peer-reviewed by a second information scientist. Known papers were used to test the effectiveness of the search strategy. Databases were searched using the following strategy: (1 OR 2) AND 3 AND 4. The full search strategy is presented in Supplementary Appendix S3. OVID Medline ALL (1946 to April 9th, 2024), OVID Embase (1974 to April 9th, 2024) and EBSCO EconLit (1996 to April 3rd, 2024) were searched on April 10th, 2024 with no date or language limits to identify studies. Deduklick was used to deduplicate studies [17]. Included reviews were used for backward and forward citation searching using citationchaser [18]. Records were exported to EndNote for deduplication and to identify systematic reviews to be screened.

All records were screened by at least one reviewer. A randomly selected 20% sample of records was independently screened on title/abstract and full text in Rayyan by E.A. and J.P. to ensure that the inclusion/exclusion criteria were applied appropriately, with disagreements resolved by consensus [19]. The remaining records were screened by J.P.

Reasons for exclusion were documented at full text review (Figure 1).

The corrected covered area (CCA) was calculated to measure the level of publication overlap in the included reviews, i.e. the same underlying studies being included in multiple reviews. The CCA divides the frequency of repeated occurrences of the index publication (i.e. the first occurrence of an underlying study) in other reviews by the product of index publications and reviews, reduced by the number of index publications [20]. The CCA can vary from 0%, no overlap of underlying studies, to 100%, complete overlap. Overlap of 0–5% is considered 'slight', 6–10% 'moderate', 11–15% 'high', and >15% 'very high', and the selection of reviews should be critically assessed when the degree of overlap is substantial [21]. Only underlying studies that contributed to the data extracted for this umbrella review were included in the CCA calculation.

Data extraction

A bespoke data extraction form was created in Excel and finalized after piloting. Specific data extraction items are reported in Supplementary Appendix S4. Data from a randomly selected 20% of included reviews was independently extracted by E.A. and J.P. to ensure that the appropriate data were extracted, with disagreements resolved by consensus. J.P. extracted data from the remaining reviews.

Data were extracted separately for each intervention–infection/organism combination reported by a review. For example, if a review reported the cost-effectiveness of hand hygiene interventions for HCAIs, environmental cleaning for HCAIs, and decolonization for meticillin-resistant *Staphylococcus aureus* (MRSA), data were extracted for each of the analyses in three separate rows in the data extraction form.

Quality assessment

The quality of included reviews was assessed using the JBI Checklist for Systematic Reviews and Research Synthesis [22]. The critical appraisal tool is made up of 11 items, each with one of four options: yes, no, unclear, not applicable. To assess the overall quality of a review, a score of 1 was awarded for a yes, 0 for a no or unclear, and non-applicable items were excluded. A review scoring >70% was classified as high quality, 50–70% medium quality, and <50% low quality [23,24].

Quality was assessed independently by E.A. and J.P. for a randomly selected 20% of included reviews to ensure the checklist was applied appropriately, with disagreements resolved by consensus. J.P. assessed the quality of the remaining reviews.

Data synthesis

A qualitative synthesis was undertaken given the heterogeneity in the geographies, settings, conditions, and interventions of interest, and the natural variation in country-specific unit costs and different cost-effectiveness guidelines.

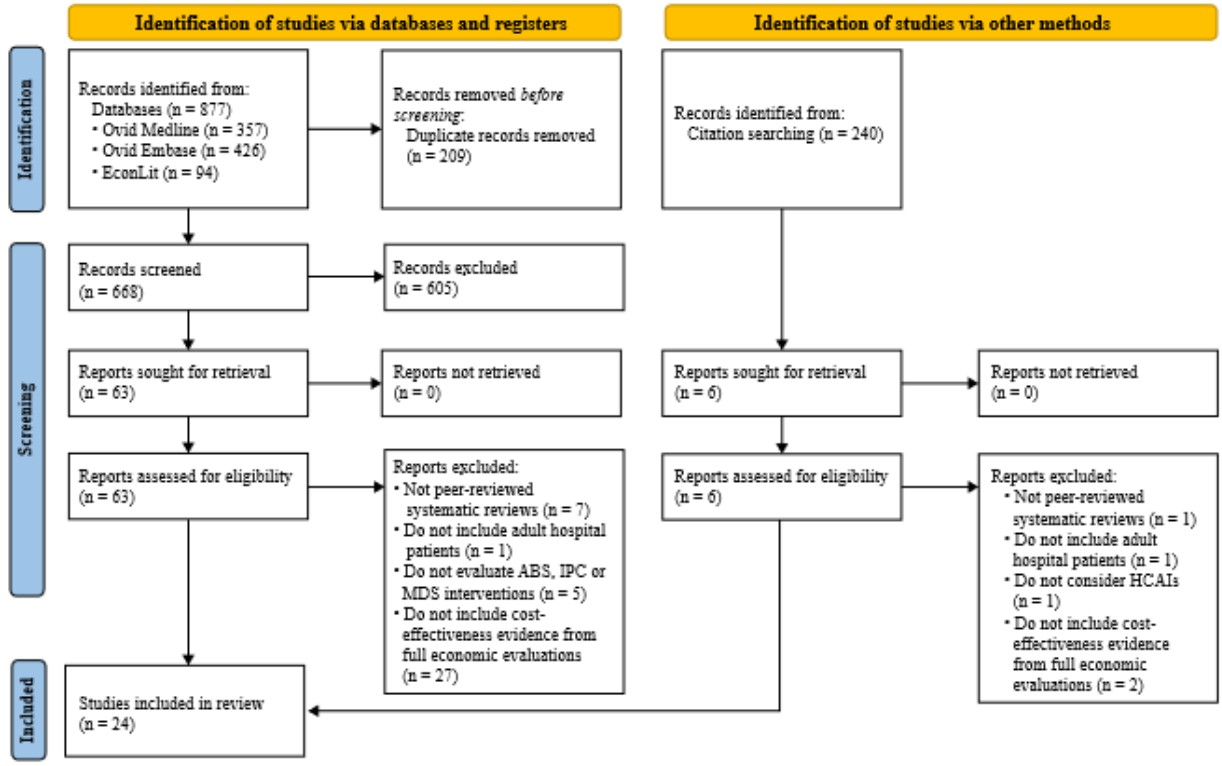

Figure 1. PRISMA flow diagram for identification of studies. ABS, antimicrobial stewardship; IPC, infection prevention and control; MDS, microbiology and diagnostic stewardship; HCAI, healthcare-associated infection.

Therefore, quantitative data analysis was not undertaken. Instead, descriptive conclusions on the cost-effectiveness of different interventions were drawn, based on the evidence presented in the included systematic reviews. Interested readers can find the quantitative data extracted from the included reviews in the full data extraction provided in Supplementary Appendix S6.

Interventions that were conceptually similar were grouped into one of 10 categories: (1) ABS; (2) screening followed by contact precautions, isolation, decolonization, or a combination of these; (3) IPC bundle (i.e. multimodal interventions incorporating more than one infection control activity); or standalone (4) decolonization (i.e. interventions to eliminate or reduce patient colonization); (5) diagnostics (i.e. method of diagnosis); (6) environmental cleaning (i.e. cleaning and disinfection of surfaces and equipment); (7) hand hygiene (i.e. cleaning hands); (8) PPE (i.e. specialist clothing or equipment); (9) surveillance (i.e. ongoing monitoring of HCAIs); and (10) education and training (i.e. material to promote appropriate behaviours and procedures).

HCAIs were grouped into one of 14 infection/organism categories, depending on the level of detail reported by the review: (1) multidrug-resistant organisms (MDROs); (2) carbapenem-resistant organisms (CROs); (3) MRSA; (4) vancomycin-resistant enterococci (VRE); (5) extended-spectrum β-lactamase-producing *Klebsiella pneumoniae* (ESBL-KP); (6) *Clostridioides difficile* infection (CDI); (7) HCAI; (8) urinary tract infection (UTI); (9) surgical site infection (SSI); (10) bloodstream infection (BSI); (11) BSI and pneumonia; (12) pneumonia; (13) influenza; (14) sepsis. HCAIs were categorized based on how the HCAI of interest was reported in the given systematic review. The infection/organism categories include various levels of detail because of the differences in reporting results across the included systematic reviews. For example, MDROs and CROs were both included as separate categories, even though CROs are a subset of MDROs, because some reviews reported results across all MDROs of interest, whereas others focused on specific subsets. Similarly, HCAI was included as a category since — although all other included categories are a subset of HCAIs — some reviews reported results across all HCAIs of interest, so it was not feasible to disaggregate the infection/organism any further.

The quantity of evidence was summarized in a heat map, with infection/organism on the vertical axis, intervention on the horizontal axis, and a count for the number of analyses extracted for each intervention–infection/organism combination (Figure 2). Darker shades signify a higher count. Although some of the intervention–infection/organism

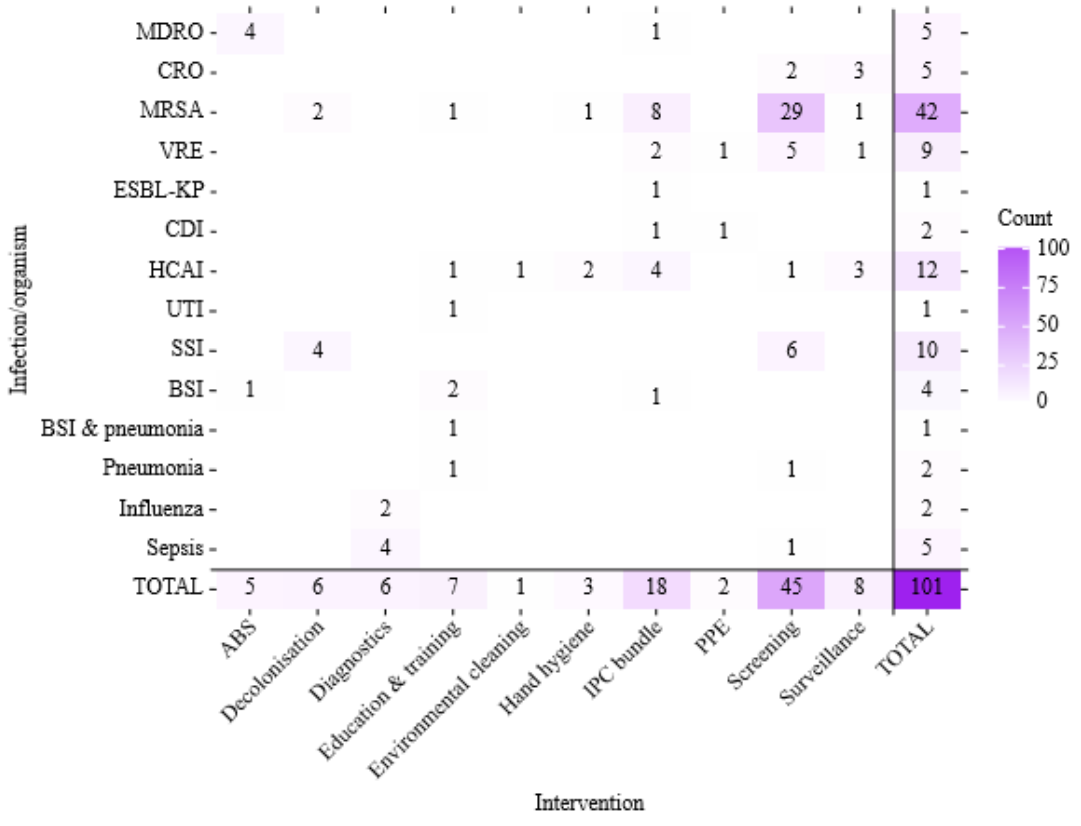

Figure 2. Quantity of evidence heatmap by intervention and infection/organism category. ABS, antimicrobial stewardship; IPC, infection prevention and control; PPE, personal protective equipment; MDRO, multidrug-resistant organism; CRO, carbapenem-resistant organism; MRSA, meticillin-resistant *Staphylococcus aureus*; VRE, vancomycin-resistant enterococci; ESBL-KP, extended-spectrum β-lactamase-producing *Klebsiella pneumoniae*; CDI, *Clostridioides difficile* infection; HCAI, healthcare-associated infection; UTI, urinary tract infection; SSI, surgical site infection; BSI, bloodstream infection.

combinations in Figure 2 are not applicable or feasible in practice (e.g. decolonization for ESBL-KP), the figure succinctly provides a comprehensive and detailed summary of the quantity of evidence identified.

The cost-effectiveness result of each extracted analysis was grouped into one of three mutually exclusive categories: yes – intervention was cost-effective; mixed – cost-effectiveness was context dependent; no – intervention was not cost-effective. The cost-effectiveness results were also summarized in a heat map, with a count of each cost-effectiveness category for the intervention–infection/organism combination (Figure 3). Darker shades signify a higher count. Analyses were excluded from the cost-effectiveness heat map if the cost-effectiveness comparator was unclear, or if the intervention and comparator were grouped in the same intervention category (e.g. polymerase chain reaction (PCR) compared to culture test for screening MRSA). If a review reported that it found no cost-effectiveness evidence for a particular intervention it was also excluded from the heat map.

The narrative write-up was presented by intervention category and organized by infection/organism within this. Information is provided on intervention, cost-effectiveness, comparator and setting, if it was more specific than hospital-wide. An overview of the synthesized findings for each intervention category is presented in Table I, which includes information on the quantity (i.e. number of reviews included) and quality (i.e. overall score on the JBI checklist) of evidence.

Results

Figure 1 shows that a total of 668 unique records were retrieved through the database searches, with 605 excluded after title/abstract screening. Of the 63 records screened on full text, 23 reviews were included. Backward and forward citation searching of these 23 reviews identified 240 unique records. Two hundred and thirty-four records were excluded after title/abstract screening and five were excluded after full text screening. Therefore, a total of 24 reviews were included for synthesis in this umbrella review.

Study characteristics

Characteristics of the included systematic reviews are presented in Supplementary Appendix S1, with full data extraction provided in Supplementary Appendix S6.

Publication year ranged from 2003 to 2024, with most (N = 13) of the reviews published from 2020 onwards. Search term periods ran up to 2023 for two reviews. Half of the reviews did

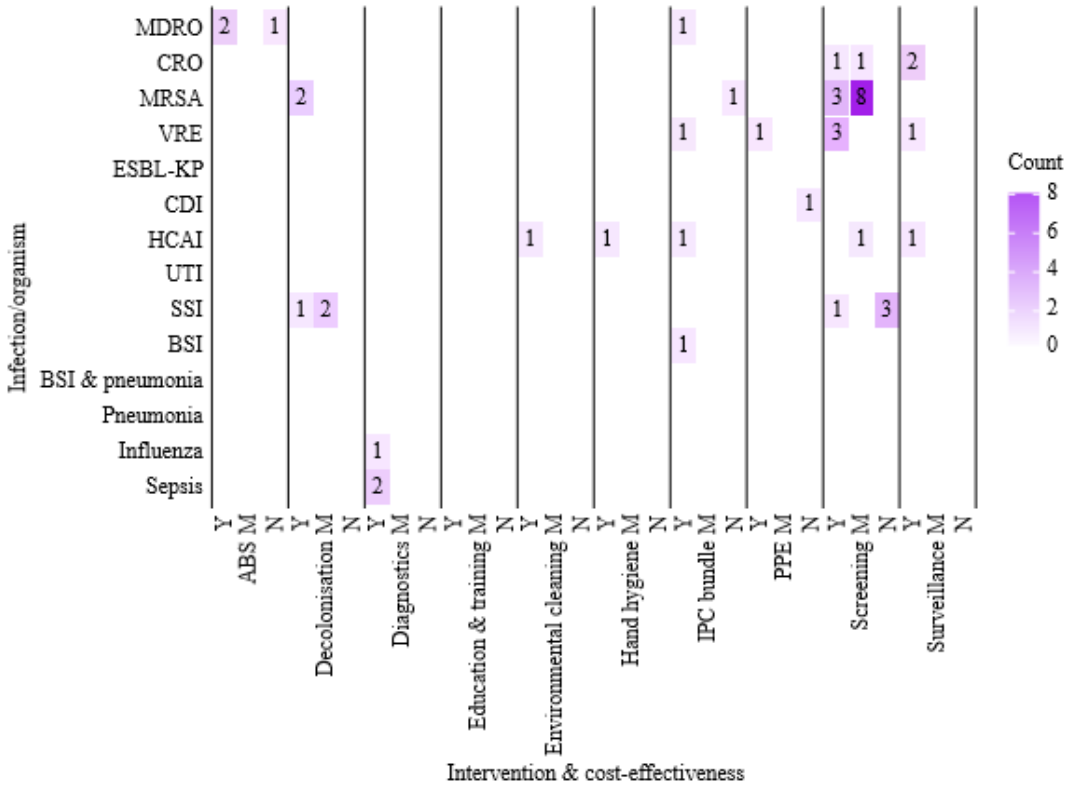

Figure 3. Cost-effectiveness heatmap by intervention and infection/organism category. Y, yes, intervention was cost-effective; M, mixed, cost-effectiveness was context dependent; N, no, intervention was not cost-effective; ABS, antimicrobial stewardship; IPC, infection prevention and control; PPE, personal protective equipment; MDRO, multidrug-resistant organism; CRO, carbapenem-resistant organism; MRSA, meticillin-resistant *Staphylococcus aureus*; VRE, vancomycin-resistant enterococci; ESBL-KP, extended-spectrum β-lactamase-producing *Klebsiella pneumoniae*; CDI, *Clostridioides difficile* infection; HCAI, healthcare-associated infection; UTI, urinary tract infection; SSI, surgical site infection; BSI, bloodstream infection.

Table I
Results of the qualitative data synthesis

Intervention	No. reviews included	Reference(s)	Synthesized findings	Quality of evidence
ABS	4	[7,8,11,26]	Generally, ABS interventions were cost-effective, including dedicated ABS team consultations. Cost-effectiveness of point-of-care test guided antibiotic prescribing compared to standard practice was context dependent.	All four reviews were rated as high quality.
Decolonization	4	[7,8,27,28]	Generally, decolonization was cost-effective, including universal decolonization for MRSA in ICU compared to no decolonization, and decolonization for MRSA compared to patient isolation. Cost-effectiveness of preoperative and perioperative decolonization for SSI compared to standard practice was context dependent.	All four reviews were rated as high quality.
Diagnostics	2	[29,30]	Diagnostic interventions were cost-effective, including molecular and procalcitonin testing compared to standard practice, and point-of-care testing compared to clinical judgement and laboratory testing.	Both reviews were rated as medium quality.
Education and training	3	[9,27,31]	Evidence on the cost-effectiveness of standalone education and training interventions was mixed, with one review not finding any studies that met their minimum quality criteria. Simulation-based education interventions were cost-effective, whereas the cost-effectiveness of local protocols, guidelines and patient safety programmes was context dependent. The cost-effectiveness of a national guideline for MRSA was not cost-effective. The comparator was unclear for all interventions.	All three reviews were rated as high quality.
Environmental cleaning	1	[9]	Environmental cleaning interventions were cost-effective compared to standard practice.	The review was rated as high quality.
Hand hygiene	3	[9,27,32]	Hand hygiene improvement interventions were cost-effective for healthcare workers and patients, compared to standard practice.	All three reviews were rated as high quality.
IPC bundle	9	[7,9,27,31–36]	IPC bundle interventions were mostly cost-effective, although there was considerable heterogeneity between the interventions that were included in the nine reviews. The comparator was also unclear in many of the analyses.	All nine reviews were rated as high quality.
PPE	2	[7,9]	Evidence on the cost-effectiveness of PPE is mixed. Gown and glove usage for VRE was cost-effective in ICU compared to glove usage alone, whereas glove and gown usage for CDI was not cost-effective for healthcare workers or visitors compared to standard practice.	Both reviews were rated as medium quality.
Screening followed by contact precaution, isolation,	18	[7–9,28,30–43]	Selective screening of high-risk patients or settings was cost-effective compared to do nothing and, in many cases, universal screening. Universal screening was cost-	Fifteen of the reviews were rated as high quality, with the other three of

decolonization, or a combination			effective in some contexts, particularly where prevalence and transmission of the infection/organism is high. Evidence on the cost-effectiveness of screening and decolonization was mixed, with results highly context dependent. Rapid diagnostic screening was cost-effective compared to standard screening techniques.	medium quality.
Surveillance	5	[7–10,12]	Surveillance interventions were cost-effective compared to standard practice and do nothing. Surveillance of CROs in ICU was cost-effective compared to do nothing, as was surveillance of VRE compared to standard practice and do nothing. Cost-effectiveness of WGS compared to standard techniques was context dependent.	All five reviews were rated as high quality.

ABS, antimicrobial stewardship; IPC, infection prevention and control; PPE, personal protective equipment; ICU, intensive care unit; MRSA, meticillin-resistant *Staphylococcus aureus*; VRE, vancomycin-resistant enterococci; CRO, carbapenem-resistant organism; CDI, *Clostridioides difficile* infection; SSI, surgical site infection; WGS, whole-genome sequencing.

not explicitly state the geographical area of interest ($N = 12$), although among those that did, most considered global evidence ($N = 8$). Most reviews considered hospital interventions across all specialties ($N = 18$), while others focused on certain specialties, such as orthopaedics and medical or surgical units. Half of the reviews only reported results for one intervention type ($N = 12$), while several considered five or more ($N = 3$).

Eighteen reviews presented results for a single infection/organism, whereas the others considered multiple ($N = 6$). Some reviews reported results for HCAIs in general, whereas others focused on specific infections or organisms. The number of underlying studies included in each review varied from five to 383. However, not all underlying studies provided cost-effectiveness evidence, as some reviews also considered other outcomes, such as clinical effectiveness. The CCA, which measures the level of overlap in the underlying studies, was 1.5%, which equates to slight overlap (i.e. 0–5%) and is therefore within acceptable limits [20,21].

Quality assessment

Results of the quality assessment of the included reviews are provided in Appendix S5, broken down by each of the 11 items of the JBI Checklist for Systematic Reviews and Research Synthesis. Twenty of the 24 reviews were classified as high quality, with six scoring 100%, five scoring 90% and nine scoring 80%. The four other reviews were of medium quality, with scores ranging from 50% to 70%.

Information on the quality assessment tool used in each review, and the overarching results of the assessment, are provided in the full data extraction (Supplementary Appendix S6). Well-established quality assessment tools were used by most reviews ($N = 18$) to assess the risk of bias in their included studies, such as the Consolidated Health Economic Evaluation Reporting Standards (CHEERS) checklist [25]. However, several created a bespoke tool ($N = 4$) and two reviews did not conduct any formal quality assessment. In summary, among the reviews that provided an overall summary of their quality assessment, the quality of the underlying studies varied. Only three reviews, all published from 2017 onwards, reported an overall high quality of evidence. Four reviews reported that the quality of evidence was generally low, and over half of the included studies in one review were excluded from the synthesis as they did not meet the minimum quality criteria. Most reviews reported that the quality of evidence was moderate, or moderate to low. Other reviews reported overall quality assessment scores but did not comment on whether it equated to low-, medium- or high-quality evidence.

Data synthesis

In total, 101 analyses were extracted from the 24 reviews, with data extracted separately for each intervention–infection/organism combination reported in each review.

Figure 2 shows the quantity of evidence identified for each intervention and infection/organism in a heat map. Screening followed by contact precautions, isolation, decolonization, or a combination of these, was the most common intervention, making up 45% of the analyses, followed by IPC bundle (18%), surveillance (8%), and education and training (7%). Evidence was most limited for environmental cleaning (1%) and PPE (2%).

The most common infection/organism was MRSA, accounting for 42% of the analyses. This was followed by analyses that reported on HCAI (12%) and SSI (10%). ESBL-KP, UTI, and BSI and pneumonia were each considered in only one analysis (1%).

A tabular summary of the qualitative data synthesis of the 101 analyses is provided in Table I. The heat map in Figure 3 presents the cost-effectiveness results graphically for the 45 analyses where the cost-effectiveness comparator was known and was not grouped in the same intervention category as the intervention.

A detailed summary of findings is presented below, organized by intervention category, and by infection/organism within this. An overarching summary of the results is then given.

Antimicrobial stewardship

Four of the 24 reviews considered the cost-effectiveness of ABS interventions, presenting five intervention–infection/organism combinations. All four reviews were rated high quality. Point-of-care (POC) test guided antibiotic prescribing was found to be cost-effective for MDROs compared to standard practice in one review [7], although cost-effectiveness varied by the country of interest in another review, with the comparator unclear [8]. The intervention was not cost-effective in a third review due to costs and antibiotic prescribing increasing in a maternity unit, when compared to standard practice [11]. Another review found that dedicated ABS team consultations were cost-effective for BSIs, although the comparator was unclear [8]. Finally, a fourth review also found that ABS interventions were cost-effective for MDROs compared to standard practice [26].

Decolonization

Four high-quality reviews presented evidence on the cost-effectiveness of standalone decolonization interventions, including six intervention–infection/organism combinations. In the context of MRSA, universal decolonization in intensive care unit (ICU) patients was cost-effective compared to interventions without decolonization, with decolonization also cost-effective compared to patient isolation [7,8]. Perioperative decolonization for SSI was cost-effective versus placebo in gastrointestinal surgery [27], although for cardiothoracic surgery the cost-effectiveness depended on willingness-to-pay (WTP) per SSI prevented, when compared to doing nothing [28]. One review also reported that preoperative decolonization for SSI was cost-effective for surgery, although the comparator was unclear, and in orthopaedic surgery it was only found to be cost-effective under certain efficacy and compliance scenarios, when compared to standard practice [28].

Diagnostics

Two reviews considered the cost-effectiveness of standalone diagnostic interventions, presenting evidence on six intervention–infection/organism combinations. Both reviews were rated medium quality. Molecular testing, including PCR, was cost-effective for sepsis when compared to standard practice, as was procalcitonin testing [29]. Similarly, POC testing for influenza and respiratory syncytial virus was cost-effective compared to clinical judgement and laboratory testing [30].

Education and training

Three high-quality reviews synthesized evidence on the cost-effectiveness of standalone education and training interventions, providing evidence on seven intervention–infection/organism combinations. One review did not find any studies that met the minimum quality criteria necessary for inclusion in their synthesis [9]. Another found that national guidelines for the prevention and control of MRSA in Germany were not cost-effective [31].

Simulation-based education interventions were cost-effective for central-line-associated BSIs in ICUs and hospitals more widely, as was an ICU patient safety programme, whereas guidelines to reduce the use of catheters were not cost-effective for urinary tract infections (UTIs) [27]. Finally, an oral care protocol in surgical ICUs was cost-effective in the context of ventilator-associated pneumonia [27]. However, the comparator was unclear for all education and training interventions.

Environmental cleaning

One high-quality review provided evidence of the cost-effectiveness of standalone environmental cleaning for HCAIs, finding that such interventions were cost-effective when compared to standard practice [9].

Hand hygiene

Three high-quality reviews summarized evidence on standalone hand hygiene interventions, each considering one intervention–infection/organism combination. One review found that hand hygiene interventions were cost-effective for HCAIs in hospitals, although the comparator was unclear [27]. Another review reported that they were cost-effective for healthcare workers (HCWs) and patients, but not for visitors, when compared to standard practice [9]. Finally, a third review found that hand hygiene interventions were cost-effective when considering MRSA in medical and surgical units, although the comparator was unclear [32].

IPC bundle

Nine high-quality reviews synthesized evidence on IPC bundles, including a total of 18 intervention–infection/organism combinations. One review reported that a proactive IPC programme – incorporating enhanced hand hygiene, environmental cleaning, increased nurse-to-patient ratio, and replacement of disposable supplies – was cost-effective compared to doing nothing for MDROs [7].

Another review found that four different IPC bundles were cost-effective for MRSA, including an ICU search-and-destroy policy, targeted surveillance alongside contact isolation, dedicated IPC personnel, pre-emptive isolation and screening without decolonization, and two other interventions combining various components of IPC [31]. A third review found that two IPC bundles for MRSA, including a search-and-destroy policy, were cost-effective, although another intervention that implemented Centers for Disease Control and Prevention contact precaution recommendations was not cost-effective, compared to glove use alone [32]. An IPC bundle made up of hand hygiene, PPE, and single room isolation to tackle MRSA

was cost-effective, as was another IPC bundle addressing MRSA in a university hospital [33,34]. However, the comparator was unclear for all but one of the analyses considering MRSA.

An IPC bundle of 15 interventions for VRE was cost-effective in an oncology ward compared to screening and contact precautions only, as was a similar IPC bundle for VRE in medical and surgical units, although the comparator was unclear [32,35]. An IPC bundle to address an outbreak of ESBL-KP was cost-effective, although the comparator was unclear [27]. Similarly, a multimodal IPC bundle for CDI was cost-saving from the hospital perspective and produced better health outcomes [36]. Again, the comparator was unclear.

With respect to HCAIs, one review found that two different IPC bundles, one including hand hygiene, oral care, and central-line catheter care, were cost-effective, although the comparator was unclear [27]. Similarly, multimodal IPC bundles tackling HCAIs were cost-effective compared to standard practice [9]. However, another review found that the cost-effectiveness of an HCAI IPC bundle including hand hygiene, cultural change, surveillance, education, and training depended on the geographical region [36]. The same review also reported that an IPC bundle incorporating a care bundle, culture change, and performance feedback was cost-effective for BSIs when compared to standard practice [36].

Personal protective equipment

Two high-quality reviews presented evidence on standalone PPE interventions, each considering one intervention–infection/organism combination. One found that gown and glove usage for VRE in ICU was cost-effective compared to glove usage alone [7]. However, another reported that glove and gown usage to tackle CDI was not cost-effective compared to standard practice, for both HCWs and visitors [9].

Screening

Eighteen reviews synthesized evidence on screening followed by contact precautions, isolation, decolonization, or a combination of these. Fifteen of the reviews were high quality and three were medium quality. These studies included 45 intervention–infection/organism combinations.

Twenty-nine of these combinations considered MRSA. Selective screening of high-risk patients or settings was the most cost-effective approach [7,8,36–38], although universal screening was cost-effective in some instances where the prevalence of MRSA was high [37,38].

One review reported that universal rapid PCR screening for MRSA at admission was not cost-effective, and neither was pre-surgical PCR screening, although the comparator for both was unclear [31]. On the other hand, another review reported that universal single-culture MRSA screening was cost-effective compared to doing nothing, as was rapid PCR screening compared to standard practice [7]. Preadmission screening was found to be cost-effective if it prevented transmission of MRSA to as few as six patients, although the comparator was unclear [34]. Another review also found that PCR screening for MRSA was cost-effective, but the comparator was unclear, while the cost-effectiveness of screening using rapid diagnostics was dependent on the willingness-to-pay for averted isolation days [32]. Furthermore, rapid diagnostic screening was reported to be cost-effective compared to laboratory testing [30].

Screening, isolation, and decolonization of MRSA was cost-effective compared to do nothing, particularly when targeted towards high-risk patients and in scenarios with high MRSA transmission rates [7]. Another review reported the same, including for selective screening and screening of ICU patients, although the comparator was unclear [31]. The same authors also found evidence that universal MRSA screening and isolation was cost-effective compared to targeted screening, as was screening and isolation for ICU patients [31]. However, a further review reported that screening and isolation of high-risk patients on admission was cost-effective, although the comparator was unclear [33].

Moreover, screening and isolation was only found to be cost-effective if the prevalence of MRSA was high enough, isolation beds were utilized, and enough MRSA infections were averted and not substituted by meticillin-susceptible infections [39]. Screening and pre-emptive isolation, before receiving the results, was not cost-effective compared to no pre-emptive isolation [31]. Results for screening and decolonization were mixed, with one review reporting that it was likely to be cost-effective [8], while another found that it was not cost-effective compared to screening without decolonization [31].

With respect to CROs, one review found that screening and decolonization for carbapenem-resistant Enterobacteriaceae (CRE) in ICU patients was cost-effective compared to do nothing, whereas universal screening for carbapenemase-producing Enterobacterales was only cost-effective compared to doing nothing if prevalence levels were high enough [7].

Another review reported that VRE screening of high-risk hospital patients was cost-effective compared to doing nothing and standard practice, as was admission screening and active surveillance of ICU patients [35]. The review also found that VRE screening, contact precaution, and gown usage for HCWs and visitors was cost-effective compared to the same intervention without gown usage.

One review synthesized results across all HCAIs, finding that selective screening of high-risk patients was likely to be cost-effective across general wards and ICU, while there was no strong evidence that universal screening was cost-effective [9]. However, there was evidence that universal screening and decolonization was cost-effective in surgical departments. On the other hand, two reviews found that universal decolonization was more cost-effective for *Staphylococcus aureus* SSI than preoperative screening and decolonization in orthopaedic wards [40,41]. Another review found that the cost-effectiveness of screening and decolonization for SSI was context dependent [28]. Specifically, universal decolonization was cost-effective compared to screening and decolonization, and compared to doing nothing, in orthopaedic wards. However, preoperative screening and decolonization in orthopaedic wards was cost-effective in other settings if the intervention reduced the relative revision rate by at least 10%. Screening and decolonization in a general ward was also cost-effective compared to doing nothing, although it was unlikely to be cost-effective on a maternity ward.

Finally, rapid PCR screening for sepsis or suspected BSI in ICU patients was likely to be cost-effective compared to blood culture screening [42], as was screening for early disease

detection in hospital patients with pneumococcal disease compared to standard practice [43].

Surveillance

Five high-quality reviews synthesized evidence on the cost-effectiveness of surveillance interventions, covering eight intervention–infection/organism combinations. One found that surveillance for HCAIs was cost-effective compared to standard practice [9]. A second found that whole-genome sequencing (WGS) was cost-effective for HCAI surveillance [12], while a third reported that such interventions were only cost-effective if they were clinically effective enough [10], and the comparator in both was unclear. In the context of MRSA, WGS for surveillance was cost-effective compared to standard practice where the infrastructure already exists and prevalence of MRSA is high enough [7].

A state-wide electronic registry for CRE was found to be cost-effective compared to do nothing, while active PCR surveillance in a surgical ICU was also cost-effective for CRE compared to no surveillance [7]. Similarly, surveillance and decontamination strategies for CRE in ICUs was cost-effective, although the comparator was unclear [8]. Finally, active surveillance of VRE was cost-effective compared to standard practice and doing nothing [7].

Summary of the results

In summary, hand hygiene and environmental cleaning was cost-effective compared to standard practice, although the evidence base was limited. Diagnostic interventions were cost-effective compared to standard practice, although the evidence base was limited, as was surveillance compared to standard practice and doing nothing. ABS and decolonization interventions were generally cost-effective, although results were context dependent for some analyses. IPC bundles were mostly cost-effective, although there was considerable intervention heterogeneity, and the comparator was often unclear. Selective screening of high-risk patients or settings was cost-effective compared to doing nothing and, in many cases, universal screening. Universal screening was cost-effective in some contexts, particularly where prevalence and transmission of the infection/organism was high. Evidence on the cost-effectiveness of standalone education and training interventions was mixed, and the comparator was unclear for each of the analyses. The cost-effectiveness of PPE was also mixed. There was considerable geographical variation in the underlying studies, with many reviews considering global evidence, which is important to note given that the cost-effectiveness of an intervention is often dependent on the context of its implementation (e.g. prevalence, healthcare setting, willingness to pay).

Discussion

Twenty-four systematic reviews were synthesized to provide a comprehensive overview of the cost-effectiveness of ABS, IPC, and MDS interventions for the prevention and clinical management of HCAIs in adult hospital patients, with a particular focus on infections resistant to antibiotics.

There was a considerable lack of cost-effectiveness evidence for most of the intervention categories. Other than IPC bundle interventions, and screening, followed by contact precautions, isolation, decolonization, or a combination of these, no intervention had more than ten analyses extracted. Moreover, except for these two categories, no intervention type was considered in more than five of the 24 reviews. Quantity of evidence does not necessarily equate to quality or strength of evidence, as one high-quality study is often more insightful than many low-quality studies. However, lack of evidence can indicate that an area is under-researched. It is worth noting that not all intervention–infection/organism combinations are applicable or feasible in practice (e.g. decolonization for ESBL-KP), but many that are feasible had no evidence.

Screening, followed by contact precautions, isolation, decolonization, or a combination of these, had the largest quantity of evidence. Selective screening of high-risk patients or settings was generally more cost-effective, although universal screening was cost-effective in some cases where prevalence and transmission were high. This is likely to be a result of the high costs associated with universal screening. If prevalence and transmission is low, the cost of universal screening is unlikely to be offset by the clinical benefit, as only a relatively small proportion of patients will test positive and receive an alternative treatment pathway and the clinical benefits associated with it.

Among screening interventions, the majority addressed MRSA. In fact, considerably more evidence was identified for MRSA than any other infection/organism across all intervention categories. This is probably because of the longevity of MRSA, which emerged in the early 1960s and remains a political priority into the 21st century [44]. CRE, for example, did not emerge until decades later [45]. Furthermore, a high proportion of economic evaluations are conducted in high-income countries, where MRSA rates are relatively high compared to other organism–drug combinations [46], which may also explain the large evidence base.

Although most of the evidence focuses on Gram-positive infections — such as MRSA and VRE — Gram-negative infections such as CRE have developed into considerable public health issues [47]. This is because of their high resistance to antibiotics and their tendency to infect already high-risk patients, such as those in ICUs [48]. Despite this, there was very little evidence on the cost-effectiveness of interventions tackling Gram-negative infections, and those that did were focused on screening and surveillance. More cost-effectiveness evidence is therefore required to identify appropriate interventions to tackle this growing concern, particularly with respect to non-screening and non-surveillance interventions.

IPC bundle interventions had the second largest quantity of evidence, with 18 analyses. Overall, they were mostly cost-effective. However, only four IPC bundle analyses were included in the cost-effectiveness heat map (Figure 3), as the comparator was unclear in most. This was also a problem with the wider evidence base, with the comparator unclear in 41 of the 101 analyses that were extracted. It is unclear whether this was an issue with primary studies not reporting their comparator, or systematic reviews not capturing the information appropriately. Regardless, it is important for the intervention and comparator to be clearly reported when presenting

cost-effectiveness evidence to enable policymakers to make informed resource allocation decisions based on the results.

Due to the nature of the IPC bundle category, defined as multimodal interventions targeting more than one infection control activity, there was considerable heterogeneity between the interventions. For example, the category included interventions ranging from comprehensive 'search-and-destroy' policies that incorporated contact precautions, dedicated IPC personnel, patient and staff cohorting, surveillance, pre-emptive isolation and screening with decolonization; to enhanced hand hygiene alongside contact precaution and PPE use. It is therefore not appropriate to assume that a potential IPC bundle will be cost-effective because of the evidence presented in this review, where most were reported to be cost-effective, given the considerable differences between the interventions.

This is part of a wider issue with the evidence base. Even when considering more homogeneous categories, interventions are typically evaluated as a single entity, despite being made up of several interventions in practice. For example, screening interventions also include contact precautions, isolation and/or decolonization, as do many surveillance interventions. Despite this, cost-effectiveness results are commonly presented for the overarching intervention, making it hard to identify specifically which components were and were not cost-effective.

Even when considering perfectly homogeneous interventions, cost-effectiveness can vary considerably depending on the context of implementation. As demonstrated by the evidence for ABS, decolonization, education and training, screening, and surveillance interventions, cost-effectiveness can vary depending on infection type, prevalence, transmission, healthcare provider, WTP for health benefits, reimbursement systems and geography. Given the heterogeneity of interventions that are often evaluated as a single entity, and the potential impact of the context of implementation, it can be difficult to draw conclusions about the cost-effectiveness of an intervention beyond the specific setting it was evaluated in. The very nature of an umbrella review (i.e. a systematic review of systematic reviews) means that we could not analyse the context of the underlying studies nor therefore the impact that this context (e.g. WTP) may have had on the cost-effectiveness results. Our review necessarily synthesized the evidence as it was reported by the included reviews, rather than examining the underlying studies. It is therefore important that future research attempts to disentangle the impact of the individual components that often make up an overarching intervention, and considers how the wider context directly influences the cost-effectiveness of an intervention, so that policymakers can better understand the circumstances under which a particular intervention may be cost-effective in their context.

Moreover, in evaluating cost-effectiveness it is important to understand the components of cost, both in terms of the cost savings generated through infections prevented, as well as the costs incurred through implementation of the intervention itself. Costs of HCAI are largely driven by additional length of hospital stay due to infection, but, due to common use of inappropriate methodologies to estimate this key cost component, infection costs have historically been overestimated [49–51]. Similarly, costs of implementing the intervention itself are often ignored. Specifically, none of the included reviews or underlying analyses explicitly considered the implementation strategy used for the intervention of interest, the costs associated with its implementation (rather than those associated directly with the intervention), or the cost-effectiveness of the implementation. Future research should therefore look to distinguish between the cost of an intervention and the cost of its implementation, to allow insights into the cost, outcome, and cost-effectiveness of different implementation strategies for a given intervention. Ideally, economic evaluations should not only report costs, but also the resource utilization that underlies them (e.g. number of diagnostic procedures, treatment duration), as these can be converted into setting-specific costs, facilitating comparisons between countries [52].

ABS interventions were generally cost-effective, although in some instances results were mixed. One potential explanation for interventions that address AMR infections not being found cost-effective is that analyses may not account for the secondary cost of antibiotic consumption, as highlighted by Painter *et al.* in their review of economic evaluations of interventions addressing AMR [8]. This cost incorporates how changes in consumption affect the risk of resistance developing and therefore the cost associated with it [53]. Therefore, it is recommended that future economic evaluations explicitly incorporate these costs into their analyses, given that most patients diagnosed with HCAI are treated with antibiotics. Even where the impact of antibiotic use on resistance has been incorporated into economic evaluations, results have been subject to considerable uncertainty regarding the evolution of resistance and its associated health outcomes and costs [54]. This is largely due to the challenges associated with estimating how antimicrobial use will affect the long-term incidence of AMR. This is therefore an important area for future research, if the full costs and outcomes associated with AMR-related interventions are to be incorporated into economic evaluations.

In their review of economic evaluations of interventions to tackle HCAIs, Rice *et al.* found that no analyses of education and training interventions met the minimum quality criteria required to be included in their synthesis [9]. Interestingly, it appeared to be the only review that explicitly drew attention to an intervention where no robust evidence was identified. Naturally, reviews focused on interventions for which they identified cost-effectiveness evidence. However, generally, they did not identify interventions where no evidence was found. Given that reporting where no evidence was identified is just as important as reporting where evidence was found, this is something future systematic reviews should incorporate in the presentation of their results.

For specific analyses to be included in their comparison of the cost-effectiveness of different interventions, Painter *et al.* required the outcome to be measured in QALYs or DALYs [8]. Economic evaluations that utilize generic outcome measures, such as QALYs and DALYs, are referred to as cost–utility analyses. Generic outcome measures allow for incremental cost-effectiveness ratios (ICERs) to be estimated, such as cost per QALY or cost per DALY, which is the ratio of the difference in costs to the difference in outcomes between two interventions. ICERs with generic outcome measures, rather than condition-specific measures, allow for the comparison of interventions across different disease areas, which is vital for decision-makers to appropriately allocate resources across healthcare systems. Condition-specific outcomes, on

the other hand, are particularly useful when decision-makers are faced with fixed infection specialist budgets. Economic evaluations with condition-specific outcomes, such as infection-specific mortality, are referred to as cost-effectiveness analyses (CEAs). Many analyses extracted for this review reported a generic outcome measure, mostly QALYs. Many also utilized condition-specific measures, including infections avoided, patients admitted, length of stay, and more. Future research should prioritize using generic measures, alongside condition-specific measures, to capture outcomes associated with the intervention and enable both CEA and CUA to be undertaken.

The major strength and originality of our review is its breadth and comprehensiveness. Evidence has been synthesized from all identified systematic reviews examining the cost-effectiveness of ABS, IPC, and MDS interventions tackling HCAIs, with a particular focus on those resistant to antibiotics. This has been achieved without compromising on quality or rigour, with best practice guidelines followed for methodology [15] and reporting [16]. As a result, a comprehensive overview of the quantity (Figure 2) and quality (Supplementary Appendix S5) of the evidence base is provided, as well as a summary of the cost-effectiveness of different interventions, by infection/organism (Figure 3 and Table I). Furthermore, 20 (83%) of the 24 reviews were high quality, with the other four (17%) medium quality. No reviews were low quality. This suggests that the results of our review have a reasonable degree of reliability.

The findings of this umbrella review should be considered in light of several limitations. First, there is no established approach to estimate an overall quality assessment score based on the JBI Checklist for Systematic Reviews and Research Synthesis. A transparent, objective approach was adopted, based on the scoring system applied in another review that used the same quality assessment tool [24]. However, the scoring system assumes that all 11 items in the checklist are of equal importance, which may not be the case. Second, given the nature of an umbrella review, we did not formally assess the quality of the underlying studies that were synthesized in the included systematic reviews. Thus, it was challenging to draw conclusions about the quality of the underlying evidence base. However, where reported, we did extract and synthesize information on the overarching quality of the underlying studies. Moreover, twenty of the included reviews were high quality, with the others medium quality. The high quality of the included reviews suggests they appropriately accounted for the quality of the underlying studies when synthesizing the evidence. Third, our search strategy used the validated built-in review filter in Medline and Embase to limit hits to systematic reviews (Supplementary Appendix S3). The filters have good specificity, meaning that most hits will be genuine systematic reviews, but lower sensitivity, meaning that studies that are not indexed as systematic reviews or do not have clear systematic review terms may be missed. This approach was taken due to time and resource constraints. However, all known papers were retrieved by this search strategy, and the included reviews were used for backward and forward citation searching to identify any systematic reviews that may have been missed by the search. Despite these limitations, this umbrella review utilizes well-established methodologies to provide a comprehensive summary of the cost-effectiveness of ABS, IPC, and MDS interventions tackling HCAIs.

Conclusion

The objective of this research was to summarize the cost-effectiveness of interventions addressing HCAIs and AMR in hospitals. Most of the cost-effectiveness evidence was focused on screening, followed by contact precautions, isolation, and/or decolonization, with selective screening generally more cost-effective. IPC bundles had the second largest evidence base and were mostly found to be cost-effective, although there was considerable heterogeneity between the interventions. The evidence also suggested that standalone environmental cleaning, hand hygiene, diagnostics, surveillance, ABS, and decolonization were mostly cost-effective. However, the evidence base was sparse, as it was for education and training, and PPE interventions, and more research is required. Importantly, cost-effectiveness can vary depending on the context of implementation (e.g. prevalence, transmission, healthcare provider, WTP); therefore policymakers should consider their setting and that of the evidence base before deciding on implementing a specific intervention. Future research should also account for this in economic evaluations, explicitly exploring how the context changes the cost-effectiveness. Even where there is a relatively large evidence base, such as with screening interventions, the majority is focused on MRSA and other Gram-positive infections. Due to the public health threat posed by Gram-negative infections, considerably more research is required in this area. Further recommendations for future research are also made based on the findings, including for economic evaluations to try to untangle the cost-effectiveness of the specific components of an overarching intervention, consider the secondary cost of antibiotic consumption, and use generic, as well as clinical, outcome measures. The intervention and comparators of interest should clearly be defined when cost-effectiveness evidence is presented, and systematic reviews of such evidence should report where no evidence is found for relevant interventions.

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Conflict of interest statement
None declared.

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Ethics statement
Not required.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jhin.2025.01.006.

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Appendix F. Co-author declarations

Due to the nature of the research, some of the works included in this thesis were sections of larger publications. Where this was the case, only sections of the publications relevant to the economic analysis were included (i.e., Appendix B. Paper 2 and Appendix C. Paper 3). All publications are multi-authored, reflecting the contributions of those in various roles, such as administration, management and research, as is common in research of this nature. I was the lead analyst of the economic analysis undertaken in each of the publications.

Co-author declarations confirming my contribution are provided below.

Co-author confirmation of candidate's contribution

Paper number: 1

Title: Estimating the cost of growing the NHS cancer workforce in England by 2029.

Candidate contribution: Contributed to the development of the research idea and led the methodological design of the research, including leading the writing of the research proposal. Led the acquisition of data from stakeholders and contributed to the collection of additional data sources through desk research. Led the development of the economic model, undertook the stakeholder interviews, and contributed to the validation workshop. Conducted the economic analysis and data visualisation. Led the writing of the first draft of the publication. All with the support and guidance of the principal investigator.

I confirm that the above statement is a fair reflection of the candidate's own original contribution to the published work.

Signed:

Full name:

EVANGELOS GKOUSIS

Date:

08/11/2024

Co-author confirmation of candidate's contribution
Paper number: 2
Title: Pregnancy research review: Policy report.
Candidate contribution: Contributed to the development of the research idea and methodological design of the research, including contributing to the writing of the research proposal. Led the identification and collection of data on the health needs and healthcare expenditure associated with pregnancy. Led the evidence summary of the costs associated with non-standard pregnancies and the creation of a bespoke tool to compare pregnancy research spend with health needs and healthcare expenditure. Conducted the economic analysis and data visualisation. Led the writing of the 'Contextualisation of research spend' sections of the publication. All with the support and guidance of the principal investigator.
I confirm that the above statement is a fair reflection of the candidate's own original contribution to the published work. **Signed:** Sarah Parkinson **Full name:** Sarah Parkinson **Date:** 10 December 2024

Co-author confirmation of candidate's contribution

Paper number: 3

Title: Outcome Evaluation of the National Model for Liaison and Diversion

Candidate contribution: Contributed to the methodological design of the difference-in-differences analysis and led the methodological design of the economic evaluation. Led the data curation of the accident and emergency (A&E), and mental health services datasets, the collection of service costs associated with the intervention, and the identification of unit cost data in the literature. Conducted the difference-in-differences analysis of A&E and mental health service utilisation, and the economic evaluation. Led the writing of the economic analysis sections of the publication and contributed to the writing of the A&E and mental health service utilisation sections. All with the support and guidance of the principal investigator.

I confirm that the above statement is a fair reflection of the candidate's own original contribution to the published work.

Signed:

Full name:

EVANGELOS GKOUSIS

Date:

08/11/2024

Co-author confirmation of candidate's contribution

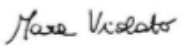
Paper number: 4

Title: Digitally augmented, parent-led CBT versus treatment as usual for child anxiety problems in child mental health services in England and Northern Ireland: a pragmatic, non-inferiority, clinical effectiveness and cost-effectiveness randomised controlled trial.

Candidate contribution: Contributed to the methodological design of the economic evaluation (e.g., cost-effectiveness and multiple imputation statistical models). Led the curation of the economic data (e.g., service utilisation, health related quality of life) collected in the trial and the identification of unit cost data in the literature, with the support of co-authors. Conducted the economic analysis and data visualisation, with the support of co-authors. Led the writing of the first draft of the economic component of the publication, which was then amalgamated with the clinical effectiveness component by co-authors for final publication. All with the support and guidance of the principal investigator of the economic evaluation.

I confirm that the above statement is a fair reflection of the candidate's own original contribution to the published work.

Signed:



.....

Full name:

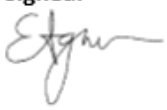
Mara Violato

.....

Date:

08/11/2024

.....

Co-author confirmation of candidate's contribution
Paper number: 5
Title: Umbrella review of economic evaluations of interventions for the prevention and management of healthcare-associated infections in adult hospital patients.
Candidate contribution: Led the development of the research idea and led the methodological design of the research. Led the creation of the eligibility criteria, identification of studies, data extraction and quality assessment, all with the support of co-authors. Conducted the data synthesis and data visualisation. Led the writing of the publication. All with the support and guidance of the principal investigator.
I confirm that the above statement is a fair reflection of the candidate's own original contribution to the published work. **Signed:**  ..... **Full name:** Emily Agnew ..... **Date:** 04/02/2025 .....

Appendix G. Forward citation analysis

Search strategy methods

Forward citations, i.e., articles or publications that have cited the publication of interest, were identified using the following sources:

- Scopus 'Cited by' forward citation tool, to identify academic literature.
- citationchaser forward citation tool (which searches PubMed, PubMed Central, CrossRef, Microsoft Academic Graph and CORE), to identify academic literature.[1]
- Google Scholar forward citation tool, to identify grey literature (e.g., guidelines, policy documents, articles) and academic literature.
- Google Scholar search of the paper title, to identify grey literature and academic literature.
- Google search of the paper title, to identify grey literature.

All hits were screened to check whether they cited the publication of interest. All grey literature and academic literature that cited the economic analysis were included in the data synthesis, except conference abstracts which were excluded. Articles that cited the publication but did not mention the economic analysis were excluded. Articles not written in English were excluded.

Data synthesis methods

A qualitative synthesis was undertaken, "whereby study findings are systematically interpreted through a series of expert judgements to represent the meaning of the collected work", to draw conclusions about how the research was utilised and how it influenced policy.[2]

[1] Haddaway, N. R., Grainger, M. J., Gray, C. T. (2021) citationchaser: An R package and Shiny app for forward and backward citations chasing in academic searching. doi: 10.5281/zenodo.4543513

[2] Bearman, M., & Dawson, P. (2013). Qualitative synthesis and systematic review in health professions education. *Medical education*, *47*(3), 252-260.

Search strategy results

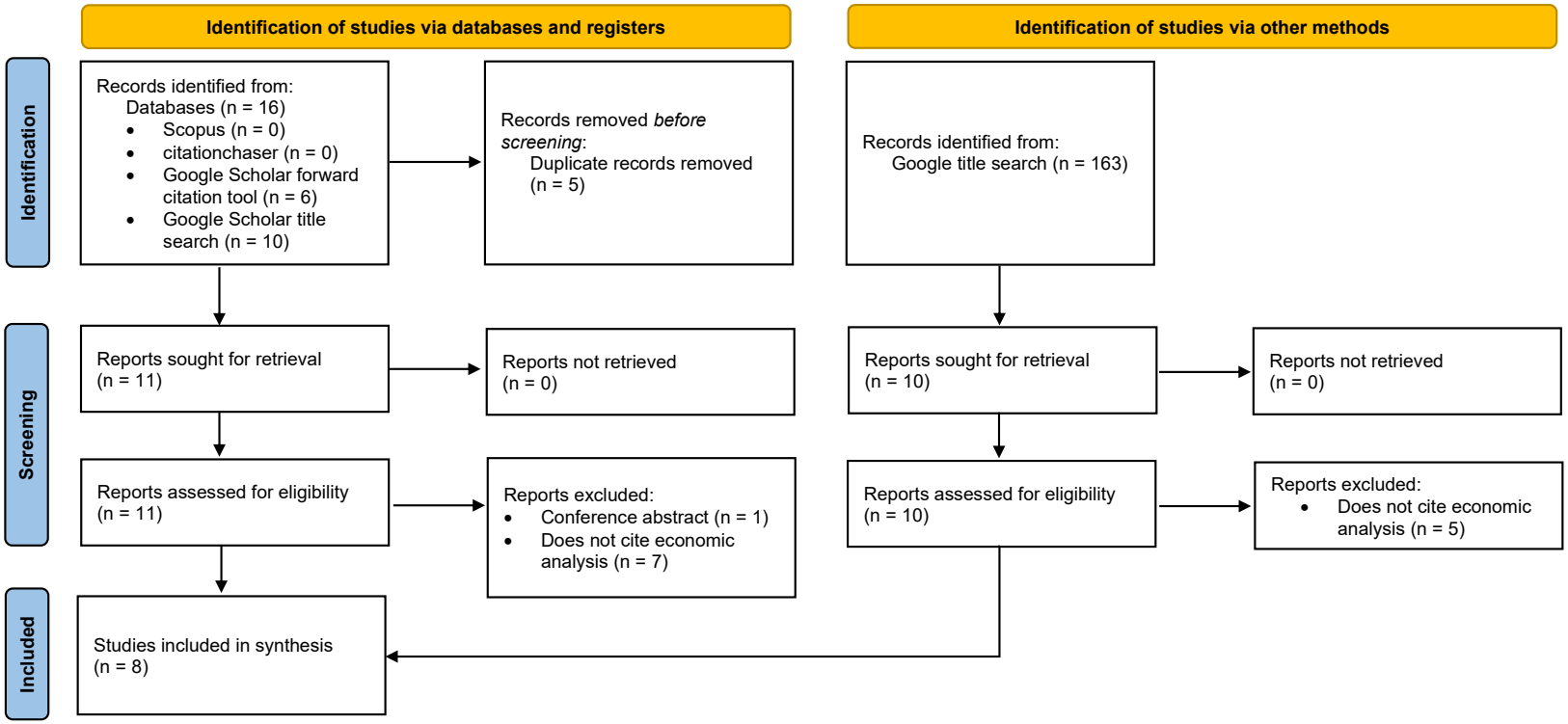


Figure G1. Paper 1 PRISMA flow diagram for forward citation search on 2 December 2024.

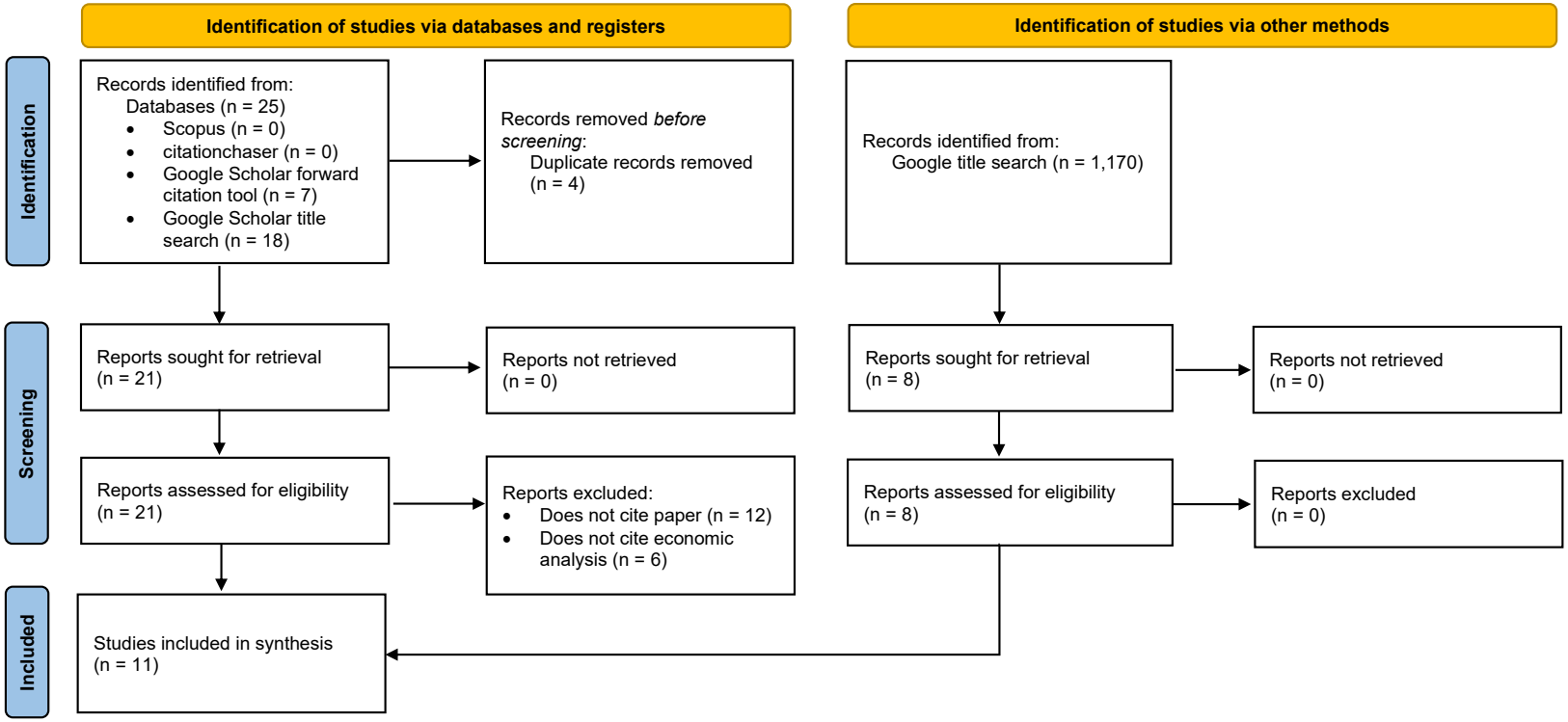


Figure G2. Paper 2 PRISMA flow diagram for forward citation search on 30 December 2024.

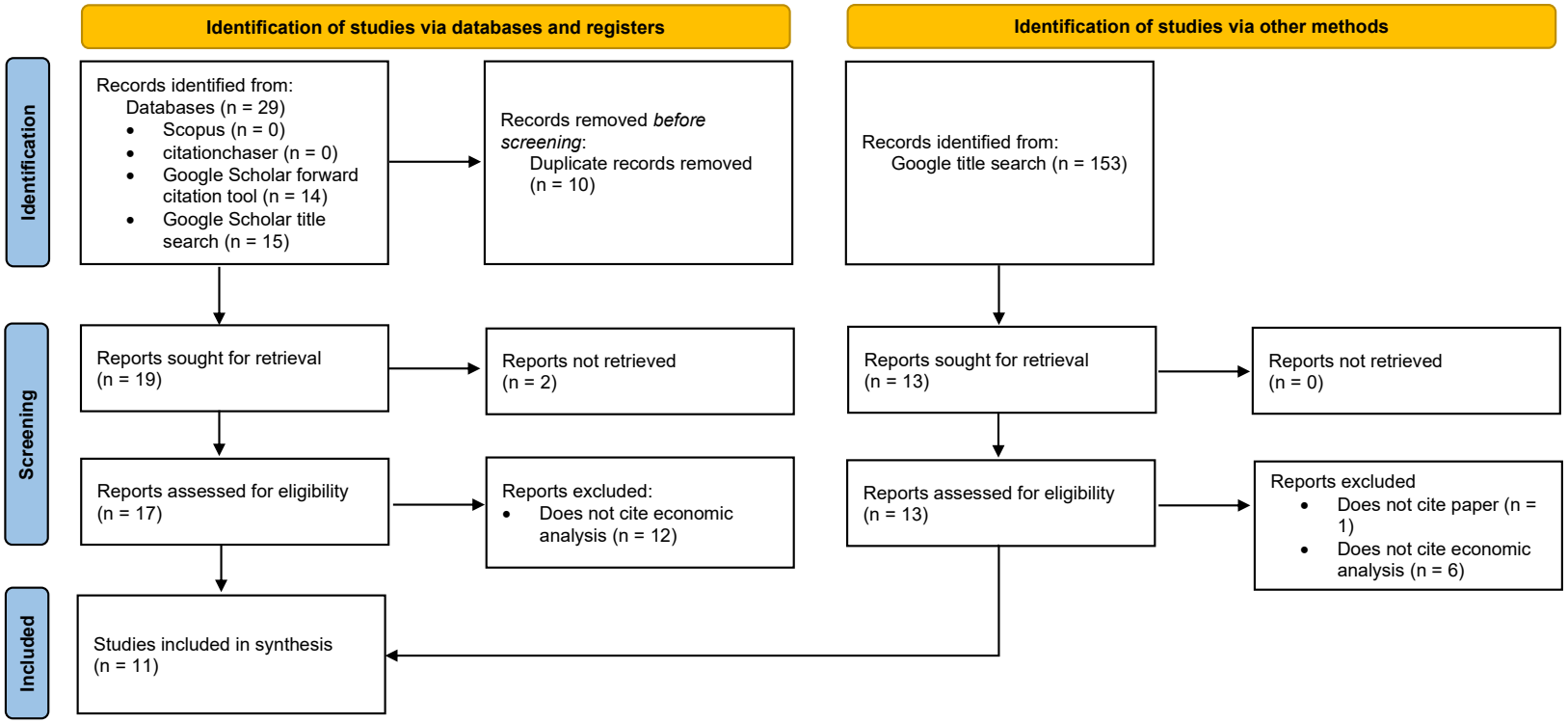


Figure G3. Paper 3 PRISMA flow diagram for forward citation search on 10 January 2025.

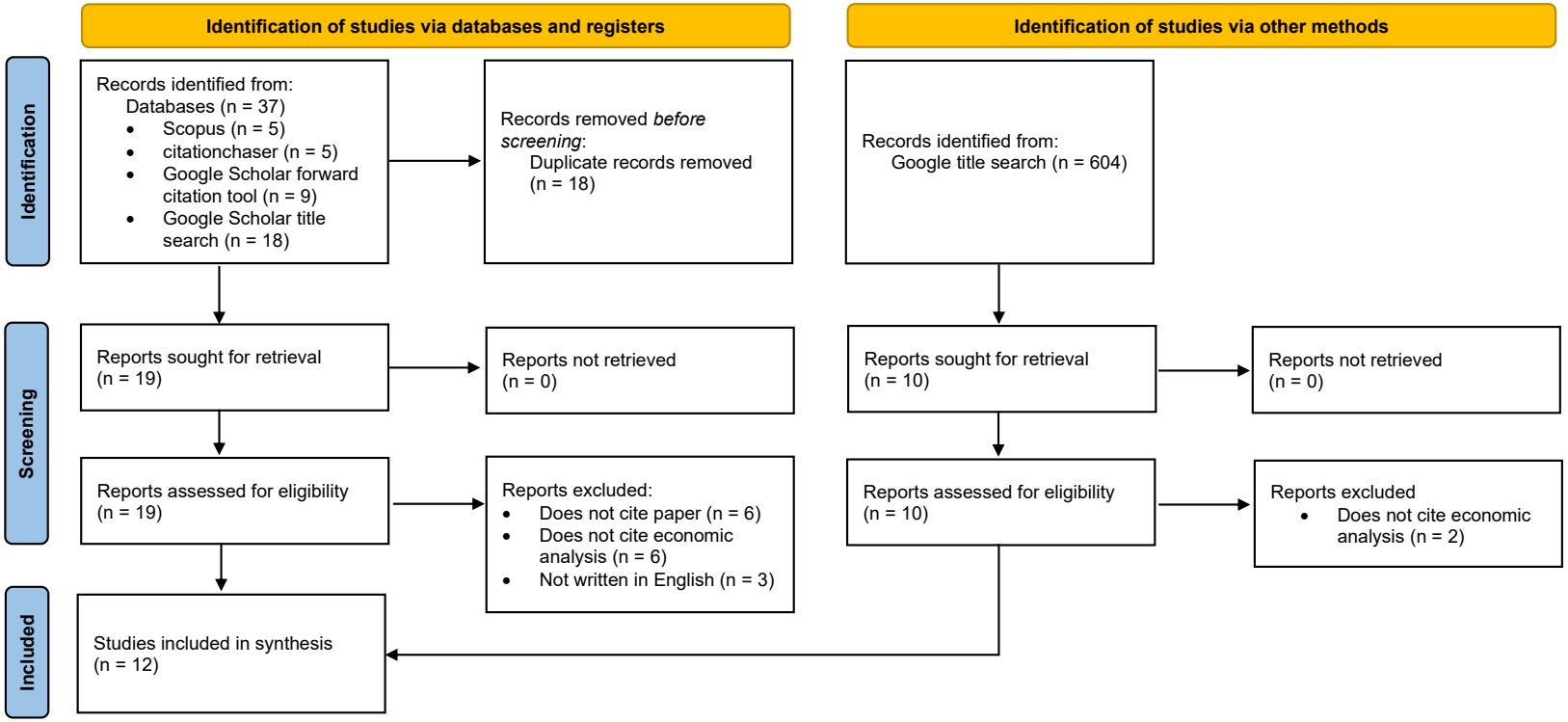


Figure G4. Paper 4 PRISMA flow diagram for forward citation search on 26 January 2025.

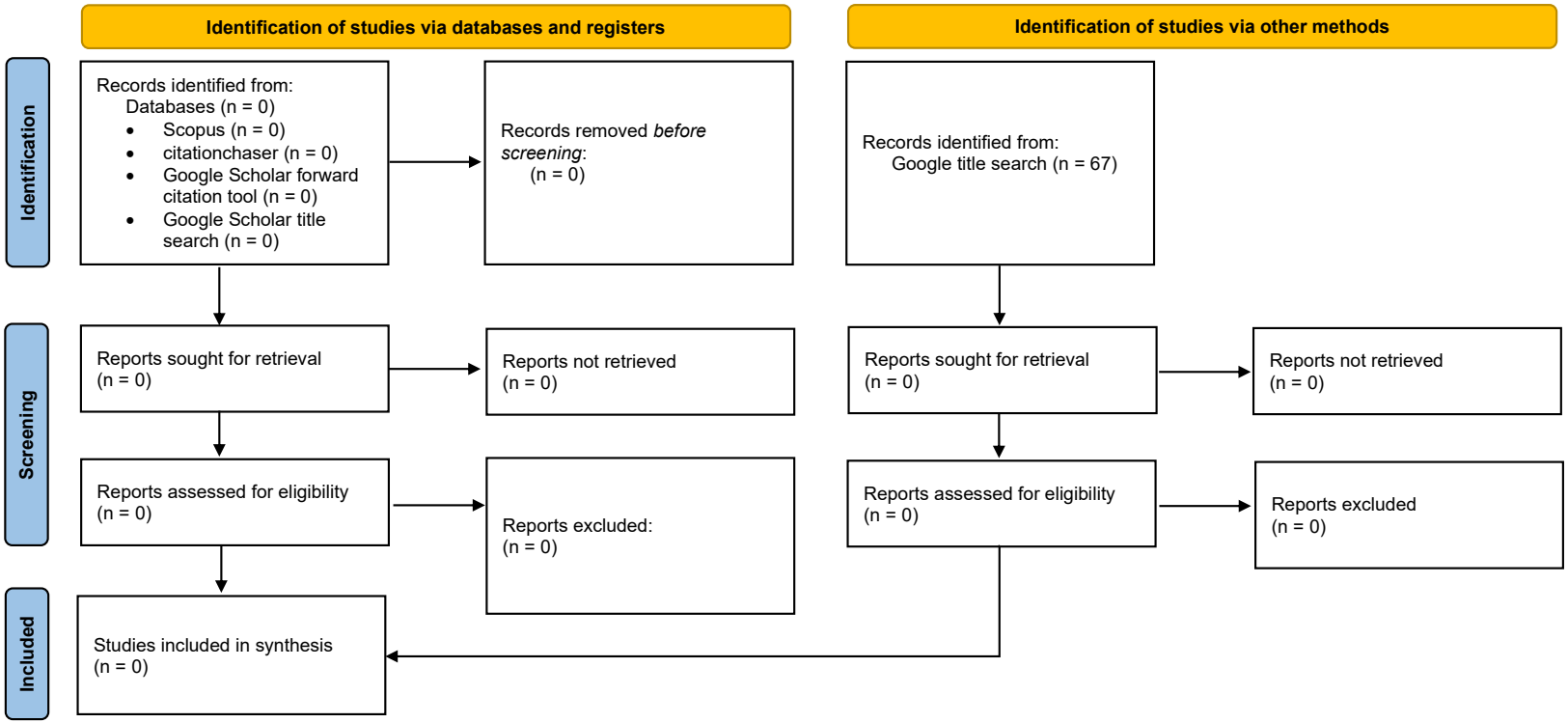


Figure G5. Paper 5 PRISMA flow diagram for forward citation search on 09 February 2025.

Appendix H. CHEERS checklist

Critical appraisal of Paper 1, 2, 3 and 4 using the Consolidated Health Economic Evaluation Reporting Standards (CHEERS) 2022 checklist

Section/topic	Item No.	Guidance for reporting	Reported in section			
			Paper 1	**Paper 2**	**Paper 3**	**Paper 4**
Title						
Title	1	Identify the study as an economic evaluation and specify the interventions being compared.	Page 1	Page i	Page i	Page 193
Abstract						
Abstract	2	Provide a structured summary that highlights context, key methods, results, and alternative analyses.	Page 8-13	Not reported, as the paper was summarised in a policy report.[3]	Page v-xii	Page 193
Introduction						
Background and objectives	3	Give the context for the study, the study question, and its practical relevance for decision making in policy or practice.	Page 15-18	Page 1-2	Page 176	Page 193-195
Methods						
Health economic analysis plan	4	Indicate whether a health economic analysis plan (HEAP) was developed and where available.	Not undertaken, as HEAPs are targeted at economic evaluations alongside randomised controlled trials and	Not undertaken, as HEAPs are targeted at economic evaluations alongside randomised controlled trials and	Not undertaken, as HEAPs are targeted at economic evaluations alongside randomised controlled trials and	Page 198

[3] Guthrie S, Pollard J, Parkinson S, Altenhofer M, Leach B, Lichten CA. (2020). Pregnancy research review: Policy report. Santa Monica, CA: RAND Corporation.

Section/topic	Item No.	Guidance for reporting	Reported in section			
			Paper 1	**Paper 2**	**Paper 3**	**Paper 4**
			were not standardised until after the research was conducted.[4]	were not standardised until after the research was conducted. [5]	were not standardised until after the research was conducted. [6]	
Study population	5	Describe characteristics of the study population (such as age range, demographics, socioeconomic, or clinical characteristics).	Page 18	Page 18	Page 15-26 & 182	Page 199-200
Setting and location	6	Provide relevant contextual information that may influence findings.	Page 21-22	Page 24	Page 176	Page 195
Comparators	7	Describe the interventions or strategies being compared and why chosen.	Page 23	Not applicable, as the analysis did not consider alternative interventions or strategies.	Page 176	Page 195-196
Perspective	8	State the perspective(s) adopted by the study and why chosen.	Page 20	Page 23	Page 176	Page 198 & 201
Time horizon	9	State the time horizon for the study and why appropriate.	Page 18	Page 18 & 23	Page 176	Page 196

[4] Thorn, J. C., Davies, C. F., Brookes, S. T., Noble, S. M., Dritsaki, M., Gray, E., ... & Hollingworth, W. (2021). Content of Health Economics Analysis Plans (HEAPs) for trial-based economic evaluations: expert Delphi consensus survey. Value in Health, 24(4), 539-547.

[5] Ibid.

[6] Ibid.

Section/topic	Item No.	Guidance for reporting	Reported in section			
			Paper 1	**Paper 2**	**Paper 3**	**Paper 4**
Discount rate	10	Report the discount rate(s) and reason chosen.	Not reported, as costs were assumed to be front-loaded to train sufficient staff by 2029. However, the use of a discount rate wasn't considered or discussed in the paper.	Not applicable, as the time horizon for the analysis was only one year. However, this wasn't stated in the paper.	Not applicable, as the time horizon for the analysis was only one year. However, this wasn't stated in the paper.	Page 198
Selection of outcomes	11	Describe what outcomes were used as the measure(s) of benefit(s) and harm(s).	Not applicable, as outcomes were not considered in the analysis.	Page 18	Page 179	Page 196
Measurement of outcomes	12	Describe how outcomes used to capture benefit(s) and harm(s) were measured.	Not applicable, as outcomes were not considered in the analysis.	Page 19-22	Page 180-182	Page 196
Valuation of outcomes	13	Describe the population and methods used to measure and value outcomes.	Not applicable, as outcomes were not considered in the analysis.	Not applicable, as this item is specific to the valuation of HRQoL outcomes.	Not applicable, as this item is specific to the valuation of HRQoL outcomes.	Page 196
Measurement and valuation of resources and costs	14	Describe how costs were valued.	Page 20	Page 20-21 & 22-23	Page 181	Page 197
Currency, price date, and conversion	15	Report the dates of the estimated resource quantities and unit costs, plus the currency and year of conversion.	Page 28	Page 23	Page 176	Page 198

Section/topic	Item No.	Guidance for reporting	Reported in section			
			Paper 1	**Paper 2**	**Paper 3**	**Paper 4**
Rationale and description of model	16	If modelling is used, describe in detail and why used. Report if the model is publicly available and where it can be accessed.	Page 21-23	Not applicable, as no modelling was undertaken.	Not applicable, as no modelling was undertaken.	Not applicable, as no modelling was undertaken.
Analytics and assumptions	17	Describe any methods for analysing or statistically transforming data, any extrapolation methods, and approaches for validating any model used.	Page 21-23	Page 22-24	Page 179-183	Page 198 & 201
Characterising heterogeneity	18	Describe any methods used for estimating how the results of the study vary for subgroups.	Page 19-20	Not reported, as analysis did not consider how results varied by subgroups.	Not reported, as analysis did not consider how results varied by subgroups.	Not reported, as analysis did not consider how results varied by subgroups.
Characterising distributional effects	19	Describe how impacts are distributed across different individuals or adjustments made to reflect priority populations.	Not reported, as the analysis did not consider the distribution of impacts.	Not reported, as the analysis did not consider the distribution of impacts.	Not reported, as the analysis did not consider the distribution of impacts.	Not reported, as the analysis did not consider the distribution of impacts.
Characterising uncertainty	20	Describe methods to characterise any sources of uncertainty in the analysis.	Page 59	Not reported, as the paper did not consider sources of uncertainty in the analysis.	Page 98 & 185	Page 201
Approach to engagement with patients and	21	Describe any approaches to engage patients or service recipients, the	Page 19-21 & 23-24	Not reported, despite engagement with a steering group	Not reported, despite preliminary results being presented to the	Page 197 & 208

Section/topic	Item No.	Guidance for reporting	Reported in section			
			Paper 1	**Paper 2**	**Paper 3**	**Paper 4**
others affected by the study		general public, communities, or stakeholders (such as clinicians or payers) in the design of the study.		throughout the project.	commissioners of the research.	
Results						
Study parameters	22	Report all analytic inputs (such as values, ranges, references) including uncertainty or distributional assumptions.	Page 26-28	Page 77-81	Page 183-187	Appendix page 5-22
Summary of main results	23	Report the mean values for the main categories of costs and outcomes of interest and summarise them in the most appropriate overall measure.	Page 58-59	Page 77-81	Page 187	Page 201
Effect of uncertainty	24	Describe how uncertainty about analytic judgments, inputs, or projections affect findings. Report the effect of choice of discount rate and time horizon, if applicable.	Page 59 & Supplement page 39-46	Not reported, as the paper did not consider sources of uncertainty in the analysis.	Page 98-99 & 185-188	Page 204
Effect of engagement with patients and	25	Report on any difference patient/service recipient, general public, community, or	Page 21 & 23	Not reported, despite engagement with a steering group	Not reported, despite preliminary results being presented to the	Not reported, despite end-users being involved in co-design, and

Section/topic	Item No.	Guidance for reporting	Reported in section			
			Paper 1	**Paper 2**	**Paper 3**	**Paper 4**
others affected by the study		stakeholder involvement made to the approach or findings of the study.		throughout the project.	commissioners of the research.	preliminary results being presented to a steering group.
Discussion						
Study findings, limitations, generalisability, and current knowledge	26	Report key findings, limitations, ethical or equity considerations not captured, and how these could affect patients, policy, or practice.	Page 60-65	Page 24	Page 188	Page 205 & 207
Other relevant information						
Source of funding	27	Describe how the study was funded and any role of the funder in the identification, design, conduct, and reporting of the analysis.	Page 3	Page iii	Page iii	Page 193 & 201
Conflicts of interest	28	Report authors conflicts of interest according to journal or International Committee of Medical Journal Editors requirements.	Not reported, as not done as standard for RAND policy reports at the time.	Not reported, as not done as standard for RAND policy reports at the time.	Not reported, as not done as standard for RAND policy reports at the time.	Page 208
Proportion of relevant items included			21/25 (84%)	15/24 (63%)	19/25 (76%)	24/27 (89%)

Appendix I. JBI Checklist for Systematic Reviews and Research Synthesis

Critical appraisal of Paper 5 using the JBI Checklist for Systematic Reviews and Research Synthesis

Item No.	Critical appraisal item	Yes	No	Unclear	Not applicable
1	Is the review question clearly and explicitly stated?	✓			
2	Were the inclusion criteria appropriate for the review question?	✓			
3	Was the search strategy appropriate?	✓			
4	Were the sources and resources used to search for studies adequate?	✓			
5	Were the criteria for appraising studies appropriate?	✓			
6	Was critical appraisal conducted by two or more reviewers independently?	✓			
7	Were there methods to minimize errors in data extraction?	✓			
8	Were the methods used to combine studies appropriate?	✓			
9	Was the likelihood of publication bias assessed?				✓*
10	Were recommendations for policy and/or practice supported by the reported data?	✓			
11	Were the specific directives for new research appropriate?	✓			
Proportion of relevant items included		10/10 (100%)			

*Not applicable because quantitative synthesis (e.g., meta-analysis) was not undertaken.