HTA Austria
Austrian Institute for
Health Technology Assessment
GmbH

VILLAGE

The economic and societal dimension of parental mental illness

Part I: Systematic review
Part II: Economic evaluation framework

Final report

AIHTA Project Report No.: 142 | ISSN: 1993-0488 | ISSN-online: 1993-0496

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Part II: Economic evaluation framework

Vienna, November 2021

Project Team

Project leader: Christoph Strohmaier (AIHTA)

Authors: Christoph Strohmaier (CS) (1st author; AIHTA)
Laura Hölzle (LH) (2nd author; Village project)

Project Support

Systematic literature searches (information specialist): Tarquin Mittermayr (TM)

Visualisation: Smiljana Blagojevic

Hand search: Christoph Strohmaier

Internal review: Ingrid Zechmeister-Koss (IZK)

Review co-investigators Village project: Annette Bauer, Melinda Goodyear, Jean Paul

External review: Nicola Brimblecombe (NB), Assistant Professorial Research Fellow,
Care Policy and Evaluation Centre (CPEC), The London School of Economics and Political Science

Correspondence: Christoph Strohmaier (AIHTA), christoph.strohmaier@aihta.at

Cover photo: © sewcream – stock.adobe.com

This report should be referenced as follows:

Strohmaier C, Hölzle L, The economic and societal dimension of parental mental illness –Systematic review and economic evaluation framework, AIHTA Project Report No.: 142; 2021. Vienna: HTA Austria – Austrian Institute for Health Technology Assessment GmbH.

Conflict of interest

All authors and the reviewers involved in the production of this report have declared they have no conflicts of interest in relation to the technology assessed according to the Uniform Requirements of Manuscripts Statement of Medical Journal Editors (www.icmje.org).

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IMPRINT

Publisher:
HTA Austria – Austrian Institute for Health Technology Assessment GmbH
Garnisongasse 7/Top20 | 1090 Vienna – Austria
https://www.aihta.at/

Responsible for content:
Priv.-Doz. Dr. phil. Claudia Wild, managing director

AIHTA Project Reports do not appear on a regular basis and serve to publicize the research results of the Austrian Institute for Health Technology Assessment.

AIHTA Project Reports are only available to the public via the Internet at http://eprints.aihta.at/view/types/hta_report.html.

AIHTA Project Report No.: 142

ISSN 1993-0488

ISSN online 1993-0496

Content

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List of abbreviations

BDI.....Beck depression index
CAU .....Care as usual
CEA.....Cost-effectiveness analysis
CEAC .....Cost-effectiveness acceptability curves
CEP .....Cost-effectiveness planes
COI.....Conflict of interest
CSRI.....Client service receipt inventory
CUA .....Cost-utility analysis
CVA.....Collaborative Village Approach
ECBI .....Eyberg Child Behaviour Inventory
ECCE .....Evaluation of Children's Centres in England
EE.....Economic evaluation
fRCT .....Feasibility randomised controlled trial
GBP .....British pounds
GDP .....Gross domestic product
Ges.ök.....Gesundheitsökonomisch
GP.....General practitioner
HCA .....Human capital approach
HEE(s) .....Health economic evaluation(s)
HFP-M.....Helping Families Programme-Modified
HRQoL .....Health related quality of life
HTA .....Health Technology Assessment
ICB(s).....Inter-sectoral cost(s) and benefit(s)
ICER .....Incremental cost-effectiveness ratios
iMTA.....Institute for Medical Technology Assessment
IY.....Incredible Years
MDP.....Multiple Determinants of Parenting
NHS .....National Health Service
NIHR .....National Institute for Health Research
OxCAP-MH...Oxford CAPabilities questionnaire-Mental Health
PBCM .....Preventive basic care management
PECUNIA.....ProgrammE in Costing, resource-use measurement and outcome valuation for Use in multi-sectoral National and International health economic evaluAtions
PSSRU .....Personal Social Services Research Unit
QALY.....Quality-adjusted life year
QoL .....Quality of life
RCT.....Randomised controlled trial
RQ.....Research question
SENSE .....Sensitive Screening
SES.....Socioeconomic statuses
SMI.....Severe mental illness
SROI .....(Social) Return on investment
TCMD.....Transgenerational cycle of mental disorders
UK.....United Kingdom
WL .....waiting list
WTP.....Willingness-to-pay
ZonMW.....Dutch organisation for health research and development

Glossary[1]

Cost-effectiveness analysis:
A form of (health) economic evaluation that compares the relative expenditures (costs) and disease-specific outcomes (effects) of two or more options. Outcomes are measured in natural units.

Cost-effectiveness acceptability curves:
A graphical illustration of the probability that an intervention is cost-effective given a willingness-to-pay threshold to gain a measure of effectiveness in health outcome.

Cost-effectiveness planes:
A graph of a coordinate system that plots the difference in effectiveness of two interventions per patient against the difference in cost of two interventions per patient. It is used to illustrate whether an intervention is cost-effective compared to an alternative.

Cost-utility analysis:
A form of cost-effectiveness analysis in which benefits are measured in terms of a generic measure or utility, such as the quality-adjusted life-year.

Decision-makers:
Institutions, policy makers or planning entities such as the ministry of health who use research evidence to make decisions on how a health care system needs to be organised.

Effectiveness:
Effectiveness is a measure of the treatment's intended effects under real-world conditions

Efficiency:
Efficiency can be defined in two ways. Either maximising the outpout (benefit) to a given input (resource expenditure) or minimising the cost of a given outpout (benefit).

Feasibility randomised controlled trial:
Feasibility randomised controlled trials are preliminary studies done before a main randomised controlled trial in order to answer the question, whether the study can be conducted.

Human capital approach:
The human capital approach is a method to estimate the indirect cost because of productivity losses. Indirect costs are income losses resulting from premature mortality, disability, and care seeking, including lost production due to work absence, or early retirement.

Health economic evaluation(s):
Study to determine whether or not an intervention achieves value for money. Definition by Drummond: *'the comparative analysis of alternative courses of action in terms of both their costs and consequences'* (Drummond et al. 2015. Methods for the Economic Evaluation of Health Care Programmes. Oxford: Oxford University Press)

Health Technology Assessment:
'*HTA is a multidisciplinary process that uses explicit methods to determine the value of a health technology at different points in its lifecycle. The purpose is to inform decision-making in order to promote an equitable, efficient, and high-quality health system.*' (from O'Rourke et al. 2020.
The new definition of health technology assessment: A milestone in international collaboration. International Journal of Technology Assessment in Health Care 36, 187 –190)

Inter-sectoral cost(s) and benefit(s):
Costs and benefits that are associated with health interventions but are incurred in other sectors as the healthcare such as education or criminal justice sector.

Incremental cost-effectiveness ratios (ICER):
The ICER is the difference between the costs of one treatment alternative and another treatment divided by the difference between the outcomes of both treatment options. It compares the additional costs and treatment effects in the experimental intervention with the control intervention.

[1] The glossary is inspired by Skivington et al. Framework for the development and evaluation of complex interventions: gap analysis, workshop and consultation-informed update. 2021; 25:57. DOI: 10.3310/hta25570.

Logic model:
A logic model illustrates 'how an intervention works' and 'which outcomes can be expected from an intervention' when implemented or when the intervention is not implemented.

Quality-adjusted life year:
A combined outcome considering both disease burden (quality of life) and survival (life expectancy). The calculation of QALYs uses number of years of life an individual receives from an intervention for the quality of life in those years that can be expected.

Randomised controlled trial:
A randomized controlled trial is an experiment to study the safety and efficacy of new treatments and to control factors that are not under direct experimental control.

Return on investment:
The return on investment is a performance measure used to evaluate the profitability of an investment or to relate profits to the (capital) amount invested.

Social return on investment:
The social return on investment is similar to the return on investment, but considers also social and environmental outcomes when evaluating whether a decision is profitable.

Sensitivity analysis:
Sensitivity analyses is an analysis to test the robustness of a statistical model by varying key assumptions in order to determine how far conclusions rely on particular assumptions.

Willingness to pay:
The willingness to pay is the maximum amount of money an individual or society is willing to pay to avoid or reduce a specific health problem or to gain a specific health benefit. Willingness to pay is one component in deciding whether an intervention is cost-effective.

Executive Summary

Introduction

children of parents with a mental illness have an increased health risk

Mental illnesses such as depression are highly prevalent and affect not only the person directly but also their family and social environment. Especially children and adolescents from so-called 'parents with a mental illness' have a potentially increased risk of developing health problems themselves on their way to adulthood, as well as experiencing further negative consequences.

intergenerational cycle of mental illness

Genetic influences but also socio-economic circumstances such as parental unemployment or (material) deprivation are causes for this intergenerational cycle of mental illness. All these factors require measures to promote the mental health of children and adolescents from parents with mental illnesses in order to break the intergenerational cycle of mental illness and its negative side effects.

family-oriented complex interventions as an option?

gaps in health economic (HE) knowledge and research

One option to mitigate or even prevent negative consequences for affected children and adolescents are family-oriented intervention programmes in combination with social support systems. These programmes usually follow a preventive approach and are complex in nature, as they involve people from different health and care professions and multiple programme components. Until recently, there was not only a lack of such programmes in everyday care. Studies on the effect(s) of these family-oriented interventions were also rare. In particular, gaps in health economic knowledge in this area is a barrier to grasp the overall benefit of those programmes

aims of the report:

source of information for (health economic) impact assessments and raising awareness

Against this background, the report is intended to inform about existing health economic evidence in this field and to serve as a source of information for (health economic) researchers to conduct sound evaluations and impact assessments of family-oriented intervention programmes in the context of children and adolescents from parent with a mental illness. Furthermore, we want to raise the general awareness in health planners and policy makers about the economic and social dimensions of mental illness in families.

4 research questions: HE evidence of family-oriented interventions and study quality?

In the course of the two report parts, four research questions were addressed. In part I, two research questions concerned the international evidence on the cost-effectiveness of family-oriented complex interventions and the associated study quality and programme transferability.

impacts of parental mental illness on children and adolescents? methodological conclusions?

In part II, we addressed the impact of parental mental illness on children's individual health, socio-economic consequences, and societal impacts (research question three). The final research question addressed methodological conclusions that can be drawn for the implementation of health economic evaluations in this field in the light of the current health economic evidence.

Methods

systematic review and risk of bias assessment

We addressed research question one and two by a systematic review of cost-effectiveness evidence of internationally implemented family-oriented complex interventions focussing on prevention in children and adolescents. The systematic review was based on a systematic and hand search of all available health economic knowledge in this field. We extracted study characteristics and narratively summarised this information. As a last step in the systematic review, we used a standard tool to critically appraise the included studies to determine the risk of bias.

empirical 'economic logic model' and analysis of relevant impacts

To address research question three, we developed an economic evaluation framework to depict short and long term effects of the parental mental illness for the child and adolescent with an underlying logic model. We used the empirical literature from the systematic search of part I for identifying the spectrum of consequences from parental mental illness and clustered them inductively. In a second step, we discussed possible budget- and resource-relevant impacts that could result for the public budget, but also for the affected children and adolescents and their parents.

critical synthesis of part I and II

Finally to answer research question four, we analysed to what extent the existing health economic studies in this context have taken the identified consequences into account and what methodical conclusions can be drawn.

Results

identified 3 HE studies analysing 3 programmes (2 UK, 1 NL)

CEA (2 studies), CUA (1 study)

clinical outcomes and quality of life outcome

In the course of the systematic search, we identified three health economic studies that analysed three different intervention programmes (HFP-M, IY and PBCM)[2]. Two programmes were implemented in the United Kingdom and one in the Netherlands. All programmes had the aim to improve parenting with a focus on child development. The studies used standard methods of health economic evaluations. Two studies took into account the use of services by parents and their children in other public sectors (e.g. education, criminal justice) and were not limited to costs and services in the health sector. In addition, two programmes mentioned some voluntary services, such as voluntary telephone counselling services. The studies used clinical outcome instruments (HOME and ECBI score)[3], but also outcomes regarding individual quality of life (QALYs)[4] were observed in one study.

1 programme clearly cost-effective

2 programmes tend to be cost-effective

perspective, subgroup, threshold matter

In terms of costs, two of the three programmes (HFP-M and IY) were less expensive than the respective comparison treatment. In two programmes (HFP-M and PBCM), the treated group showed a greater improvement in symptoms or outcomes than the comparison group. However, one programme (IY) showed mixed results depending on the analysis perspective chosen. In summary, two programmes (HFP-M and IY) showed mixed results in terms of cost-effectiveness depending on the perspective, subgroup considered or decision-relevant threshold, but cost-effectiveness (efficiency) tended to be present. In contrast, the PBCM programme was cost-effective from all perspectives. The modelling of the long-term savings of a programme (IY) showed for one modelling scenario that the IY programme is promising in relation to monetary returns from the resources used.

low to moderate risk of bias

Because the studies followed the standard methods of health economic evaluations, they also met most of the quality standards and had only a low to moderate risk of bias.

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[2] The abbreviations stand for Helping Families Programme-Modified (HFP-M), Incredible Years® Basic Parenting Programme (IY) and Preventive Basic Care Management (PBCM).

[3] Home Observation for Measurement of the Environment (HOME) is a measurement instrument for measuring the child's environment; Eyberg Child Behaviour Inventory (ECBI) measures and assesses the frequency and severity of behavioural disorders in the home and school environment.

[4] A quality-adjusted life year (QALY) is a measure to evaluate a life year in relation to health.

39 studies considering impacts of the underlying parental mental illness

In the course of answering research question three, we were able to identify 39 studies that could be categorised into four impact categories:

- Physical and psychological consequences for the children and adolescents,
- Effects on social functioning,
- Socio-economic consequences for the children and adolescents, and
- Societal implications

that materialise due to the parental illness.

parental mental illness has not only health consequences

wide range of possible consequences for children and adolescents

also societal consequences

The identified studies showed that a wide range of possible consequences for children and adolescents at the individual level, but also at the societal level are associated with the parental mental illness. In addition to mental health issues, effects on physical health have also been observed, such as unhealthy nutrition or oral health. Parental mental illness also potentially affects the children's and adolescent's social competence and consequently their social life. Furthermore, parental mental illness also seems to have a negative influence on the children's and adolescents' attendance at school in some cases, but also on their general academic performance. From a societal point of view, there are also macroeconomic effects such as loss of income or productivity, but also the immediate social environment of the children and adolescents, such as friends or teachers, is affected.

follow-up costs also incurred in other public sectors

education, social care, and criminal justice sector

public and private costs

All these potentially negative consequences have at first glance an impact on the available resources, the necessary personnel, and entail subsequent costs in the health sector. However, a large number of impacts also affect other public sectors and may materialise at a later point in time. Follow-up costs are also incurred in the education sector, as some of the children and adolescents affected show poorer school performance and these have to be mitigated by socio-political measures such as special educational support programmes. Another affected sector is the criminal justice sector. Costs of incarceration or other measures related to offences, such as probation assistance, are potential consequences. The negative consequences of the parent's mental illness can also lead to private costs such as co-payments for treatment, costs for tutoring, reduced income, or costs due to waiting and travelling times for treatment. Possible early retirement leads to both public and private costs.

Discussion

discrepancies between current HE study situation and relevant costs/outcomes

When comparing the results of both parts of the report, discrepancies emerged with regard to the relevant outcome instruments and relevant costs and resources. None of the applied outcome instruments in the existing economic evaluations address dimensions of social functioning such as social competence, empathy, and tolerance. No instrument captured short-term socio-economic dimensions at the individual level (e.g. school attendance, level of education) or influences on socio-economic dimensions in later life (employment, income level, etc.). If health consequences were measured they were usually only capturing one dimension of health (e.g. mental health) or one diagnosis (e.g. conduct disorders).

many questions to answer and topics to address by researchers and policy makers

On the basis of these findings researchers and policy makers need to address a number of issues and open questions with regard to the current health economic evidence of interventions in the field of parental mental illness. For example, cost and outcome parameters in existing health economic studies do not capture the full range of possible costs and benefits. Thus, although there have been few methodological concerns about quality, the current qual-

ity assessment questionnaires do not appear to be designed to capture these types of limitations. The gaps in the methods may in turn lead to limited decision support for policy makers and health planners.

methodological conclusions:

A number of methodological conclusions and recommendations for future health-economic evaluations of preventive interventions in the field of parental mental illness can be drawn from our report. The perspective of evaluation should go beyond the boundaries of the health sector. This means that public sectors such as the social care sector, education and the criminal justice sector should also be taken into account. But also individual costs and expenses of parents, children, and adolescents should not be neglected. Outcome parameters should be chosen carefully. For example, QALYs are not necessarily appropriate for capturing the multiple impacts of a programme, especially in the long term and outcomes need to be measured in the long run. Common checklists for reviewing study quality in the field of mental health should be revised. Furthermore, cost-effectiveness and cost-utility analyses are usually limited to only one outcome parameter. This is probably not sufficient to capture the whole picture of the consequences for the children and adolescents concerned. Cost-consequence analysis may be a more appropriate study design.

broader perspective and comprehensive outcome parameters necessary

revising quality appraisal checklists

standard HE methods not sufficient?

specific recommendations for the Village project

In addition to the general methodological considerations for health economic evaluations in this area, we have also elaborated specific recommendations for a health economic study in the course of the 'Village Programme' which is currently implemented in Tyrol to identify children who have a parent with a mental disorder and support them in everyday life. These recommendations mainly concern resources that should be taken into account for the implementation of the programme, costs resulting from the negative consequences of parental mental illness that can potentially be prevented, and outcomes that should be considered in the course of a (health economic) evaluation, in particular a cost-consequence analysis.

Conclusion

increasing evidence on the effectiveness of family-oriented interventions

In recent years, there has been increasing evidence that family-oriented complex interventions have significant preventive and therapeutic effects on the health of children and adolescents. In the meantime, these family-oriented interventions are also being implemented in German-speaking countries, as the 'Village Project' in Austria or the programme "Nicht von schlechten Eltern" (NischE) in Germany show. However, as resources in health and social care are limited, evidence of effectiveness alone is not sufficient to make decisions about the use of resources to maximise the health of the population in general, and more specifically of those affected within a given budget. However, standard methods of health economic evaluation are limited in assessing intervention programmes of a complex nature.

BUT: HE evidence to support decision-making is scarce and partially has limitations

report proposes solutions to improve HE evaluations and illustrates the broad spectrum of (economic) impacts of parental mental illness

This report attempts to address these limitations and proposes solutions to improve the health economic evaluation methodology in the field of children and adolescents from families with mental illnesses. The application of the methods in their current form could lead to a misjudgement of the cost-effectiveness of such programmes and thus to wrong decisions regarding funding. Last but not least, the report highlights the broad spectrum of economic effects of parental mental illness and the need to prevent them through targeted prevention.

Zusammenfassung

Einleitung

Warum werden psychische Erkrankungen über Generationen weitergetragen?

Kinder aus psychisch belasteten Familien haben erhöhtes gesundheitliches Risiko

Psychische Erkrankungen wie beispielsweise Depression sind alles andere als selten und beeinflussen nicht nur die davon direkt betroffene Person. Speziell Kinder und Jugendliche aus so genannten „psychisch belasteten Familien[5]“, in denen zumindest ein Elternteil eine diagnostizierte psychische Erkrankung erlebt, haben deshalb auf dem Weg zum Erwachsenwerden ein potentiell erhöhtes Risiko selbst gesundheitliche Probleme zu entwickeln sowie weitere negative Folgen zu erleben.

generationenübergreifender Kreislauf psychischer Erkrankungen erfordert therapeutische und präventive Maßnahmen

Auch wenn dieses Risiko nicht zwingend bei allen betroffenen Kindern und Jugendlichen erhöht ist, ist ein nicht zu vernachlässigender Anteil von Kindern und Jugendlichen betroffen. Die Ursachen für diesen generationenübergreifenden Kreislauf psychischer Erkrankungen sind vielfältig und stehen meist in Wechselwirkung. In der Fachliteratur gibt es Hinweise darauf, dass neben genetischen Einflüssen auch sozioökonomische Umstände wie Arbeitslosigkeit der Eltern, materielle Benachteiligung oder geringere Chancen auf höhere Bildung Auswirkungen auf die Entwicklung von Kindern und Jugendlichen haben. All diese Faktoren erfordern Maßnahmen zur Förderung der psychischen Gesundheit der Kinder und Jugendlichen aus psychisch belasteten Familien, um den generationenübergreifenden Kreislauf der psychischen Erkrankungen und deren negative Begleiterscheinung zu durchbrechen.

Durch welche Interventionen kann der generationenübergreifende Kreislauf psychischer Erkrankungen durchbrochen werden?

familienorientierte komplexe Interventionen als Option?

Lücken im gesundheitsökonomischen (ges.ök.) Wissen

Eine Option, um negative Folgen auf betroffene Kinder und Jugendliche abzumildern oder sogar zu verhindern, sind familienorientierte Interventionsprogramme in Verbindung mit der Aktivierung sozialer Unterstützungssysteme[6]. Diese Programme verfolgen zumeist einen präventiven Ansatz und sind von komplexer Natur, da sie über einzelne klassische Psychotherapieformen wie der Verhaltenstherapie hinausgehen und Personen aus unterschiedlichen Gesundheits- und Pflegeberufen miteinbinden. Bis vor kurzem fehlte es nicht nur am Angebot solcher Programme im Versorgungsalltag. Auch Studien über die Wirkung(sweise) dieser familienorientierten Interventionen waren rar. Insbesondere die Lücke an gesundheitsökonomischem Wissen in diesem Bereich ist eine Hürde, um die Komplexität dieser Programme zu erfassen.

[5] In der Fachliteratur werden Kinder und Jugendliche aus psychisch belasteten Familien als Children of Parents with a Mental Illness (COPMI) bezeichnet. Der Begriff COPMI schließt Kinder und Jugendliche mit ein.

[6] Mit sozialer Unterstützung ist die emotionale oder materielle Unterstützung, die einer Person durch das soziale Netzwerk (andere Personen, Gruppen, größere Gemeinschaften) zur Verfügung steht, gemeint.

Ziele des Berichts

Ziele: Informationen für Forscher*innen für Folgenabschätzung bereitstellen …

Im Zuge dieses Berichts versuchten wir, auf diese bestehenden Schwächen hinzuweisen. Der Bericht soll zudem als Informationsquelle für (gesundheitsökonomische) Forscher*innen dienen, um fundierte Bewertungen und Folgenabschätzungen von familienorientierten Interventionsprogrammen im Kontext von Kindern und Jugendlichen aus belasteten Familien durchzuführen.

… Bewusstsein für wirtschaftliche und gesellschaftliche Dimension der Erkrankung schaffen

Schließlich wollen wir auch das allgemeine Bewusstsein von Gesundheitsplaner*innen und politischen Entscheidungsträger*innen für die wirtschaftlichen und sozialen Dimensionen von psychischen Erkrankungen in Familien schärfen.

Forschungsfragen:

Folgende Forschungsfragen (FF) werden im Bericht adressiert:

internationale ökonomische Evidenz familienorientierter komplexer Interventionen?

FF1. Welche internationalen Belege gibt es für die Kosteneffektivität familienorientierter komplexer Interventionsprogramme für Kinder und Jugendliche von psychisch erkrankten Eltern, die sich auf die Prävention konzentrieren? Welche Methoden werden zur Bewertung der Kosteneffektivität verwendet? Welche Programme, Kostenkategorien und Ergebnisse werden in den Studien behandelt?

Qualität der Studien?

FF2. Wie ist die Qualität der identifizierten gesundheitsökonomischen Studien und inwieweit sind die Daten (Effekte, Population, Leistungen, Kosten etc.) der Studien für den österreichischen Kontext verallgemeinerbar?

Wirkung der elterlichen Erkrankung auf die Kinder?

FF3. Wie wirkt sich eine psychische Erkrankung eines oder beider Elternteile auf die individuelle Gesundheit und auf sozioökonomische Rahmenbedingungen der Kinder und Jugendliche aus? Welche Auswirkungen ergeben sich für die Gesellschaft?

methodische Schlussfolgerungen für künftige ges.ök Studien?

FF4. Inwieweit berücksichtigen die in FF1 und FF2 identifizierten gesundheitsökonomischen Studien diese Auswirkungen? Welche methodischen Schlussfolgerungen lassen sich für die Durchführung von gesundheitsökonomischen Evaluationen im Bereich der Kinder und Jugendlichen aus psychisch belastenden Familien ableiten?

Methoden

Teil 1: Systematische Übersicht

Methoden – Teil 1: systematische Übersicht von ges.ök. Studien: Zusammenfassung und kritische Bewertung aller verfügbaren ges.ök. Erkenntnisse

Im ersten Teil des Berichts haben wir eine systematische Übersicht von gesundheitsökonomischen Studien im Kontext familienorientierter Interventionen mit Schwerpunkt auf Prävention bei Kindern und Jugendlichen im Alter zwischen dem 4. und 19. Lebensjahr[7] durchgeführt. Gesucht wurde in acht verschiedenen gesundheitswissenschaftlichen und gesundheitsökonomischen Datenbanken. Das Ziel war es, einen systematischen Überblick über die internationale gesundheitsökonomische Studienlage von solchen Interventionen zu geben. Darüber hinaus wurde in weiteren Datenbanken nach zentralen Begriffen zum Thema gesucht, um auch solche Studien zu finden, die nicht in Fachjournalen publiziert wurden.

[7] Diese Altersspanne wurde festgelegt, da die Ergebnisse dann zum einen auf die im Village-Projekt angewandte Maßnahme übertragbar wäre. Zudem ist die Literatur, die jüngere Kinder einbeziehen oder den perinatalen Bereich betreffen, besser erforscht im Gegensatz zu dem vorliegenden Kontext.

tabellarische Darstellung der Studiencharakteristika:

Im Zuge der systematischen Übersicht haben wir unter anderem folgende studienrelevanten Daten tabellarisch dargestellt und narrativ zusammengefasst:

Personengruppen, Programme, Analyseperspektiven, Ressourcen, Kosten, Nutzen etc.

- Personengruppen, die an den Programmen teilnahmen und deren demographische Charakteristika (bspw. Alter und Geschlecht des Kindes etc.)
- eingenommene Analyseperspektive aus der die Kosten berechnet wurden (bspw. Gesundheitssystem, Gesellschaftsperspektive etc.)
- Analysemethode
- Kosten der Programme,
- verwendete Ressourcen,
- Nutzen der Programme etc.

Qualitätsbewertung der Studien

In einem weiteren Schritt haben wir die Studien hinsichtlich ihrer Qualität mit Hilfe eines geläufigen gesundheitsökonomischen Fragenkatalogs bewertet, um die Aussagekraft und das Verzerrungsrisiko der Studien festzustellen.

Teil 2: Ökonomisches Wirkmodell

Teil 2: ökonomisches Wirkmodell auf Basis der Literatur aus Teil 1

Im zweiten Teil des Berichts griffen wir auf die ursprüngliche Literaturrecherche aus Teil 1 zurück und suchten nach Studien, die sich mit den Auswirkungen der elterlichen psychischen Erkrankung auf die Kinder und Jugendlichen beschäftigen. Um die kurz- und langfristigen Auswirkungen der elterlichen psychischen Erkrankung darzustellen, verwendeten wir die Methode des Wirkmodells (Logic-Modell). Wirkmodelle dienen zur grafischen oder narrativen Darstellung von Wenn-Dann-Beziehungen (Kausalbeziehungen) von Interventionen oder wie in unserem Beispiel Auswirkungen von der elterlichen psychischen Erkrankung auf die Kinder und Jugendlichen. Die unterschiedlichen Auswirkungen, die in den Studien empirisch beschrieben wurden, haben wir anschließend kategorisiert in …

kurz- und langfristigen Auswirkungen der elterlichen psychischen Erkrankung auf die Kinder dargestellt und beschrieben

Zuordnung der Auswirkungen in 4 Kategorien

- körperliche und psychische Folgen,
- Auswirkungen auf die soziale Funktionsfähigkeit (bspw. soziale Kompetenz und Interaktion mit Gleichaltrigen),
- sozioökonomische Konsequenzen, welche sich aufgrund der elterlichen psychischen Erkrankung für das einzelne Kind und Jugendlichen ergeben, und
- gesellschaftliche Effekte der elterlichen psychischen Erkrankung.

Fokus auf Kinder und Jugendliche

Die identifizierten Studien wurden nach diesen vier verschiedenen Kategorien gruppiert, diskutiert und grafisch dargestellt. Es gilt zu betonen, dass jede einzelne Auswirkung Rückkopplungseffekte auf die betroffenen Eltern haben kann. Da sich das Modell auf Kinder und Jugendliche konzentriert, werden diese Zusammenhänge nicht abgebildet.

budget- und ressourcenrelevante Auswirkungen beschrieben und Teil 1 und 2 kontrastiert

In einem zweiten Schritt haben wir mögliche budget- und ressourcenrelevante Auswirkungen besprochen, die sich für den öffentlichen Haushalt, aber auch für die betroffenen Kinder und Jugendliche und deren Eltern ergeben könnten. Zuletzt haben wir noch analysiert, inwieweit die bestehenden gesundheitsökonomischen Studien in diesem Kontext die identifizierten Folgen berücksichtigt haben.

Ergebnisse

Systematische Übersicht

3 ges.ök. Studien bzw. Programme identifiziert

Im Zuge der systematischen Suche identifizierten wir drei gesundheitsökonomische Studien, die drei unterschiedliche Interventionsprogramme (HFP-M, IY und PBCM)[8] analysierten.

2 aus Großbritannien, 1 aus Niederlande

unterschiedliche Programmansätze

Zwei der drei gesundheitsökonomischen Studien (HFP-M, IY) kamen aus dem Vereinigten Königreich, das zu den Ländern mit einer langen Tradition im Bereich von gesundheitsökonomischen Evaluationen gehört. Ein Programm (PBCM) ist im niederländischen Gesundheitskontext eingebettet. Obwohl sich die drei identifizierten Programme in Bezug auf die erbrachten gesundheitsrelevanten Leistungen, Gesundheitsbereiche und beteiligten Berufsgruppen teilweise überschneiden, liegen ihnen unterschiedliche konzeptionelle Ansätze und Kernziele zugrunde.

Ziel aller Programme: Verbesserung der Elternschaft mit Fokus auf die Kindesentwicklung

Was sie gemeinsam haben: Alle drei in der Studie untersuchten Programme zielten darauf ab, die Elternschaft zu verbessern, wobei die Entwicklung des Kindes im Mittelpunkt stand. Ein Programm (IY) konzentrierte sich noch zusätzlich auf sozioökonomisch benachteiligte Familien in denen bspw. ein nicht zu vernachlässigender Anteil der Eltern von Arbeitslosigkeit betroffen war. Eine Form der Regelversorgung[9] diente als Vergleichsgruppe in allen drei Studien.

Standardmethoden der ges.ök. Evaluation und eine Modellierung langfristiger Einsparungen

tlw. Berücksichtigung von mehreren öffentlichen Sektoren

Die drei identifizierten Studien verwendeten Standardmethoden der gesundheitsökonomischen Evaluation in Form von Kosteneffektivitätsanalysen (zwei Studien), Kostennutzwertanalysen[10] (eine Studie) und einer ökonomischen Modellierung von langfristigen Einsparungen (eine Studie) aus verschiedenen Perspektiven. Zwei Studien berücksichtigen die Inanspruchnahme von Leistungen durch Eltern und deren Kinder in anderen öffentlichen Sektoren (z. B. im Bildungssektor, Bereich der Strafjustiz) und beschränkten sich nicht nur auf die Kosten und Leistungen im Gesundheitssektor. Zusätzlich wurden in zwei Programmen einige Freiwilligendienste wie etwa ehrenamtliche telefonische Beratungsdienste erwähnt.

klinische Ergebnisinstrumente (2 Studien) und QALYs (1 Studie)

Die Studien verwendeten sowohl klinische Ergebnisinstrumente (HOME- und ECBI-Score)[11], aber auch Ergebnisse hinsichtlich der individuellen Lebensqualität (QALYs)[12] wurden beobachtet.

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[8] Die Abkürzungen stehen für Helping Families Programme-Modified (HFP-M), Incredible Years® Basic Parenting Programme (IY) und Preventive Basic Care Management (PBCM).

[9] In der englischsprachigen Literatur wird der Begriff Care As Usual oder Usual Care verwendet, was so viel bedeutet wie standardmäßige Versorgung, Regel- oder Routineversorgung.

[10] Die Kosten-Nutzwertanalyse ist eine Art von Kosten-Effektivitätsanalyse. Bei der Kosten-Nutzwertanalyse wird ein generisches, die Lebensqualität betreffendes Ergebnismaß wie qualitäts-angepasstes Lebensjahr und nicht ein klinisches Ergebnismaß herangezogen.

[11] Home Observation for Measurement of the Environment (HOME) ist ein Messinstrument zur Messung des kindlichen Umfelds; Eyberg Child Behavior Inventory (ECBI) misst und bewertet die Häufigkeit und den Schweregrad von Verhaltensstörungen im häuslichen und schulischen Umfeld.

[12] Ein qualitätskorrigiertes Lebensjahr (QALY) ist eine Maßzahl, um ein Lebensjahr in Relation zur Gesundheit zu bewerten.

Hinsichtlich der Kosten waren zwei der drei Programme[8] (HFP-M und IY) günstiger als die jeweilige Vergleichsbehandlung. In zwei Programmen (HFP-M und PBCM) zeigte die behandelte Gruppe eine stärkere Verbesserung der Symptome bzw. der Ergebnisse als die Vergleichsgruppe. Ein Programm (IY) wies jedoch je nach gewählter Analyseperspektive gemischte Resultate auf.

2 Programme kostengünstiger und besser, 1 Programm mit gemischten Ergebnissen

Zusammenfassend lässt sich sagen, dass zwei Programme (HFP-M und IY) je nach Perspektive, betrachteter Untergruppe (bspw. Vergleich Buben vs. Mädchen) oder entscheidungsrelevanten Schwellenwert unterschiedliche Ergebnisse in Bezug auf ihre Kosteneffektivität zeigten. Im Gegensatz dazu war das PBCM-Programm aus allen Blickwinkel kosteneffektiv. Die Kosten des PBCM-Programm waren zwar aus allen Perspektiven höher als die der Vergleichsintervention. Allerdings gab es in der PBCM-Gruppe im Vergleich zur Vergleichsgruppe ausschließlich Verbesserungen im HOME-Score und die relevante Kennzahl, um die Kosten-Effektivität zu bewerten, ICER[13], lag unter dem entscheidungsrelevanten Schwellenwert. Die Modellierung der langfristigen Einsparungen eines Programms (IY) zeigte für ein Modellierungsszenario, dass das IY-Programm im Verhältnis zum aufgewandten Ressourceneinsatz erfolgsversprechend ist.

1 Programm eindeutig kosteneffektiv, 2 Programme tendenziell kosteneffektiv (abhängig von Perspektive, Schwellenwert, Subgruppe)

Modellierung zeigt langfristige Einsparungen für ein Szenario

Aufgrund dessen, dass sich die Studien an die Standardmethoden gesundheitsökonomischer Evaluationen hielten, erfüllten sie auch einen Großteil der Qualitätsstandards und wiesen nur ein geringes bis moderates Verzerrungsrisiko auf.

geringes bis moderates Verzerrungsrisiko

Ökonomisches Wirkmodell: Welche Einflüsse der elterlichen Erkrankung auf die Kinder und Jugendliche konnten gefunden werden?

Im Zuge des zweiten Teils des Berichts konnten wir 39 Studien zu den vier definierten Kategorien…

- körperliche und psychische Folgen,
- Auswirkungen auf die soziale Funktionsfähigkeit,
- sozioökonomische Konsequenzen, und
- gesellschaftliche Implikationen,

welche sich für die Kinder und Jugendliche aufgrund der elterlichen Erkrankung ergeben, finden.

39 Studien zu gesundheitlichen Folgen, Auswirkungen auf soziale Funktionsfähigkeit, sozioök. Folgen, und gesellschaftliche Implikationen

Die Resultate bestätigen, dass Kinder und Jugendliche aus psychisch belasteten Familien ein erhöhtes Risiko haben, eine psychische Erkrankung zu entwickeln, beispielsweise eine Depression oder Verhaltensauffälligkeiten wie aggressives Verhalten. Neben der psychischen Gesundheit wurden aber auch Auswirkungen auf die körperliche Gesundheit beobachtet, wie etwa ungesunde Ernährung oder Zahngesundheit. Diese Umstände stellen die Kinder und Jugendlichen nicht nur vor individuelle Probleme, sondern wirken sich auch auf ihre soziale Kompetenz und folglich auch auf ihr soziales Leben aus. Des Weiteren scheint die elterliche psychische Erkrankung auch einen negativen Einfluss auf die Anwesenheit der Kinder und Jugendlichen in der Schule, aber auch auf die allgemeinen schulischen Leistungen zu haben.

mögliche Folgen auf …

psychische und physische Gesundheit,

soziale Kompetenz, Anwesenheit in der Schule und schulische Leistungen

[13] Das inkrementelle Kosteneffektivitätsverhältnis (ICER) ist eine Kennzahl zur Bewertung der Kosteneffektivität. Liegt dieses Verhältnis unter einem gewissen Schwellenwert, dann ist die Intervention kosteneffektiv.

Arbeitslosigkeit, prekäre Arbeitssituation im Erwachsenenalter

Einkommens- und Produktivitätsverluste

Zudem besteht ein erhöhtes Risiko, dass die betroffenen Kinder und Jugendlichen in ihrem Erwachsenenleben vermehrt von Arbeitslosigkeit oder prekären Berufssituationen betroffen sind. In einigen Fällen wurden auch vermehrte kriminelle Delikte beobachtet. Gesellschaftlich gesehen ergeben sich gesamtwirtschaftliche Auswirkungen wie Einkommens- oder Produktivitätsverlust aber auch das unmittelbare soziale Umfeld der betroffenen Kinder und Jugendliche wie Freund*innen oder Lehrer*innen ist betroffen. Zudem ergeben sich auch Konsequenzen für das Gesundheits- und Sozialsystem, da diese Kinder und Jugendlichen in ihrer Kindheit aber auch im Laufe ihr Erwachsenenleben vermutlich eine höhere Nutzung von gesundheitlichen und sozialen Leistungen wie vermehrte Krankenhausaufenthalte haben.

Welche weiteren Folgekosten ergeben sich aus diesen Zusammenhängen?

Folgekosten:

Gesundheitssektor: mehr medizinische Leistungen → mehr budgetäre Ressourcen und Personal nötig

All diese potenziell negativen Konsequenzen haben Einfluss auf die verfügbaren Mittel und das nötige Personal in verschiedenen gesellschaftsrelevanten Sektoren. Bspw. müssen aufgrund nicht adäquater oder fehlender Maßnahmen in der Kindheit, betroffene Kinder und Jugendliche in ihrem Erwachsenenleben mehr psychiatrische aber auch andere medizinische Leistungen beanspruchen. Das bedeutet aber auch, dass mehr budgetäre Ressourcen aufgewendet und Personal im Gesundheitssektor benötigt werden, um den Auswirkungen entgegenzuwirken.

Bildungssektor: sonderpädagogische Förderungsprogramme

Justizsektor: bspw. Bewährungshilfe

private Kosten: Selbstbeteiligung, Warte- und Fahrtzeiten

Zudem fallen auch Folgekosten im Bildungssektor an, da einige der betroffenen Kinder und Jugendlichen schlechtere Schulleistungen aufweisen und diese durch gesellschaftspolitischen Maßnahmen wie sonderpädagogischen Förderungsprogramme abgeschwächt werden müssen. Ein weiterer betroffener Sektor ist das Justizsystem, das die Kosten für Inhaftierung oder andere Maßnahmen im Zusammenhang mit Delikten, wie etwa Bewährungshilfe, trägt. Die negativen Folgen der psychischen Erkrankung der Eltern können auch zu privaten Kosten wie bspw. Selbstbeteiligungen für die Behandlung, Kosten für Nachhilfe, verringertes Einkommen oder Kosten aufgrund von Warte- und Fahrtzeiten für Behandlungen führen. Mögliche Frühpensionierungen führen sowohl öffentliche als auch private Kosten herbei.

Diskussion

Diskrepanzen zwischen Studien aus Teil 1 und 2

Bei der Gegenüberstellung der Ergebnisse von beiden Berichtsteilen ergaben sich Diskrepanzen hinsichtlich der relevanten Ergebnisinstrumente und relevanten Kosten und Ressourcen.

soziale Funktion nicht berücksichtigt

- Keines der angewandten Ergebnisinstrumente befasst sich mit Dimensionen der sozialen Funktionsfähigkeit wie soziale Kompetenz, Empathie, Toleranz etc.

individuelle und langfristige sozioökon. Dimension nicht erfasst

- Kein Instrument erfasst die kurzfristigen sozioökonomischen Dimensionen auf individueller Ebene (z. B. Schulbesuch, Bildungsniveau)
- Einflüsse des Programms auf sozioökonomische Dimensionen im späteren Leben (Beschäftigung, Einkommenshöhe usw.) werden auch nicht erfasst.

gesamtgesellschaftliche Auswirkungen werden nicht erfasst

Die Instrumente erfassen auch nicht die gesamtgesellschaftlichen Produktivitätsgewinne, die sich aus den positiven langfristigen Auswirkungen der Programme auf Krankenstand, Frühpensionierung oder vorzeitige Sterblichkeit der Kinder und Jugendlichen ergeben können. Zudem liegen auch generelle methodische Schwächen der gesundheitsökonomischen Standardmethoden auf der Hand.

offene Themen und Fragen

Von Forscher*innen und Entscheidungsträger*innen sind ausgehend von den Ergebnissen aus Teil 1 und 2 des Berichts und der Gegenüberstellung der Ergebnisse im vorliegenden Kontext, eine Reihe an Themen und offenen Fragen zu adressieren:

heterogene Programme und fehlende ges.ök. Evidenz

- Heterogene Interventionsprogramme und fehlende gesundheitsökonomische Evidenz: Obwohl die Zahl der Programme für Kinder und Jugendliche aus psychisch belasteten Familien in den letzten Jahren beträchtlich gestiegen ist, besteht eine erhebliche Wissenslücke in Bezug auf die gesundheitsökonomische Studienlage vieler bereits durchgeführter Interventionen.

Übertragbarkeit der Studien heikel

- Angelsächsische Dominanz begrenzt Übertragbarkeit: Die Unterschiede zwischen den Gesundheitssystemen des Vereinigten Königreichs und Österreichs schränken die Übertragbarkeit der Ergebnisse auf den österreichischen Kontext ein.

teils eingeschränkte Interpretation der Ergebnisse

- Die Evidenz der Kosteneffektivität ist auf bestimmte Untergruppen beschränkt und die Ansätze zur gesundheitsökonomischen Bewertung schränken die Interpretation ein, da in den vorliegenden Studien meist willkürliche Schwellenwerte für die Bewertung der Kosteneffektivität verwendet wurden.

Ergebnisse teils abhängig von gewählten Parametern

- Die Kosteneffektivität hängt zudem von verschiedenen Parametern wie der Perspektive, der betrachteten Subgruppe oder den angewandten Schwellenwerten ab.

Vergleichbarkeit und Verallgemeinerbarkeit der Kosten/Studien nur bedingt zulässig

- Die Kosten der vorliegenden Programme variieren und sind zwar insgesamt niedrig, aber die Vergleichbarkeit der Kosten über die Studien hinweg ist eingeschränkt (Ursache: keine standardisierten Kosten oder Leistungen). Zusätzlich ist die vollständige Umlegung der in den Studien berücksichtigten Kosten über den vollen Krankheitsverlauf aufgrund des kurzen Beobachtungszeitraums nur bedingt zulässig.

elterliche psychische Erkrankung hat nicht nur gesundheitliche Folgen

Unser ökonomisches Wirkmodell zeigte eine breite Palette möglicher Konsequenzen für Kinder und Jugendliche auf individueller, aber auch auf gesellschaftlicher Ebene. Die Auswirkungen auf die Gesundheit beschränken sich nicht nur auf die psychische Gesundheit, wie der Begriff "intergenerationaler Kreislauf psychischer Erkrankungen" vorläufig vermuten lässt. Die Daten zeigten auch eine Reihe von Auswirkungen auf die soziale Funktionsfähigkeit. Zudem können diese Kinder und Jugendliche im späteren Leben auch sozioökonomische Nachteile erfahren. Insgesamt können diese Implikationen zu umfassenderen gesellschaftlichen Kosten führen, z. B. in Form eines erhöhten Bedarfs an psychosozialer Betreuung. Die Auswirkungen können früh, aber auch auf dem Weg zum Erwachsenenalter auftreten.

breite Palette an möglichen Konsequenzen für Kinder und Jugendliche

andere öffentliche Sektoren neben dem Gesundheitssektor betroffen

Viele der identifizierten Auswirkungen fallen auf den ersten Blick zwar in den Zuständigkeitsbereich des Gesundheitssektors. Eine große Zahl betrifft jedoch auch andere öffentliche Sektoren. Wir haben Beispiele für den Sozial-, Bildungs- und Strafrechtssektor aufgeführt. Darüber hinaus können auch private Kosten entstehen, zum Beispiel durch private Zuzahlungen für Behandlungen. Das bedeutet, dass bei erfolgreichen Präventionsprogrammen nicht nur Kosten in der (psychischen) Gesundheitsversorgung, sondern möglicherweise auch in anderen Bereichen vermieden werden können.

auch private Kosten

Kosten- und Ergebnisparameter in ges.ök. Studien teils nicht erfasst obwohl Studien gute Qualität aufweisen

Bei der Gegenüberstellung der Studien aus Teil 1 mit dem Wirkmodell wurde deutlich, dass die Kosten- und Ergebnisparameter in bestehenden gesundheitsökonomischen Studien nicht das gesamte Spektrum möglicher Kosten und Nutzen erfassen. Obwohl es hinsichtlich der Qualität kaum methodische Bedenken gab, scheinen die derzeitigen Fragenkataloge zur Qualitätsbewertung

→ Auswirkung auf die Entscheidungsunterstützung

daher nicht darauf ausgelegt zu sein, diese Art von Limitationen zu erfassen. Die Lücken in den Methoden können wiederum dazu führen, dass die Entscheidungsunterstützung für politische Entscheidungsträger*innen und Gesundheitsplaner*innen wie das Gesundheitsministerium mangelhaft ist und als schlimmste Folge nötige Präventionsprogramme nicht finanziert werden.

methodische Schlussfolgerungen und Empfehlungen

Aus den Ergebnissen beider Berichtsteile lassen sich eine Reihe von methodischen Schlussfolgerungen und Empfehlungen für zukünftige (gesundheitsökonomische) Evaluationen von präventiven Intervention im Bereich der elterlichen psychischen Erkrankung ableiten:

breite Perspektive

- Betrachtungsweise sollte über die Grenzen des Gesundheitssektors hinausgehen. Das bedeutet, dass auch öffentliche Sektoren wie das Sozialwesen, Bildungswesen, Strafrechtssektor berücksichtigt werden. Aber auch individuelle Kosten und Aufwendungen der Eltern, Kinder und Jugendlichen dürfen nicht vernachlässigt werden.

langfristige Betrachtung

- Langfristige Betrachtungen im Bereich psychischer Gesundheit mithilfe von Modellierungsstudien oder Längsschnittstudien sind essenziell, um langfristige (Kosten)Konsequenzen einzufangen und die Gesundheitsplanung zu unterstützen.

Einbindung der gesundheitsökonomischen Perspektive

- Gesundheitsökonom*innen sollten von Beginn an miteingebunden werden und auch die Entwicklung eines eigenständigen ökonomischen Wirkmodells zusätzlich zu den Wirkmodellen der Wirksamkeitsstudien ist sinnvoll. Mithilfe dieser beiden Ansätze können nötige Ressourcen und Folgen besser eingeschätzt werden und relevante sozioökonomische Informationen der Eltern aber auch die der Kinder und Jugendlichen werden so bei den Datenerhebungen besser berücksichtigt.

Ergebnisparameter sorgfältig wählen

- Die Ergebnisparameter sollten sorgfältig gewählt werden. Bspw. sind QALYs nicht unbedingt geeignet, um die vielfältigen Auswirkungen eines Programms zu erfassen, vor allem nicht auf lange Sicht.

Doppelzählung vermeiden

- Vorsicht ist auch bei der Bewertung von immateriellen Kosten- und Nutzenkennziffern gegeben. Hier kann es zu einer Doppelzählung kommen, was zu verzerrten Ergebnissen führen kann.

gebräuchliche Qualitäts-Checklisten angemessen?

Kosten-Konsequenz-Analyse besser geeignet?

- Gebräuchliche Checklisten zur Überprüfung der Studienqualität im Bereich der psychischen Gesundheit sollten überarbeitet werden. Darüber hinaus beschränken sich Kosten-Effektivitäts- und Kosten-Nutzwert-Analysen meist nur auf einen einzigen Ergebnisparameter. Dies ist wahrscheinlich nicht ausreichend, um das gesamte Bild der Folgen für die betroffenen Kinder und Jugendliche zu erfassen. Aus diesem Grund sollte eher auf eine Kosten-Konsequenz-Analyse zurückgegriffen werden.

Verteilungseffekte berücksichtigen!

- Außerdem sollten auch mehrere Subgruppen miteinbezogen werden. Das ist wichtig, um mögliche unterschiedliche Wirkungsweisen des Programms auf unterschiedliche teilnehmende Gruppen, sogenannte Verteilungseffekte, aufzudecken.

Village-spezifische Vorschläge

Neben den allgemeinen methodischen Überlegungen für gesundheitsökonomische Evaluationen in diesem Bereich haben wir auch noch spezifische Vorschläge für eine gesundheitsökonomische Studie des Village-Progamms herausgearbeitet.

Diese Vorschläge betreffen vor allem …

- die Ressourcen, welche für die Kostenberechnung des Programms selbst berücksichtigt werden sollten (Personalbedarf, Materialien, Infrastruktur inklusive Transportmittel, Zeitressourcen etc.), **notwendige Ressourcen?**
- die Kosten, welche sich durch die negativen Konsequenzen der elterlichen psychischen Erkrankung ergeben und potentiell durch das Village-Programm verhindert werden können, **Kosten der elterlichen psychischen Erkrankung?**
- die Endpunkte, welche im Zuge einer (gesundheitsökonomischen) Evaluation oder Kosten-Folgen-Analyse herangezogen werden sollten. **relevante Endpunkte?**

Schlussfolgerung

In den letzten Jahren gibt es immer mehr Belege dafür, dass familienorientierte komplexe Interventionen signifikante präventive und therapeutische Erfolge auf die Gesundheit der Kinder und Jugendliche haben. Mittlerweile kommen diese familienorientierten Interventionen auch in deutschsprachigen Ländern zum Einsatz wie das „Village Projekt“ in Österreich oder das Programm „Nicht von schlechten Eltern“ (NischE) in Deutschland beweisen. Da jedoch die Ressourcen im Gesundheits- und Sozialwesen begrenzt sind, reichen einfache Nachweise über die Wirksamkeit allein nicht aus, um Entscheidungen über die Ressourcenverwendung zu treffen, die die Gesundheit der Bevölkerung im Allgemeinen und im speziellen der Betroffenen im Rahmen eines bestimmten Budgets maximieren. Standardmethoden der gesundheitsökonomischen Bewertung sind jedoch limitiert, um Interventionsprogramme von komplexer Natur zu bewerten.

zunehmende Evidenz zur Wirksamkeit von familienorientierten Interventionen

ABER: ges.ök. Studien zur Entscheidungs-unterstützung ist rar und weisen teils Limitationen auf

Der vorliegende Bericht versucht auf diese Limitationen hinzuweisen und schlägt Lösungsansätze vor, um die gesundheitsökonomische Bewertungsmethodik und die gesundheitsökonomische Aussagekraft von familienorientierten Interventionsprogrammen im Rahmen von Kindern und Jugendlichen aus psychisch belasteten Familien zu verbessern. Die Anwendung der Methoden in der derzeitigen Form könnte zu einer Fehleinschätzung der Wirtschaftlichkeit von derartigen Programmen und somit zu Fehlentscheidungen bezüglich Finanzierung führen. Der Bericht verdeutlicht nicht zuletzt das breite Spektrum ökonomischer Auswirkungen elterlicher psychischer Erkrankungen und die Notwendigkeit, diesen durch gezielte Prävention entgegenzusteuern.

Bericht zeigt Lösungsansätze auf und verdeutlicht das breite Spektrum an ökonomischen Auswirkungen der elterlichen psychischen Erkrankung

1 Introduction

1.1 Background

ca. eines aus 5 Kindern lebt mit einem Elternteil zusammen, der eine psychische Krankheit erlebt

Population estimates indicate that over 50% of people with a lifetime diagnosis of mental illness are parents and up to 60% of people with a severe mental illness (SMI)[14] live with one or more children [5, 6]. In Austria, the latest prevalence study on mental illness demonstrated that from the 22% of study participants (aged 18 to 65) who had a mental illness in the previous year, more than a fifth had at least one child below the age of 18 [7]. Additionally, international estimates show that one in four to one in five children live with a parent who experiences a mental illness [8-11].

Kinder aus psychisch belasteten Familien haben erhöhtes gesundheitliches Risiko

Children who grow up with a parent who has a mental health problem[15] have an increased risk of developing physical and mental disorders themselves [10] (described as the transgenerational transmission of mental disorders or transgenerational cycle of mental disorders) [12] and/or of developing other types of problems (e.g. educational) due to a range of genetic, environmental, and psychosocial factors. In addition, these children are more likely to utilise mental health services more often compared to children that do not have a parent with a mental illness [13].

präventive Interventionen zeigen positive Wirkungen, aber vergleichsweise wenig komplexe familienorientierte Präventionsprogramme

A number of meta-analyses and systematic reviews demonstrated significant positive outcomes of preventive interventions such as risk reduction to develop the same illness as the parent or mental illness in general, for internalizing symptoms, or the children's' psychopathology [14-17]. However, interventions for which robust evaluation results are available are mainly focussing on (cognitive behaviour) therapy approaches and psycho-education and more studies on parents with small children or new-borns than with older children exist [14]. Knowledge gaps exist on more complex family-oriented interventions (e.g. including different components for parents and children) or on interventions targeting older children.

Forderung nach Ausbau Präventions-/ Unterstützungsmaßnahmen zur Förderung der psychischen Gesundheit von Kindern und Jugendlichen

Several policy documents address the importance of child health at the global and national level, such the UN Convention on the Rights of the Child [18], the Austrian Health Targets ('Gesundheitsziele'), which include the commitment to support healthy growing up for children and adolescents, while ensuring health equity and fostering psychosocial health in the best possible way [19] or the Austrian child and adolescent health strategy ('Kinder- und Jugendgesundheitsstrategie'), which includes the aim to early identify health risks and support of children [20]. This calls for more preventive action and effective support interventions to foster mental health of children and adolescents by interrupting the transgenerational cycle of mental disorders (TCMD).

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[14] SMI is defined as a mental, behavioural, or emotional disorder resulting in severe functional impairment and is understood as a clear deviation from the social or medical norm of mental functions according to the International Classification of Diseases (version 10) codes F00-F99. A SMI substantially interferes with or limits at least one activity in life. In the US context, the term 'serious mental illness' instead of 'severe mental illness' is more common [2-4].

[15] When we are referring to children with a mental disorder in the remainder of the report we will use the term ‚children' including children and adolescents of all age until 19.

1.2 The need for economic research and current limitations

umfassende gesundheitsökonomische (ges.ök) Studien sind selten und beschränken sich meist auf die perinatale psychische Gesundheit

Given that resources are scarce and that one aim of health and social policy is to maximize health and well-being, economic analyses are needed alongside effectiveness evidence for decision-makers to identify the best options in using available resources. Although, there is some evidence on costs [21] and cost-effectiveness [6, 22] of interventions in the area of family mental health, comprehensive economic assessments are rare and mostly limited to perinatal mental health, such as postpartum depression [23-25]. The general scarcity of health economic information calls for more health economic research to inform decision makers on the economic dimensions and for supporting mental health care planning and reimbursement decisions.

Bedarf an ges.ök. Auseinandersetzung im Themengebiet der elterlichen psychischen Erkrankungen

Recently, a programme addressing children who have a parent with a mental illness has been implemented in Tyrol alongside a research project [19]. There will be a need to generate economic evidence for this programme, however, limitations regarding valid data (e.g. cost data) for conducting economic evaluations have been observed in Austria [26]. There is therefore a need to address methodological issues of conducting health economic evaluations (HEEs) in the field of parental mental illness in general, but also considering the Austrian situation specifically.

themenspezifische aber auch wissenschaftstheoretische und methodische Herausforderungen existieren

Generally, two challenges exist regarding economic research related to parental mental illness: One issue relates to limited mental health services research, and the other issue relates to the philosophy of science and applies generally. However, both issues are interrelated. Firstly, there is a lack of estimates on the direct and indirect economic burden of parental mental illnesses and the broader economic impacts: Direct estimates for this specific context are not ready at hand [27]. In most of the cases, data on the global (economic) or regional burden of mental illnesses are used to extrapolate country-specific estimates [27, 28].

meistens werden für diesen Kontext Daten aus anderen Bereichen der psychischen Gesundheit herangezogen

Some information can be derived from available economic evidence in other mental health fields such maternal mental health, child and adolescent mental health, mental health in older persons, and studies investigating the economic dimension of depression or other (severe) mental illnesses [25]. Especially, the economic evidence on perinatal mental health serves as a proxy for wider economic impacts of parental mental illness because of comparable spill over effects into the family and the wider community [25]. For example, a modelling study in the United Kingdom estimated that the total costs of adverse impacts caused by maternal illness during the perinatal period are £ 8.1 billion (~€ 9.65 billion) for each one-year cohort of births [29, 30]. Of these total costs, 72% can be attributed to the child, but costs arose also in other sectors such as the social care sector. [25, 29].

ges.ök Studien vernachlässigen meist breitere sozioökonomische Auswirkungen

Waldmann et al. (2021) published one of the few studies on the costs of health and social services use in children who have a parent with a mental illness for Germany [21]. While total costs per child with a psychiatric diagnosis amounted to € 5691.93 (95% CI: € 4146.27–7451.38), children without a psychiatric diagnosis generated a total of € 1245.01 (95% CI: € 657.44–1871.49) of costs per person. The study only addressed direct costs for services but did not estimate broader socio-economic consequences.

However, such estimates based on other regions' data are not transferrable to other jurisdictions [28]. For Austria, separate studies are needed, taking into account the epidemiological situation and local Austrian costs.

Übertragbarkeit von "fremden" Daten meist nicht möglich

The second, philosophy of science issue concerns ontological and epistemological questions. In health sciences, including health economics, these questions regard choices related to value judgements, the perspective taken, and outcome measurement in the assessment. The debate about value judgements, representative outcomes, or output measures in (social) sciences has been ongoing since the 'positivism dispute' [31]. There have been debates, whether the gross national income and gross domestic product (GDP) as a production output measure, originally conceptualised by Keynes and Kuznets, is a representative measure for well-being and welfare [32]. This debate and resulting issues also affect the application of HEEs.

Fragen bezüglich Werturteilen, der eingenommen Perspektive und der Messung der Endpunkte sind bei der Anwendung von ges.ök. Evaluationen allgegenwärtig

Health economics and associated social value judgements are based on welfare economics (originating in mid-20th century) and its underlying and strong reliance on utilitarianism [33]. Welfare economics, for its part, is strongly influenced by logical positivism, which strictly limits the occurrence of knowledge to observations and feelings. However, the world is constituted not only by single empirical observations in a strict positivist manner, but also by preliminarily unobserved entities[16], underlying structures, and mechanisms that possibly materialise at some point in time by scientific discourse [34]. This issue is shaping health economic research and health decision-making. It has implications on which costs and benefits are considered in HEEs, the evaluation framework applied and the interpretation of the results and, thus, on the priority setting for service funding [33]. As such, the philosophy underlying the HEE methods applied may indirectly discriminate some illnesses or population groups against others.

Gesundheitsökonomie ist maßgeblich beeinflusst von der Wohlfahrtsökonomik und der Nutzenethik (Utilitarismus)

Perspective in health economic studies

HEEs can be conducted from different perspectives. While the health care system perspective covers only those costs that are relevant for the health care payers, the societal perspective includes costs beyond the health care system such as costs in other sectors or productivity losses. In many cases, HEEs fail to capture the socio-economic dimension, benefits beyond immediate health outcomes or benefits and costs outside the health care sector [23] – so-called inter-sectoral costs and benefits (ICBs) [35, 36]. Reasons for this reductionist approach are difficulties to evaluate such interventions with the appropriate evaluation tool and metric [37, 38], but also because standards in decision-making are often to use a health care system perspective, which is ignoring broader economic consequences and or consequences in other sectors.

ges.ök. Evaluationen können aus mehreren Perspektiven gemacht werden, da ökonomische Konsequenzen in verschiedenen Sektoren anfallen

Moreover, a societal perspective in HEEs does not guarantee that all benefits and costs incurred in other sectors, such as the criminal justice sector or educational sector, are addressed in the studies. Yet, costs as well as benefits within these sectors can be substantial [35, 36, 39]. This is especially the case in the area of mental health. [36].

Gesellschaftsperspektive garantiert keine vollständige Kosten- und Nutzenerfassung

[16] Absence of evidence ≠ evidence of absence

in der Praxis wird aber meist eine enge Perspektive eingenommen

In addition to the insufficient consideration of ICBs, methods to calculate the value of all outcomes are limited [36]. Furthermore, accurate benefit and cost accounting is time- and resource-consuming and researchers have a limited amount of both [39]. Consequently, researchers often apply a more narrow evaluation perspective in practice.

Komplexität fordert die traditionellen ges.ök. Methoden heraus

Social systems can be defined as so-called open systems, which are characterised by a multiplicity of mechanisms [34]. The complexity of the social system in which the interventions operate, makes it difficult to accurately capture impacts that go beyond short-term economic consequences. Furthermore, complexity can also arise from the intervention. Hence, such complexities challenge the traditional (health) economic evaluation methods [40].

methodische Weiterentwicklungen sind im Gange

Some methods to assess the societal value of an intervention exist (e.g. the concept of social return on investment (SROI) [41]). Furthermore, methodological development of new standards of economic evaluation that consider impacts beyond health-related outcomes or multi-sectoral impacts from a societal perspective is an on-going agenda [23, 37, 39, 42-44]. Although those methods have their own limitations, they may be useful to capture the economic dimension of relevant aspects and long-term opportunity costs related to parental mental illness and their children.

Measuring benefits

herkömmliche Nutzenparameter und Endpunkte untererfassen weitgehende Auswirkungen auf das Wohlbefinden

With regard to the benefit side, conventional outcome measures leave out the wider benefits closely associated to health and the health concept of well-being. There is an ongoing debate whether outcome measurement in a HEE should be augmented by the capability approach[17] initially developed by Sen [46] and further developed by Nussbaum [47] and whether health related quality of life (HRQoL) measures are sufficient for measuring the multidimensionality of well-being. Well-being consists of both health and non-health outcomes that are important to consider when evaluating broader impacts of health interventions [48]. Measures that capture a broader set of patient impacts are especially relevant for mental health interventions and even more for preventive interventions, because these interventions are not only expected to improve health outcomes, but impact also social relationships, meaning of life, or optimism about the future [48]. Social indicators such as empowerment, social participation, feeling safe, dignity, self-respect, social support, or improvement in help-seeking play an important role in the everyday-life of people.

Measuring costs

Methoden zur Feststellung der Kosten abstrahieren von der sozialen Reproduktion und unbezahlten Arbeit

Whereas direct costs are 'directly' measurable and 'visible', indirect costs are more difficult to capture. Direct costs are for example utilised services such as general practitioner (GP) visits, therapy sessions, hospital admissions, medication etc. Indirect costs are income losses resulting from premature mortality, disability, and care seeking, including lost production due to work absence, or early retirement. In health economics, the most commonly used approach to determine indirect costs is the human capital approach (HCA). One of the flaws in using the HCA is that it only considers production in form of

[17] The capability approach is an alternative to standard utilitarian welfare economics: Outcomes should not serve as the only object of welfare assessment, but should be extended by capabilities when evaluating well-being [45].

lost income and completely abstracts from social reproduction and unpaid work in the informal care economy [49]. These issues arise because the HCA is closely associated to the GDP and the GDP is 'only' an outcome measure of production output and not a pure indicator of well-being. Alternatively, studies employ the friction cost approach. However, this approach is even more restrictive as it just considers cost of productivity loss for employers [50].

1.3 Methodological developments to overcome limitations

Limitation der Methoden können Entscheidungen fehlleiten

The previous sections have demonstrated that the current standards in HEEs may be insufficient to capture the complexity of interventions related to parental mental illness. These insufficiencies bear the risk that decision support from HEEs in programmes is of limited value and decision makers base their decision on incomplete evidence. This drawback in turn affects patients, medical staff, and the population in general. Developments with this regard are on the agenda.

Lösungsansätze und Rahmenwerke zur Vermeidung der Limitationen

To overcome specific limitations regarding the perspective, Drost et al. (2020) [39] propose five recommendations on the way to a more complete consideration of wider impacts on society and well-being (see Table 1-1). The recommendations are not restricted to the health economics of mental health, but apply for health economics in general. Some of the recommended pillars were already discussed above.

Table 1-1: Recommendations for the societal perspective by Drost et al. [39]

Pillars for the societal perspective
1. Irrelevance who bears the impact: Measuring transmission of impacts and spill overs are relevant, not the individual bearing the impact
2. Considering impacts outside the healthcare and informal care sector such as the educational and criminal sector, and move beyond labour productivity/GDP
3. Include high frequent costs, high unit prices (outside the healthcare sector) possibly relevant to the evaluation, and conduct according scenario and sensitivity analyses to target uncertainty
4. Include outcomes beyond clinical outcome measures or generic health outcomes
5. Critical reflection on included costs and benefits

für den Bereich der psychischen Gesundheit existieren spezielle ges.ök. Ansätze wie bspw. PECUNIA und OxCAP-MH

In addition to recommendations for conducting general HEEs, specific research projects investigating evaluation approaches for broader costs and benefits exist for mental health topics. One of the most recent research projects is the 'ProgrammE in Costing, resource-use measurement and outcome valuation for Use in multi-sectoral National and International health economic evaluAtions' (PECUNIA) project [51]. The PECUNIA project aimed to develop new standardised, harmonised, and validated methods for the assessment of costs and outcomes including broader impacts such as ICBs for the mental health domain. Authors of the associated studies conducted systematic reviews on the basis of previous work on ICBs [52] in order to identify further potentially relevant ICBs in the educational [36] and criminal sector

[35]. The result of these studies was a list of ICBs that are incurred in the educational[18] and criminal[19] sector and are potentially relevant when a societal perspective is taken.

OxCAP-MH basiert auf Sens Befähigungsansatz

Another approach that is mental health domain specific and mainly addresses measuring benefits is the Oxford CAPabilities questionnaire-Mental Health (OxCAP-MH). The OxCAP-MH is based on pillars of Sen's capability theory and includes Nussbaum's proposed 10 central human capabilities. The capabilities relate to underpinnings of basic political principles and can be anchored to constitutional rights [47, 53]. The proposed capabilities in Table 1-2 are relevant for truly human functioning.

Table 1-2: Nussbaum's central human capabilities [47], p. 41f

Central human functional capabilities	
1. Life	2. Bodily health
3. Bodily integrity	4. Senses, imagination, and thought
5. Emotions	6. Practical reason
7. Affiliation	8. Other species
9. Play	10. Political and material control over one's environment

es existiert auch eine OxCAP-Version in deutscher Sprache

The OxCap-MH was initially applied in the UK mental health context and has been used in several studies [48]. A German version is also available [54]. Based on the central human capabilities by Nussbaum [47], domains of the German OxCAP-MH are the following: daily activities, social networks, losing sleep over worry, enjoying social and recreational activities, having suitable accommodation, feeling safe, likelihood of discrimination and assault, influencing local decisions, freedom of expression, appreciation of nature, respecting and valuing people, friendship and support, self-determination, imagination, and creativity and access to interesting activities.

OxCAP-Ansatz scheint Wohlergehen besser zu erfassen als EQ-5D

Researchers investigated to what extent the OxCAP-MH measures broader impact categories of well-being compared to the EQ-5D-5L in the course of routine mental health services [48]. The OxCAP-MH seems to be superior compared to the EQ-5D-5L. The tool captures more aspects with regard to well-being than conventional HRQoL measures and is not just complementary [48].

[18] Input: Special education school, additional education services, educational therapy, special needs diagnostics, student counselling, counselling of legal guardians, student transport to education facility, student-related financing, training and support services for teachers; Throughput: Home education; Output: Reduced school readiness, problems with school entry, learning disabilities, reduced school adaption/competence/participation/engagement/attainment/productivity/performance, grade retention, negative school experiences, school (re-)integration, disruptive school behaviour

[19] Input: Police services, fire and rescue services, legal services, services for children/spouse of incarcerated, victim support services, services provided in correctional facilities; Throughput: Organised transport, other costs of correctional facilities, shelters; Output: Pain and suffering, lost freedom of offender, material losses

OxCAP-Ansatz misst ökonomische und gesellschaftliche Auswirkungen umfassender

Although the OxCAP-MH questionnaire focusses on the benefit side, the included capability approach and its categories may also be helpful to identify impacts on the cost side. Hence, for measuring broader economic and societal impacts in the mental health and specifically in the context of parental mental illness, the OxCAP-MH questionnaire may serve as a valid option for methodological development.

Vorsicht vor Doppelzählung: einige ges.ök. Ergebnisse können entweder als Kosten oder Nutzen gemessen werden

Some general pitfalls need to be considered: Two main criteria that evaluation instruments need to fulfil in order to truly measure outcomes or impacts are validity and reliability. That means, instruments need to assure that they actually measure the relevant outcome (validity) and if so, then ideally very precise (reliability) [55]. One issue to consider is that some outcomes can be measured in two ways. On the one hand, the item can be treated as a cost, but it can be assigned to the benefit side in form of quality of life (QoL) as well. This phenomenon is called double counting or cross-domain counting. It mainly concerns intangible impacts (costs as well as benefits) and needs to be avoided [35, 36]. Double counting or cross-domain counting can occur in evaluating (mental) health interventions. If an intervention reduces morbidity, but does not affect life-expectancy then we can show on the basis of a possible consequence of the intervention such as change in productivity or income that double counting can occur. If patients consider the change in income in the assessments of quality weights in QoL-measures, then this it would lead to double-counting if the change in income is also included in the costs [56].

ein universeller Ansatz zur Bewertung immaterieller Effekte scheint bis heute nicht zu existieren

Furthermore, an obstacle in evaluating intangible impacts is whether these impact categories are deemed relevant for the targeted population or society. Differences in the institutional context, culture, and value conceptions of illness, health, medicine, efficiency, equity, and responsibility across countries affect chosen endpoints in decision-making and even determine the relevance of HEE approaches [57, 58]. Consequently, a universal approach to evaluate intangible outcomes does not seem to exist to date [58].

2 Project aims and Research Questions

2.1 Project aims

Part I of the report is supposed to give a systematic overview of HEEs of family-oriented complex interventions, which aim to improve outcomes in children who have a parent with a mental illness and to describe methodological characteristics of the identified studies.

Projektziele:
1. systematische Übersicht ges.ök. Studien

Part II aims to develop an economic framework for future economic evaluation in this field, in particular for guiding the HEE of the currently running 'Village programme' in Austria as described in section 3.2.1.

2. ges.ök. Rahmenwerk für zukünftige ges.ök. Evaluationen

The report should aid as an information source for health (economic) researchers in order to conduct economic evaluations and impact assessments for interventions in the field of parental mental illness. Not least, we aim to raise general awareness in health planners and decision makers on the economic dimensions of mental health problems in families and the need to take economic evidence into account in decision making.

Bericht adressiert Gesundheitswissenschaftler*innen und Entscheidungsträger*innen

2.2 Research questions

The following research questions (RQ) will guide part I of the report:

Forschungsfragen:

RQ1 What is the international evidence on the cost-effectiveness of family-oriented complex interventions focussing on prevention or early intervention in children? Which methods are used to evaluate cost-effectiveness? What programmes, cost categories, and outcomes are addressed in the studies?

internationale ökonomische Evidenz familienorientierter komplexer Interventionen?

RQ2 What is the quality of the identified health economic studies and to what extent is the data (effects, population, services, costs etc.) of the studies generalisable for the Austrian context?

Qualität der ges.ök.Studien?

Part II of the report will answer the following RQs:

RQ3 What is the impact of parental mental illness on individual health and socio-economic outcomes in children and what is the societal impact?

RQ4 To what extent do the identified HEEs from Part I address these impacts and what methodological conclusions can be derived for conducting a HEE in the field of parental mental illness?

Wirkung der elterlichen Erkrankung auf die Kinder und methodische Schlussfolgerungen für künftige ges.ök Studien

3 Methods

3.1 Part I: Systematic review of health economic evaluations

3.1.1 Inclusion and exclusion criteria

Ein- und Ausschlusskriterien:

Fokus auf familienorientierte komplexe Interventionen präventiver Natur für Kinder ab 4 Jahren

As a first step, we addressed RQ1 to RQ2 by a systematic literature search for cost-effectiveness evidence [59] of internationally implemented family-oriented complex interventions focussing on prevention or 'early' intervention in children. In addition, we undertook a hand search to identify relevant grey literature. The scope was limited to interventions including children from four years onwards. The rational for this restriction was to identify HEE targeting a similar age group as in the Village project (see section 3.2.1) and to focus on age groups where less economic knowledge is available (e.g. in comparison to perinatal mental health). In summary the following inclusion criteria guided the search and were are applied for selecting relevant studies:

Table 3-1: Inclusion criteria – PICO-Analysis for part I

Population	■ Children or adolescents of parents with a mental illness aged 4 to 18 and their parents[20] Mental illnesses include all mental and behavioural disorders (F00-F99), e.g. affective disorders, schizophrenia, psychosis, with or without substance misuse
Intervention	Family-oriented[21] complex[22] interventions focussing on prevention or 'early' intervention in children, e.g. ■ Programmes supporting parenting in parents with mental health problems ■ Mental health interventions establishing child-focused support networks Not[23]: Psychotherapeutic interventions, general mental health care services for adults with a mental illness, child and adolescent mental health care services, interventions for perinatal mental health problems, or early interventions starting from pregnancy to the child's age of 3
Control	Any alternative approach including no intervention

[20] This age range was defined to identify economic evaluations on interventions that are similar to the intervention applied in the Village project which included children aged 4 to 18. Interventions including younger children require different approaches and often primarily address parents or mothers.

[21] Refers to interventions which view the person with a mental illness in the context of their family relationships and thus additionally address the social environment (e.g. other family members) rather than just the individual patient experiencing a mental illness.

[22] Complexity arises from the intervention, the context in which an intervention an intervention is implemented, and the interplay of the two. We orient our understanding of a complex intervention on the definition by Skivington et al. (2021): *"Interventions become more complex in line with increasing the number of intervention components and the interactions between them, the range of behaviours, expertise and skills (e.g. particular techniques and communication) required by those delivering or receiving the intervention, the number of groups, organisational levels or settings that are targeted by the intervention, and the level of flexibility or tailoring of the intervention or its components that is permitted (i.e. how dynamic or adaptive the intervention is)."*
(Skivington, Matthews, et al. [60], p.17)

[23] Individual evidence-based interventions such as specific forms of psychotherapy were not the focus of this report. These interventions could, however be part of the family-oriented complex interventions that were included in this review.

Outcomes	■ Health economic outcomes (resource use, costs, effects and cost-effectiveness ratios) … with respect to child and/or parent quality of life and psychopathology; health and well-being outcomes (depression improvement, social functioning shame, stress, self-confidence, improvement in QALYs)
Study design(s)	■ Health economic evaluations (Cost-utility analyses, cost-effectiveness analyses, cost-benefit analyses) ■ Impact analysis (Budget impact analysis, social/societal impact analysis, equality impact assessment/analysis) ■ Other relevant study designs that may be identified (e.g. Social value (assessment), social return on investment, cost-minimisation analyses, cost-consequence analyses, cost of illness studies, beneficiary assessment)
Language	■ English/German
Type of publication	■ (un)Published journal articles and research reports
Time period	■ Beginning from 2010

QALY … quality-adjusted life year

3.1.2 Literature search and selection

Literatursuche

The information specialist (TM) performed systematic literature searches between the 17th-21st of May 2021:

Suche in 8 gesundheitswissenschaftlichen Datenbanken

- Cochrane,
- Centre for Research and Dissemination
- EconLit,
- Embase,
- Medline,
- PsycInfo,
- CINAHL, and
- Web of Science database.

zusätzliche Handsuche

Additionally, we carried out a hand search for relevant literature in the following sources:

- Trip database (Turning research into practice)
- G-I-N database (Guidelines network international), and
- Exploratory web-based literature searches in combination with Google, Google Scholar, and PubMed

Publikationszeitraum: 2010 bis 2021

The systematic search was limited to the years 2010 to 2021 and to articles published in English or German. After deduplication, overall 1,621 citations were available. We attached the search strategy in the Appendix.

insgesamt 1.624 Treffer von denen 3 ges.ök. Studien eingeschlossen wurden

By hand search, we identified three potential publications [61-63]. In total, we identified 1,624 records for abstract screening. Two researchers (CS, LH) independently screened the references. All cases of disagreement were resolved through discussion or by consulting a third person (IZK). In total, 113 publications were eligible for a full text screening. Of the 113 full texts, we included three publications [22, 64, 65] for the systematic review. Eighty-four publications served as an information source for background information. The flow chart in Figure 3-1 displays the selection process.

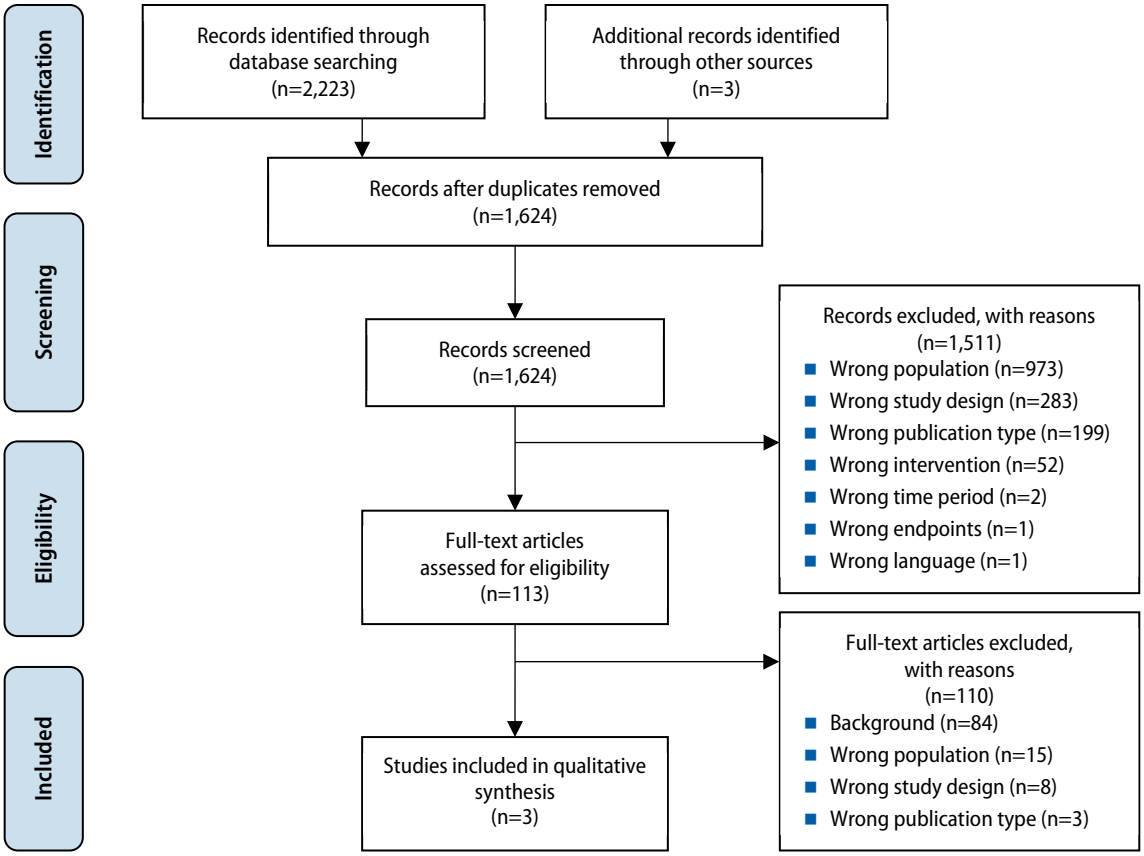

Figure 3-1: Flow chart of study selection (PRISMA Flow Diagram) for part I (systematic review)

3.1.3 Data extraction, analysis, synthesis and risk of bias assessment

Datenextraktion und Beschreibung durch eine Person und Kontrolle durch eine weitere

One author (CS) extracted relevant characteristics of the studies such as population, study period, perspective of analysis etc., and economic categories, such as costs and utilised services in tables. The second author (LH) verified the extracted data. The extracted data was narratively summarised by information category.

kritische Bewertung der ges.ök. Studien mit einem Fragebogen, um das Verzerrungsrisiko zu bewerten

In the course of answering RQ2, we critically appraised the quality of the identified studies independently by applying the (extended) CHEC checklist [66, 67] for assessing (health) economic evaluations[24]. The checklist consists of 20 questions addressing key methodological standards in HEEs, which can be answered by a YES (indicating low risk of bias) and a NO (indicating high risk of bias) category. We augmented the risk of bias judgement categories by adding a new category 'Partly fulfilled (some concerns)'. This adaptation is in line with judgement categories of Cochrane's revised risk of bias tool 2 [68], and enabled us a more differentiated assessment.

Auswertungen der Ergebnisse des Fragebogens

After grading the risk of bias of each economic study for every question in the checklist, we compared the number of fulfilled, unfulfilled, or partly fulfilled categories between studies. Sums of answers to each categories per study are calculated.

[24] The CHEC-extended checklist includes a question on modelling studies. The list is therefore also relevant for modelling studies.

3.2 Part II: Developing a framework for economic evaluation in the field of parental mental illness

3.2.1 Economic evaluation framework

Darstellung von kurz- und langfristigen Folgen mithilfe eines ökonomischen Wirkmodells

The economic evaluation (EE) framework to be developed will illustrate short-term and long-term impacts from parental mental illness in the form of an economic logic model.

Introduction to logic models

Wirkmodell (Logic Model) = Wie funktioniert (k)eine Intervention?

Simply put, a logic model illustrates 'how an intervention works' and 'which outcomes can be expected from an intervention' when implemented or not implemented. Logic models are part of programme theory and overlap with theory of change [69]. A logic model typically defines the following aspects of an intervention:

Teilkomponenten eines Wirkmodells

- the specific context,
- utilised inputs and available resources,
- implemented/planned activities and tasks,
- (expected/desired) outcomes and impacts (in the short-, medium-, and long-run), and
- inter-relationships of these domains.

Wirkmodelle helfen im Evaluierungsprozess, da sie wichtige Aspekte einer Intervention entflechten

These aspects are relevant when evaluating health care or social interventions. In particular, logic models offer a clear overview when interventions are of complex nature, while simultaneously having effects in complex system or so-called open-systems [34]. Hence, an underlying logic model supports the (complex) evaluation process of health care interventions, because these models help to unravel essential aspects and interrelations of the respective intervention. Relevant data associated with the intervention can be collected and analysed according to the model's priority. Furthermore, logic models do not only show why and how interventions work, but also why an intervention does not work with regard to an endpoint.

Wirkmodelle sollten komplexe Zusammenhänge berücksichtigen, aber gleichzeitig praktikabel in der Anwendung sein

The majority of logic models postulate linear paths from utilised inputs to (un)desired or (un)expected impacts. Hence, also the evaluation process of interventions considers only linear (causal) relationships. For 'simple' interventions in clearly described systems, the assumption of a linear relationship is sufficient. Feedback effects and complex relationships – so-called non-linearities – between the relevant aspects are characteristic in complex contexts, and should not be neglected. Offered services in one part of the system can lead to (un)intended consequences in other parts of the system. Therefore, considering interconnectedness in logic models of complex interventions and depicting health services in the context of a complex system that is in constant flux are key when evaluating interventions and their impacts [69]. Furthermore, a logic model can help to decide whether a key variable should be chosen as a moderator and/or a mediator variable in a specific evaluation context[25]. Such models deliver a picture of the blind spots of health and med-

[25] Mediator variables are intermediate variables in a causal chain between two other variables. A mediator variable can establish causation between two otherwise unrelated variables to be related. If two variables depend on the value of a third variable, the moderator variable, then we talk about moderation [70].

ical research, where a deeper understanding of the issue itself and causal relationships in form of more research are necessary.

However, well-devised logic models should always hold the balance between simplicity, tractability, and reflecting the complexity of the real world.

Example for a logic model in the field of parental mental illness

Entwicklung und Einsatz eines Wirkmodells im Village-Projekt

One example is a logic model developed as part of the Village project [1]. It was produced to guide the design, implementation, and evaluation of one part of the programme provided to the families within the project, which was the sensitive screening to early identify children (SENSE). The logic model demonstrates the mechanisms and outcomes of the screening.

Wirkmodell und Teilkomponenten entwickelt auf Basis von Expert*innen-Interviews

bildet 3 Wirkungsdimensionen bei Kindern, Eltern und Fachkräften ab

The primary data source for developing the logic model were interview data from international qualitative expert interviews. The details of the model are demonstrated in Figure 3-1. The model addresses three dimensions of outcomes, the programme may influence: knowledge, emotions and behaviour. Furthermore, it addresses outcomes in three different groups: in children, parents and practitioners. Long-term outcomes for the children that may follow from the intermediate outcomes and their interrelations are: improved relationship with parent, social relationships within and outside the family, academic performance, resilience, and coping. This demonstrates the complexity of mechanisms and outcomes in family-oriented programmes, which are to be taken into account in HEEs as well.

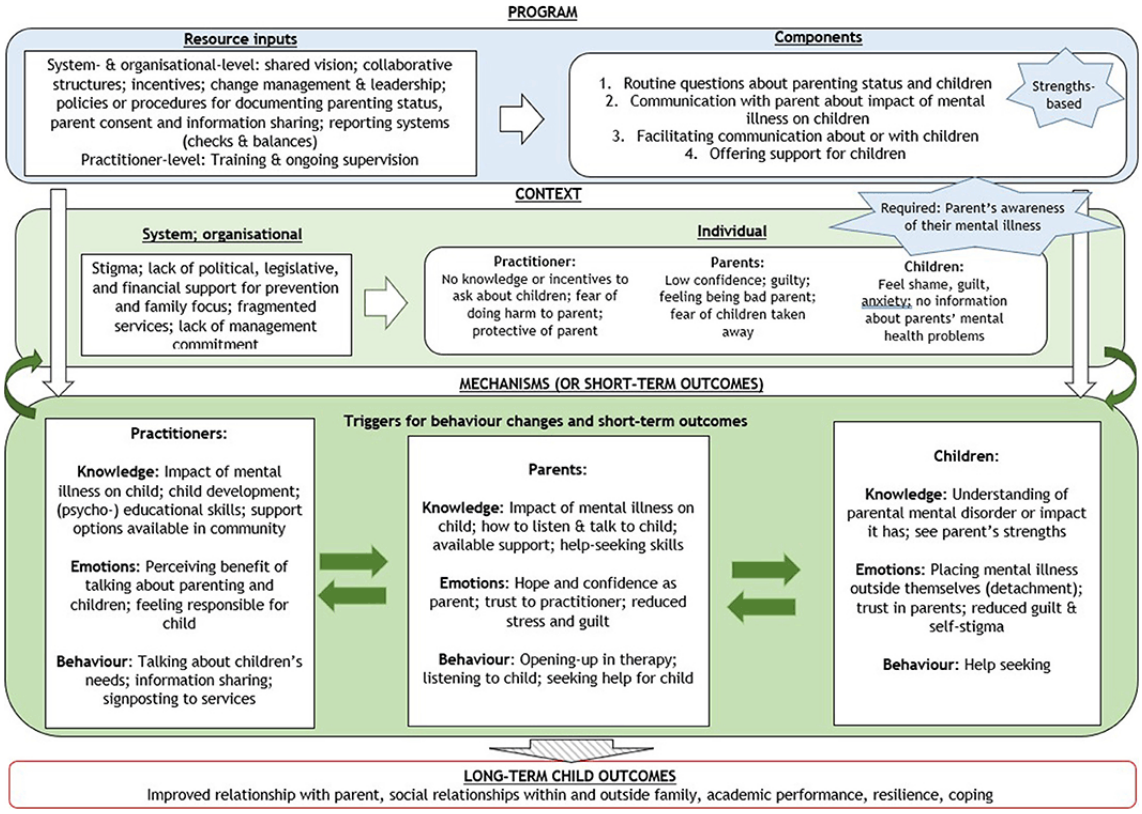

Figure 3-2: Logic model of the Village programme from [1]

A logic model for guiding economic evaluations

Village-Wirkmodell wird durch ein ökonomisches Wirkmodell ergänzt

The logic model presented in the previous section is (among other sources) informing the evaluation of the Village programme. The short-term outcomes that have been identified (which are on the pathway to long-term outcomes), as well as long-term outcomes support the selection of appropriate instruments for data collection (e.g. validated and standardised questionnaires), (qualitative and quantitative) data collection methods, data collection time points and the subjects for data collection (e.g. practitioners, participating children). Overall, the model is used for a realist evaluation regarding what works for whom, why and under which circumstances [1]. However, the model was not designed to guide the HEE, which is planned in addition to the realist evaluation. In this report, we therefore aim to complement the existing Village logic model with another model that can be used to guide the HEE.

Herausforderungen für ges.ök.Evaluationen im vorliegendem Kontext

As discussed above, traditional HEE frameworks do not necessarily account for all possible economic and social impacts. Recent developments have already addressed those limitations (see section 1.3). While mental health was at the core of those developments, we argue that parental mental illness has some unique characteristics. These special characteristics makes it worthwhile to draw specific attention to in terms of methodological challenges for HEEs.

spezielle Charakteristika elterlicher psychischer Erkrankung erfordern gezielte methodische Herangehensweise

For example, children of parents with a mental illness are not necessarily ill themselves and may take up services or receive types of support that may not have been covered by the tools developed in previous projects. Additionally, support for parents may be beyond what is usually part of the treatment for mentally ill patients (e.g. support in parenting skills, support in how to talk to the children about their mental illness). Furthermore, support might also not directly be applied to the child, but more the child's surroundings, and is not necessarily focused on individuals but on family systems as the 'core client'. This challenges the calculation of costs in HEEs. The outcomes of a programme for the children, on the other hand, may also be unique and potentially different from those programmes, which directly address mentally ill persons (programmes may for example prevent a lack of well-being). As demonstrated in the Village logic model, the programme addresses at least three groups (the children themselves, the parents, and the practitioners). Thus, the recent methodological HEE advances may still not fully grasp the outcome dimensions of preventive programmes in our field.

das ökonomische Wirkmodell soll umfassende Berücksichtigung von Kosten und Outcomes unterstützen

The economic logic model to be developed will therefore lay the foundation to comprehensively cover cost and outcome dimensions in HEEs that evaluate programmes for families where a parent has a mental illness. This will be done by developing a generic model, which demonstrates the multiple consequences of parental mental illness, both for the individual child and for the society and by identifying the public sectors that may be affected as well as how they might be affected. The model does not include a specific preventive programme at this stage and can therefore described as an impact inventory or economic evaluation framework rather than a full logic model as defined in 3.2.1 [60].

„empirisches" Wirkmodell

The consequences of parental mental illness for children in the model will be based on the literature identified in the systematic search described in section 1.2. Thus, the model is empirically driven by evidence from published studies.

This strategy refers to the 'deductive approach' in the literature on building logic models [71]. We want to stress that the adverse consequences described in the model do not occur in all children as not all will go on to develop an illness or other problems [72]. The model is rather developed to demonstrate potential costs and timeframes on when they occur, which may have to be considered in a HEE on preventive programmes.

negative Konsequenzen betreffen nicht alle Kinder

The model will include the following dimensions and impact categories:

Modelldimensionen:

- individual consequences from parental mental health for children regarding physical and mental conditions, social functioning and socio-economic consequences,
- societal consequences of parental illness in economic terms,
- the complexity and possible interrelationships of the model elements,
- public sectors (e.g. health care, social care) that are affected by those consequences, and
- types of private costs for children that may occur.

individuelle gesundheitliche und gesellschaftliche Auswirkungen, Verflechtungen der Modellelemente, Konsequenzen für die öffentliche Hand, private Kosten

The presented 'evidence-fed' logic model does not assume mono-linear causal relationships. The models assumption is rather of associational nature as the links between socioeconomic statuses (SES) and health are multi-related. Concretely, there are two hypotheses in (mental) health research establishing relationships between SES and (mental) health outcomes: 1) the social causation hypothesis, i.e. SES determines health outcomes and 2) the downward drift hypothesis that posits people are more likely to be susceptive to a lower socioeconomic status because of the mental illness. Both models are not mutually exclusive, but there is consensus that the social causation theory generally has more evidence [73]. In the light of transgenerational transmission of mental illnesses, the social causation hypothesis seems to be more valid.

dargestellte Zusammenhänge der Faktoren sind nicht kausal, aber assoziativ

In the discussion in chapter 5, we are contrasting our model with the existing HEE evidence on preventive family-oriented interventions from part I (section 4.1). We are identifying which elements from our economic logic model have been addressed by the existing HEE evidence and whether knowledge gaps regarding the full spectrum of economic impacts in those prevention programmes exist.

Gegenüberstellung von beiden Berichtsteilen und kritische Reflexion der ges.ök. Methoden

3.2.2 Data sources

For constructing the economic model, we used the literature identified in the systematic search from part I, and selected those studies that demonstrated an association between parental mental illness and potential consequences for their children. We included any study that presented information on health outcomes, but explicitly also those studies that addressed non health-related outcomes or societal consequences according to the inclusion and exclusion criteria in Table 3-2.

Datenbasis: Literatur aus der systematischen Suche

Two researchers (CS, LH) independently screened the references. All cases of initial disagreement were resolved through discussion. In case of subsequent disagreement, a third person (IZK) would be consulted. In total, 113 publications were eligible for a full text screening. Of the 113 full texts, we included 39 publications [10, 123-160] for the model. The flow chart in Figure 3-3 displays the selection process.

Auswahlprozess der Literatur

39 Publikationen eingeschlossen

Table 3-2: Inclusion criteria – PICO-Analysis for part II

Population	Children or adolescents of parents with a mental illness aged 4 to 18 and their parents[26] Mental illnesses include all mental and behavioural disorders (F00-F99), e.g. affective disorders, schizophrenia, psychosis, with or without substance misuse
Intervention	Not applicable
Control	Not applicable
Outcomes	All types of adverse consequences (health and non-health) from parental mental illness for children (short term and long term)
Study design(s)	No restriction on study design
Language	English/German
Type of publication	(un)Published journal articles and research reports
Time period	Beginning from 2010

3.2.3 Data extraction, analysis and synthesis

Datenextraktion, Analyse und Synthese: narrative Zusammenfassung der Auswirkungen auf die Kinder

keine Qualitätsbeurteilung der Studien

We summarised the findings in a narrative form. The Village logic model in section 3.2.1, the guidance for assessing complex technologies from the INTEGRATE-HTA project and its proposed system approach [40] served as a theoretical underpinning, in order to describe the causal pathways by which parental mental illness is thought to impact health, further outcomes, or well-being in general. The economic logic model is empirically driven, as it is based on currently available evidence. However, we did not assess the quality of the studies, nor did we extract quantitative information on the potential impact (e.g. prevalence of mental illness in adult children), because the aim was to illustrate the variety and types of potential adverse consequences rather than their magnitude.

2 'Wirkebenen' und mehrere Subkategorien:

We inductively clustered the spectrum of identified consequences into the following categories on overall two levels, whereby the first (individual level) is divided into three sub-categories:

Auswirkungen auf das Kind

- Individual child level:
  - Mental and physical health impact
    - Short-term
    - Long-term
  - Social functioning
  - Socio-economic impact

gesellschaftliche Auswirkungen

- Societal level: demonstrates societal consequences that may result from the individual impacts.

It needs to be emphasised that each impact in all categories can have feedback effects on affected parents. Since the model focusses on children, those correlations are not depicted in the model.

[26] This age range was defined to identify economic evaluations on interventions that are similar to the intervention applied in the Village project which included children aged 4 to 18. Interventions including younger children require different approaches and often primarily address parents or mothers.

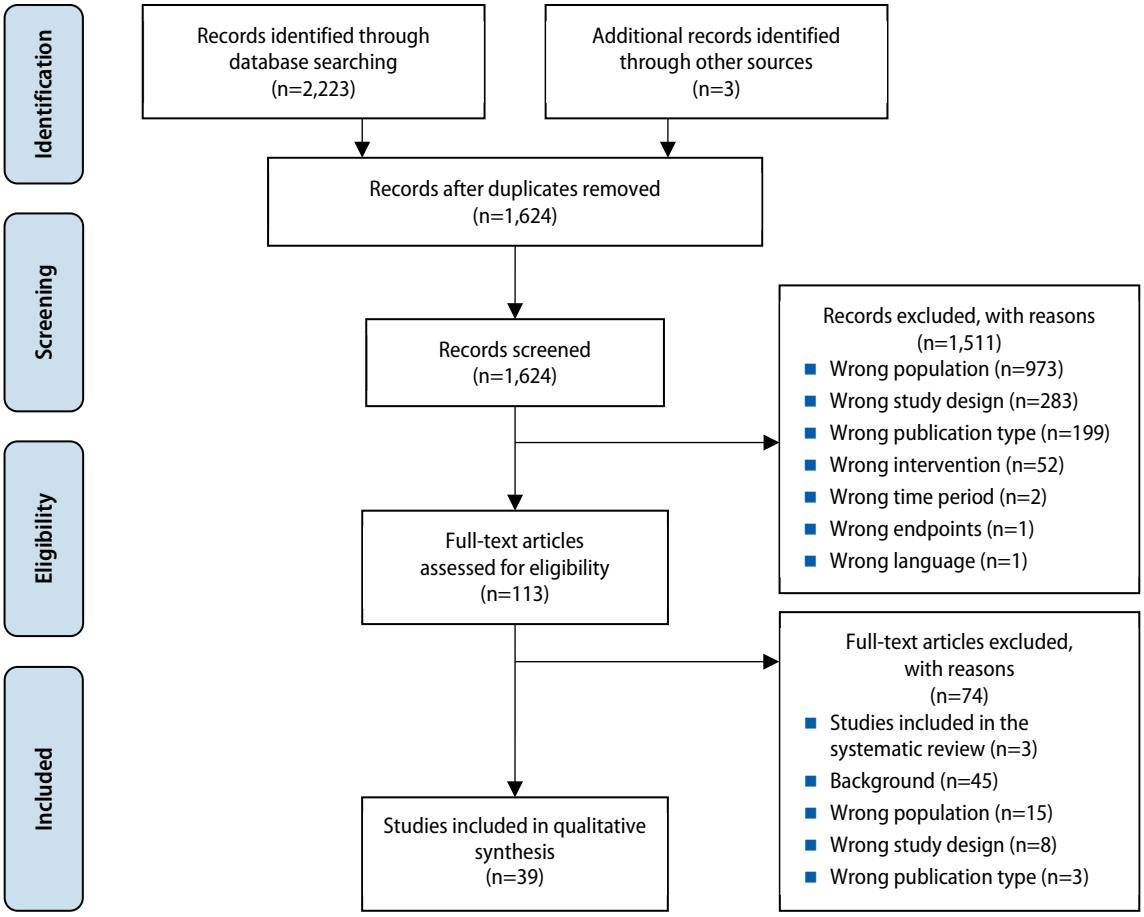

Figure 3-3: Flow chart of study selection (PRISMA Flow Diagram) for part II (economic evaluation framework)

3.3 Quality assurance

Qualitätssicherung des Berichts durch eine interne und eine externe Begutachterin

zusätzliche Begutachtung durch Investigatorinnen des Village-Projekts

One internal reviewer (IZK), Village co-investigators and one external reviewer (NB) independently reviewed the report. The latter was asked for the assessment of the following quality criteria:

- Technical correctness: Is the report technically correct (evidence and information used)?
- Does the report consider the latest findings in the research area?
- Adequacy and transparency of method: Is the method chosen adequate for addressing the research question and are the methods applied in a transparent manner?
- Logical structure and consistency of the report: Is the structure of the report consistent and comprehensible?
- Formal features: Does the report fulfil formal criteria of scientific writing (e.g. correct citations)?

The AIHTA considers the external assessment by scientific experts from different disciplines a method of quality assurance of scientific work. The final version and the policy recommendations are under full responsibility of the AIHTA.

4 Results

4.1 Systematic review of health economic evaluations

4.1.1 Characteristics and features of the programmes and their evaluation studies

Programme characteristics

In total, three [22, 64, 65] publications from 1,624 hits fulfilled the eligibility criteria defined in our PICO scheme. All three programmes in the studies pursued different approaches and had different underlying concepts (see Table 4-1).

Einschluss: 3 Publikationen von 1.624 Treffern

3 verschiedene Programme

Table 4-1: Studies and programmes

Studies and name of the programme:
■ Day et al. (2020) : Helping Families Programme-Modified (HFP-M) [65]
■ Gardener et al. (2017): Incredible Years® (IY) basic parenting programme[27] [64]
■ Wansink et al. (2016): Preventive basic care management (PBCM) [22]

The 'Helping Families Programme-Modified' (HFP-M) [65] is a specialised psychoeducational programme to improve parent-child relationships, interpersonal conflicts, promote effective parenting and parental coping with daily stress, and build family social support. It is based on 'The Multiple Determinants of Parenting (MDP) conceptual model'. The MDP model specifies how the interaction between child characteristics and parenting is influenced by the multiple impacts of parents' personality, couple relations, family and social networks, and work experiences.

HFP-M: psychoedukatives Programm

The 'Incredible Years' (IY) programme is a parenting programme [64]. Its content is derived from social learning and attachment theory, and addresses the following topics: relationship-building, providing praise and rewards as reinforcement of positive behaviour, effective limit-setting, adequate disciplining techniques, and coaching children in social, emotional and academic skills.

IY: Programm mit Fokus auf die Elternschaft

The 'Preventive basic care management' (PBCM) in the third study [22] is a family-focused strength-oriented rehabilitation model, focussing on strengthening positive parenting and providing community and network support.

PBCM: stärkenorientiertes Familienrehabilitationsprogramm

All three programmes in the study, generally aimed to improve effective parenting with the focus on the child's development. One programme focused on socioeconomically disadvantaged families [64]. The three programmes partly overlapped with regard to subtasks, involved professions, and services that were additionally utilised besides the specific programme tasks itself[28].

Ziel aller Programme: Verbesserung der Elternschaft mit Fokus auf Kindesentwicklung

[27] Although the pooled study focuses on (primary, secondary, and tertiary) prevention of child disruptive behaviour and not explicit on children who have a parent with a mental illness, we included the study, because it has also a focus on family mental health and parental mental health and functioning.

[28] A complete list of subtasks, components, and involved professions can be inferred from Table A-3 in the Appendix.

neben Gesundheitsleistungen wurden Leistungen in anderen Sektoren wie dem Bildungssektor eingesetzt

The mode of delivery was usually in the form of a limited number of sessions with the parent/family. In the PBCM programme, the number of sessions depends on the needs of the family. Hence, duration varied and ended when parenting and the children's development were sufficient according to the PBCM coordinator. In all programmes a number of different (mental) health care services were used, but also service use in other public sectors was described (e.g. from the educational sector). Additionally, some voluntary services were mentioned in two programmes.

Specific characteristic that distinguish the programmes are listed in Table 4-2.

Table 4-2: Specific characteristics of each programme

Programme:	Approach	Mode of delivery	Professions/Services involved

Helping Families Programme-Modified (HFP-M) [65]	HFP-M is intended to augment rather than replace care as usual (CAU) and uses a relational, goal-orientated helping process (e.g. reduction of parental alienation and stigma, **assess and strengthen parent-child relationship, child's emotional and behavioural difficulties**[29], and manage wider family and life circumstances, generate hope, and encourage parents' use of learnt skills in daily life etc.).	Between eight and 12 sessions and up to 16 weekly sessions of psychoeducational parenting intervention delivered one-to-one by trained and supervised mental health therapists	■ Health care
			  ■ professionals (e.g. GPs, psychologists, mental health nurses, paediatrician, optician, occupational therapist, drug/alcohol adviser, community mental health worker, dentist, hearing specialist, family therapist),
			  ■ specific teams (e.g. home treatment/crisis teams, assertive outreach teams, early intervention team, accident and emergency service team, drug/alcohol service team)
			■ Education
			  ■ professionals (e.g., adult educational teachers, school nurse, speech therapist)
			■ Social care
			  ■ social workers, home help/care worker
			■ Voluntary sector (e.g. self-help/support group)

Incredible Years® (IY) basic parenting programme30 [64]	Parents learn to break coercive cycles of **parent–child interaction** in which parents and children reinforce negative and aggressive behaviour in each other. Parents (not therapists) are seen as the experts on their own children. Parents are guided to set weekly goals, which fit with their cultural and personal needs and values; video-taped scenes showing examples of parent–child interactions are central in the sessions and parents are guided to identify key parenting behaviours or principles that might be useful for their own family context	12- to 14-sessions delivered to groups of between six and 15 parents, in weekly sessions of 2-2.5 hours by a IY therapists	■ Health care professionals (e.g. GPs, nurses, speech therapist, child and adolescent mental health care worker, inpatient personnel) ■ Social care (e.g. child placement workers, social worker) ■ Voluntary sector (voluntary helpers)
Preventive basic care management (PBCM) [22]	Intervention consists of five steps: 1.) the enrolment procedures (referral by parent's therapist), 2.) Systematic assessment of strengths and vulnerabilities of parenting and **children's development**, 3.) Design of an integrated preventive plan/tailored preventive care, 4.) Linking families to and **coordinating services for childcare/ children** clubs/community health services, services for debt restructuring and financial resources, 5.) Monitoring of the implementation/evaluation of effects in **regular meetings with parents** and **children**.	Home visits, face-to-face contact with parents or the family and other forms of contact (telephone calls, e-mail exchanges) by a PBCM coordinator; frequency is tailored to the needs of the family; duration varied and ended when parenting and the children's development were sufficient according to the PBCM coordinator	■ Health care professionals (GPs, mental health care workers, ■ Social care (day care workers, babysitters, debt restructuring personnel, preventive family support workers, youth care workers) ■ Criminal justice system (criminal justice personnel)

CAU…Care as usual, GP…General practitioner, HFP-M…Helping Families Programme-Modified, IY…Incredible Years, PBCM…Preventive basic care management

[29] The **bold** parts of the text indicate the role of the child in the programme.

[30] Although the pooled study focuses on (primary, secondary, and tertiary) prevention of child disruptive behaviour and not explicit on children who have a parent with a mental illness, we included the study, because it has also a focus on family mental health and parental mental health and functioning.

General characteristics of programme evaluations

2 der 3 ges.ök. Studien basierten auf einer randomisierten kontrollierten Studie (RCT)

Two [64, 65] of the three publications embedded the HEE within a Health Technology Assessment (HTA) report. One [65] of the two HTA reports was based on a two arm, parallel feasibility randomised controlled trial (fRCT). The second HTA report [64] pooled individual-level data from 14 randomised controlled trials (RCTs) [74-89] for the effectiveness domain and pooled clinical data from five trials [81, 83-85, 87, 89] for the HEE. The third study [22] was published as a journal article in a health services research journal. An RCT served as the basis for the HEE.

2 Studien sind aus Großbritannien, eine aus den Niederlanden

The HFP-M in the first HTA [65] was conducted in the United Kingdom (UK) in two centres. The IY programme covered in the second HTA [64] was implemented at multiple sites across Europe (UK, NL, IRE, NOR, SWE, POR). The HEE of this study only covered data of included trials conducted at UK sites [81, 83-85, 87, 89], and the PBCM in the third study was implemented in the Netherlands (western urban area) [22].

„Regelversorgung" war die Vergleichsintervention in allen 3 Studien

All three publications [22, 64, 65] used some form of care as usual (CAU) as comparator. One additionally included a waiting list (WL) and a minimal intervention (MI) as comparator [64]. Follow-up time for the EEs ranged from six to 18 months, with data collection at different time points in between. One study additionally modelled long-term savings of the intervention for a time span of 25 years (child age 5 to 30) [64].

The effectiveness data is based on randomised trial designs in all three studies.

öffentliche Finanzierung

Autor*innen berichten Interessenskonflikte

All three studies reported on the received funding. Two studies [64, 65] received funding from the National Institute for Health Research (NIHR) and one study [22] received funding from the Dutch organisation for health research and development (ZonMw). All authors of each study reported on competing interests. In two studies [64, 65], authors were involved in the design of the intervention programme and conducted and/or evaluated the studies.

Population characteristics

Einschlusskriterien

Ausschlusskriterien variierten zwischen den Studien

In two studies, parental (severe) personality difficulties [65] or psychiatric disorders [22] were an explicit inclusion criterion. All studies reported on psychiatric diagnosis [64, 65] or the Beck Depression Index (BDI) score of parents at baseline [22]. In one of these two studies [22], families included needed to meet three or more risk factors of a list of sixteen risk factors for parenting difficulties?[31]. The eligibility age range for included children was between 1 and 12 years. One study [65] included children that experienced mental health issues. Exclusion criteria varied between the studies.

[31] In the HTA with the pooled data [64] mental illnesses of parent was not an explicit inclusion criteria, but of the 1,799 families included, 1,131 parents in 11 out of 14 trials experienced depressive symptoms. With regard to the trials included in the economic evaluation [81, 83-85, 87, 89], three out of five trials reported on depressive symptoms in parents, two out of five reported on parenting stress, and two out of five reported on self-efficacy. The study was therefore included in our review.

In the trials, between 24 and 372 families[32] and between 24 and 236 families participated in the intervention and comparison groups respectively. The percentage of single parents ranged from 33% to more than three quarter of the participating families. Where recorded, the majority of participating parents were mothers (in one study 100% [65]). Parental age was only reported in one study where the mean age was 35 years in both groups.

Anzahl Familien: zwischen 24 und 372 (Intervention) bzw. 24 und 236 Familien (Vergleichsgruppe)

Where reported, the median number of children ranged from 2 to 2.16 per family. Two studies [22, 65] reported on the age of the index child, which ranged from 5.6 to 7.9 years. One study [64] reported the general child age, whereby the mean age was 4.7 and 4.9 years in the intervention and control group respectively. There was a slightly higher overall proportion of male than female index children in one study (60% in the intervention and 63% in the control group) and in the comparison group versus the intervention group in the other two studies (60% versus 50%).

Median-Anzahl der Kinder 2 bis 2.16

Altersspanne der Kinder: 5,6 bis 7,9 Jahre

Out of 608 children in one study [64], 389 children (64%) stem from families with a low income. In the same study, a third of the children came from families where the parent was unemployed. Two studies [22, 64] report about the ethnic background of children. In one study, 114 out of 608 (19%) children belong to an ethnic minority. Another study [22] reports about the ethnic background of families, whereby 39 of 49 families in the intervention arm and 27 of 50 families in the comparator arm (in total ~66.67%) belong to ethnic minorities.

demographische und sozioökonomische Charakteristika der Familien

Health economic evaluation framework and methods

Two HEEs [22, 65] are so-called 'piggy-backed' onto a clinical-effectiveness study, whereas the third study [64] conducted the health economic analysis based on the pooled clinical data from several RCTs. All of the three included studies carried out standard HEEs. Two studies [22, 64] performed a cost-effectiveness analysis (CEA) and one study [65] implemented a cost-utility analysis (CUA). One study [64] additionally modelled the long-term savings of the programme in addition to the CEA.

2 Kosten-Effektivitäts-Analysen, 1 Kosten-Nutzwert-Analyse und eine Modellierung von langfristigen Einsparungen

Two studies depicted costs from a health care perspective [22, 65], two from a social care [22] and public sector perspective[64], and two studies additionally implemented a societal perspective [22, 65].

unterschiedliche Analyseperspektiven

One study used complete cases of five trials for cost analysis (differences in total costs). Two studies [22, 64] used imputation methods for deriving missing values. Cost data often show outliers and it is important how studies dealt with them. One study [22] did not find any outlier. The two other studies [64, 65] did not report on outliers. Only in two studies [22, 64], authors controlled for baseline costs.

Berücksichtigung von Baseline-Kosten und Ausreißern

The time horizon in the HEE was identical to the follow-up period in the clinical studies and therefore ranged from six months [64] over 10 months [65] to 18 months [22]. One study additionally modelled expected savings from the intervention over a 25-year time horizon.

Beobachtungszeitraum: 6 bis 10 Monate bzw. 25 Jahre in der Modellierung

[32] As all programmes are parent-children interventions, we refer to families as a compound patient in the report. We use the term family without regard of the actual composition of the family.

ICERs wurden mithilfe von 1.000 – 10.000 Bootstrapwiederholungen kalkuliert

All three studies present incremental cost-effectiveness ratios (ICER) and used bootstrap methods to calculate them. Bootstrap replications ranged from 1,000 [65] to 10,000 [64] repetitions. All of the included studies presented cost-effectiveness planes (CEP), cost-effectiveness acceptability curves (CEAC), and utilised willingness-to-pay (WTP) thresholds, but only one study [65] was explicit about the threshold value below which a programme would be rated as cost-effective. This was at £ 30,000 per quality-adjusted life year (QALY).

2 Modellierungsszenarien von langfristigen Einsparungen

One study [64] conducted economic modelling of long-term savings depicting two models of cost variation (model 1 and 2). Cost variations at follow-up were adjusted for baseline costs and other demographic covariates (model 1) and the final model (model 2) was adjusted for child age, gender, and treatment condition covariates via linear regression estimation.

Subgruppenanalysen in zwei Studien, Sensitivitätsanalyse nur in einer Studie

Two studies [22, 64] conducted cost-effectiveness analyses for different subgroups[33]: One study [64] analysed subgroups by the categories gender, baseline Eyberg Child Behaviour Inventory Intensity Scale (ECBI-I)[34] score (<134 vs ≥134), child age (<5 years vs ≥5 years), and parental depression at baseline (BDI score ≥20 vs <20). The other study [22] did a subgroup analysis on those study participants who actually received the full programme. Only one study conducted a sensitivity analysis [22] considering uncertainty by: 1.) Exclusion of cost outliers (e.g. high-cost families), 2.) Analysis of complete cases without imputed data, and 3.) Calculation of ICERs adjusted for baseline costs.

keine Diskontierung aufgrund des kurzen Zeithorizonts

None of the three studies discounted costs or benefits to account for the present value in the cost-effectiveness and cost-utility analysis. Two studies argued with the short time horizon – six months [65] and 10 months [64]. One study [22] gave no reason for not discounting. The study modelling long-term savings of the intervention applied a discount rate of 3.5%.

Cost categories and unit costs

unterschiedliche Instrumente zur Ermittlung der Ressourcen

Different instruments were used to elicit resource use in different sectors: the Client Service Receipt Inventory (CSRI) [90] in [64, 65], Evaluation of Children's Centres in England (ECCE) report (Strand 6) [91], and the list of services from the PBCM manual [92] in [22]).

Two studies [22, 64] listed unit costs (e.g. costs per GP contact) for valuing the resources used. Units for presenting the costs were per participant (family) per number of contact/days utilised, per visit, per 'unit', per duration, and per hour, per professional and meeting, and per consultation[35]. The third study [65] estimated mean costs per average number of contacts or days.

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[33] We strictly distinguish between sensitivity analyses and subgroup analyses as recommended in the Cochrane handbook. Subgroup analyses have the aim to estimate effects of the same intervention for each subgroup in order to make formal statistical comparisons across subgroups. The goal of sensitivity analyses is to make informal comparisons between estimated outcomes on the basis of different (input) assumptions [68].

[34] The ECBI-I is a 36-item questionnaire that assesses intensity and number of disruptive behaviour problems in children (0-36 problem scale, 36-252 intensity scale).

[35] Unit costs represent the total expenditure incurred to produce one unit of output. The unit in per 'unit' is meant as defined in the Unit Costs of Health and Social Care by the PSSRU (e.g. in the case of GP surgery/clinic, it is meant per patient contact lasting 11.7 minutes excluding travel). Per duration means that costs are standardised across trials with regard to per average duration of the service – a 'typical' duration for each service.

Studien verwendeten unterschiedliche Quellen für Stückkosten

For identifying unit costs, different secondary data sources were consulted: Personal Social Services Research Unit (PSSRU) unit costs of health and social care [93, 94] and the National Health Service (NHS) Improvement reference cost data [95, 96]. Additionally, one study [22] used the Trimbos/Institute for Medical Technology Assessment (iMTA) questionnaire for costs associated with psychiatric illness (TiC-P) [97, 98], the iMTA Questionnaire Intensive Youth Care [99], Dutch guidelines for health economic research to derive prices for professional and informal childcare [100], the standard pricing research for youth care and parenting support Noord-Brabant [101], standards and rates for outpatient care [102], and the Dutch manual for ICBs [103] for inter-sectoral costs and benefits. Authors of one study [64] conducted own calculations for unit costs regarding one service (the telephone help line).

unterschiedliche Darstellung der Kosten in den Studien

One study [22] reported costs for the following sectors: health care sector, childcare sector, educational sector, criminal justice sector, and 'other' sectors. The other two did not explicitly categorise costs in cost sectors but reported costs of single services [64] or service categories such as day-care services or inpatient admissions [65]). Overall, the full service spectrum, for which costs were calculated, varied across studies[36].

unterschiedliche Referenzjahre der Kosten

The two studies embedded in the British context [64, 65] presented the costs in British pounds (GBP), based on 2017 and 2016 as reference years respectively. The study from the Netherlands [22] presented costs in EURO referring to the year 2012.

Outcome parameters in the economic evaluations

krankheitsspezifische Ergebnismessungen als primäre Endpunkte in 2 Studien (HOME und ECBI)

1 Studie: QALYs

Two studies [22, 64] used disease specific outcome measures as primary outcome measure for the HEE: the ECBI-I[37] which measures outcomes in children reported by their parents, and the Home Observation for Measurement of the Environment (HOME) inventory[38] for children. The HOME inventory captures the quality of parenting by measuring the qualitative and quantitative aspects of stimulation and support available to a child in the home environment [104, 105]. One study calculated QALYs (for parents/carers and children) as the primary outcome parameter using the generic instrument EuroQoL-5 Dimensions (EQ-5D-5L for parents and EQ-5D-Y for children) [65].

Modelling parameters

Modellierung berechnet Return on Investment (ROI)

One study [64] additionally modelled the long-term savings and return on investment (ROI) assuming successful implementation of the programme. The model was based on a previous publication that used a decision-analytic Markov model [106]. The comparator in the model was a simulated control group with 'no intervention'. The study estimated the present value of savings from providing the IY programme per child with conduct disorder problems at the age of five for a time horizon of 25 years (age 5 to 30 years). The authors modelled two scenarios:

[36] For a complete overview of the cost parameters, I refer to Table A-5.

[37] The ECBI-I is a 36-item questionnaire that assesses intensity and number of disruptive behaviour problems in children (0-36 problem scale, 36-252 intensity scale).

[38] The HOME inventory measures following aspects with regard to quality of parenting: availability and impact of objects, events and interactions with parents. It covers four dimensions (responsiveness, learning materials, stimulation, harsh parenting).

2 Ansätze der Modellierung: Annahmen aus der Fachliteratur und Annahmen aus den zugrundeliegenden Daten

- Scenario 1 ('literature' scenario): The trajectory of the 'no intervention' group was modelled with a 60% chance that a child with behavioural problems at the age of 5 still showed problems at 16 years of age. In the intervention group this chance was 54%
- Scenario 2 ('data' scenario): Data from the pooled IY study sample for the control group with a 12% decrease in probability of scoring above the ECB-I cut-off point in year 1 were taken to model the trajectory of the 'no intervention' group. The probability of behaviour problems persisting past the age of 16 was 60% with a steeper decrease in year 1 compared to scenario 1. The probability that conduct problems persist beyond the age of 16 in the programme group was reduced to 52%.

zusätzlich Berechnung eines Hoch- und Niedrigkostenszenarios

Both scenarios were modelled with a high- and low-cost approach. The authors did not provide a clear description of the high- and low-cost scenarios. They reported that they drew on additional literature for calculating the cost-of-illness in the model. The cost categories in their calculation are related to services provided by the NHS, social service departments, department for education, voluntary sector, criminal justice system, health impacts of crime, and benefits payments. Programme effects in the model are based on the results of the CEA. Further assumptions of the model are the following: Drop-out is already accounted for in the overall effectiveness figures (intention-to-treat basis) and the modelled control group that does not receive the intervention experiences a 'natural' trajectory of the illness.

4.1.2 Results

Resource use

Anzahl der Sitzungen: ~13 bis 16 Einheiten à 1 bis 2,5 Stunden

Two studies reported on the number of sessions of the programmes consumed. The number of sessions ranged on average from 12.7 weekly sessions [64] to 16 sessions [65] basis for a 10 months treatment interval. The duration per session ranged from 60 minutes to two and a half hours.

Costs

programmspezifische Kosten wurden in 2 Studien berichtet

Two studies reported on the programme-specific cost [22, 64]. The IY programme-specific cost [64] for the full follow-up period amounted to £ 2,414. The PBCM programme-specific service cost were € 1,685. Authors evaluating the HFP-M programme did not report on programme-specific cost [65].

Kostenunterschiede aufgrund der gewählten Perspektive

Total costs from a societal perspective for the groups receiving the three different programmes were £ 1,135 (£ 6,971)[39] [65] and € 19,805 [22][40]. In the study, which additionally measured costs from a healthcare and social care perspective [22], the total costs for the programme amounted to € 13,012 and € 17,717 for the two perspectives respectively.

Kosten in der Vergleichsgruppe variierten

For the comparator groups, total costs ranged from £ 501 [64] to € 19,209 [22] from the societal perspective [22]. The total costs in the comparator group from a healthcare perspective and social care perspective amounted to € 11,219 and € 16,979 respectively in one study.

[39] Standard deviation in parentheses

[40] The study reported also total costs for time intervals T0-T1 and T1-T2, but we report only total costs over the full time horizon in this report.

Overall, in two studies, the costs in the intervention group were lower than in the comparison group in the base case analysis [64, 65]. In the third study [22], total costs in those receiving the programme were higher than in the comparator group for all perspectives.[41]

Kosten der Interventionsgruppe in 2 Studien niedriger als in Vergleichsgruppe

Effectiveness

The programmes showed better outcomes than the comparators in two of three studies [22, 65], However, one of these studies [65] showed mixed results depending on the perspective. For children, the intervention provided more QALYs (0.0297) in analysis scenario 1, but less (–0.125) in analysis 2. The other study [65] reported on the change in HOME scores in the base case scenario. The HOME score changes amounted to 1.93 for the intervention and –1.89 for the comparator for all three perspectives. In the third study [64], changes or differences in benefits were not reported separately.

in 2 Studien waren Resultate des Programms im Vergleich zur Kontrollgruppe besser

Cost-effectiveness

Regarding HPF-M [65], the results for both, parents and children differed depending on the analysis: in the parental analysis 1[42], the programme dominated the comparator from both the societal and NHS perspective. In parental analysis 2, ICER were £ 102,083 per QALY for the NHS perspective and £ 96,155 per QALY for the societal perspective. For children, the ICER ranged from £ 16,466 per QALY for the NHS perspective to £ 15,191.21 per QALY for the societal perspective in analysis 1. However, in analysis 2, CAU dominated HFP-M from both a NHS and societal perspective. Only ICER for children were used to explicitly state probabilities to be cost-effective at certain thresholds. At a threshold of £ 30,000 per QALY, the probability for the programme of being cost-effective was 52%.

bei Schwellenwert von £ 30.000 pro QALY lag Wahrscheinlichkeit, dass HPF-M kosteneffektiv ist, bei 52 %

The IY-study [64] did not report explicitly on ICER, but provided CEAC with respective probabilities to be cost-effective conditional on the WTP threshold. In the base case analysis, the IY programme was cost-effective compared to the comparator with probabilities of 50%, 80%, 95%, and 99% for threshold values of £ 109, £ 121, £ 134, and £ 145 per ECBI-I score improvement respectively.

IY ab einem Schwellenwert von £ 121 pro ECBI-Score zu 80 % kosteneffektiv

For the subgroup analyses, the study reported only qualitative results:

- Baseline ECB-I score: IY less likely to be cost-effective with ECBI <134
- Child age: IY less likely to be cost-effective for age <5 years
- Gender: IY more likely to be cost-effective for male children
- Parental depression at baseline: IY more likely to be cost-effective for children whose parents have a moderate level of depression (BDI score ≥20)

Ergebnisse unterschiedlich für verschiedene Subgruppen

The ICER for PBCM [22] were € 461 (health care perspective), € 215 (social care perspective), and € 175 (societal perspective) per HOME score improvement respectively. With a WTP threshold € >2,500, PBCM was cost-effective with a probability of almost 100% from all three perspectives.

PBCM ab einem Schwellenwert von € 2.500 zu 100 % kosteneffektiv

[41] Individual results in two studies were mixed because of the different analyses settings.

[42] Due to the substantial number of missing data at T3, authors conducted two CEA. Analysis 1 is based on data from T1 and T2 (more participants had complete cost data) and analysis 2 is based on data from T1 and T3.

gemischte Resultate für 2 Programme aber tendenziell kosteneffektiv, ein Programm in jeder Berechnungsvariante kosteneffektiv

In summary, two programmes (HFP-M and IY programme) showed mixed results in terms of being cost-effective depending on the perspective, considered subgroup, or thresholds [64, 65], but had a tendency on being cost-effective. On the contrary, PBCM [22] was cost-effective above a threshold of € 2,500 across perspectives. Overall, reported ICER ranged from € 175 per HOME score [22] to £ 16,466 per QALY [65]. Reported thresholds and probabilities for being cost-effective ranged from £ 109 [65] to £ 30,000 [64] and 20% to ~100% respectively [22].

Outcomes of modelling studies

ROI von 3 (Szenario 1) bzw. 4 (Szenario 2)

The study modelling the present value of long-term savings of the IY programme [64] calculated a savings range for scenario 1 of £ 1,023-7,565 per child with a ROI of three or 300%[43] and for scenario 2, of £ 1,254-9,408 per child with a ROI of four or 400%. The average net savings (savings without intervention costs) were between £ 5,000 and £ 7,000 per child in the high-cost scenario. In the low-cost scenario, the IY intervention would not result in long-term savings once the intervention cost is subtracted.

Sensitivity analysis

ab einem Schwellenwert von € 3.500 pro HOME-Score war PBCM zu 100 % kosteneffektiv

Sensitivity analyses for the PBCM programme [22] showed that cost-effectiveness is primarily dependent on the WTP threshold applied. Regarding the other parameters addressed in the sensitivity analysis, the programme was dominant (and thus superior) if only complete cases are used for the societal perspective calculation. In all the other scenarios for all the three perspectives, the programme is both, more effective and more costly than the comparator. Overall, for a WTP threshold >€ 3,500 per HOME score, the programme was cost-effective with a probability of 100%.

2 Studien: keine Sensitivitätsanalysen

No results on sensitivity analysis are available for the other two studies [64, 65].

Authors' conclusion

Schlussfolgerungen der Autor*innen der Studien:

According to the conclusions from the authors, in most of analyses undertaken, programmes seem to improve the outcome measured, but also result in higher costs compared to the control groups. Whether the resulting ICER are to be classified as favourable strongly depends on the WTP threshold applied.

alle Programme tendenziell effektiver aber auch teurer als Alternative

Kosteneffektivität hängt vom Schwellenwert ab

For the IY programme, authors concluded that the intervention is less likely to be considered cost-effective for children with clinical levels of disruptive behaviour (ECB I<134) and for children aged <5 years, but more likely to be cost-effective for male children and children whose parents have a moderate level of depression (BDI score ≥20). Additionally, return on investment may be substantial if a long-term perspective is applied and if their assumption on the high cost for persistent conduct disorders holds. In their conclusion, authors noted that the children in their sample generally used few services and, thus the potential for immediate savings from the IY intervention is reduced.

Hinweise auf Wichtigkeit einer breiten Perspektive

Authors from the PBCM study point to the importance of ICBs and choosing a broad societal perspective.

[43] A ROI of 4 or 400% means that one unit of money invested brings additionally four units of money in return.

4.1.3 Quality appraisal

Zielgruppe, Vergleichsinterventionen und Forschungsfrage klar definiert

All three studies described target populations and comparators clearly (see Table A-2, Table A-1, and Table A-3). Furthermore, authors presented a well-defined research question either in separate sections [64, 65] or in the background section [22]. The study design of each economic evaluation was appropriate for the stated objective in each study. Authors of the study with the additional economic model calculating long-term savings of the intervention stated model assumptions. Authors reported that they used a model that was previously used for modelling economic impact of conduct disorder [106], but they did not explicitly report that a Markov model was used.

Modellannahmen angegeben

kurzer Zeithorizont → Vernachlässigung intersektoraler Kosten- und Nutzenparameter

All authors reported on the time-horizon, but the time-horizon was overall too short for depicting long-term inter-sectoral costs and benefits that often appear in the mental health care context. This may bias the cost-effectiveness of programmes in both directions. The chosen perspective was appropriate in all three studies to answer the stated research question(s).

validierte Kosteninstrumente und relevante Endpunkte angewendet

Only in one study [22], all important and relevant costs for each intervention were identified. Authors of two studies [64, 65] missed to depict inter-sectoral costs in their trial based economic evaluations. All three studies reported costs in physical units, and valued costs appropriately with validated costing tools. Each of the studies identified relevant and important outcomes for each alternative conditional on the used study design. Authors of all three studies measured and valued outcomes appropriately. All three studies implemented incremental analysis. However, only one study [65] applied and reported commonly used WTP thresholds for decision-making situations. The two other studies [22, 64] applied incremental analyses, but explicitly stated that official thresholds for the outcomes they used (e.g., £ per ECBI-I improvement) were not available. Hence, because of the lack of clear WTP thresholds for those clinical outcome measures in both studies, interpretation of the health economic results and costs per unit of effect are limited compared to standard outcome measures such as the QALY.

inkrementelle Analysen durchgeführt

Schwellenwerte tlw. willkürlich

Sensitivitätsanalyse nur in einer Studie

The study with the economic model [64] discounted all employed future values that were included. Only one study [22] explicitly conducted a sensitivity analysis.

Schlussfolgerungen, Generalisierbarkeit und Limitationen größtenteils angeführt

The conclusions by study authors are in line with the reported and analysed data in all three studies (see Table A-9). Generalisability to other subpopulations or countries was discussed in two studies. One study [22] concluded that the data from the real world setting strengthens the generalizability of the results. The other study [64] provided a large sample that is possibly transferable to other countries, service contexts, and subpopulations. The study that did not report generalisability [65] stated that the review synthesis did not intend to determine the generalisability of findings.

kein relevanter Interessenskonflikt

All three studies reported on authors' conflict of interest (COI). It seems that there is no concerning COI of study researchers and funders (see Table A-1).

eine Studie diskutierte explizit ethische und verteilungstechnische Fragen, zwei Studien nur am Rande

Ethical and distributional issues were considered in one study, meaning that these issues were fully discussed [64]. The two other studies [22, 65] only indirectly addressed ethical and distributional topics. One of these two studies [65] conducted a moderator analysis in the course of the effectiveness analysis to determine whether all subpopulations benefit from the intervention (e.g. 'social and socioeconomic disadvantaged': low income, low educational level, lone parent, teenage parent etc.). Authors in the third study [22] included families being exposed to three or more of a list of sixteen risk factors for

poor parenting. Risk factors relevant for distributional questions were among others single parenthood, housing problems, poverty or debts. However, the study did not specifically discuss ethical or distributional issues in relation to the results of their HEE.

Studien weisen niedriges bis moderates Verzerrungspotential auf

When evaluating risk of bias categories with the CHEC checklist [66, 67]. The picture in Table 4-3 shows that studies exhibited a rather low to moderate risk of bias. Between 13 and 15 criterion are fulfilled in each study. Between one and a maximum of four questions per study could not be answered positively. Across all studies, 42 from overall 60 questions are answered positively. Nine criterion are not fulfilled, six are partly fulfilled, and four categories are not available or applicable.

Table 4-3: CHEC-list for assessing the risk of bias (RoB)

Authors and year		Day et al. (2020) [65]	Gardener et al. (2017) [64]	Wansink et al. (2016) [22]
#	Question (category)			
1.	Is the study population clearly described?	+	+	+
2.	Are competing alternatives clearly described?	+	+	+
3.	Is a well-defined research question posed in answerable form?	+	+	+
4.	Is the economic study design appropriate to the stated objective?	+	+	+
5.	Are the structural assumptions and the validation methods of the model properly reported?	NA	+/-[44]	NA
6.	Is the chosen time horizon appropriate in order to include relevant costs and consequences?	-[45]	-[45]	-[45]
7.	Is the actual perspective chosen appropriate?	+	+	+
8.	Are all important and relevant costs for each alternative identified?	-[46]	-[47]	+
9.	Are all costs measured appropriately in physical units?	+	+	+
10.	Are costs valued appropriately?	+	+	+
11.	Are all important and relevant outcomes for each alternative identified?	+	+	+
12.	Are all outcomes measured appropriately?	+	+	+
13.	Are outcomes valued appropriately?	+	+	+
14.	Is an incremental analysis of costs and outcomes of alternatives performed?	+	+/-[48]	+/-[49]
15.	Are all future costs and outcomes discounted appropriately?	NA[50]	+/-[51]	NA[52]
16.	Are all important variables, whose values are uncertain, appropriately subjected to sensitivity analysis?	-[53]	-[53]	+

[44] Authors completely reported assumptions in the HTA report, but the study missed to report what type of economic model was used. The authors referred to the underlying modelling study that was previously used [106].

[45] Short time horizon

[46] Analysis from a societal perspective did not consider some inter-sectoral costs and benefits.

[47] Analysis from a public sector perspective did not consider some inter-sectoral costs and benefits.

[48] No explicit WTP threshold analysis for ECBI-I scores

[49] No explicit WTP threshold analysis for HOME T-scores

[50] Short time horizon: no discounting applied

[51] Short time horizon in the CEA: no discounting applied;
Model: discounting rate of 3.5%

[52] Short time horizon: no discounting applied

[53] No sensitivity analysis applied

Authors and year		Day et al. (2020) [65]	Gardener et al. (2017) [64]	Wansink et al. (2016) [22]
#	Question (category)			
17.	Do the conclusions follow from the data reported?	+	+	+
18.	Does the study discuss the generalizability of the results to other settings and patient/client groups?	-[54]	+	+
19.	Does the article indicate that there is no potential conflict of interest of study researcher(s) and funder(s)?	+	+	+
20.	Are ethical and distributional issues discussed appropriately?[55]	+/-[56]	+	+/-[57]
	# of Yes/No/Partly fulfilled/NA	13/4/1/2	14/3/3/0	15/1/2/2

+ *Yes (low risk of bias)* - *No (high risk of bias)* +/- *Partly fulfilled* NA *not available/applicable*

4.2 Economic evaluation framework

In the following, the identified literature on adverse consequences from parental mental illness for children and for society will be summarised. The literature has been clustered according to the different impact categories described in section 3.2.1. Next, we describe the affected public sectors and the implications for private costs in children. Figure 4-1 summarises the results in the form of an economic logic model and illustrates each category with some examples.

Rahmenwerk für ökonomische Evaluationen in Form eines ökonomischen Wirkmodells

4.2.1 Potential consequences of parental mental illness for children

Health impacts for children

Determinants of children's health

Exposure to parental mental illness of children particularly increases the risk for mental illness in children [107, 108], but potentially also leads to worse physical health outcomes [109]. The reasons for the adverse health consequences are complex and related to an interaction of genetic, environmental, and psychosocial factors as presented in developmental models of transgenerational transmission of psychopathology [110]. In contrast to genetic factors,

gesundheitliche Folgen

Determinanten der Gesundheit: genetische, umweltbedingte und psychosoziale Faktoren

[54] The review synthesis did not intend to determine the generalisability of findings.

[55] The item on discussion of distributional and ethical issues is general and refers to aspects that arise regarding the intervention, context, included population, subgroups or outcomes.

[56] The study conducted a moderator analysis in the course of the effectiveness analysis to determine whether the intervention benefits all subpopulations (e.g. 'social and socioeconomic disadvantaged': low income, low educational level, lone parent, teenage parent etc.) the same in terms of clinical outcomes. The authors did not explicitly discuss ethical or distributional implications in the health economic analysis.

[57] Authors included families being exposed to three or more of a list of sixteen risk factors for poor parenting. Risk factors relevant for distributional questions were among others single parenthood, housing problems, poverty or debts. The study did not specifically discourse ethical or distributional issues.

environmental and psychosocial factors can be mediated by preventive measures. Some examples for potential environmental and psychosocial factors that have been described in the literature are:

fehlende emotionale Unterstützung, Spannung zwischen den Eltern, Verlust des Sorgerechts

Parents with mental illness may sometimes have difficulties in providing adequate emotional support [111]. Furthermore, impacts on the family and adverse family functioning (tension between parents, custody loss, and child protection interventions [112, 113]) can lead to family disruption and divorce [108]. Custody loss and child protection interventions makes reunification [113] more unlikely as both characteristics are more frequent in parents with mental illness.

familiäre Gewalt und psychische Misshandlung

Other serious adversities of parental mental illness experienced by children in affected families, although relatively rare but not negligible, are family violence [114], maltreatment [113, 115, 116] including physical (e.g. sexual abuse) [108, 109] and psychological abuse [117].

sozioökonomische Faktoren: geringes Haushaltseinkommen, Wohnungs- und Ernährungsunsicherheit

Regarding the socio-economic dimensions in families where a parent has a mental illness, the literature describes lower household income [118], food insecurity (household and children specific) [111, 119-121] and housing issues. These socioeconomic adversities potentially feedback on other impacts such as the child's perceived stigma or self-confidence (see below).

Short-term health impact

kurzfristige gesundheitliche Auswirkungen:

The following potential immediate adverse health outcomes for children have been described:

Verhaltensschwierigkeiten/ -störungen

soziale Isolation, Selbstverletzung, suizidale Gedanken

Regarding mental health, behavioural difficulties and conduct disorders [108, 122-124], feelings of being left alone, being devalued, being excluded by other people, and loss of own sense of self have been described as common [108]. Social or self-isolation can be consequences of these feelings that reinforce the intergenerational cycle of mental illness [108]. Additionally, children face an increased risk of self-harm and suicide ideation [125].

Essstörungen, Übergewicht

Schlaflosigkeit, Suchtverhalten, reduziertes visuelles Gedächtnis, Zahngesundheit

Some studies have shown that eating disorders [121] such as obesity [126] that could lead to nutritional issues [111, 121] are more prevalent in children of mentally ill parents compared to children without mentally ill parents. Further observed health-related issues in children are sleeplessness [120], addictive behaviour triggered by stress (e.g. internet addiction) [127, 128], significantly lower visual memory performance [129], and reduced oral health [130]. Furthermore, a study has shown that children growing up in a household with a mentally ill parent are more likely to grow up in smoking households. Consequently, children are more likely to be exposed to second hand smoke [131]. Furthermore, there is some evidence suggesting that the risk of injuries is increased in the children [132].

Long-term health impact

Langfristige gesundheitliche Auswirkungen: psychische/physische Erkrankungen auf dem Weg zum Erwachsensein

Negative health consequences can also occur in the long-run: (Health) impacts experienced in childhood, can have direct impacts on the mental health, functioning, and physical health on the path to adulthood. Exposure of parental illness in childhood may contribute to psychological illnesses and issues among the middle-aged and elderly, such as depression [109], anxiety disorder, self-harming behaviour, or negative physical conditions in later life such as migraine, sleep problems [108], or increased risk of being obese [126].

Social functioning in children

soziale Funktionsfähigkeit:

The literature has been described that some children may experience adverse effects regarding social competency, which in turn can impact the development of empathy, solidarity, and tolerance [108]. Further potential consequences that have been described are impaired social relationships, missing integration in later life, and social competency issues in general [108, 133]. Furthermore, potential identity issues have been identified such as some children developing an ambivalence between self-responsibility, the social self and responsibility for the parent or others in adulthood. It has been observed that some children develop a pathological form of the 'helper syndrome' in later life [108, 133].

elterliche psychische Erkrankung hat Einfluss auf soziale Kompetenz, Empathie, Solidarität, Toleranz der Kinder

Schuld, Scham, Stigmatisierung

Additionally, children often experience guilt and shame [134, 135], because of the (perceived) societal stigmatisation [108, 136, 137] of (parental) mental illnesses. These experiences and perceptions increase the risk of developing internalizing or externalizing problems [134]. As mentioned before, anti-social behaviour, conduct problems [108, 122-124], social isolation [108] are more prevalent in the affected children compared to children without mentally ill parents. Reduced self-esteem, (perceived) stigmatisation, shame, and guilt for the parental mental illness can extend into adulthood and influence the social functioning later in life [108, 133, 134, 137].

Schwierigkeiten langfristige Beziehungen herzustellen

Another observation with regard to social and familial relationship is that children may experience difficulties in establishing long-lasting partnerships in adulthood and family cohesion seems to be fragile [108, 109, 120]. However, that does not mean that they cannot have functioning partnerships or marriages [138]. Furthermore, some studies indicate that they are less likely to have children on their own, although they desire to have children, because of worries of transgenerational transmission of mental illnesses and the feeling that they are not able to be a good parent [108, 139].

Socio-economic impact on children

sozioökonomische Auswirkungen

A number of potential socio-economic disadvantages for the children have been described:

Anwesenheit In der Schule, Bildungserfolg/fehlende Bildung → Arbeitslosigkeit, prekäre Arbeitsverhältnisse, Abhängigkeit Sozialhilfe

Firstly, the higher use of health services and health adversities experienced by the children directly influence school attendance [140] and in second order educational attainment [108, 109]. Educational attainment is not only a protective factor, and plays a central role as a mediator and moderator of physical and mental health, but the educational status also drives individual and societal economic impacts [25, 122]. As such, the children are more likely to experience unemployment, lower wages, precarious job situations, welfare dependence and dependence on public programmes and a poor individual economic status in adulthood [109, 119, 122, 141].

Betreuung der Eltern durch die Kinder

Since the children sometimes care for their sick parents from childhood or adolescence into adulthood, self-realisation is limited and subsequent income losses are to be expected [142].

erhöhtes Risiko für Straftaten

Furthermore, some studies described that parental mental illness may have an impact on criminal conviction of children such as drunk driving, serious, and minor offenses [107, 143].

Societal and economic impact

langfristige gesellschaftliche und ökonomische Auswirkungen:

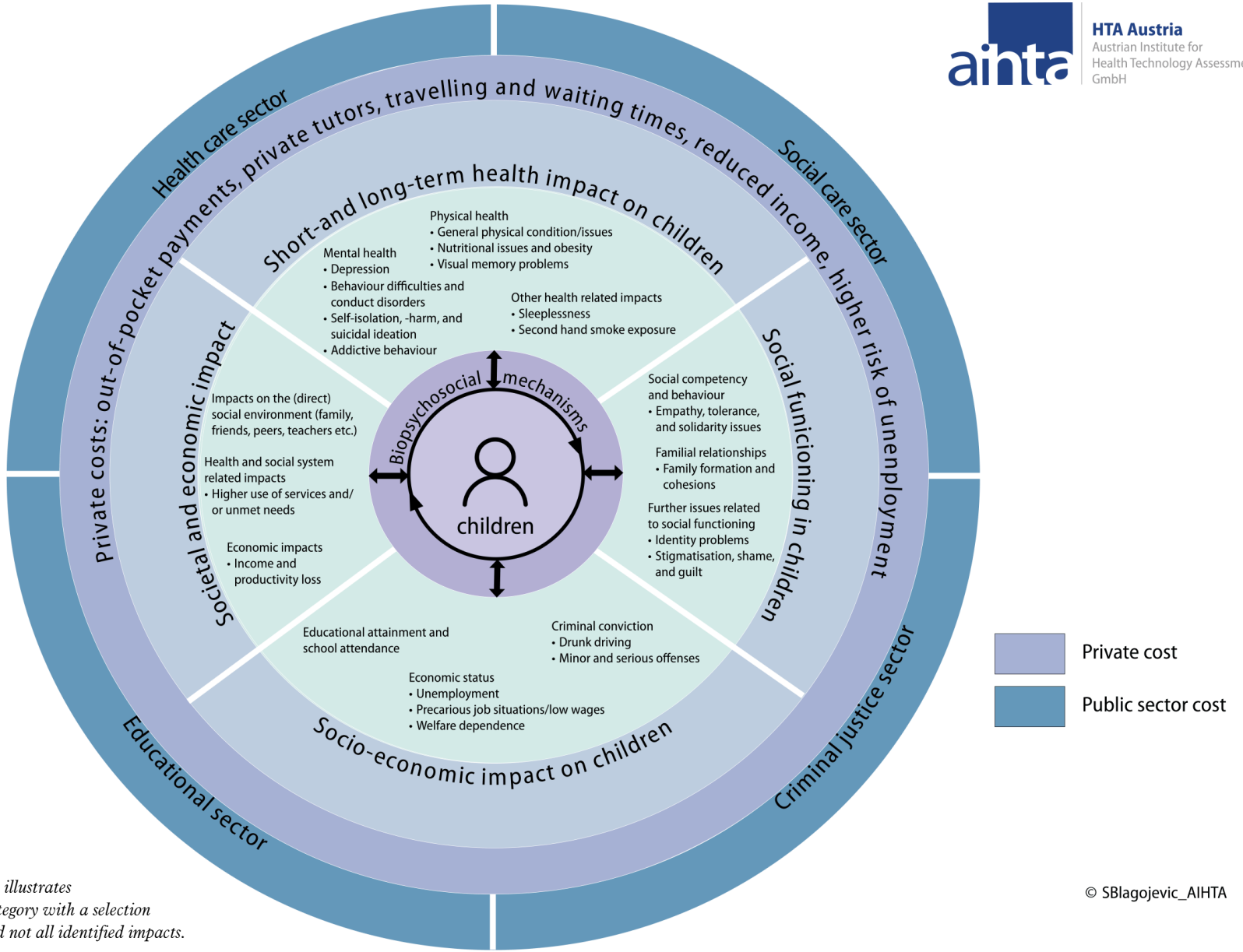
Einfluss auf das (unmittelbare) soziale Umfeld der betroffenen Kinder

The impacts described possibly have feedback effects on the parent, the social environment of the children, and the parents/entire family, and entail medium-term and long-term consequences at a societal level. The following societal consequences have been described: Firstly, the risk of self-harm and suicide ideation [125] may indirectly affect relatives, friends and other people in the direct social environment. Secondly, because of the increased prevalence of health and social adversities in children, they are more likely to utilise more acute health and social care services that could be reduced by an adequate intervention framework [123, 138, 144, 145]. Although the children have a higher need for health and social care services, simultaneously the (perceived) stigma may affect help seeking behaviour, thus leading to delayed support and treatment. Stigma is a potential barrier to problem recognition and constitutes substantial cost on families and society [146].

aggregierte Einkommens- bzw. Produktivitätsverluste

Krankenstände, Frühpension

Further (macro)economic impacts on the societal level are income and productivity loss, as the children could be impeded by the aforementioned mental, physical, and social functioning outcomes as adults [138]. Time spent in treatment and acute service (which is often associated with increased rates of unemployment, sick leave and early retirement [28]) potentially contributes further to productivity losses [123, 138, 144, 145].



★*Visualisation illustrates each impact category with a selection of examples and not all identified impacts.*

Figure 4-1: Economic evaluation framework (logic model) ★

4.2.2 Affected public sectors

betroffene öffentliche Sektoren:

The impact dimensions identified above demonstrate that costs for children can incur in the following sectors:

Gesundheitssektor

- *Health care (including services and drug treatments):*
  - use of child and adolescent mental health care to treat mental health problems in children
  - use of health care to treat physical health problems in children
  - use of adult (mental) health care in case of health problems in adulthood

Sozialbereich inkl. Geldleistungen

- *Social care and benefits:* foster care and other forms of out of home placements (e.g. youth welfare office etc.), vocational services, housing support, cash benefits (e.g. unemployment benefits, early retirement pensions), etc.

Bildungssektor

- *Educational services:* support that address conduct problems, anti-social behaviour or social isolation in the education sector, etc.

Strafrechtssektor

- *Criminal justice services:* police service, prison, probation service in case of crime conviction

4.2.3 Potential private costs

private Kosten: Selbstbeteiligung

The adverse consequences from parental mental health can also result in private costs for children. These may include

private Bildungsausgaben

- costs for private co- or complete out-of-pocket-payments for treatment in child and adolescent or adult (mental) health care

Kosten für Reisetätigkeit und Wartezeiten

- costs for private tutors compensating for reduced school attendance
- travelling and waiting times for treatments

reduziertes Einkommen und weitere ökonomische Konsequenzen

- reduced income due to lower educational attainment, higher risk for unemployment, early retirement or reduced employment due to long-term care responsibilities for their mentally ill parent

5 Discussion

An increasing body of evidence is showing that family-oriented preventive interventions for children who have a parent with a mental illness have significant positive effects on child outcomes [14-16, 147] and even can mitigate already materialised adversities [6, 15, 16, 147, 148].

zunehmende Evidenz für Wirksamkeit familienorientierter Programme

Since resources in health and social care are limited, evidence on effectiveness alone is not sufficient to make resource allocation decisions that maximise population health within a given budget. Health economic research of family-oriented interventions can help to determine whether monetary resources and human labour are efficiently utilised to achieve value for health and social care systems and the population in general.

ges.ök. Forschung unterstützt effiziente Ressourcenallokation

Therefore, part I of the report summarised the current economic evidence of family-oriented complex interventions in form of a systematic review of HEEs.

Berichtsteil 1: aktuelle ges.ök. Evidenz

Part II developed a framework for future economic evaluations in this field by structuring the potential adverse impacts from parental mental illness for their children at the individual level and for society that may be relevant to address in HEEs.

Berichtsteil 2: ges.ök. Rahmenwerk für zukünftige ges.ök. Studien

In the following, the results are summarised and discussed and some concluding suggestions for the HEE of the Village programme are presented.

5.1.1 Summary of the systematic review of health economic evaluations

The systematic search and selection process yielded three health economic evaluations, from which two were cost-effectiveness studies and one was a cost-utility study.

"nur" 3 ges.ök. Studien

Heterogeneous programmes and lack of economic evidence

Although all three programmes partly overlapped with regard to subtasks, sectors, and professional groups involved, they had different underlying conceptual approaches and core aims. The programmes were focussing on improving parent-child relationships and strengthening positive parenting. Although many programmes addressing children who have a parent with a mentally illness focus on parenting and psycho-education, they are generally heterogeneous and many more approaches than those provided in the three HEE exist (see 4.1). Given that the number of programmes has risen considerably over the last years, the number of published HEEs is surprisingly low. Therefore, there is a substantial knowledge gap regarding the cost-effectiveness of many interventions that have already been implemented.

allgemeiner Mangel an ges.ök. Evidenz von familienorientierten Interventionen

→ Wissenslücke zur Kosteneffektivität dieser Programme

Although included studies describe that programmes focused on child's development, conceptually, the three programmes focus primarily on the parent regarding core recipients of the programme. This limits comparability with the Village approach which is more family-oriented and actively works with children in addition to their parents. From the three HEEs identified, the content of the PBCM programme [22] from the Netherlands seems to come closest to the Village programme delivered in Tyrol. However, the two programmes are still too different to transfer cost-effectiveness results from the PBCM to the Village approach.

Vergleich mit dem Programm im Village-Projekt ist limitiert → anderer Fokus

British dominance limits transferability

begrenzte Übertragbarkeit auf den österreichischen Kontext

Two of the HEEs came from the UK, which belongs to those countries with a long tradition on HEEs. On the contrary, none of the countries with a longer history on implemented programmes (e.g., Australia, Finland, USA) have published HEEs. The differences in health care systems between the UK and Austria limit transferability of the results to the Austrian and even more to the Tyrolean local context where the Village project is located and where specific regional system characteristics including regional cost patterns and price levels exist.

Cost-effectiveness evidence limited to specific subgroups

Studienfokus auf Subgruppen, u.a. vulnerable Personengruppen (alleinerziehende Mütter, ethnische Minderheiten)

In the three HEEs, single parents and mothers affected by mental illnesses accounted for a significant proportion of all included parents. In one study, almost all participating parents were women [65]. That means that we know little on the cost-effectiveness of interventions addressing fathers or dual parent families.

→ allerdings Wissenslücke zu universellen Programmen

In those studies, where socio-economic data were collected, a substantial proportion of children are from families with low income or from families where the parent is currently unemployed. Ethnic minorities also seem to be overrepresented relatively to the total population in two studies. While this provides information on specific target groups and may even confirm that the intended groups have been successfully reached, a knowledge gap exists on the cost-effectiveness of more universal programmes that includes other subgroups.

Health economic evaluation approaches limit interpretation

methodische Limitationen hindern tlw. Interpretation der Ergebnisse

All included studies utilised a standard health economic approach, taking perspectives beyond the health care system perspective and they fulfilled current methodical standards of health economic evaluations to a large extent. However, the cost-effectiveness thresholds applied to evaluate whether the programme would be cost-effective, were mostly arbitrary and the outcome parameters used in some cost-effectiveness ratios (e.g. per unit of HOME score) are unusual and not comparable to other diseases which makes interpretation and resource allocation decisions across different indications difficult. Sensitivity analysis, a standard in HEEs to address uncertainty, was missing in two studies. This limits interpretations on the robustness of the results.

Costs of interventions vary but are overall low, cost comparability limited

Programmkosten variieren, sind aber insgesamt niedrig

The total costs of the intervention groups (receiving the programmes) varied substantially between the studies. One natural cause is the chosen perspective. However, this finding is also an indicator that the programmes delivered varied in unit costs and/or quantities such as number of session, although some similarities in tasks across programmes were observed. Overall, costs of the programmes themselves are low compared to interventions in other medical fields (e.g. oncology [149]).

Vergleichbarkeit der Kosten ist eingeschränkt

Cost results are limited in comparability, even if they have been conducted in the same country. For example, the two UK studies used different approaches for eliciting unit costs. Furthermore, the time horizons differ (in two studies < 1 year, in one up to 25 years) which also limits comparability.

Cost-effectiveness depends on different parameters

Kosteneffektivität ist stark von Perspektive, Subgruppe, und Schwellenwert abhängig

With regard to cost-effectiveness, results depend heavily on the perspective, considered subgroup, or WTP thresholds applied. This is particular the case for the HFP-M and IY programme [64, 65], whereby the IY showed that at a threshold value of £ 145 per ECBI-I score improvement, the probability for being cost-effective would be 100%. For PBCM [22] cost-effectiveness for all perspectives was demonstrated for a threshold of € >2,500. However, in both cases the WTP thresholds are hard to interpret. As demonstrated by one study, definite return on investment may only be demonstrated in a long-term perspective.

5.1.2 Summary on the economic evaluation framework

39 Studien mit Nachweisen zu negativen Konsequenzen der elterlichen psychischen Erkrankung für Kinder

A total of 39 publications [107-109, 111-146] were identified that provided empirical evidence on potential short-term and long-term adversities from parental mental illness for children (see 4.2.1). Our economic model demonstrated a broad variety of possible consequences for children individually, but also at the societal level. The most frequently mentioned adversities were negative health impacts, whereby studies most often identified mental health issues. The mental health problems cover a broad range of diagnoses in child and adolescent and adult mental health. Additionally, some of the problems mentioned are risk factors for mental illness rather than manifest diagnoses (e.g. social and self-isolation).

Gesundheitsfolgen sind nicht nur auf die psychische Gesundheit beschränkt

However, health impacts are not only restricted to mental health, as the term 'intergenerational cycle of mental illness' may suggest, but parental mental illness can also affect the physical health of the child (e.g. obesity). Beyond health, a number of consequences on social functioning but also socio-economic disadvantages in later life have been shown in the data. Overall, this results in broader societal economic impacts, e.g., in the form of increased need for mental health care. Impacts can occur early but also on the path to adulthood or in adult children of parents with a mental illness.

viele Auswirkungen betreffen Gesundheitssektor, aber auch andere Sektoren sind betroffen

Many of the impacts identified fall within the responsibility of the health sector. Yet, a large number affects other public sectors. We have presented examples for the social, educational and criminal justice sector. Additionally, costs can incur privately, for example due to private co-payments for treatment. This is in line with evidence on HEEs in child and adolescent mental health interventions, which showed that the majority of costs are incurred outside the health care sector. For example, one review showed that 90% of costs fell to the educational sector [24, 25].

erfolgreiche Prävention senkt Kosten außerhalb des Gesundheitssektors

In turn, this means that if preventive programmes are successful, costs are not just avoided in (mental) health care but potentially even more in other public realms. Furthermore, private cost burdens for the individuals may be reduced.

5.1.3 Contrasting existing health economic evidence with the evaluation framework

Outcome measures

nur tlw. Begründung für verwendete Endpunkte

The three HEEs identified in part I have used the following outcome parameters to inform the incremental cost-effectiveness ratios: (1) Quality adjusted life years (QALYs), (2) the Eyeberg Child Behaviour Inventory Scale (ECBI-I), and (3) the Home Observation for Measurement of the Environment (HOME) inventory. The outcomes were mainly retrieved from parents for their children, but one study [65] additionally collected QALYs from children. The HEEs only partially provide a rational for their selection of the outcome parameters and rather used the parameter from the clinical studies.

QALYs, ECBI-I- und HOME-Score

QALYs represent a generic utility measure, which combines changes in quality of life and life expectancy into one measurement figure. The ECBI-I assesses intensity and number of disruptive behaviour problems in children. The HOME inventory measures several dimensions of parenting quality.

QALYs vernachlässigen eine Reihe von potenziellen Nutzenkategorien im Bereich der psychischen Gesundheit

QALYs have been widely applied in economic evaluations across diseases and have been defined as the standard for outcome measurement in many guidelines on economic evaluation [150]. One of the core arguments to use QALYs is that they enable comparison of cost-effectiveness results across different diseases and across prevention programmes and treatments. As outlined in section 1.2, capturing quality of life and life expectancy is neglecting a number of potential further benefits from a programme. With regard to the impact dimensions identified in section 4.2, QALYs may capture the numerous health impacts indirectly, since health detriments likely influence QoL. However, the nuances and the broad spectrum of health impacts are likely not captured.

langfristige Erhöhung der Lebenserwartung in QALY-Berechnung nicht berücksichtigt

Firstly, the instrument that has been used to assess QoL was the EQ-5D. The EQ-5D includes only one domain on mental health, while our model has identified a broad variety of different mental health impacts. Secondly, the physical domains in the EQ-5D are unlikely appropriate to capture the very specific physical health impacts that has been described (e.g. oral health, injuries, health impact from parental smoking). Furthermore, while the preventive programmes may in the long-run increase life expectancy, this was not captured by the QALY measure in the studies, mainly due to the short time horizon. This potentially discriminates preventive programmes in the field of parental mental illness against treatments for physical health problems that have a more short-term life expectancy impact (e.g. in oncology [149]) in resource allocation decisions.

QALYs im Themengebiet elterlicher psychischer Erkrankung problematisch

Other authors have already addressed some of the methodological limitations of QALYs. For example, health economists have discussed QALYs controversially, in particular in relation to evaluating mental health care interventions [151]. Our analysis indicates that the application of QALYs are at least as problematic in studies addressing parental mental illness.

HOME-Score ist Surrogatparameter für kindliches Wohlergehen, ECBI-Score berücksichtigt nur eine Dimension des kindlichen Wohlergehens

The HOME inventory is only a surrogate parameter for the children's well-being as it addresses parenting quality. The study which applied this measure [22] may capture possible spill overs, but potentially fails to capture direct effects on children. The ECBI-I, on the other hand, only addresses behaviour problems in children. Both instruments address only one dimension of outcome and in the case of the ECBI-I only one dimension of mental health issues in children. Applying them as an outcome measure in HEEs ignores the numerous further (mental) health impacts that have been described in our model and that may be prevented by the programme.

soziale Funktionsfähigkeit und sozioökonomische Aspekte vernachlässigt

None of the outcome instruments applied address dimensions of social functioning and none of them captured short-term socio-economic dimensions on the individual level (e.g. school attendance, educational attainment) or influences of the programme on socio-economic dimensions later in life (employment, wages, etc.). The instruments also do not capture productivity gains that may result from positive long-term influences of the programmes on sick leave, early retirement, or premature mortality.

Ergebnisse nicht immer direkt bei Kindern gemessen → mögliche Verzerrung der ges.ök. Ergebnisse

While our economic logic model demonstrates that there may be numerous consequences from programmes directly in children, not all studies measured outcomes directly in children. Knapp and Wong (2020) [25] demonstrated for the field of perinatal mental health that a large proportion (in their example study 72%) of the total costs of perinatal mental health problems in parents is related to the child. These costs may be positively affected by preventive programmes. It is likely that this also applies for family programmes where children are older. If outcomes in children are not addressed, this may bias the cost-effectiveness results against the intervention.

Resource use and costs

Studien berechneten direkte Kosten aus mehreren Perspektiven

The studies calculated costs from different perspectives (health care, social care, public health sector). Apart from the cost for delivering the programme itself, the covered cost categories mostly relate to direct costs in the health care sector (e.g. costs for GP contacts, costs for psychotherapy sessions etc.).

allerdings nur für kurzen Beobachtungszeitraum

Two studies took a societal perspective and included inter-sectoral costs for the criminal justice sector, educational sector, and social care sector. However, the studies only depicted costs that materialise in the (short) period of observation. Private costs or out-of-pocket payments by patients associated with utilisation of 'other' services and not listed in clinical service inventories may be potentially excluded in studies although they may be a burden for some programme recipients (e.g. those with low income).

informelle Betreuung und Versorgung größtenteils vernachlässigt

→ Standardmethoden ignorieren Sorgearbeit

A general challenge concerns measuring resource use and costs arising from informal care in parental mental illness. The standard approaches (such as the human capital approach to measure productivity losses) abstract from social reproduction, unpaid work in the informal care economy or other sectors, and only consider production in form of income from paid work [49]. Costs for informal care in adults are usually measured by valuing the time spent on informal care with the income loss from forgone paid work or by applying salaries from professional carers. However, as our economic logic model has shown, in our cases the carers are children who do not have income from paid work. Their informal care cannot therefore be easily valued in monetary terms.

Economic evaluation framework and methods

ges.ök. Standardmethoden erfassen nicht die gesamte Bandbreite der Kosten und Effekte

The studies applied the standard study types, i.e. cost-effectiveness and cost-utility analyses using a time horizon below one year, except for one study, which extrapolated the long-term social return on investment based on a decision model. As has already been described, the time-horizon had consequences for the type of costs and outcomes included in the studies and a number of consequences identified in the logic model in section 4.2 have therefore not been captured by the studies. Cost-effectiveness and cost-utility analysis present the result as an incremental cost-effectiveness ratio based on one outcome parameter. These evaluation frameworks unlikely reflect the full range of costs and consequences of the programmes.

gesellschaftliche Perspektive fokussiert sich fast ausschließlich auf Kosten

The current standard in many health economic guidelines is to apply a societal perspective and two studies adequately followed this guideline. However, as we demonstrated, this is not sufficient to cover the full picture of costs and outcomes. This could be for a number of reasons: Firstly, the perspective chosen primarily influences the costs to be included in a HEE but less the outcomes. Yet, as shown above, it was the outcome instruments in the three HEEs, which resulted in the exclusion of a number of potential further relevant outcomes.

es fehlt oft an belastbaren empirischen Belegen für die Berechnung der Parameter

Secondly, even if cost categories are covering a broad variety of costs across sectors, robust empirical evidence to populate the parameters is often lacking. This limitation is especially the case for long-term consequences, which often need to be calculated and modelled based on assumptions rather than high quality direct empirical evidence. Thirdly, two of the three HEEs were piggy-back studies, where health economic data is collected alongside a clinical trial. Clinical studies are usually only powered for one primary outcome, which – as demonstrated – is not necessarily sufficient for the HEE. Even if more outcomes were assessed in the clinical study as it was for example the case in the HFP-M programme [65], in a cost-effectiveness or cost-utility analysis only one can be used for calculating the cost-effectiveness threshold.

derzeitige Qualitätschecklisten haben Limitationen

Despite these limitations, the quality appraisal showed that there were little methodological concerns in the studies. Hence, the current standard quality checklists seem not be designed to capture specific shortcomings that may apply in the field of parental mental illness.

Schlussfolgerung: Studien als valide Entscheidungsgrundlage möglicherweise nicht adäquat

The consequence from these findings is that the results from the HEEs may be misleading for decision makers. If the three studies in our review had included a broader spectrum of outcomes or long-term consequences, it could easily be the case that the programmes would have demonstrated less ambiguous results regarding value for money. For example, Knapp et al. [152] have shown in their overview on mental health promotion and mental illness prevention that many interventions were outstandingly good value for money if a broad spectrum of costs and outcomes is addressed and a long term time horizon is applied.

5.1.4 Methodological conclusions for economic evaluation in the field of parental mental illness

General methodological considerations

Schlussfolgerung für ges.ök. Evalutionen im vorliegendem Kontext:

From the results, we can derive a number of conclusions and inspirations for methodological approaches in HEEs of programmes in the field of parental mental illness and some research needs:

breite Betrachtungsweise und Berücksichtigung aller öffentlicher Sektoren

- It should become a standard for HEEs in this field to identify costs not just in the health sector, but also in the social care, education, and criminal justice sector. Additionally, private costs for children and their families should be collected. The recent methodological developments in HEEs (e.g. Drost et al. [39], PECUNIA [35, 36, 43, 51]) should be taken into account and will be helpful for doing that appropriately.

langfristige Betrachtung und Berücksichtigung von Kosten und Nutzen sind essentiell

- HEEs in addressing programmes in the field of parental mental illness should always address a long-term perspective. This is difficult to achieve because the effectiveness studies that are used for populating the outcome parameters in HEEs, usually have short time-horizons (for various reasons, e.g. because long follow-ups are expensive, decision

makers cannot wait for years to decide which programme to fund). One solution is to conduct more decision modelling studies where long-term consequences are mathematically modelled based on the best available evidence on pathways and (health) impacts. An example is the report on mental health promotion and mental illness prevention by Knapp et al. (2011) [152] or the study by Gardner et al. (2017) [64] in our review. In parallel, research activities should be initiated to collect more robust empirical primary data on long-term impacts. One way forward could be to collect primary data on parameters that are related to long-term consequences of parental mental illness (health status, social functioning, socio-economic status, educational attainment etc.) in a sample of adult children and compare them with adults who did not grow up with a mentally ill parent. Another approach would be to do a panel study to follow participating children in studies of prevention programmes over many years to get more robust evidence on their outcomes and status in the long run. This has been done in other areas on early intervention programmes, such as the longitudinal cohort study focussing on mental health of adolescents with and without mild intellectual disabilities in England [153].

→ meist nur im Zuge von Modellierungen möglich

Längsschnittstudien als weitere ergänzende Option

- It will be helpful for HEEs of prevention programmes if the socio-economic characteristics of the study population that are collected in the effectiveness study are coordinated with the health economists who will do the HEE, so that all relevant socio-economic information will be available from participating parents as well as their children.

Einbindung Gesundheitsökonom*innen von Beginn an

- Not all impacts from our model would necessarily be influenced by a single programme. There is therefore a need for thinking carefully which of the impacts in our model may be affected by the preventive programme in question. As sensible approach to address this challenge has been suggested by Skivington et al. (2021) [60] in the context of evaluating complex interventions. They recommend developing a separate logic model for the economic evaluation in addition to the one for the effectiveness evaluation. In contrast to our economic logic model in this report, such a model would additionally include the programme to be evaluated and specify the economic consequences from this specific programme.

Konzeption eines eigenständigen ökonomischen Wirkmodells der Intervention

- The decision on outcome parameters to be used in the HEE needs to be made carefully. The examples in our review have shown that QALYs are not necessarily appropriate to capture the multiple consequences a programme may have, especially not in the long run. While this means to deviate from existing standards and restrict comparability of cost-effectiveness with other diseases, it will increase chances to demonstrate a fuller picture of the outcomes than the existing studies were able to do. There is evidence that family-oriented complex interventions can empower the affected parent [154] as well as the child regarding self-efficacy [108, 155], self-reliance, and autonomy [156]. Others have shown to alleviate feelings of guilt and shame triggered by the parental mental illness [147] and to foster the acceptance of the parental illness [137]. Outcome instruments that have been developed for HEEs in mental health to overcome limitations of standard tools (e.g., the Oxford CAPabilities questionnaire-Mental Health/OxCAP-MH [47, 53]) should be assessed for their capacity to capture these type of outcomes in studies addressing parental mental illness validly.

sorgfältige Wahl der Outcome-Parameter in der ges.ök. Evaluation

Vermeidung von Doppelzählung von Kosten/Nutzen

- Attention needs to be paid to double counting or cross-domain counting that mainly concern intangible impacts (costs as well as benefits), as some outcomes can be treated as a cost, but also assigned to the benefit side [35, 36].

"Huckepack"-Analysen durch Modellierung ergänzen

- The analysis has also shown that piggy-back designs seem rather insufficient to cover the full spectrum of outcomes in the HEE, at least, if they are not combined with some modelling later on.

Kosten-Folgen-Analyse scheint am besten geeignetste Studienform

- From the available economic evaluation study types (cost-effectiveness, cost-utility, cost-benefit, cost-consequence analysis), cost-consequence analysis seems to be most appropriate to cover the complexity in this field. As we have seen, cost-effectiveness and cost-utility studies are restricted to one outcome parameter only, to present a single cost-effectiveness ratio. This is unlikely sufficient to address the full picture. This suggestion is in line with the recent recommendations on the economic evaluation of complex interventions which recommend to use cost-consequence analysis, thereby

  "… revealing the resource cost/outcome trade-offs and causal connection … as much as producing a ratio or a number" (Skivington et al., 2021, p.7).

Berücksichtigung von Verteilungseffekten und Subgruppen

- Subgroup analysis and assessing distributional impacts seem to be required in order to identify target groups for whom the programme would be most cost-effective while at the same time avoiding resource allocation that would reinforce existing health and socio-economic inequalities.

evt. Anpassung Qualitätschecklisten

- It may be worthwhile to evaluate in more detail whether the current standard checklists for quality appraisal of HEEs are sufficiently addressing quality issues that are specifically relevant in parental mental illness interventions and whether specific appraisal tools would have to be developed.

Conclusions for the economic evaluation of the Village programme

Schlussfolgerung für die ges.ök. Evaluation des Village-Programms

This section is to be seen as a starting point for a design of the HEE of the Village programme and a detailed data analysis plan.

The Village programme

Ziele des Village-Programms sind vielfältig und sollen durch 2 Praxisansätze erreicht werden

As described above (section 3.2.1), the Village programme aims to improve child development and wellbeing outcomes for children by two practices combined into a programme. The first pillar is the 'Sensitive Screening' (SENSE) approach that facilitates identification of children by determining the parental status, care obligations, family constellation, details of children, and by finding out strengths and challenges of the child when coping with the affected parent. Essential steps of the second pillar, the 'Collaborative Village Approach' (CVA), are to understand daily perceptions and experiences of the child (from the parent and child's perspective), the development of a plan to strengthen and sustain the support of the children via activating the child's social and professional network. Further goals of the intervention are to overcome barriers such as the current lack of skills and knowledge of practitioners.

Expected Outcomes

Programmkomponenten sollen Wissen verbessern, Verhalten und Emotionen verändern

In section 3.2.1, it was illustrated that components of the programme are expected to positively change knowledge, behaviour and emotions in children, parents and practitioners. For example, Village investigators hypothesise to improve knowledge in practitioners regarding available support options, in parents concerning the impact of mental illness on the child and children may better understand parental mental illness. Emotional changes in practitioners can be that they feel more responsible for the children, parents possibly increase trust to practitioners and children may feel less guilt and self-stigma. Behaviour changes in practitioners may be better signposting of support needs of children to services, parents may better listen to their child and children may more actively seek help.

mögliche langfristige Verbesserungen betreffen die Eltern-Kind-Beziehung, soziale Beziehungen, schulischen Leistungen und Resilienz des Kindes

Investigators of the Village project assume that these mechanisms can have the following favourable outcomes in children in the long run:

- Improved relationship with the parent
- Social relationships within and outside the family
- Academic performance,
- Resilience and coping.

This would eventually maintain mental health and wellbeing (in healthy children) or reduce health burden and increase QoL (in children already experiencing problems).

Änderungen im Verhalten führen auch zu ökonomischen Konsequenzen

From an economic point of view, changes in behaviour are most directly linked to economic consequences. For example, increased help seeking in children has a direct impact on resource use of public services.

Health economic study type for the Village programme

Vielfalt der Ergebnisse erfordert eine Kosten-Folgen-Analyse

The methodological considerations above and the diversity of outcomes that may result from the programme confirm the original plan to conduct a cost-consequence analysis rather than a cost-effectiveness or a cost-utility analysis.

Costs to be measured

zu berücksichtigende Kosten und Ressourcen

The HEE needs to cover the following cost components:

a) Resource inputs to deliver the programme

To deliver the full programme, the following resource inputs are needed:

- Personnel resources for providing the programme
  - Medical staff: Physicians and (mental) health care professionals for providing the screening component of the programme
  - Village facilitators (different professionals such as social workers, psychologists or social pedagogues) working with the children and their parents to provide the CVA-component of the programme
  - Administrative/management staff to organise logistics, standardised documentation, coordination among village facilitators, training of screening practitioners and village facilitators, fidelity and quality assurance
  - Supervision staff
  - Professionals offering professional support for the children and/or families if need is identified by the village coordinator

Bedarf an medizinischem Personal und nicht-medizinischem Personal wie Sozialarbeiter*innen etc.

administratives Personal und professionelle Unterstützung für die Kinder

materielle Ressourcen

- Materials and resources to support the work of professionals (e.g. books, manuals, screening tools)

Infrastruktur

- Infrastructure (e.g. room to meet with families, online platform to work online with families)

Transportmöglichkeiten und benötigte Zeit

- Transport (to travel to families)
- Time resources by families (for participation in the programme and for travelling)

sonstige Zeitbedarfe

- Extra hours used by Village facilitators to provide support beyond the programme (e.g. psycho-education, supporting disease management of parents and daily life structures, telephone advice)

b) Costs related to adverse impacts of parental mental illness

Berücksichtigung der Kosten, welche durch die zugrundeliegende Krankheit verursacht werden und nicht in Standardinstrumenten Niederschlag finden

In order to address costs in all relevant sectors identified in section 4.2.1, available services and cash benefits that may be used by children in health care, social care, education and criminal justice need to be identified and covered in the resource use inventory. The services included in the inventories may go beyond those typically addressed in standard resource use measurement tools, because they include very likely services for healthy children (e.g. increased use of child care to support the mentally ill parent) that would not be relevant in HEEs on programmes targeting only mentally ill persons. The challenge will, however be, where to draw the line, i.e., how to define which services have been used by children because of having a parent with a mental illness and which would have been used anyway. Childcare is a good example for this.

Berücksichtigung der negativen Effekte der elterlichen Erkrankung auf der Folgenseite

In order to avoid double counting, as described above, it seems to make most sense in the cost-consequence analysis framework, to address all costs related to adverse effects from parental mental illness on the consequence side of the equation. The cost side would then only cover the costs for delivering the programme (including overhead costs for management etc.).

Umsicht ist geboten bei der Interpretation und Zuordnung von Folgen/Konsequenzen

For example, the costs from using health and social services due to own health problems in children can be described as an outcome (hence a consequence) from the programme. Hereby, it will be challenging how to interpret changes in services use. For example, an increase in use increases costs for the public sector but it is at the same time a desired outcome because children and adolescents with mental health problems substantially underuse services and often do not seek help despite obvious needs for professional support [157, 158]. In the absence of long term data, it will be difficult to see whether an increase of costs in the short term will be offset by reduced needs for services and other benefits in the long run. Furthermore, the increase in use may partly concern inadequate services due to a lack of available needs-based services. For example, children are sometimes admitted to inpatient paediatric care in Tyrol in a crisis situation of the parent, because there is no other service available [159]. Hospital care is the most expensive care element and may be used more often if more children are identified as a result of the programme.

eventuelle Modellierung von langfristigen Folgen/Konsequenzen

Long-term data to quantify impacts in adulthood will not be available in our study participants. It needs to be considered, whether some modelling activities would be possible to account for the long-term impacts identified in section 4.2.1. or to what extent and in which direction the results would be biased if long term consequences are neglected in our HEE.

Outcomes to be measured

QALYs nicht als alleiniger Ergebnisparameter

In a cost-consequence analysis, we would refrain from measuring QALYs as a single parameter. For demonstrating the outcomes that are illustrated in the Village logic model, a number of validated and standardised questionnaires are used in the Village project. Furthermore, qualitative data is collected from participating parents and children. Information is collected on short-term outcomes.

bedachte Auswahl der erhobenen Ergebnisparameter für die Kosten-Folgen-Analyse ist essentiell

Not all outcomes for which data have been collected, can be used in the cost-consequence analysis and a selection will have to be made. One way to prioritise outcomes depicted in the cost-consequence analysis would be to choose those with the best established link to the impacts identified in section 4.2.1, or those that address some of the components in the economic model directly. An example for the former would be if some participating children placed parental mental illness outside themselves (detachment), thus not feeling responsible anymore, after they receive the programme, which may have an impact on school attendance. An example for the latter would be help seeking in children which is related to the societal economic impact dimension if professional services are used. It should also be reflected whether information on some of the impacts identified in section 4.2.1 might be derived from the qualitative data collected in programme participants. For example, these data could give insight whether the programme influenced school attendance or caring responsibilities of children.

Berücksichtigung von Verhaltensänderungen bei Fachkräften?

One question to be discussed is whether outcomes measured in practitioners also needs to be addressed in the HEE. For example, if psychiatrists increase referrals of children to other professionals because they have a better knowledge on which services are available, their behaviour has resource use consequences. However, these services would be probably directly measured in children via the service receipt inventory.

5.1.5 Limitations

möglicherweise fehlen Studien, da Interventions- und Populationsbegriff nicht eindeutig definiert

Our review on HEEs aimed to identify HEEs on family-oriented complex interventions that are preventive in nature for children. However, as there is no standard term for such programmes and authors use different notions and definitions for the programmes, further studies may be available. The same limitation applies for the population term. The primary term we used was the frequently used expression ‘children of parents with a mental illness’ (COPMI), but we did also look for similar terms in this context. We still may have missed studies that used terms that were not included in our search strategy. For example, there may be overlaps with the ‘young carer’ literature, which we have not actively searched for.

Beschränkung hinsichtlich des Altersspektrums der Kinder, keine Berücksichtigung der perinatalen Population

The age of the children in families participating in the programmes was restricted to the age range 4 to 18, hence this report does not include HEEs on perinatal mental health or on programmes targeting mentally ill parents with children up to 3 years of age. This may reinforce the artificial dichotomy between perinatal parental mental illness and parental mental illness that affects older children. However, we were primarily interested in preventive programmes that targeted a similar age group as the intervention in the Village project. This way, the international health economic evidence would be more relevant for the Village programme and the methods applied would more likely be of interest for evaluating the Village programme. For example, child out-

come measures in perinatal mental health programmes would be very different from those in programmes where children can already talk themselves.

keine explizite Bewertung der Übertragbarkeit mit validiertem Instrument

Another limitation concerns the transferability of the studies to the Austrian context. We did not carry out an explicit transferability appraisal. However, even without, it is clear that transferability of studies is limited due to differences in the health care systems and prices/costs (two are from the UK, which has a Beveridge system, whereas Austria has a Bismarck system). Some characteristics of the programme (e.g. type of staff involved) may not be able to implement in Austria (e.g. due to different occupational laws). Furthermore, programmes in the three HEEs differed from the Village programme, which also limits transferability.

ökonomisches Wirkmodell beschränkt sich auf die in der Suche identifizierte Literatur

The economic logic model only takes into account available empirical data and considers only those impact categories that have been addressed in the studies identified. There may be further adverse consequences that are not illustrated in our model. For example, we did not identify studies measuring socio-economic and health inequality or poverty resulting from the parental illness. Furthermore, we did not assess the quality of the studies and therefore some of the associations between parental mental illness and child outcomes may be uncertain.

Schwerpunkt liegt „nur" auf adversen Effekten der elterlichen Erkrankung, nicht auf möglichen positiven Effekten durch Prävention

Another limitation is that we restricted the model to the adverse impact from parental mental illness while potential positive impacts from preventive interventions may also be valuable for identifying important parameters for HEEs. In other words, the benefits from prevention may not just be a reversal of the adverse impacts but go beyond. For example, an intangible benefit from offering social support could be social cohesion, which is not directly addressed in our model. The suggestion above to create a separate logic model for HEEs would help to overcome this limitation. In addition, we have not taken into account possible resilience factors that may develop as a result of the difficult family situation.

nicht alle Kinder sind von negativen Effekten betroffen

The model describes potential adverse impacts from parental mental illness and does not account for the fact that not all children are experiencing these consequences. Caution is therefore required not to generalise these results to all children. The aim of our report was not to identify the magnitude of adverse effects or specific groups of children at risk but to demonstrate the range and types of potential adverse impacts to illustrate where costs may occur and, thus, which types of costs to include in a HEE.

eine Literatursuche für beide Berichtsteile

We used one literature search strategy for both parts. Due to the lack of standards in search terms in this field, our search was rather broad, yielding more than 1,600 hits. While this demonstrates low specificity for the search for existing HEEs (part I), we increased sensitivity to identify as many studies as possible for part II.

Fokus auf englisch- und deutschsprachige Literatur

Only sources in English and German could be included. The search period was from 2010 onwards. While this seems unproblematic for part I (because HEE research activities in this field mainly happened recently) research on the consequences of parental mental illness for their children may have been published before. Our model may therefore not illustrate all consequences on which evidence is available.

6 Conclusion

This report summarises the evidence on health economic studies in the context of parental mental illness and family-oriented complex interventions (addressing children ≥ 4 years of age). Furthermore, this report tried to identify the multiple potential adverse impacts of parental mental illness for children, and thus to illustrate which dimensions need to be addressed in a HEE so that impacts are covered as comprehensive as possible. Results on both should support a HEE of the intervention programme used in the Village project but also on other preventive programmes in the field of parental mental illness.

Berichtsfokus: ges.ök. Evidenz von Interventionen im Kontext elterlicher psychischen Erkrankungen inkl. deren Auswirkungen auf die Kinder

The results demonstrate that there is extremely little economic knowledge on preventive family-oriented interventions addressing children in our age range available, even though many countries have started and evaluated preventive programmes. Decision makers therefore lack decision support whether or not to invest in such programmes and which programmes would offer the best value for money.

wenig ges.ök. Studien für familienorientierte präventive Interventionen

Evidence on the cost-effectiveness of programmes in other fields of mental health promotion and prevention of mental illness has shown that many interventions have demonstrated outstandingly good value for money and are even self-financing over time. The lack of HEEs of programmes in the field of parental mental illness therefore forgoes an opportunity to make the economic case for investing in preventive programmes.

ges.ök. Evaluationen könnte ökonomische Sinnhaftigkeit von Investition in Prävention zeigen

At the same time, our analysis showed that a number of methodological challenges need to be taken into account in conducting robust HEEs in this field. They go beyond current methodological standards of HEEs. Because of that, the confidence in the results from some of the available studies is limited even though they have been rated with a high quality according to existing assessment tools. This bears the risk to make decisions on biased cost-effectiveness results and, according to our examples, it seems more likely that the value for money from those programmes is underestimated than overestimated. We have specified needs for data and research that may help to overcome some of the limitations. These needs should be considered in future applications for research grants.

zahlreiche methodische Limitationen trotz Einhaltung von Qualitätskriterien

→ Wirtschaftlichkeit präventiver Interventionen möglicherweise unterschätzt

7 Literature

[1] Bauer A., Best S., Malley J., Christiansen H., Goodyear M. J., Zechmeister-Koss I., et al. Towards a program theory for family-focused practice in adult mental health care. Front Psychiatry – Public Mental Health. 2021. DOI: Submitted on: 14 Jul 2021, DOI: 10.3389/fpsyt.2021.741225 (in press).

[2] World Health Organisation (WHO). The ICD-10 classification of mental and behavioural disorders. World Health Organization; 1993.

[3] National Institute of Mental Health (NIH). Mental Illness – Definition. 2021 [cited 14/04/2021]. Available from: https://www.nimh.nih.gov/health/statistics/mental-illness.

[4] ICD10Data.com. Mental, Behavioral and Neurodevelopmental disorders F01-F99. 2021 [cited 14/04/2021]. Available from: https://www.icd10data.com/ICD10CM/Codes/F01-F99.

[5] Nicholson J., Biebel K., Katz-Leavy J., Williams V. and Williams. The Prevalence of Parenthood in Adults with Mental Illness: Implications for State and Federal Policymakers, Programs, and Providers. Psychiatry Publications and Presentations Paper. 2002;153.

[6] Bee P., Bower P., Byford S., Churchill R., Calam R., Stallard P., et al. The clinical effectiveness, cost-effectiveness and acceptability of community-based interventions aimed at improving or maintaining quality of life in children of parents with serious mental illness: a systematic review. Health Technol Assess. 2014;18(8):1-250. Epub 2014/02/08. DOI: 10.3310/hta18080.

[7] Wancata J. Prävalenz und Versorgung psychischer Krankheiten in Österreich. Klinische Abteilung für Sozialpsychiatrie, Medizinische Universität Wien,, [cited 15/09/2021] 2017. Available from: https://www.meduniwien.ac.at/hp/fileadmin/sozpsychiatrie/pdf/Abschlussbericht_WIssenschaft_Wancata.pdf.

[8] Maybery D. and Reupert A. E. The number of parents who are patients attending adult psychiatric services. Curr Opin Psychiatry. 2018;31(4):358-362. Epub 2018/05/31. DOI: 10.1097/yco.0000000000000427.

[9] Östman M. and Hansson L. Children in families with a severely mentally ill member. Social Psychiatry and Psychiatric Epidemiology. 2002;37(5):243-248. DOI: 10.1007/s00127-002-0540-0.

[10] Pretis M. and Dimova A. Vulnerable children of mentally ill parents: towards evidence-based support for improving resilience. Support for Learning. 2008;23(3):152-159. DOI: https://doi.org/10.1111/j.1467-9604.2008.00386.x.

[11] Radicke A., Barkmann C., Adema B., Daubmann A., Wegscheider K. and Wiegand-Grefe S. Children of Parents with a Mental Illness: Predictors of Health-Related Quality of Life and Determinants of Child-Parent Agreement. International journal of environmental research and public health. 2021;18(2):379. DOI: 10.3390/ijerph18020379.

[12] Goodyear M., McDonald M., von Doussa H., Cuff R. and Dunlop B. Meeting the Intergenerational Needs of Families Where a Parent Has a Mental Illness. Journal of Parent and Family Mental Health. 2018;3(2), 1-3. [1]. DOI: https://doi.org/10.7191/parentandfamily.1011.

[13] Furlong M., McGilloway S., Bywater T., Hutchings J., Smith S. M. and Donnelly M. Behavioural and cognitive-behavioural group-based parenting programmes for early-onset conduct problems in children aged 3 to 12 years. Cochrane Database Syst Rev. 2012(2):Cd008225. Epub 2012/02/18. DOI: 10.1002/14651858.CD008225.pub2.

[14] Lannes A., Bui E., Arnaud C., Raynaud J.-P. and Revet A. Preventive interventions in offspring of parents with mental illness: a systematic review and meta-analysis of randomized controlled trials. Psychological Medicine. 2021;51(14):2321-2336. Epub 2021/08/26. DOI: 10.1017/S0033291721003366.

[15] Siegenthaler E., Munder T. and Egger M. Effect of preventive interventions in mentally ill parents on the mental health of the offspring: systematic review and meta-analysis. J Am Acad Child Adolesc Psychiatry. 2012;51(1):8-17.e18. Epub 2011/12/20. DOI: 10.1016/j.jaac.2011.10.018.

[16] Thanhäuser M., Lemmer G., de Girolamo G. and Christiansen H. Do preventive interventions for children of mentally ill parents work? Results of a systematic review and meta-analysis. Curr Opin Psychiatry. 2017;30(4):283-299. Epub 2017/05/16. DOI: 10.1097/yco.0000000000000342.

[17] Cuijpers P., Weitz E., Karyotaki E., Garber J. and Andersson G. The effects of psychological treatment of maternal depression on children and parental functioning: a meta-analysis. Eur Child Adolesc Psychiatry. 2015;24(2):237-245. Epub 2014/12/20. DOI: 10.1007/s00787-014-0660-6.

[18] United Nations Children's Fund (UNICEF). Convention on the Rights of the Child text. 2021 [cited 14/04/2021]. Available from: https://www.unicef.org/child-rights-convention/convention-text.

[19] Bundesministerium für Soziales, Gesundheit, Pflege und Konsumentenschutz (BMSGPK). Gesundheitsziele Österreich. 2021 [cited 14/04/2021]. Available from: https://gesundheitsziele-oesterreich.at/website2017/wp-content/uploads/2018/08/gz_langfassung_2018.pdf.

[20] Bundesministerium für Soziales, Gesundheit, Pflege und Konsumentenschutz (BMSGPK). Kinder- und Jugendgesundheitsstrategie. 2021 [cited 14/10/2021]. Available from: https://www.sozialministerium.at/Themen/Gesundheit/Kinder--und-Jugendgesundheit/Kinder--und-Jugendgesundheitsstrategie.html.

[21] Waldmann T., Stiawa M., Dinc Ü., Saglam G., Busmann M., Daubmann A., et al. Costs of health and social services use in children of parents with mental illness. Child and Adolescent Psychiatry and Mental Health. 2021;15(1):10. DOI: 10.1186/s13034-021-00360-y.

[22] Wansink H. J., Drost R. M. W. A., Paulus A. T. G., Ruwaard D., Hosman C. M. H., Janssens J. M. A. M., et al. Cost-effectiveness of preventive case management for parents with a mental illness: a randomized controlled trial from three economic perspectives. BMC health services research. 2016;16:228-228. DOI: 10.1186/s12913-016-1498-z.

[23] Lysdahl K. B., Mozygemba K., Burns J., Brönneke J. B., Chilcott J. B., Ward S., et al. Comprehensive assessment of complex technologies: Integrating various aspects in Health Technology Assessment. Int J Technol Assess Health Care. 2017;33(5):570-576. Epub 2017/08/07. DOI: 10.1017/S0266462317000678.

[24] Knapp M., Ardino V., Brimblecombe N., Evans-Lacko S., Iemmi V., King D., et al. Youth mental health: new economic evidence. 2016.

[25] Knapp M. and Wong G. Economics and mental health: the current scenario. World Psychiatry. 2020;19(1):3-14. DOI: https://doi.org/10.1002/wps.20692.

[26] Zechmeister-Koss I., Stanak M. and Wolf S. The status of health economic evaluation within decision making in Austria. Wien Med Wochenschr. 2019;169(11-12):271-283. Epub 2019/03/15. Stand der gesundheitsökonomischen Evaluation bei der Entscheidungsfindung in Österreich. DOI: 10.1007/s10354-019-0689-8.

[27] Mihalopoulos C. Assessing the economic costs of parental mental illness. In: Reupert A., Maybery D., Nicholson J., Seeman M. V. and Göpfert M., editors. Parental Psychiatric Disorder: Distressed Parents and their Families. 3 ed. Cambridge: Cambridge University Press; 2015. p. 42-50.

[28] Łaszewska A., Wancata J., Jahn R. and Simon J. The excess economic burden of mental disorders: findings from a cross-sectional prevalence survey in Austria. The European Journal of Health Economics. 2020;21(7):1075-1089. DOI: 10.1007/s10198-020-01200-0.

[29] Bauer A., Parsonage M., Knapp M., Iemmi V. and Adelaja B. The costs of perinatal mental health problems. 2014.

[30] Bauer A., Knapp M. and Parsonage M. Lifetime costs of perinatal anxiety and depression. J Affect Disord. 2016;192:83-90. Epub 2015/12/29. DOI: 10.1016/j.jad.2015.12.005.

[31] Keuth H. The positivist dispute in German sociology: A scientific or a political controversy? Journal of Classical Sociology. 2015;15(2):154-169. DOI: 10.1177/1468795x14567825.

[32] Coyle D. GDP – A Brief but Affectionate History – Revised and expanded Edition: Princeton University Press; 2015.

[33] Charlton V. and Weale A. Exorcising the positivist ghost in the priority-setting machine: NICE and the demise of the 'social value judgement'. Health Economics, Policy and Law. 2021:1-7. Epub 2021/02/11. DOI: 10.1017/S1744133121000049.

[34] Bhaskar R., Danermark B. and Price L. Interdisciplinarity and Wellbeing: A Critical Realist General Theory of Interdisciplinarity. 1 ed. London2018.

[35] Janssen L. M. M., Pokhilenko I., Evers S. M. A. A., Paulus A. T. G., Simon J., König H.-H., et al. Exploring the identification, validation, and categorization of the cost and benefits of criminal justice in mental health: the PECUNIA project. Int J Technol Assess Health Care. 2020;36(4):418-425. Epub 2020/07/27. DOI: 10.1017/S0266462320000471.

[36] Pokhilenko I., Janssen L. M. M., Evers S. M. A. A., Drost R. M. W. A., Simon J., König H.-H., et al. Exploring the identification, validation, and categorization of costs and benefits of education in mental health: The PECUNIA project. Int J Technol Assess Health Care. 2020;36(4):325-331. Epub 2020/05/28. DOI: 10.1017/S0266462320000203.

[37] Evers S. and Dircksen C. Towards standardization of economic evaluation research in the youth psychosocial care sector: A broad consultation in the Netherlands. Global & Regional Health Technology Assessment. 2020;7:117-123. DOI: 10.33393/grhta.2020.2143.

[38] Shiell A., Hawe P. and Gold L. Complex interventions or complex systems? Implications for health economic evaluation. BMJ. 2008;336(7656):1281-1283. DOI: 10.1136/bmj.39569.510521.AD.

[39] Drost R. M. W. A., Paulus A. T. G. and Evers S. M. A. A. Five pillars for societal perspective. Int J Technol Assess Health Care. 2020;36(2):72-74. Epub 2020/01/31. DOI: 10.1017/S026646232000001X.

[40] Lysdahl K., Mozygemba K., Burns J., Chilcott J., Brönneke J. and Hofmann B. Guidance for assessing effectiveness, economic aspects, ethical aspects, socio-cultural aspects and legal aspects in complex technologies2016.

[41] Fischer S. and Stanak M. Social return on investment: outcomes, methods, and economic parameters. LBI-HTA Project report No. 95b, [cited 14/04/2021] 2017. Available from: https://eprints.aihta.at/1141/.

[42] Drost R. M. W. A., van der Putten I. M., Ruwaard D., Evers S. M. A. A. and Paulus A. T. G. Conceptuatlizations of the societal perspective withing economic evaluations: A systematic review. Int J Technol Assess Health Care. 2017;33(2):251-260. Epub 2017/06/23. DOI: 10.1017/S0266462317000526.

[43] PECUNIA. Programme in costing, resource use measurement and outcome valuation for use in multi-sectoral national and international health economic evaluation (PECUNIA). 2021 [cited 26/04/2021]. Available from: https://www.pecunia-project.eu.

[44] Duevel J. A., Hasemann L., Peña-Longobardo L. M., Rodríguez-Sánchez B., Aranda-Reneo I., Oliva-Moreno J., et al. Considering the societal perspective in economic evaluations: a systematic review in the case of depression. Health Economics Review. 2020;10(1):32. DOI: 10.1186/s13561-020-00288-7.

[45] Simon J., Anand P., Gray A., Rugkåsa J., Yeeles K. and Burns T. Operationalising the capability approach for outcome measurement in mental health research. Social Science & Medicine. 2013;98:187-196. DOI: https://doi.org/10.1016/j.socscimed.2013.09.019.

[46] Sen A. Equality of What? In The Tanner Lecture on Human Values, I, 197-220: Cambridge: Cambridge University Press; 1980.

[47] Nussbaum M. Capabilities as fundamental entitlements: Sen and social justice. Feminist Economics. 2003;9(2-3):33-59. DOI: 10.1080/1354570022000077926.

[48] Łaszewska A., Schwab M., Leutner E., Oberrauter M., Spiel G. and Simon J. Measuring broader wellbeing in mental health services: validity of the German language OxCAP-MH capability instrument. Quality of Life Research. 2019;28(8):2311-2323. DOI: 10.1007/s11136-019-02187-9.

[49] Bowles S. and Gintis H. The Problem with Human Capital Theory--A Marxian Critique. The American Economic Review. 1975;65(2):74-82.

[50] Encyclopedia of Public Health. Friction Cost Method. In: Kirch W., editor. Encyclopedia of Public Health. Dordrecht: Springer Netherlands; 2008. p. 465-465.

[51] PECUNIA Group. PECUNIA Reference Unit Cost Compendium (PECUNIA RUC Compendium) (Version 1.0/2021). 2021.

[52] Drost R. M., Paulus A. T., Ruwaard D. and Evers S. M. Inter-sectoral costs and benefits of mental health prevention: towards a new classification scheme. J Ment Health Policy Econ. 2013;16(4):179-186. Epub 2014/02/15.

[53] Nussbaum M. C. Women and Human Development: The Capabilities Approach. Cambridge: Cambridge University Press; 2000.

[54] Simon J., Łaszewska A., Leutner E., Spiel G., Churchman D. and Mayer S. Cultural and linguistic transferability of the multi-dimensional OxCAP-MH capability instrument for outcome measurement in mental health: the German language version. BMC psychiatry. 2018;18(1):173. DOI: 10.1186/s12888-018-1762-3.

[55] Heale R. and Twycross A. Validity and reliability in quantitative studies. Evidence Based Nursing. 2015;18(3):66-67. DOI: 10.1136/eb-2015-102129.

[56] Johannesson M. Avoiding Double-Counting in Pharmacoeconomic Studies. PharmacoEconomics. 1997;11(5):385-388. DOI: 10.2165/00019053-199711050-00001.

[57] Landwehr C. and Klinnert D. Value congruence in health care priority setting: social values, institutions and decisions in three countries. Health Economics, Policy and Law. 2015;10(2):113-132. Epub 2014/12/01. DOI: 10.1017/S1744133114000437.

[58] Torbica A., Tarricone R. and Drummond M. Does the approach to economic evaluation in health care depend on culture, values, and institutional context? Eur J Health Econ. 2018;19(6):769-774. Epub 2017/12/07. DOI: 10.1007/s10198-017-0943-1.

[59] van Mastrigt G. A., Hiligsmann M., Arts J. J., Broos P. H., Kleijnen J., Evers S. M., et al. How to prepare a systematic review of economic evaluations for informing evidence-based healthcare decisions: a five-step approach (part 1/3). Expert Rev Pharmacoecon Outcomes Res. 2016;16(6):689-704. Epub 2016/11/03. DOI: 10.1080/14737167.2016.1246960.

[60] Skivington K., Matthews L., Simpson S. A., Craig P., Baird J., Blazeby J. M., et al. Framework for the development and evaluation of complex interventions: gap analysis, workshop and consultation-informed update. 2021;25:57. DOI: 10.3310/hta25570.

[61] Edwards R. T., Céilleachair A., Bywater T., Hughes D. A. and Hutchings J. Parenting programme for parents of children at risk of developing conduct disorder: cost effectiveness analysis. BMJ. 2007;334(7595):682. DOI: 10.1136/bmj.39126.699421.55.

[62] Muntz R., Hutchings J., Edwards R.-T., Hounsome B. and O'Céilleachair A. Economic Evaluation of Treatments for Children with Severe Behavioural Problems. J Ment Health Policy Econ. 2005;7:177-189.

[63] O'Neill D., McGilloway S., Donnelly M., Bywater T. and Kelly P. A cost-effectiveness analysis of the Incredible Years parenting programme in reducing childhood health inequalities. The European Journal of Health Economics. 2013;14(1):85-94. DOI: 10.1007/s10198-011-0342-y.

[64] Gardner F., Leijten P., Mann J., Landau S., Harris V., Beecham J., et al. Could scale-up of parenting programmes improve child disruptive behaviour and reduce social inequalities? Using individual participant data meta-analysis to establish for whom programmes are effective and cost-effective. Southampton (UK): NIHR Journals Library; 2017.

[65] Day C., Briskman J., Crawford M. J., Foote L., Harris L., Boadu J., et al. An intervention for parents with severe personality difficulties whose children have mental health problems: a feasibility RCT. Health Technol Assess. 2020;24(14):1-188. Epub 2020/03/17. DOI: 10.3310/hta24140.

[66] Evers S., Goossens M., de Vet H., van Tulder M. and Ament A. Criteria list for assessment of methodological quality of economic evaluations: Consensus on Health Economic Criteria. Int J Technol Assess Health Care. 2005;21(2):240-245. Epub 2005/06/01.

[67] Wijnen B., Van Mastrigt G., Redekop W. K., Majoie H., De Kinderen R. and Evers S. How to prepare a systematic review of economic evaluations for informing evidence-based healthcare decisions: data extraction, risk of bias, and transferability (part 3/3). Expert Rev Pharmacoecon Outcomes Res. 2016;16(6):723-732. Epub 2016/10/21. DOI: 10.1080/14737167.2016.1246961.

[68] Higgins J. P. T., Thomas J., Chandler J., Cumpston M., Li T., Page M. J., et al. Cochrane Handbook for Systematic Reviews of Interventions: Wiley; 2019.

[69] House A., Guthrie E., Walker A., Hewsion J., Trigwell P., Brennan C., et al. A programme theory for liaison mental health services in England. BMC health services research. 2018;18(1):742. DOI: 10.1186/s12913-018-3539-2.

[70] Fritz M. S. and Arthur A. M. Moderator Variables. Oxford University Press; 2017.

[71] Funnell S. and Rogers P. Purposeful Program Theory: Effective Use of Theories of Change and Logic Models2011.

[72] Collishaw S., Hammerton G., Mahedy L., Sellers R., Owen M. J., Craddock N., et al. Mental health resilience in the adolescent offspring of parents with depression: a prospective longitudinal study. The Lancet Psychiatry. 2016;3(1):49-57. DOI: 10.1016/S2215-0366(15)00358-2.

[73] Mossakowski K. N. Social Causation and Social Selection. The Wiley Blackwell Encyclopedia of Health, Illness, Behavior, and Society2014. p. 2154-2160.

[74] Larsson B., Fossum S., Clifford G., Drugli M. B., Handegård B. H. and Mørch W.-T. Treatment of oppositional defiant and conduct problems in young Norwegian children. European Child & Adolescent Psychiatry. 2009;18(1):42-52. DOI: 10.1007/s00787-008-0702-z.

[75] Axberg U. L. F. and Broberg A. G. Evaluation of "The Incredible Years" in Sweden: The transferability of an American parent-training program to Sweden. Scandinavian Journal of Psychology. 2012;53(3):224-232. DOI: https://doi.org/10.1111/j.1467-9450.2012.00955.x.

[76] Homem T. C., Gaspar M. F., Santos M. J. S., Azevedo A. F. and Canavarro M. C. Incredible Years Parent Training: Does it Improve Positive Relationships in Portuguese Families of Preschoolers with Oppositional/Defiant Symptoms? Journal of Child and Family Studies. 2015;24(7):1861-1875. DOI: 10.1007/s10826-014-9988-2.

[77] Azevedo A. F., Seabra-Santos M. J., Gaspar M. F. and Homem T. C. The Incredible Years Basic Parent Training for Portuguese Preschoolers with AD/HD Behaviors: Does it Make a Difference? Child & Youth Care Forum. 2013;42(5):403-424. DOI: 10.1007/s10566-013-9207-0.

[78] Fernandes Azevedo A., Seabra-Santos M. J., Gaspar M. F. and Homem T. A parent-based intervention programme involving preschoolers with AD/HD behaviours: are children's and mothers' effects sustained over time? Eur Child Adolesc Psychiatry. 2014;23(6):437-450. Epub 2013/09/04. DOI: 10.1007/s00787-013-0470-2.

[79] McGilloway S., Mhaille G. N., Bywater T., Furlong M., Leckey Y., Kelly P., et al. A parenting intervention for childhood behavioral problems: a randomized controlled trial in disadvantaged community-based settings. J Consult Clin Psychol. 2012;80(1):116-127. Epub 2011/12/14. DOI: 10.1037/a0026304.

[80] Menting A. T. A., de Castro B. O., Wijngaards-de Meij L. D. N. V. and Matthys W. A Trial of Parent Training for Mothers Being Released From Incarceration and Their Children. Journal of Clinical Child & Adolescent Psychology. 2014;43(3):381-396. DOI: 10.1080/15374416.2013.817310.

[81] Leijten P., Raaijmakers M. A. J., Orobio de Castro B., van den Ban E. and Matthys W. Effectiveness of the Incredible Years Parenting Program for Families with Socioeconomically Disadvantaged and Ethnic Minority Backgrounds. Journal of Clinical Child & Adolescent Psychology. 2017;46(1):59-73. DOI: 10.1080/15374416.2015.1038823.

[82] Hutchings J., Bywater T., Daley D., Gardner F., Whitaker C., Jones K., et al. Parenting intervention in Sure Start services for children at risk of developing conduct disorder: pragmatic randomised controlled trial. BMJ. 2007;334(7595):678. DOI: 10.1136/bmj.39126.620799.55.

[83] Morpeth L., Blower S., Tobin K., Taylor R. S., Bywater T., Edwards R. T., et al. The effectiveness of the Incredible Years pre-school parenting programme in the United Kingdom: a pragmatic randomised controlled trial. Child Care in Practice. 2017;23(2):141-161. DOI: 10.1080/13575279.2016.1264366.

[84] Little M, Berry V, Morpeth L, Blower S, Axford N and Taylor R. The impact of three evidence-based programmes delivered in public systems in Birmingham, UK. Int J Conf Violence. 2012(6):260–272.

[85] Scott S., Webster-Stratton C., Spender Q., Doolan M., Jacobs B. and Aspland H. Multicentre controlled trial of parenting groups for childhood antisocial behaviour in clinical practice. BMJ. 2001;323(7306):194. DOI: 10.1136/bmj.323.7306.194.

[86] Hutchings J., Griffith N., Bywater T. and Williams M. E. Evaluating the Incredible Years Toddler Parenting Programme with parents of toddlers in disadvantaged (Flying Start) areas of Wales. Child: Care, Health and Development. 2017;43(1):104-113. DOI: https://doi.org/10.1111/cch.12415.

[87] Scott S, Sylva K, Kallitsoglou A and T. F. Which Type of Parenting Programme Best Improves Child Behaviour and Reading? Follow-Up of the Helping Children Achieve Trial. London: Nuffield Foundation. 2014.

[88] Scott S., Sylva K., Doolan M., Price J., Jacobs B., Crook C., et al. Randomised controlled trial of parent groups for child antisocial behaviour targeting multiple risk factors: the SPOKES project. Journal of Child Psychology and Psychiatry. 2010;51(1):48-57. DOI: https://doi.org/10.1111/j.1469-7610.2009.02127.x.

[89] Scott S., O'Connor T. G., Futh A., Matias C., Price J. and Doolan M. Impact of a parenting program in a high-risk, multi-ethnic community: the PALS trial. Journal of Child Psychology and Psychiatry. 2010;51(12):1331-1341. DOI: https://doi.org/10.1111/j.1469-7610.2010.02302.x.

[90] Beecham J. and Knapp M. Costing psychiatric interventions. In: Thornicroft, Graham, (ed.) Measuring Mental Health Needs. Gaskell, London, UK. 2001:pp. 200-224.

[91] Gaheer S. and Paull G. Evaluation of Children's Centres in England (ECCE) Strand 6. London: Department for Education; 2016.

[92] Wansink HJ, Hosman CMH and Verdoold CJ. Basiszorg, een handleiding: Preventieve zorgcoördinatie voor ouders met psychiatrische problemen. [Basic care, a manual: Preventive basic care manamgement for parent with psychiatricproblems.]. The Hague: Parnassia Bavo Group, Prevention Department. 2010.

[93] Curtis L. and Burns A. Unit Costs of Health and Social Care 2017. Canterbury: Personal Social Services Research Unit, University of Kent2017.

[94] Curtis L. Unit Costs of Health and Social Care 2014. Canterbury: University of Kent, PSSRU; 2014.

[95] NHS Improvement. Reference Costs. [cited 09/07/2021] 2018. Available from: https://improvement.nhs.uk/resources/reference-costs/.

[96] Department of Health (DH). NHS Reference Costs 2013–14. London: DH; 2014.

[97] institute for Medical Technology Assessment (iMTA). Trimbos/iMTA questionnaire for costs associated with psychiatric illness (TiC-P adults). 2012 [cited 09/07/2021]. Available from: https://www.imta.nl/questionnaires/.

[98] Bouwmans C., De Jong K., Timman R., Zijlstra-Vlasveld M., Van der Feltz-Cornelis C., Tan Swan S., et al. Feasibility, reliability and validity of a questionnaire on healthcare consumption and productivity loss in patients with a psychiatric disorder (TiC-P). BMC health services research. 2013;13:217. Epub 2013/06/19. DOI: 10.1186/1472-6963-13-217.

[99] institute for Medical Technology Assessment (iMTA). iMTA Questionnaire Intensive Youth Care. [cited 11/07/2021]. Available from: https://www.imta.nl/questionnaires/.

[100] Hakkaart-van Roijen L, Tan SS and Bouwmans CA. Handleiding voor kostenonderzoek. Methoden en referentieprijzen voor economische evaluaties in de gezondheidszorg. Geactualiseerde versie 2010. [Manual for studying costs. Methods and reference prices for economic evaluations in health care. Update 2010]. Diemen: College voor Zorgverzekeringen2011.

[101] Nederlands Jeugdinstituut. Normprijzenonderzoek jeugd & opvoedhulp Noord-Brabant, [Standardpricing research for youth care and parenting support Noord-Brabant.]. [cited 11/07/2021]. Available from: https://www.nji.nl/nl/Download-NJi/Publicatie-NJi/Normprijzen_jeugd_opvoedhulp_Brabant.pdf.

[102] NZa (Dutch Healthcare Authority). Prestatiebeschrijvingen en tarieven extramurale zorg 2012. [Standards and rates for outpatient care in 2012.]. In: Policy letter CA-300-4872012.

[103] Drost RMWA, Paulus ATG, Ruwaard D and Evers SMAA. Handleiding intersectorale kosten en baten van (preventieve) interventies: Classificatie, identificatie en kostprijzen [Manual on intersectoral costs and benefits of (preventive) interventions: Classification, identification and cost prices.]. Maastricht University, Department of Health Services Research: 2014 [cited 11/07/2021]. Available from: http://www.google.nl/url?sa=t&rct=j&q=&esrc=s&source=web&cd=2&ved=0CCsQFjAB&url=http%3A%2F%2Fwww.maastrichtuniversity.nl%2Fweb%2Ffile%3Fuuid%3D140bf3cc-a0e8-41ea-be66-545663a93f62%26owner%3D5b078ea8-4508-4eee-bb18-80a308782b25&ei=ox4AVeqnBMLe7AbWiYHICA&usg=AFQjCNE5yChWLMkNLW2nDm5teJ4yompQxg.

[104] Bradley R. H. and Caldwell B. M. Using the home inventory to assess the family environment. Pediatr Nurs. 1988;14(2):97-102. Epub 1988/03/01.

[105] Vedder P, Eldering L and Bradley RH. The home environments of at risk children in the Netherlands, Advances in family research. Amsterdam: Thesis Publishers, pp. 69–76 1995.

[106] Bonin E.-M., Stevens M., Beecham J., Byford S. and Parsonage M. Costs and longer-term savings of parenting programmes for the prevention of persistent conduct disorder: a modelling study. BMC Public Health. 2011;11(1):803. DOI: 10.1186/1471-2458-11-803.

[107] Aebi M., Giger J., Plattner B., Metzke C. W. and Steinhausen H.-C. Problem coping skills, psychosocial adversities and mental health problems in children and adolescents as predictors of criminal outcomes in young adulthood. European Child & Adolescent Psychiatry. 2014;23(5):283-293. DOI: 10.1007/s00787-013-0458-y.

[108] Metz D. and Jungbauer J. "My Scars Remain Forever": A Qualitative Study on Biographical Developments in Adult Children of Parents with Mental Illness. Clinical Social Work Journal. 2021;49(1):64-76. DOI: 10.1007/s10615-019-00722-2.

[109] Zheng X., Shangguan S., Fang Z. and Fang X. Early-life exposure to parental mental distress and adulthood depression among middle-aged and elderly Chinese. Economics & Human Biology. 2021;41:100994. DOI: https://doi.org/10.1016/j.ehb.2021.100994.

[110] Hosman C. M. H., van Doesum K. T. M. and van Santvoort F. Prevention of emotional problems and psychiatric risks in children of parents with a mental illness in the Netherlands: I. The scientific basis to a comprehensive approach. Australian e-Journal for the Advancement of Mental Health. 2009;8(3):250-263. DOI: 10.5172/jamh.8.3.250.

[111] Guerrero N., Wagner K. M., Gangnon R., Valdez C. R., Curtis M. A., Ehrenthal D. B., et al. Food Insecurity and Housing Instability Partially Mediate the Association Between Maternal Depression and Child Problem Behavior. The journal of primary prevention. 2020;41(3):245-259. DOI: 10.1007/s10935-020-00588-y.

[112] Katy Kaplan, Ph.D. „ Eugene Brusilovskiy, M.U.S.A. „ Amber M. O'Shea, Ph.D. , and Mark S. Salzer, Ph.D. Child Protective Service Disparities and Serious Mental Illnesses: Results From a National Survey. Psychiatric Services. 2019;70(3):202-208. DOI: 10.1176/appi.ps.201800277.

[113] Roscoe J. N., Lery B. and Chambers J. E. Understanding child protection decisions involving parents with mental illness and substance abuse. Child Abuse Negl. 2018;81:235-248. Epub 2018/05/18. DOI: 10.1016/j.chiabu.2018.05.005.

[114] Hartney E. and Barnard D. K. A framework for the prevention and mitigation of injury from family violence in children of parents with mental illness and substance use problems. Aggression and Violent Behavior. 2015;25:354-362. DOI: https://doi.org/10.1016/j.avb.2015.09.004.

[115] Afifi T. O., Fortier J., MacMillan H. L., Gonzalez A., Kimber M., Georgiades K., et al. Examining the relationships between parent experiences and youth self-reports of slapping/spanking: a population-based cross-sectional study. BMC Public Health. 2019;19(1):1345-1345. DOI: 10.1186/s12889-019-7729-6.

[116] Westad C. and McConnell D. Child welfare involvement of mothers with mental health issues. Community Ment Health J. 2012;48(1):29-37. Epub 2011/01/19. DOI: 10.1007/s10597-011-9374-0.

[117] Lane W. G., Dubowitz H., Frick K. D., Semiatin J. and Magder L. Cost effectiveness of SEEK: A primary care-based child maltreatment prevention model. Child Abuse Negl. 2021;111:104809. Epub 2020/11/19. DOI: 10.1016/j.chiabu.2020.104809.

[118] Pierce M., Abel K. M., Muwonge J., Jr., Wicks S., Nevriana A., Hope H., et al. Prevalence of parental mental illness and association with socioeconomic adversity among children in Sweden between 2006 and 2016: a population-based cohort study. The Lancet Public Health. 2020;5(11):e583-e591. DOI: 10.1016/S2468-2667(20)30202-4.

[119] Noonan K., Corman H. and Reichman N. E. Effects of maternal depression on family food insecurity. Economics & Human Biology. 2016;22:201-215. DOI: https://doi.org/10.1016/j.ehb.2016.04.004.

[120] Fekadu W., Mihiretu A., Craig T. K. J. and Fekadu A. Multidimensional impact of severe mental illness on family members: systematic review. BMJ Open. 2019;9(12):e032391. Epub 2020/01/02. DOI: 10.1136/bmjopen-2019-032391.

[121] Lippert A. M. and Lee B. A. Adult and Child Food Insecurity Among Homeless and Precariously-Housed Families at the Close of the Twentieth Century. Population Research and Policy Review. 2021;40(2):231-253. DOI: 10.1007/s11113-020-09577-9.

[122] Knapp M., King D., Healey A. and Thomas C. Economic outcomes in adulthood and their associations with antisocial conduct, attention deficit and anxiety problems in childhood. J Ment Health Policy Econ. 2011;14(3):137-147. Epub 2011/11/26.

[123] McGilloway S., NiMhaille G., Bywater T., Leckey Y., Kelly P., Furlong M., et al. Reducing child conduct disordered behaviour and improving parent mental health in disadvantaged families: a 12-month follow-up and cost analysis of a parenting intervention. Eur Child Adolesc Psychiatry. 2014;23(9):783-794. Epub 2014/09/04. DOI: 10.1007/s00787-013-0499-2.

[124] Lindsay G., Strand S. and Davis H. A comparison of the effectiveness of three parenting programmes in improving parenting skills, parent mental-well being and children's behaviour when implemented on a large scale in community settings in 18 English local authorities: the parenting early intervention pathfinder (PEIP). BMC Public Health. 2011;11:962-962. DOI: 10.1186/1471-2458-11-962.

[125] Jung J. and Cho S. Y. The Effects of Depression, Anxiety, and Parents' Support on Suicide Ideation and Attempts by Gender among Korean Adolescents. Journal of Child and Family Studies. 2020;29(5):1458-1466. DOI: 10.1007/s10826-020-01697-2.

[126] Fahrenkamp A. J., Darling K. E., Ruzicka E. B. and Sato A. F. Maternal Depressive Symptoms Mediate the Association between Socio-economic Status and Adolescent Weight Outcomes: A Longitudinal Analysis. Maternal and Child Health Journal. 2018;22(10):1462-1469. DOI: 10.1007/s10995-018-2541-y.

[127] Lam L. T. The Roles of Parent-and-Child Mental Health and Parental Internet Addiction in Adolescent Internet Addiction: Does a Parent-and-Child Gender Match Matter? Frontiers in public health. 2020;8:142-142. DOI: 10.3389/fpubh.2020.00142.

[128] Sahanapriya G. K., Bijulakshmi P., Rajendhiran G., Mathumathi S., Ramasubramanian V. and Kannan M. A Cross-Sectional Study on Internet Addiction Among Adolescent Children of Parents With Mental Illness. Indian Journal of Psychological Medicine. 2021:0253717621999761. DOI: 10.1177/0253717621999761.

[129] Howes Vallis E., MacKenzie L. E., Rempel S., Stephens M., Howard C., Drobinin V., et al. Visual memory in offspring of parents with mental illness. Psychiatry Research. 2020;286:112813. DOI: https://doi.org/10.1016/j.psychres.2020.112813.

[130] Renzaho A. M. N. and de Silva-Sanigorski A. The importance of family functioning, mental health and social and emotional well-being on child oral health. Child: Care, Health and Development. 2014;40(4):543-552. DOI: https://doi.org/10.1111/cch.12053.

[131] Padrón A., Galán I., García-Esquinas E., Fernández E., Ballbè M. and Rodríguez-Artalejo F. Exposure to secondhand smoke in the home and mental health in children: a population-based study. Tob Control. 2016;25(3):307-312. Epub 2015/03/27. DOI: 10.1136/tobaccocontrol-2014-052077.

[132] Nevriana A., Pierce M., Dalman C., Wicks S., Hasselberg M., Hope H., et al. Association between maternal and paternal mental illness and risk of injuries in children and adolescents: nationwide register based cohort study in Sweden. BMJ. 2020;369:m853. DOI: 10.1136/bmj.m853.

[133] Murphy G., Peters K., Wilkes L. M. and Jackson D. Adult Children of Parents with Mental Illness: Losing Oneself. Who am I? Issues in Mental Health Nursing. 2016;37(9):668-673. DOI: 10.1080/01612840.2016.1178359.

[134] Bosch A., Riebschleger J. and van Loon L. Dutch youth of parents with a mental illness reflect upon their feelings of guilt and shame. International Journal of Mental Health Promotion. 2017;19(3):159-172. DOI: 10.1080/14623730.2017.1315955.

[135] Bosch A., van de Ven M. and van Doesum K. Development and Validation of the Guilt and Shame Questionnaire for Adolescents of Parents with a Mental Illness (GSQ-APMI). Journal of Child and Family Studies. 2020;29(4):1147-1158. DOI: 10.1007/s10826-019-01671-7.

[136] Levy-Frank I., Hasson-Ohayon I., Kravetz S. and Roe D. Family psychoeducation and therapeutic alliance focused interventions for parents of a daughter or son with a severe mental illness. Psychiatry Research. 2011;189(2):173-179. DOI: https://doi.org/10.1016/j.psychres.2011.02.012.

[137] Tabak I., Zabłocka-Żytka L., Ryan P., Poma S. Z., Joronen K., Viganò G., et al. Needs, expectations and consequences for children growing up in a family where the parent has a mental illness. Int J Ment Health Nurs. 2016;25(4):319-329. Epub 2016/06/10. DOI: 10.1111/inm.12194.

[138] Stabile M. and Allin S. The economic costs of childhood disability. Future Child. 2012;22(1):65-96. Epub 2012/05/04. DOI: 10.1353/foc.2012.0008.

[139] Patrick P. M., Reupert A. E. and McLean L. A. A cross-sectional study on intergenerational parenting and attachment patterns in adult children of parents with mental illness. Child & Family Social Work. 2019;24(4):601-609. DOI: https://doi.org/10.1111/cfs.12641.

[140] Allen C. W., Diamond-Myrsten S. and Rollins L. K. School Absenteeism in Children and Adolescents. Am Fam Physician. 2018;98(12):738-744. Epub 2018/12/12.

[141] Homlong L., Rosvold E. O., Sagatun Å., Wentzel-Larsen T. and Haavet O. R. Living with mentally ill parents during adolescence: a risk factor for future welfare dependence? A longitudinal, population-based study. BMC Public Health. 2015;15(1):413. DOI: 10.1186/s12889-015-1734-1.

[142] Sruamsiri R., Mori Y. and Mahlich J. Productivity loss of caregivers of schizophrenia patients: a cross-sectional survey in Japan. Journal of Mental Health. 2018;27(6):583-587. DOI: 10.1080/09638237.2018.1466048.

[143] Dean K., Mortensen P. B., Stevens H., Murray R. M., Walsh E. and Agerbo E. Criminal conviction among offspring with parental history of mental disorder. Psychological Medicine. 2012;42(3):571-581. Epub 2011/08/16. DOI: 10.1017/S0033291711001395.

[144] LeCloux M., Maramaldi P., Thomas K. and Wharff E. Family Support and Mental Health Service Use Among Suicidal Adolescents. Journal of Child and Family Studies. 2016;25(8):2597-2606. DOI: 10.1007/s10826-016-0417-6.

[145] Turner E. A. and Liew J. Children's Adjustment and Child Mental Health Service Use: The Role of Parents' Attitudes and Personal Service Use in an Upper Middle Class Sample. Community Mental Health Journal. 2010;46(3):231-240. DOI: 10.1007/s10597-009-9221-8.

[146] Villatoro A. P., DuPont-Reyes M. J., Phelan J. C., Painter K. and Link B. G. Parental recognition of preadolescent mental health problems: Does stigma matter? Social Science & Medicine. 2018;216:88-96. DOI: https://doi.org/10.1016/j.socscimed.2018.09.040.

[147] Tapias E., Coromina M., Grases N. and Ochoa S. Psychological Treatments with Children of Parents with Mental Illness: A Systematic Review. Child & Youth Care Forum. 2021. DOI: 10.1007/s10566-021-09608-2.

[148] Bee P. Community preventive interventions for children of parents with serious mental illness: a commentary on the evidence base. Advances in Mental Health. 2015;13(2):165-167. DOI: 10.1080/18387357.2015.1066945.

[149] Neyt M., Devos C., Thiry N., Silversmit G., De Gendt C., Van Damme N., et al. Do innovative medicines against cancer always have a real added value? Health Technology Assessment (HTA). Brussels: Belgian Health Care Knowledge Centre (KCE), 2021.

[150] Heintz E., Gerber-Grote A., Ghabri S., Hamers F. F., Rupel V. P., Slabe-Erker R., et al. Is There a European View on Health Economic Evaluations? Results from a Synopsis of Methodological Guidelines Used in the EUnetHTA Partner Countries. PharmacoEconomics. 2016;34(1):59-76. DOI: 10.1007/s40273-015-0328-1.

[151] Rand L. Z. and Kesselheim A. S. Controversy Over Using Quality-Adjusted Life-Years In Cost-Effectiveness Analyses: A Systematic Literature Review. Health Aff (Millwood). 2021;40(9):1402-1410. Epub 2021/09/09. DOI: 10.1377/hlthaff.2021.00343.

[152] Knapp M., McDaid D. and Parsonage M. Mental Health Promotion and Mental Illness Prevention: The Economic Case2011.

[153] Hatton C., Emerson E., Robertson J. and Baines S. The mental health of adolescents with and without mild/moderate intellectual disabilities in England: Secondary analysis of a longitudinal cohort study. J Appl Res Intellect Disabil. 2018;31(5):768-777. Epub 2017/11/25. DOI: 10.1111/jar.12428.

[154] Ayton D. and Joss N. Empowering vulnerable parents through a family mentoring program. Australian Journal of Primary Health. 2015;22. DOI: 10.1071/PY14174.

[155] Plass-Christl A., Ravens-Sieberer U., Hölling H. and Otto C. Trajectories of health-related quality of life in children of parents with mental health problems: results of the BELLA study. Quality of Life Research. 2021. DOI: 10.1007/s11136-021-02783-8.

[156] Im S. and Heo M. L. Children in the Blind Spot of Social Medical Concerns: Supporting Children with Mentally Ill Parents in Korea. Community Mental Health Journal. 2021. DOI: 10.1007/s10597-020-00772-8.

[157] Klasen F., Meyrose A. K., Otto C., Reiss F. and Ravens-Sieberer U. Psychische Auffälligkeiten von Kindern und Jugendlichen in Deutschland. Monatsschrift Kinderheilkunde. 2017;165(5):402-407. DOI: 10.1007/s00112-017-0270-8.

[158] Wagner G., Zeiler M., Waldherr K., Philipp J., Truttmann S., Dür W., et al. Mental health problems in Austrian adolescents: a nationwide, two-stage epidemiological study applying DSM-5 criteria. Eur Child Adolesc Psychiatry. 2017;26(12):1483-1499. Epub 2017/05/26. DOI: 10.1007/s00787-017-0999-6.

[159] Zechmeister-Koss I., Goodyear M., Tüchler H. and Paul J. L. Supporting children who have a parent with a mental illness in Tyrol: a situational analysis for informing co-development and implementation of practice changes. BMC health services research. 2020;20(1):326. DOI: 10.1186/s12913-020-05184-8.

[160] Wansink H. J., Janssens J. M., Hoencamp E., Middelkoop B. J. and Hosman C. M. Effects of preventive family service coordination for parents with mental illnesses and their children, a RCT. Fam Syst Health. 2015;33(2):110-119. Epub 2015/03/10. DOI: 10.1037/fsh0000105.

[161] Curtis L and A N. Unit Costs of Health and Social Care 2004. Canterbury: University of Kent, PSSRU; 2005.

[162] Rutter J. Childcare Costs Survey 2015. London: Family and Childcare Trust; 2016.

Appendix

Extraction tables

Table A-1: Study and programme characteristics

Authors (Year)	Day et al. (2020) [65]	Gardener et al. (2017) [64]	Wansink et al. (2016) [22]
Programme name (Intervention)	Helping Families Programme-Modified (HFP-M)	Incredible Years® (IY) basic parenting programme[58, 59]	Preventive basic care management (PBCM)
Country	UK	UK, NL, IRE, NOR, SWE, POR[60]	NL (western urban area)
Comparator	Usual care including the Being a Parent session (CAU)	Waiting list (WL), minimal intervention (MI), or care as usual (CAU)	Care as usual (CAU)
Follow-up (months), data collection	~10 months[61], data collection at baseline (T1), after four months (T2), and after 10 months (T3) from baseline; because of missing data at T3, cost-utility results are based on two separate analyses[62]	Clinical follow-up/Duration randomisation to end of intervention: five months (10 trials) [74-85], five to eight (one trials) [87], eight months (two trials) [88, 89], 12 months (one trial) [86] Follow-up of economic studies[63]: six months [81, 83-85, 87, 89]	18 months, data collection at baseline (T0), after nine months (T1), and after 18 months (T2)[64] from baseline

[58] Although the pooled study focuses on (primary, secondary, and tertiary) prevention of child disruptive behaviour and not explicit on children who have a parent with a mental illness, we included the study, because it has also a focus on family mental health and parental mental health and functioning. Of the 1799 families included, 1131 parents in 11 out of 14 trials had depressive symptoms, 502 parents in five trials experienced parenting stress, 384 parents in four trials had feelings of self-efficacy (all numbers post-test). Seven out of 14 trials reported on depressive symptoms in parents, six out of 14 reported on parenting stress, and six out of 14 reported on feelings of self-efficacy. Furthermore, all seven studies reporting on depressive symptoms used the BDI to as a measure to assess parental depressive symptoms. Other studies used reduced instruments such as Brief Symptom Inventory – depression subscale, Symptoms checklist – depression subscale, the Depression Anxiety Stress Scale, or the General Health Questionnaire, that have only limited explanatory power for a broad spectrum of mental health issues. With regard to the trials included in the economic evaluation [81, 83-85, 87, 89], three out of five trials reported on depressive symptoms in parents, two out of five reported on parenting stress, and two out of five reported on feelings of self-efficacy.

[59] For the economic analysis, data from only five trials were merged [81, 83-85, 87, 89]. The use of this limited set of the trials is because trials are from different countries and health care systems. Only relevant trials from the UK were pooled and evaluated from a health economic perspective. Hence, there are some notable differences between the full analysis sample and the sample for economic analysis with regard to the data set. The following extraction tables illustrate mainly the data on pooled health economic evaluations.

[60] The health economic evaluation only included the studies conducted in the UK.

[61] The feasibility randomised control trial comprised of three phases: 1) Screening and intervention development: nine months, 2) Pre-trial feasibility study: 13 months, and 3) the trial itself: 23 months

[62] Analysis 1: Sample with CSRI and intervention costs at T2, and EQ-5D at T1 and T2 (more participants at these two time points), Analysis 2: 'complete case' analysis, CSRI and intervention costs at T2 and T3, and EQ-5D at T1, T2, and T3 (reduced sample size: results have to be treated with caution)

[63] Trial numbers in the HTA report: 4 [79], 7 [82], 9 [83, 84], 10 [87], 12 [88]

[64] Recruitment took place between September 2010 and April 2012; the last follow-up was between March 2012 and November 2013.

Authors (Year)	Day et al. (2020) [65]	Gardener et al. (2017) [64]	Wansink et al. (2016) [22]
Research design for effectiveness evaluation	Two arm, parallel Feasibility RCT in the course of a report with a pragmatic, mixed-methods design to develop and pilot HFP-M, randomisation: 1:1 ratio in HFP-M and CAU; multicentre study (two centres)	Pooled individual level-data from 14 RCTs in Europe, randomisation: 1:1 ratio (five trials), 1:1:1 ratio (one trial), 2:1 (eight trials), randomisation unit: individual (12 trials), cluster (two trials), in eight trials the randomisation ratio changed during the trial[65]; for the EE/analysis of service use and costs: number and percentage of trial participants reporting use of services by randomisation group at baseline and follow-up for the pooled sample	RCT, randomisation: 50:50 (1:1 ratio)
Underlying clinical study and publication type	Health technology assessment [65]	Health technology assessment [64] with pooled individual level-data from 14 IY RCTs in Europe [74-89]	Journal article [160]
Reporting of sources of funding/competing interest	Yes/Yes[66]	Yes/Yes[67]	Yes/Yes[68]

CAU…Care as usual, CSRI…Client service receipt inventory, HFP-M…Helping Families Programme-Modified, IY…Incredible Years, MI…Minimal intervention, NL…Netherlands, PBCM…Preventive basic care management, RCT…Randomised controlled trial, UK…United Kingdom, WL…Waiting list

[65] One of the 14 RCTs had three intervention arms [88]. The rest had two intervention arms each. Two trials had a minimal intervention control, 10 trials had a waiting list control, and two other RCTs had CAU. One trial used the IY – toddler version of the IY programme, three trials the IY basic parenting programme combined with a literacy intervention, and ten trials used the IY basic parenting programme.

[66] The National Institute for Health Research (NIHR) Health Technology Assessment funded this project. One author (Day) is the lead developer and another (Harris) is the co-developer of the parenting programme. One further author (McMurran) also contributed with previous work to the HFP-M. Another author (Crawford) received research grant funding from the NIHR. Another author (Moran) reports personal fees from a talk and led the development of the Standardised Assessment of Personality – Abbreviated Scale used in the study.

[67] The NIHR funded the programme. One author (Hutchings) was principal investigator in two included trials. Furthermore, she is a certified trainer for the Incredible Years® and receives personal fees from the Incredible Years® company. A second author (Landau) reports grants from the NIHR during the conduct of the study. Another three authors (Scott, Gardner, Leijten) were involved in trials included in this pooled study.

[68] The underlying RCT was supported by the Dutch Organisation for Health Research and Development (ZonMw), The Hague, Grant 157003002.
The economic study was supported by Grant 200400010 from ZonMw. All authors declared that they have no competing interests.

Table A-2: Population characteristics

Authors (Year)	Day et al. (2020) [65]		Gardener et al. (2017) [64]		Wansink et al. (2016) [22]	
I/C	I	C	I	C	I	C
Sample size	n=24[69] participants/families	n=24 participants/families	n=372 children/families[70]	n=236 children/families	n=49[71] families	n=50 families
Family structure	Single parent[72]: 14 (58.3) Female parent: 24 (100%) Number of children at home, median (range): 2 (1-5)	Single parent: 17 (77.3) Female parent: 23 (95.8%) Number of children at home, median (range): 2 (1-5)	Single parent: 135[73] (38%)	Single parent: 74 (33%)	Single mother: 28 (57%) Two parent family: 21[74] – Mother/Father: 15 (31%)/2 (4%) Mother and Father: 4 (8%) Number of children, mean (sd): 2.10 (0.98)	Single mother: 18 (36%) Two parent family: 32 – Mother/Father: 26 (52%)/2 (4%) Mother and Father: 4 (8%) Number of children, mean (sd): 2.16 (1.02)
Age children and age parent	Index child, mean (sd): 7.7 (2) Parent, mean (sd): 34.7 (7.5)	Index child, mean (sd): 7.9 (2.2) Parent, mean (sd): 35 (6.9)	Child (in months), mean (sd): 56.17 (16.63) Parent: NA	Child (in months), mean (sd): 59.23 (16.63) Parent: NA	Index child, mean (sd): 6.53 (2.19) Age distribution: 0-3y: 27 4-12y: 61 13-20y: 13 Parent: NA	Index child, mean (sd): 5.64 (1.76) Age distribution: 0-3y: 35 4-12y: 63 13-20y: 9 Parent: NA
Gender children	Gender (male) index child: 12 (50%)	Gender (male) index child: 14 (60.9%)	Gender (male) child: 224[75] (60%)	Gender (male) child: 149 (63%)	Gender (male) index child: 25 (51%)	Gender (male) index child: 30 (60%)

[69] Centre 1: n=36, Centre 2: n=12. In total 36 participants (n=21 participants in HFP, n=15 participants in CAU) accepted the interventions at T1 (baseline), at T2 30 participants (n=18 participants in HFP, n=12 participants in CAU) were still enrolled, and at T3 19 participants (n=13 participants in HFP, n=6 participants in CAU) remained.

[70] The studies' pooled sample comprised of 1,799 families in total. The economic analysis' sample consisted of 608 participants (families) in total. Data on certain demographic characteristics was not fully available for the whole sample. For the demographic characteristics, authors reported always the total sample size of families with available data including the mean, standard deviation or the proportion in percentage (%) of the particular characteristic. Number of families with a certain characteristic are based on own calculations and are not necessarily representative for the whole sample, e.g. for the low-income characteristic of the main intervention (IY), data on 359 families was available, 62% or 223 (=359*0.62) families were low-income families.

[71] Eligible and interested families: 256; dropouts were low in both arms (lost to follow-up) n=4 (PBCM), n=3 (CAU), 22 families (44%) of the 50 in the CAU arm used the consultation service with the team and two were referred to the PBCM group. In total n=38 families (77%) actually received PBCM. At T1 86 files and at T2 88 files were available. A total sum of 82 families (83%) had complete datasets for the outcome.

[72] Data on certain demographic characteristics was not fully available for the whole sample. Hence, the percentage numbers in parentheses do not relate to the full sample.

[73] The number of 'single' parents was derived from the available data on the number of total parents, e.g. 38% from 355 parents of the IY sample, hence for IY we have n=135=355 * 0.38. The available number of individuals with demographic characteristics (gender child, single parent, low income, unemployed, ethnic minority) need not correspond to the 'full economic' sample population.

[74] In some families, both parents experience mental illnesses. The number of two parent families in the intervention arm is the sum of 15+2+4=21 (the same goes for the comparator arm).

Authors (Year)	Day et al. (2020) [65]		Gardener et al. (2017) [64]		Wansink et al. (2016) [22]	
Other characteristics	Received psychiatric diagnosis, n (%): 16 (88.9%)	Received psychiatric diagnosis, n (%): 19 (81.8%)	BDI parents, mean (sd): 13.48 (10.37) Low income, n (%): 223 (62%) Unemployed, n (%): 128 (43%) Ethnic minority, n (%): 74 (20%)	BDI parents, mean: 11 (10.37) Low income, n (%): 166 (72%) Unemployed, n (%): 70 (36%) Ethnic minority, n (%): 42 (18%)	Diagnosis Mothers/Fathers, n (%): Depressive/anxiety disorders: 36 (77%)/4 (67%) Other Axis I disorders: 8 (17%)/2 (33%) Personality disorders: 3 (7%)/ Ethnic minority, n (%): 39 (80%)	Diagnosis Mothers/Fathers, n (%): Depressive/anxiety disorders: 36 (75%)/4 (67%) Other Axis I disorders: 9 (19%)/2 (33%) Personality disorders: 3 (6%)/- Ethnic minority, n (%): 27 (54%)
Eligibility criteria	■ Inclusion criteria: Parent (aged 18-65 years) having severe personality difficulties (DSM-4 and Standardised Assessment of Personality – Abbreviated Scale score of ≥3), being proficient in English, and having capacity to consent; Index child (aged 3-11 years[76]), living at home with index parent, having mental health issues (Development and Well-Being Assessment/Pre-School Age Psychiatric Assessment; Strengths and Difficulties Questionnaire Total Difficulties score of ≥17] ■ Exclusion criteria: Parent with presence of psychosis, being currently engaged in structured psychotherapy and/or another parenting intervention, receiving inpatient care and/or language/cognitive difficulties affecting consent, and participation in research procedures; Index child with presence of a neurodevelopmental disorder, not residing with index parent and/or currently being considered for/subject to local authority care/supervision proceedings.		■ Inclusion criteria[77]: all completed RCTs of the IY programme (or IY in combination with a literacy intervention) in Europe for children aged 1-12 years[78], no restrictions on year, follow-up, outcome measures ■ Exclusion criteria: Non-RCTs, trials with additional non-parenting programme, trials with programmes much more minimal than the IY programme (<12-14 sessions)		■ Inclusion criteria: Outpatients treated for a psychiatric disorder, being a caregiver for a child aged between three and 10 years of age (age range 1-10 years[79]), the parents being interested in PBCM, and the family being exposed to three or more of a list of sixteen risk factors for poor parenting[80] ■ Exclusion criteria: Children with a mental health diagnosis (e.g. ADHD, or conduct disorder), expected duration of less than three months for further therapy, living outside the catchment areas, and previous help utilising PBCM	

BDI…Beck depression index, C…Control intervention/comparator, CAU…Care as usual, HFP-M…Helping Families Programme-Modified, I…Intervention, IY…Incredible Years, MI…Minimal intervention, NA…not available, PBCM…Preventive basic care management, RCT…Randomised controlled trial, sd…standard deviation, UK…United Kingdom, WL…Waiting list

[75] The number of children was derived from the number given by the child gender number, e.g. 60% or in absolute numbers 372 of children of the IY sample were of male gender of single parents, hence for IY we have n=935=355 * (1/0.38).

[76] Although the eligibility criteria for the present report includes only children between the age 4-18, this study with the age range of 3-11 years was still included as it overlaps with the eligible age range.

[77] Because the present study is a pooled RCT analysis, inclusion and exclusion criteria are restricted to trial characteristics and not necessarily to population characteristics. However, trials investigating the IY parenting programme have similar inclusion and exclusion criteria with regard to the population. Henceforth, within this present report it is assumed that inclusion and exclusion criteria match across studies.

[78] Although the eligibility criteria for the present report includes only children between the age 4-18, this study with the age range of 1-12 years was still included as it overlaps with the eligible age range.

[79] Although the eligibility criteria for the present report includes only children between the age 4-18, this study with the age range of 1-10 years was still included as it overlaps with the eligible age range.

[80] Risk factors: single parenthood, little support from spouse, little network support, relational problems, partner with mental health problems, children with poor health/handicaps/difficult temperament, changes in family structure/housing, two or more life events in the past two years, housing problems, poverty or debts, parents having been abused as a child, severe psychiatric symptoms, low compliance with psychiatric treatment, impulse control problems, alcohol or drug problems, low intelligence

Table A-3: Specific programme and intervention characteristics

Authors (Year)	Day et al. (2020) [65]	Gardener et al. (2017) [64]	Wansink et al. (2016) [22]
Programme name (abbrev.) and underlying concept	HFP-M, Multiple Determinants of Parenting (MDP) conceptual model that specifies how the interaction between child characteristics and parenting is influenced by the multiple impacts of parents' personality, couple relations, family and social networks and work experiences	IY, content of the programme is derived from social learning and attachment theory and comprises of the following topics: relationship-building, providing praise and rewards as reinforcement of positive behaviour, effective limit-setting, adequate disciplining techniques, and coaching children in social, emotional and academic skills	PBCM, family-focused strength-oriented rehabilitation model, focussing on strengthening positive parenting and providing community and network support
Programme aims	HFP-M aims to identify the ways that parent personality traits and functioning impact individual parenting and impact the index child's development, identify mutually agreed goals for parenting change, use a range of evidence-based parenting methods to improve parenting, child mental health problems and parent-child relationships, use a range of evidence-based strategies to improve parental emotion regulation and coping, improve parents' social resources	In the course of the IY programme, techniques parents learn are designed to break coercive cycles of parent-child interaction in which parents and children reinforce negative and aggressive behaviour in each other. IY focuses on socioeconomically disadvantaged families. The group format of IY further allows for normalisation and social support.	Preventive programme targeting and supporting effective parenting, promoting socio-emotional development of children, and reducing risk of developing behavioural problems.
Subtask, components, and involved professions	■ Face-to-Face contact with professionals: primary and secondary care (GP, psychiatrist, psychologist, drug and alcohol advisor, home treatment/ crisis team, assertive outreach team, early intervention team, social worker, mental health nurse, occupational therapist, accident and emergency service) ■ Day-care services: Drug/alcohol service, community mental health centre, day-care centre/hospital, drop-in centre, self-help/support group, class/ group at a leisure centre, adult education class, other day-care activities ■ Inpatient admission/care ■ Child section services: School nurse, health visitor, dentist, GP, paediatrician, optician, child development centre, child and adolescent mental health services, speech therapy out of school, hearing specialist ■ Counselling, support, and other services: Family therapist, individual therapy, home help/care worker, social services nursery school place, after-school club, prescribed medication, police contacts	■ Hospital care: accident and emergency (A&E) department, ambulance, outpatient service, inpatient stay, other hospital services ■ Community health care: GP, GP nurse, health visitor, speech and language therapies, other community health care services ■ Mental health care: Child and Adolescent Mental Health Services (CAMHS), other MH services ■ Social care: social worker, other social care services ■ Accommodation: Child placements (foster care and children's homes) ■ Voluntary services/sector: voluntary sector support, self-help	■ Health care/services[81]: primary and secondary care (mental health care, GP, paramedical services, youth care (care agencies), preventive family support; ■ Child care services: Day care (professional childcare), babysitter (informal childcare); ■ Inter-sectoral services: educational sector services (special education), criminal justice sector services (court proceedings, police services), debt restructuring services
Other characteristics	HFP-M is intended to augment rather than replace CAU and uses a relational, goal-orientated helping process (e.g. reduction of parental alienation and stigma, assess and strengthen parent-child relationship, child's emotional and behavioural difficulties, and manage wider family and life circumstances, generate hope, and encourage parents' use of learnt skills in daily life etc.).	In the IY programme, parents not therapists are seen as the experts on their own children. Parents are guided to set weekly goals, which fit with their cultural and personal needs and values. Moreover, as opposed to the therapist 'talking' about the kind of parenting behaviour that is considered to be appropriate, video-taped scenes showing examples of parent–child interactions are central in the sessions and parents are guided to identify key parenting behaviours or principles that might be useful for their own family context.	Five steps in the intervention: 1.) the enrolment procedures (referral by parent's therapist), 2.) Systematic assessment of strengths and vulnerabilities, 3.) Design of an integrated preventive plan/tailored preventive care, 4.) Linking families to and coordinating services for childcare/children clubs/community health services, services for debt restructuring and financial resources, 5.) Monitoring of the implementation/evaluation of effects in regular meetings with parents and children.

C…Controll intervention/comparator, CAU…Care as usual, GP…General practitioner, HFP-M…Helping Families Programme-Modified, I…Intervention, IY…Incredible Years, MI…Minimal intervention, NA…not available, PBCM…Preventive basic care management, RCT…Randomised controlled trial, WL…Waiting list

[81] Full list of services, professions, and facilities for 1) Health care: mental health care (primary mental health care, community mental health services, psychiatric clinics); other primary health care (GP, paramedical services, logopedics, dietician, health and safety service, social welfare work), alternative medicine; other secondary care (somatic hospitals, emergency room, revalidations centres, specialised clinics for obesity, specialised burns department); Youth care/preventive family support services (youth and family centres, domestic services, preventive home-based family care, preventive orthopedagogical services, parenting classes and parenting education, preventive mental health education for children and parents, family coaches of Youth and family centres, home-based family support by non-professionals); Youth care/specialised youth care services (semi-residential and residential care, foster care and secure care, child protection and probation services, youth care agencies, intensive ambulatory home-based specialised support for multi-problem families, care for youth with mental and/or cognitive disabilities), for 2) Child care: informal child care (child care given by non-professionals, i.e. babysitter, granny); professional child care (child care services, such as 'kindergarten'), and 3) other sectors: educational sector (school attendance officer, interne special education teacher, specials education, specialised educational services); criminal/justice sector (lawyers, police, court); Debts restructuring services.

Table A-4: Evaluation characteristics of the studies and programmes

Authors and Year	Day et al. (2020) [65]	Gardener et al. (2017) [64]	Wansink et al. (2016) [22]
Type of health economic or impact analysis	Cost-utility analysis (piggy-backed[82])	Cost-effectiveness analysis and economic modelling of long-term savings	Cost-effectiveness analysis (piggy-backed)
Perspective	UK NHS/Personal Social Services perspective and societal (SCT) perspective	Public sector perspective	Health care (HC) perspective, social care (SC) perspective, and societal (SCT) perspective[83]
Data preparation, analysis and handling uncertainty	General: ■ Imputation of missing outcomes was not applied; the original analysis used multilevel models to estimate the likely range of intervention effects, because of a small number of follow-ups at T3 ■ the analysis could not control for baseline costs ■ QALY gains were calculated using area under the curve methods with linear interpolation between scores, controlling for baseline utility ■ if both costs and outcomes were higher for the intervention than the comparator group, then the ICER was calculated ■ ICERs: non-parametric bootstrapping using 1000 Cost-QALY combinations ■ CEACs and CE planes were illustrated ■ Explicit WTP threshold of £ 30,000 per QALY, but probabilities for CE was also calculated for other thresholds Sensitivity analysis: ■ not applied and no control for baseline costs	General: ■ Data from five trials were merged; cost analyses (differences in total costs) used complete cases (CC) and included participants only when they had completed CSRI at baseline and follow-up; service contacts were imputed when exact number of contacts were missing (mean of service contacts) ■ Skewed distribution of cost data, the hierarchical structure of the data and potential clustering by cluster and strata were accounted (p-values derived by using a clustered regression model) ■ Cost-effectiveness analysis: Participants additionally needed a completed ECBI-I assessment; focus on the children's use of services and support; the probability that the intervention is cost-effective was assessed by ICERs and CEACs ■ ICERs: 10,000 bootstrap replications of the treatment effect were generated for each cost-outcome combination ■ No explicit threshold available for ECBI-I outcome ■ Economic modelling of long-term savings from the IY intervention was conducted ■ Cost variations at follow-up adjusted for baseline costs and other demographic covariates (model 1) and final model (model 2) with child age, gender and treatment condition covariates via linear regression estimation Sensitivity analysis: ■ Subgroup analyses by gender, baseline ECBI score (<134 vs ≥134), child age (<5 years vs ≥5 years), and parental depression at baseline (BDI score ≥20 vs <20) for cost-effectiveness, but no separate sensitivity analysis[84] ■ Two models of cost variation (model 1 and model 2)	General: ■ Missing outcome values were analysed and imputed (<5%), no outliers were found, missing costing data was imputed and measured costs were extrapolated, descriptive statistics, t- and chi-squared-tests were used ■ Calculation of absolute costs and change in HOME scores of the base case scenario for all three perspectives ■ ICERs were calculated for all perspectives and presented in CE planes and CEACs ■ ICERs: Median of the 5000 bootstrap replications ■ Wide range of WTP thresholds ■ Subgroup analysis on the population that actually received PBCM was conducted Sensitivity analysis: ■ Cost outliers (e.g. high cost families) excluded ■ Only complete cases without imputed data were analysed ■ ICERs were calculated based on adjustment of baseline cost differences of interventions (mean differences)

BDI…Beck depression index, CE…Cost-effectiveness, CEAC…Cost-effectiveness acceptability curves, CSRI…Client service receipt inventory, ECBI…Eyberg child behaviour inventory, HC…Health care perspective, HFP-M…Helping Families Programme-Modified, ICER…Incremental cost-effectiveness ratio, IY…Incredible Years, PBCM…Preventive basic care management, QALY...Quality-adjusted life year, SC…Social care perspective, SCT…Societal perspective , WTP…Willingness-to-pay

[82] An economic piggy-back analysis is conducted onto a clinical effectiveness study (i.e. RCTs or observational study), whereas a model-based economic evaluation uses data from a wide range of resources (RCTs, observational studies, trial based economic evaluations, other literature and reports).

[83] The healthcare perspective includes costs for health and child/family support services, social care perspective additionally includes costs for childcare, and the societal perspective includes all measured use of services including ICBs within the educational sector, the criminal justice system, and services for debt restructuring.

[84] We strictly distinguish between sensitivity analyses and subgroup analyses. The latter estimates are produced for each subgroup in order to make a comparison across subgroups. In sensitivity analyses, informal comparisons are applied between different estimation approaches of the same thing [68].

Table A-5: Cost categories, parameters, unit costs, and instruments including inter-sectoral cost parameters

Authors (Year)	Cost sector or type	Cost parameter (task and activity)	Unit or mean costs (sd)		Unit				Cost instrument, tariffs and [Source]	Currency, reference year and discount rate
Intervention/Control			I	C		I	C			
Day et al. (2020) [65]	Face-to-face contact	General practitioner[85]	£ 431.9 (444.8)	£ 444.8 (744.7)	per participant per	9.6 (13.9)	8.9 (12.7)	contacts/ days	modified Client Service Receipt Inventory (CSRI): includes primary, secondary health care, social care, school-related services, early years help, and youth and criminal justice services, time spent by parents, days off work, and days out of school [90], Personal Social Services Research Unit (PSSRU) unit costs of health and social care [93], NHS Improvement 2016-2017 reference cost data [95]	GBP, 2017, not applied because of short time period (10 months)
		Psychiatrist	£ 220.8 (588.8)	£ 636 (1,867.8)		7.4 (9.6)	26.5 (36.1)	-		
		Other medical professional	£ 72.2 (165.7)	£ 293.3 (654.8)		3.5 (3)	8.5 (10.6)			
		Psychologist	£ 497.6 (1,155.9)	£ 306.2 (918.7)		26.6 (31.2)	52 (0)			
		Drug and alcohol advisor	£ 140 (447.7)	-		10 (7.1)	-			
		Other counsellor/therapist	£ 233.80 (550.5)	£ 235.60 (467.4)		25 (9.5)	24 (0)			
		Home treatment/crisis team member	£ 66.7 (188.60)	£ 0.4 (1.1)		27.5 (17.7)	1 (0)			
		Assertive outreach team member	£ 11.2 (46.1)	-		5 (0)	-			
		Early intervention team member	£ 59.2 (132.7)	-		5.7 (3.7)	-			
		Social worker	£ 395.3 (705.6)	£ 140.9 (251.4)		14 (14.5)	9.3 (8.9)			
		Mental health nurse	£ 51.8 (213.4)	£ 29.3 (88)		20 (0)	6 (0)			
		Occupational therapist	£ 5.3 (21.8)	£ 30 (90)		2 (0)	8 (0)			
		Accident and emergency service	£ 69.60 (157.8)	£ 16.40 (49.3)		2 (1.4)	1 (0)			
	Day-care services	Drug/alcohol service	£ 77.6 (220.2)	-		5.5 (0.7)	-			
		Day-care centre/day hospital	£ 854.4 (3,498.4)	£ 680.3 (2,041)		185.5 (260.9)	157 (0)			
		Drop-in centre	£ 59.6 (236.1)	-		13 (17)	-			
		Self-help/support group	£ 59.6 (153)	-		8.7 (5.5)	-			
		Class/group at a leisure centre	£ 44 (181.40)	-		17 (0)	17 (0)			
		Adult education class	£ 224.80 (528.70)	£ 73.70 (221)		24.5 (19.2)	17 (0)			
		Other day-care activity	£ 55.10 (227)	£ 73.70 (221)		24 (0)	17 (0)			
	Inpatient admissions	Days as an inpatient	£ 332.50 (1,370.80)	£ 907.10 (2,721.30)		9 (0)	13 (0)			
	Prescribed medications	Receipt of prescription	£ 136.40 (328.40)	£ 413.20 (355.60)		-	-			
	Employment	Days off work (owing to health problems)	£ 30.20 (85.20)	£ 56.90 (118.80)		1.3 (1.2)	4 (1.4)			

[85] The authors did not list specific information on unit costs for specific services. Instead, authors estimated mean costs and estimated mean utilization of services for T2 and T3. In Table A-5 mean (unit) costs are depicted only for time point T2, because findings that are more robust in the study are those reported at T2.

Authors (Year)	Cost sector or type	Cost parameter (task and activity)	Unit or mean costs (sd)		Unit		Cost instrument, tariffs and [Source]	Currency, reference year and discount rate
Day et al. (2020) [65] *(continuation)*	Child section services	School nurse	£ 736.30 (2,138.20)	£ 32.80 (59.80)	23.6 (48.3)	1.7 (1.2)		
		Health visitor	£ 6.90 (19.60)	-	1 (0)	-		
		Dentist	£ 77.40 (102.80)	£ 195.10 (406.80)	1.5 (0.5)	3.5 (4.4)		
		General practitioner	£ 134.10 (112.80)	£ 93.30 (115.30)	4.1 (2.4)	2.8 (1.6)		
		Paediatrician	£ 77.70 (165.60)	£ 66.10 (99.10)	1.7 (0.9)	1 (0)		
		Optician	£ 63.70 (66.90)	£ 48.10 (57.10)	1.1 (0.3)	1 (0)		
		Child development centre	£ 149.60 (422.40)	-	24 (0)	-		
		Child and adolescent mental health services	£ 147.90 (290.80)	£ 144.20 (332.60)	5.3 (4)	5.7 (7.2)		
		Speech therapy out of school	£ 11.50 (32.50)	-	1 (0)	-		
		Hearing specialist	£ 9.80 (33.30)	-	1 (0)	-		
	Counselling	Family therapist	£ 25.9 (106.7)	£ 19.6 (58.7)	5 (0)	2 (0)		
		Individual therapy	£ 227.8 (851)	£ 123.90 (339.8)	15.7 (21.2)	7.5 (9.2)		
		Other counselling	£ 38.8 (86.4)	£ 756.1 (1,458.6)	1 (0)	34 (14)		
	Support	Home help/care worker	£ 85 (232.7)	£ 583.3 (1,750)	15.3 (15.5)	30 (0)		
		Day-care centre	£ 52.6 (216.8)	-	24 (0)	-		
		Social services nursery school place	£ 3.5 (14.3)	-	1 (0)	-		
		After-school club	NA	NA	78.8 (78.2)	13.8 (13.7)		
		Other service	NA	-	29 (15.6)	-		
	Hospital services	Hospital services/Contacts	£ 80.6 (195.1)	£ 1,873.3 (5,115.9)	1 (0)	46 (45.3)		
Gardener et al. (2017) [64]			Unit costs reported for both intervention or comparator		per 'unit'/per duration/per hour[86]		Client Service Receipt Inventory (CSRI) [90]; PSSRU, 2014 [94]	GBP, 2016, for CEA not applied because of short time period (six months), for modelling 3.5% discount rate
		GP surgery/clinic	£ 38	-	-			
		GP home/other	£ 76	£ 23.4	-			
		Nurse/GP nurse surgery/clinic	£ 17	£ 19.75	£ 50.5			

[86] Unit costs represent the total expenditure incurred to produce one unit of output. The unit in per 'unit' is meant as defined in the Unit Costs of Health and Social Care by the PSSRU (e.g. in the case of GP surgery/clinic, it is meant per patient contact lasting 11.7 minutes excluding travel). Per duration means that costs are standardised across trials with regard to per average duration of the service – a 'typical' duration for each service. Structural differences across trials sufficed to harmonise service duration.

Authors (Year)	Cost sector or type	Cost parameter (task and activity)	Unit or mean costs (sd)			Unit	Cost instrument, tariffs and [Source]	Currency, reference year and discount rate
Gardener et al. (2017) [64] *(continuation)*		Nurse other	£ 27	£ 31.75	£ 50.5			
		Health visitor surgery/clinic	£ 26	£ 24	£ 65			
		Health visitor other	£ 54	£ 50	£ 65			
		Hearing problems/audiologist	£ 16	£ 30	£ 32			
		Speech and language therapist surgery	£ 24	£ 45	£ 32			
		Speech and language other	£ 30	£ 57	£ 32			
		Other primary	£ 16	£ 30	£ 32			
		Physiotherapist surgery	£ 24	£ 45	£ 32			
		Physiotherapist other	£ 30	£ 57	£ 32			
		Community paediatrician	£ 310	-	-			
		Social worker	£ 55	£ 60	£ 55			
		Sessional worker	£ 50	£ 60	£ 50			
		Home help/home care worker	£ 24	£ 60	£ 24			
		CAMHS	£ 236	-	-		NHS Reference Costs 2013-14 [96]	
		Child guidance centre/psychiatric worker	£ 22	£ 90	£ 14		Evaluation of Children's Centres in England (ECCE) Strand 6 [91]	
		Child development centre	£ 310	-	-		PSSRU 2014 [94]	
		A&E/casualty	£ 141	-	-		NHS Reference Costs 2013–14 [96]	
		Ambulance	£ 180	-	-			
		Outpatient appointment	£ 189	-	-		PSSRU 2014 [94]	
		Inpatient stay	£ 1,095	-	-		NHS Reference Costs 2013–14 [96]	
		Day hospital/day care centre	£ 296	-	-		PSSRU 2014 [94]	
		Other (hospital)	£ 189	-	-			
		Home Start	£ 18	-	-		PSSRU 2004, [161]	
		Day care centre	£ 22	-	-		Childcare Cost Survey 2015, [162]	
		Drop-in centre	£ 3	-	-			
		Counselling/advice services	£ 50	£ 60	£ 50		PSSRU 2014 [94]	
		Support group	£ 14	£ 60	£ 13.8			
		Telephone help line	£ 16	-	-		Author's calculations	
		Web pages	£ 0	-	-		-	
		Voluntary agency – home based	£ 18	-	-		PSSRU 2004 [161]	

Authors (Year)	Cost sector or type	Cost parameter (task and activity)	Unit or mean costs (sd)			Unit	Cost instrument, tariffs and [Source]	Currency, reference year and discount rate
Gardener et al. (2017) [64] *(continuation)*		Other	£ 18	-	-		PSSRU 2014 [94]	
		Respite foster care	£ 700	-	-	per week		
		Children's home	£ 2,995	-	-			
		Foster home	£ 700	-	-			
Wansink et al. (2016) [22]	Intervention specific	Time spent by the coordinator (telephone/e-mails)	€ 23.90		NA	per contact	Price rate of PBCM: tariff as billed by the organisation for integral costs (gross salary costs plus overhead) [no source], list of services from the PBCM manual [92]	EUR, 2012, NA
		Series of several telephone and e-mail contacts	€ 95.61		NA	three or more contacts		
		Face-to-face contact	€ 119.51		NA	per contact		
		Home visits including traveling time	€ 191.22		NA	per visit		
		Coordination meetings	€ 191.22		NA	per meeting		
		Standard tariff for professionals in coordination meetings	€ 95.61		NA	per professional and meeting		
		Optional consultation	NA		€ 95.61	per consultation		
		Participation in groups by parents or children	NA		€ 350	per meeting		
		Intervention	€ 1,685		€ 229	mean per-family costs over full 18 months		
	Health care	Mental health care	€ 4,511		€ 3,303	mean per-family costs over full 18 months (T2)	Trimbos/iMTA questionnaire for Costs associated with Psychiatric Illness (TiC-P) [97, 98], list of services from the PBCM manual [92] , iMTA Questionnaire Intensive Youth Care [99], Dutch guidelines for health economic research [100], Standard pricing research for youth care and parenting support Noord-Brabant [101], Standards and rates for outpatient care [102]	
		Primary care (other)	€ 1,259		€ 1,106			
		Secondary care (other)	€ 2,277		€ 2,677			
		Preventive family support	€ 2,749		€ 3,559			
		Specialised child services	€ 531		€ 345			
	Child care	Informal childcare	€ 2,284		€ 2,627		Dutch guidelines for health economic research [100]	
		Professional child care	€ 2,421		€ 3,133			
	Educational	Educational sector costs	€ 1,238		€ 1,409		iMTA Questionnaire Intensive Youth Care [99], Dutch manual for ICBs [103]	
	Criminal justice	Criminal justice sector costs	€ 290		€ 207			
	Other sectors	Debt restructuring	€ 558		€ 614			

CSRI…Client service receipt inventory, EUR…Euro, GBP…British pound, HFP-M…Helping Families Programme-Modified, iMTA…Institute for Medical Technology Assessment, IY…Incredible Years, NA…Not available, NHS…National health service, PBCM…Preventive basic care management, PSSRU…Personal Social Services Research Unit (PSSRU), sd…standard deviation

Table A-6: Clinical and economic outcome parameters including inter-sectoral benefit parameters

Authors (Year)	Day et al. (2020) [65]	Gardener et al. (2017) [64]	Wansink et al. (2016) [22]
Clinical outcome parameters	Clinical outcome parameters in the underlying HTA/RCT: ■ Concerns About My Child (CAMC): visual analogue scale (0-100) ■ Eyberg Child Behaviour Inventory (ECBI): 36-item questionnaire that assesses intensity and number of disruptive behaviour problems in children (0-36 problem scale, 36-252 intensity scale) ■ Child Behaviour Checklist-Internalising Scale (CBCL-Int): 32-item questionnaire that assess internalising problems ■ Arnold-O'Leary Parenting Scale (PS): 30-item questionnaire that assesses dysfunctional discipline styles of children ■ Kansas Parental Satisfaction Scale (KPSS): three-item scale that provides measure of parenting stress and satisfaction ■ Symptom Checklist-27 (SCL-27): 27-item questionnaire that assess psychological symptoms in adults ■ Working Alliance Inventory-Short Revised (WAI-SR): 12-item questionnaire that assess participants' rating of therapeutic relationships and intervention acceptability	Primary outcome in the HTA/RCTs and EE[87]: ■ Eyberg Child Behaviour Inventory Intensity Scale (ECBI-I): 36-item measure to assess parent-reported frequency of disruptive child behaviour on a 7-point scale Secondary outcomes: ■ Strengths and Difficulties Questionnaire (SDQ): Instrument to assess parent-reported comorbid ADHD symptoms and emotional problems in children ■ Beck Depression Inventory (BDI): 21-item measure to assess parental depressive symptoms ■ Parental Stress Index Short Form (PSI-SF): 36-item measure of parental stress ■ Parental Sense of Competence (PSOC) scale: 16-item scale to assess parental self-efficacy ■ Self-reported positive and negative parenting practices: Parenting Practices Inventory (PaPi), Alabama Parenting Questionnaire (APQ), Parenting Scale (PS), interview version 1	Primary outcome in the RCT and HEE: ■ Home Observation for Measurement of the Environment (HOME) inventory [104]: Infant-Toddler, Early Childhood, Middle Childhood, and Early Adolescent version of Vedder, Eldering and Bradley [105]
Miscellaneous	-	-	Quality of parenting: measures availability and impact of objects, events and interactions with parents and covers four dimensions (responsiveness, learning materials, stimulation, harsh parenting)
Economic or generic outcome parameter	QALYs derived form EuroQol-5 Dimensions (EQ-5D)	NA	NA
Miscellaneous	Two versions of the EQ-5D questionnaire are used; the five level version (EQ-5D-5L) for parents/carers, and the EQ-5D-Y for children	-	-

HEE…Health economic evaluation, HTA…Health technology assessment, NA…Not available, QALY…Quality-adjusted life year, RCT…Randomised controlled trial

[87] Also in the cost-effectiveness analysis, the primary outcome measure was the ECBI-I. A secondary analysis used a binary variable indicating whether a child is moved below the ECBI-I clinical cut-off point (score ≥131 points) following the intervention.

Table A-7: Modelling assumptions and parameters

Authors (Year)	Day et al. (2020) [65]	Gardener et al. (2017) [64]	Wansink et al. (2016) [22]
Modelling technique	no model	Decision-analytic Markov model based on a previous publication [106]: Estimation of the present value of long-term savings/ return on investment from providing the IY intervention per child with conduct disorder problems at age 5 years by agency	no model
Comparator	no model	Simulated control group with no intervention	no model
Number and type of health or cost states (scenarios)	no model	Two trajectory scenarios ('literature' and 'data' scenario) with each having a high-cost and a low-cost sub-scenario: ■ Scenario 1 ('literature' scenario): trajectory of the 'no' intervention group with a 60% chance that a child with behavioural problems at age 5 years still show problems at age 16 years; in the intervention group this chance is 54% ■ Scenario 2 ('data' scenario): data from the pooled IY sample for the control group, with a 12% decrease in probability of scoring above the ECBI cut-off point in year 1; probability of behaviour problems persisting past age 16 years is 60% with a steeper decrease in year 1; probability that conduct problems persist beyond age 16 years in the intervention group is reduced to 52%	no model
Time horizon or cycle length	no model	25 years (age 5 to 30)	no model
Discount rate	no model	3.5%	no model
Handling of uncertainty	no model	Two trajectory and two cost scenarios[88]	
Cost categories	no model	■ NHS, ■ Social service departments, ■ Department for Education, ■ Voluntary sector, ■ Criminal justice system, ■ Health impacts of crime, and ■ Benefits payments	no model
Benefit/Effect categories	no model	Intervention effect is based on the results of the CEA: odds ratio of a child falling into the non-clinical range of the ECBI-I	no model
Further assumptions	no model	■ Take-up data are not considered ■ Dropout is already accounted for in the overall effectiveness figure (intention-to-treat basis) ■ Control group: Trajectory of remission in the absence of intervention is estimated	no model

ECBI…Eyberg child behaviour inventory, IY…Incredible Years, NHS…National health service

[88] The authors did not provide clear distinction characteristics between the low- and high-cost scenarios. They reported only that they drew on additional literature to model both cost scenarios and that costs are with regard to the cost-of-illness.

Table A-8: Intervention costs, components and quantities

Authors (Year)	Service category		Cost		Unit		Quantities/Amount		Duration (e.g. weeks)		Total costs	
Intervention/Control	**I**	**C**	**I**	**C**	**I**	**C**	**I**	**C**	**I**	**C**	**I**	**C**
Day et al. (2020) [65]	see Table A-5[89]						16 appointments, over 10 months	NA	one session à 60-90 minutes once a week	NA	£ 6,058.7[90]	£ 8,273.6
Gardener et al. (2017) [64]												
Baseline, mean (sd)	Hospital		£ 406 (2,065)	£ 292 (1,938)	per intervention or control group		-	NA	-	NA	-	-
	Community health		£ 159 (283)	£ 131 (191)	-	-	-	-	-	-	-	-
	Mental health		£ 62 (455)	£ 25 (145)	-	-	-	-	-	-	-	-
	Social services		£ 31 (169)	£ 10 (96)	-	-	-	-	-	-	-	-
	Accommodation		£ 470 (6,618)	£ 37 (47)	-	-	-	-	-	-	-	-
	Voluntary sector		£ 6 (54)	£ 5 (37)	-	-	-	-	-	-	-	-
	Total costs		-	-	-	-	-	-	-	-	£ 1,135 (6,971)	£ 501 (2,064)
Follow-up, mean (sd)	IY	-	£ 2,414 (1,248)	NA	-		12.7/8.7[91] sessions offered/attended	NA	weekly sessions of 2-2.5 hours	NA	£ 2,414	NA
	Hospital		£ 215 (857)	£ 193 (1,182)	-	-	-	-	-	-	£ 621	£ 324
	Community health		£ 115 (238)	£ 84 (155)	-	-	-	-	-	-	£ 274	£ 109
	Mental health		£ 11 (67)	£ 29 (161)	-	-	-	-	-	-	£ 73	£ 39
	Social services		£ 23 (184)	£ 22 (139)	-		-	-	-	-	£ 54	£ 59
	Accommodation		£ 2 (36)	-	-		-	-	-	-	£ 472	£ 5
	Voluntary sector		£ 5 (55)	£ 9 (85)	-		-	-	-	-	£ 11	£ 9
	Total costs		-	-	-		-	-	-	-	£ 2,766 (1,594)	£ 338 (1,261)
Baseline + Follow-up											£ 3,901[92]	£ 839
Wansink et al. (2016) [22]	PBCM	CAU	€ 1,685	€ 229	mean per-family costs over full 18 months, n=49 (PBCM) vs n=50 (CAU)		-	-	over 18 months (T0-T2)[93]		€ 13,012[94]	€ 11,219
	Healthcare services		€ 11,327	€ 10,990			-	-				
	Child care services		€ 4,705	€ 5,760			-	-			€ 17,717[94]	€ 16,979
	Inter-sectoral services		€ 2,086	€ 2,230	-		-	-			€ 19,805[94]	€ 19,209

[89] See footnote[85]. The authors did not list total costs, but only listed estimated mean (unit) costs for utilised services.

[90] Own calculations: the total costs are the sum of mean costs of each task and activity per 'average' participant/family for the whole intervention or comparator from Table A-5.

[91] The range of offered sessions amounted to 11-19 session.

[92] Own calculations: the total costs are the sum of estimated baseline and follow-up costs.
The results do not take into account any dispersion or 'uncertainty' measure in form of standard deviations.

[93] The study reported also total costs for time intervals T0-T1 and T1-T2, but we report only total costs over the full horizon in this report.

[94] TC HC perspective = Intervention costs + Costs healthcare services; TC SC perspective = TC HC perspective + Costs child care services;
TC SCT perspective = TC SC perspective + Costs outside care sector

Table A-9: Results of the economic evaluation analysis (cost-effectiveness ratios, economic impact measures etc.)

Authors (Year)	Day et al. (2020) [65]	Gardener et al. (2017) [64]	Wansink et al. (2016) [22]
Cost and benefits/ effects	Incremental costs and benefits (QALYs) of the two separate analyses[95] separately for parent and child, (95% CI)	Differences in average (total) costs between treatment group at baseline and follow-up: p-values derived by using a clustered regression model	Absolute costs and change in HOME scores of the base case scenario[96] for all three perspectives (Healthcare, social care, societal), PBCM vs CAU
Cost	Analysis 1/Analysis 2: ■ NHS PSS perspective: ■ Parent: £ -2,302 (-10,300 to 4,480)/2,547 (-8,584 to 10,837.7) ■ Child: £ 488 (-6,279 to 5,827)/3,654 (-5,636 to 10,867) ■ SOC perspective: ■ Parent: £ -2,388 (-10,284 to 4,391)/£ 2,399 (-8,577 to 10,571) ■ Child: £ 451 (-6,300 to 5,769)/£ 3,534 (-5,661 to 10,791)	Total costs/service category: p-value baseline/follow-up ■ Total costs: 0.175/<0.001 ■ Hospital: 0.497/0.792 ■ Community health: 0.184/0.046 ■ Mental health: 0.224/0.046 ■ Social services: 0.086/0.989 ■ Accommodation: 0.316/0.426 ■ Voluntary sector: 0.967/0.499	■ HC perspective: € 13,012 vs 11,219 ■ SC perspective: € 17,717 vs 16,979 ■ SOC perspective: € 19,805 vs 19,209
Benefit/Effect	Analysis 1/Analysis 2: ■ NHS PSS perspective: ■ Parent: 0.1367 (–0.0393 to 0.3334)/0.0249 (–0.1982 to 0.2318) ■ Child: 0.0297 (–0.1155 to 0.1961)/–0.125 (–0.3847 to 0.1381) ■ SOC perspective: ■ Parent: 0.1367 (–0.0393 to 0.3334)/0.0249 (–0.1982 to 0.2318) ■ Child: 0.0297 (–0.1155 to 0.1961)/–0.125 (–0.3847 to 0.1381)	Changes or differences in benefits specifically for the cost-effectiveness analyses were not reported separately	HC/SC/SOC perspective: 1.93 vs -1.89
Health economic outcome measures and thresholds	ICERs (non-parametric bootstrapping using 1,000 Cost-QALY combinations) of the two separate analyses	ICERs (10,000 bootstrap replications of the treatment effect were generated for each cost-outcome combination) willingness-to-pay thresholds for IY being cost-effective, CEACs, and economic modelling of long-term savings	ICERs (median of the 5000 bootstrap replications), willingness-to-pay thresholds for PBCM being cost-effective, and CEACs
Final outcome measures I	Analysis 1/Analysis 2: ICERs ■ NHS PSS perspective: ■ Parent: HFP dominates/102083 ■ Child: 16466/CAU dominates ■ SOC perspective: ■ Parent: HFP dominates/96155 ■ Child: 15191.21/CAU dominates	Main cost-effectiveness analysis: ■ No explicit ICERs reported, but CEAC (see Thresholds) Subgroup analysis: ■ Baseline ECBI score: IY less likely to be cost-effective with ECBI <134 ■ Child age: IY less likely to be cost-effective for age <5 years ■ Gender: IY more likely to be cost-effective for male children ■ Parental depression at baseline: IY more likely to be cost-effective for children whose parents have a moderate level of depression (BDI score ≥20)	■ HC perspective: € 461 ■ SC perspective: € 215 ■ SOC perspective: € 175

[95] Analysis 1: Sample with CSRI and intervention costs at T2, and EQ-5D at T1 and T2 (more participants at these two time points), Analysis 2: 'complete case' analysis, CSRI and intervention costs at T2 and T3, and EQ-5D at T1, T2, and T3 (reduced sample size: results have to be treated with caution)

[96] Includes imputed data and cost outliers

Authors (Year)	Day et al. (2020) [65]	Gardener et al. (2017) [64]	Wansink et al. (2016) [22]
Final outcome measures II (economic modelling)	NA	Savings per child : Present value of savings (savings range): ■ Scenario 1: £ 1,023-7,565 ■ Scenario 2: £ 1,254-9,408 Return on investment: ■ Scenario 1: threefold the investment ■ Scenario 2: fourfold the investment ■ Average net savings between £ 5,000 and £ 7,000 per child in the high-cost scenario	NA
Thresholds (probability to be cost-effective in %)	■ NHS PSS perspective/SOC perspective: ICERs ■ Child: £ 30,000 (52%)	■ £ 109 (50%) ■ £ 121 (80%) ■ £ 134 (95%) ■ £ 145 (99%)	■ HC perspective: € 0/>2,500 (20%/→100%) ■ SC perspective: € 0/>2,500 (37%/→100%) ■ SOC perspective: € 0/>2,500 (39%/→100%)
Author's conclusions	*'Ideally, cost-effectiveness would be assessed over the entire follow-up* [10 months], *but here the more robust findings are those reported at T2* [4 months]. *When considering parental QALYs, the HFP-M intervention dominated usual care. When QALYs for children were used, the HFP-M resulted in higher costs and more QALYs.'*	*'There were no differences in costs between the treatment and the control groups, other than the cost of the IY intervention.'* *'[…] the IY intervention is less likely to be considered cost-effective for children who scored below the clinical threshold on the ECBI-I at baseline and children aged <5 years.'* *'[…] the IY intervention would be more likely to be considered cost-effective for boys than for girls, and for children whose parents had a BDI-II score at baseline indicating at least a mild level of depression.'* *'[…] our findings appear to be similar to a previous cost-effectiveness analysis of the IY intervention […] we estimate it to be 99% at £ 145 (2014 prices).'* *'The findings from our longer-term economic modelling indicate that if the costs associated with persistent conduct disorder are in fact lower than previously thought, at the current price and effectiveness, the IY intervention would not result in longer-term savings. However, in our 'high-cost' scenario […] the return on investment is substantial, with average net savings of between £ 5,000 and £ 7,000 per child associated with persistent conduct disorder.'* *'As few services were used, costs were low and, thus, the potential for immediate savings from the IY intervention is reduced. This raises the question of whether or not children with mental health problems are adequately supported by mainstream services, and how access and engagement can be improved.'*	*'The results of this study show, from both a healthcare and a societal perspective, that the intervention* [PBCM] *is both more costly and more effective than CAU. Since no WTP study was conducted, no conclusive 'yes' or 'no' can be provided to the question whether the intervention is cost-effective. However […] the CEACs provide decision supportive information.' 'It* [the study] *underscores the importance of evaluating costs and benefits in other sectors and planning and evaluating innovative integrative services for children or families at risk.'* *'This study punctuates the importance of choosing a broad societal perspective in economic evaluations. ICBs should be and already are increasingly considered in underpinning (the financing of) health policies.'*

BDI…Beck depression index, CAU…Care as usual, CEAC…Cost-effective acceptability curve, ECBI…Eyberg child behaviour inventory, HC…Health care perspective, HFP-M…Helping Families Programme-Modified, HOME…Home observation measurement of the environment, ICBs...Inter-sectoral costs and benefits, ICER…Incremental cost-effectiveness ratio, IY…Incredible Years, NA…Not available, NHS…National health service, QALY…Quality-adjusted life year, PBCM…Preventive basic care management, SC…Social care perspective, sd…standard deviation, SOC…Societal perspective, WTP…Willingness-to-pay

Table A-10: Sensitivity Analysis

Authors and Year	Day et al. (2020) [65]	Gardener et al. (2017) [64]	Wansink et al. (2016) [22]	
Cost and benefits/effects	NA[97]	NA[98]	PBCM vs CAU ■ Scenario A (n=47 vs 47): imputed data, excluding cost outliers ■ Scenario B (n=41 vs 41): complete cases, including cost outliers ■ Scenario C (n=38 vs 48): imputed data, including cost outliers, PBCM-families who received the intervention ■ Scenario D (n=49 vs 50): imputed data, including cost outliers, mean difference adjustment	
Cost measure	NA	NA	■ Scenario A – HC/SC/SOC: € 11,564 vs 8,969/16,138 vs 14,422/18,194 vs 16,634 ■ Scenario B – HC/SC/SOC: € 13,480 vs 11,475/18,375 vs 17,765/19,621 vs 20,242 ■ Scenario C – HC/SC/SOC: € 14,579 vs 10,933/19,522 vs 16,140/20,736 vs 18,458 ■ Scenario D – HC/SC/SOC: € 13,012 vs 8,981/17,717 vs 12,613/19,804 vs 15,647	
Benefit/Effect measure	NA	NA	■ Scenario A: 1.70 vs -1.28/1.70 vs -1.40/1.70 vs -1.82 ■ Scenario B: 2.34 vs -2.06 ■ Scenario C: 2.24 vs -1.65 ■ Scenario D: 1.93 vs -1.89	
Outcome measures and thresholds	NA	NA	ICERs (median of the 5000 bootstrap replications) and WTP thresholds for PBCM being cost-effective	
Final outcome measures	NA	NA	■ Scenario A – HC/SC/SOC: € 776/517/410 ■ Scenario B – HC/SC/SOC: € 446/133/dominant (-143)[99] ■ Scenario C – HC/SC/SOC: € 897/843/558 ■ Scenario D – HC/SC/SOC: € 1,031/1,313/1,059	
Thresholds	NA	NA	Scenario A[100]: ■ HC perspective: € 0/>3,500 (~0%/→100%) ■ SC perspective: € 0/>3,000 (~18%/→100%) ■ SOC perspective: € 0/>2,500 (~22%/→100%) Scenario B: ■ HC perspective: € 0/>2,500 (~20%/→100%) ■ SC perspective: € 0/>3,000 (~40%/→100%) ■ SOC perspective: € 0/>1,500 (~60%/→100%)	Scenario C: ■ HC perspective: € 0/>3,500 (~5%/→100%) ■ SC perspective: 0/>3,500 (~5%/→100%) ■ SOC perspective: 0/>3,000 (~20%/→100%) Scenario D: ■ HC perspective: € 0/>3,500 (~5%/→100%) ■ SC perspective: € 0/>3,500 (~5%/→100%) ■ SOC perspective: € 0/>3,500 (~20%/→100%)

CAU…Care as usual, CEAC…Cost-effective acceptability curve, HC…Health care perspective, HFP-M…Helping Families Programme-Modified, ICER…Incremental cost-effectiveness ratio, IY…Incredible Years, NA…Not available, NHS…National health service, QALY…Quality-adjusted life year, PBCM…Preventive basic care management, SC…Social care perspective, sd…standard deviation, SOC…Societal perspective, WTP…Willingness-to-pay

[97] Two separate analyses were conducted, but no specific sensitivity analysis.

[98] A subgroup analysis for the cost-effectiveness and two separate analyses for the estimation of long-run savings were conducted, but no specific sensitivity analysis.

[99] Lower incremental costs and positive incremental effect of PBCM vs CAU (negative ICER): PBCM is superior to CAU on cost-effectiveness.

[100] No explicit data was available for these thresholds. Instead, the supplementary material of the study (Additional file 2) provides the CE planes and CEACs [22].

Literature search strategies

Search strategy for Centre for Research and Dissemination

Search name: COPMI economics	
Search Date: 28/05/201	
#1	MeSH DESCRIPTOR Mothers EXPLODE ALL TREES
#2	MeSH DESCRIPTOR Fathers EXPLODE ALL TREES
#3	MeSH DESCRIPTOR Parents EXPLODE ALL TREES
#4	MeSH DESCRIPTOR Family EXPLODE ALL TREES
#5	MeSH DESCRIPTOR Caregivers EXPLODE ALL TREES
#6	(Mother*) IN NHSEED
#7	(Father*) IN NHSEED
#8	(Parent*) IN NHSEED
#9	(Family) IN NHSEED
#10	(Families) IN NHSEED
#11	(Care*giver*) IN NHSEED
#12	(Care-giver*) IN NHSEED
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#15	(mental* OR psychiatr*)
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#21	#18 OR #19 OR #20
#22	MeSH DESCRIPTOR Child EXPLODE ALL TREES
#23	(child*)
#24	(Daughter*)
#25	(Son)
#26	(Sons)
#27	(Offspring)
#28	#22 OR #23 OR #24 OR #25 OR #26 OR #27
#29	#21 AND #28
#30	MeSH DESCRIPTOR Child of Impaired Parents EXPLODE ALL TREES
#31	(COPMI)
#32	#29 OR #30 OR #31
#33	(#32) FROM 2010 TO 2021
#34	(#33) IN NHSEED
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Search strategy for Embase

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#2	'mother'/exp/mj
#3	'father'/exp/mj
#4	'family'/exp/mj
#5	'caregiver'/exp/mj
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#8	mental*:ab,ti OR psychiatr*:ab,ti
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#22	'progeny'/exp/mj
#23	offspring:ti,ab
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#25	#14 AND #24
#26	'child of impaired parents'/exp
#27	copmi:ti,ab
#28	#25 OR #26 OR #27
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#30	preventive NEAR/3 management
#31	pbcm:ti,ab
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#44	#38 OR #43
#45	letter:it
#46	editorial:it
#47	note:it
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#49	#44 NOT #48
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#51	((energy OR oxygen) NEAR/1 cost):ti,ab
#52	((energy OR oxygen) NEAR/1 expenditure):ti,ab
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#57	'nonhuman'
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Total: 557 hits	

Search strategy for Medline

Search name: COPMI economics	
Search Date: 26/05/201	
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#2	*Mothers/ (35095)
#3	*Fathers/ (6411)
#4	*Family/ (40418)
#5	*Caregivers/ (35307)
#6	*Parent-Child Relations/ (18651)
#7	1 or 2 or 3 or 4 or 5 or 6 (165760)
#8	(mental* or psychiatr*).ti,ab. (743140)
#9	*Mental Disorders/ (147398)
#10	8 or 9 (789872)
#11	7 and 10 (19143)
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#14	11 or 12 or 13 (44151)
#15	*Child/ (3425)
#16	child*.ti,ab. (1795674)
#17	daughter*.ti,ab. (32568)
#18	son.ti,ab. (24596)
#19	sons.ti,ab. (26419)
#20	offspring.ti,ab. (98110)
#21	15 or 16 or 17 or 18 or 19 or 20 (1943684)
#22	14 and 21 (19601)
#23	exp "Child of Impaired Parents"/ (6317)
#24	COPMI.ti,ab. (40)
#25	22 or 23 or 24 (24554)
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#27	(preventive adj3 management).mp. (1775)
#28	PBCM.ti,ab. (153)
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#30	25 and 29 (7833)
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#33	exp "costs and cost analysis"/ (282320)
#34	Economics, Dental/ (1937)
#35	exp economics, hospital/ (27905)
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#37	Economics, Nursing/ (4027)
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#41	value for money.ti,ab. (2460)
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#43	32 or 33 or 34 or 35 or 36 or 37 or 38 or 39 or 40 or 41 or 42 (1357762)

#44	((energy or oxygen) adj cost).ti,ab. (5500)
#45	(metabolic adj cost).ti,ab. (1996)
#46	((energy or oxygen) adj expenditure).ti,ab. (33276)
#47	44 or 45 or 46 (39569)
#48	43 not 47 (1348521)
#49	letter.pt. (1339797)
#50	editorial.pt. (726542)
#51	historical article.pt. (385056)
#52	49 or 50 or 51 (2428305)
#53	48 not 52 (1304996)
#54	exp animals/ not humans/ (5296551)
#55	53 not 54 (1224392)
#56	30 and 55 (820)
#57	impact analys#s.mp. (2534)
#58	impact assessment*.mp. (6754)
#59	social return* on investment*.mp. (100)
#60	SROI.ti,ab. (101)
#61	beneficiary assessment*.mp. (1)
#62	57 or 58 or 59 or 60 or 61 (9338)
#63	30 and 62 (7)
#64	31 or 56 or 63 (830)
#65	limit 64 to yr="2010 -Current" (650)
#66	limit 65 to (english or german) (641)
#67	remove duplicates from 66 (405)
Total: 405 hits	

Search strategy for PsycInfo

Search name: COPMI economics	
Search Date: 27/05/201	
#1	*PARENTS/ (22736)
#2	parent*.ti,ab. (280146)
#3	*MOTHERS/ (22988)
#4	mother*.ti,ab. (126805)
#5	*FATHERS/ (6665)
#6	father*.ti,ab. (48315)
#7	*Family/ (31242)
#8	family.ti,ab. (308966)
#9	families.ti,ab. (143403)
#10	*Caregivers/ (23030)
#11	care$giver*.ti,ab. (50653)
#12	care-giver*.ti,ab. (1140)
#13	*Parent Child Relations/ (23351)
#14	1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 (659348)
#15	(mental* or psychiatr*).ti,ab. (610774)
#16	*Mental Disorders/ (70163)
#17	"Health & Mental Health Treatment & Prevention ".cc. (239615)

#18	15 or 16 or 17 (794115)
#19	14 and 18 (141735)
#20	((mental* or psychiatr*) adj5 (parent* or mother* or father* or family or families or care?giver* or care-giver* or carer* or maternal or paternal)).mp. (39512)
#21	19 or 20 (148988)
#22	child*.ti,ab. (713958)
#23	daughter*.ti,ab. (13011)
#24	son.ti,ab. (11001)
#25	sons.ti,ab. (7071)
#26	offspring.ti,ab. (20639)
#27	22 or 23 or 24 or 25 or 26 (744842)
#28	21 and 27 (73717)
#29	(child* adj5 ((impaired or mental* or psychiatric*) adj3 (parent* or mother* or father* or family or families or care?giver* or care-giver* or carer*))).mp. (8659)
#30	COPMI.ti,ab. (28)
#31	28 or 29 or 30 (74740)
#32	(program* or intervention* or inititative* or network$1 or collaborat* or cooperative* or co-op*).mp. (988007)
#33	(prevent*ive adj3 management).mp. (273)
#34	PBCM.ti,ab. (5)
#35	32 or 33 or 34 (988161)
#36	31 and 35 (26654)
#37	"costs and cost analysis"/ (17487)
#38	"Cost Containment"/ (653)
#39	(economic adj2 evaluation$).ti,ab. (1862)
#40	(economic adj2 analy$).ti,ab. (1609)
#41	(economic adj2 (study or studies)).ti,ab. (856)
#42	(cost adj2 evaluation$).ti,ab. (361)
#43	(cost adj2 analy$).ti,ab. (3942)
#44	(cost adj2 (study or studies)).ti,ab. (925)
#45	(cost adj2 effective$).ti,ab. (16239)
#46	(cost adj2 benefit$).ti,ab. (3663)
#47	(cost adj2 utili$).ti,ab. (1373)
#48	(cost adj2 minimi$).ti,ab. (387)
#49	(cost adj2 consequence$).ti,ab. (124)
#50	(cost adj2 comparison$).ti,ab. (193)
#51	(cost adj2 identificat$).ti,ab. (26)
#52	(pharmacoeconomic$ or pharmaco-economic$).ti,ab. (326)
#53	37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49 or 50 or 51 or 52 (36970)
#54	(task adj2 cost$).ti,ab,id. (695)
#55	(switch$ adj2 cost$).ti,ab,id. (1431)
#56	(metabolic adj cost).ti,ab,id. (106)
#57	((energy or oxygen) adj cost).ti,ab,id. (298)
#58	((energy or oxygen) adj expenditure).ti,ab,id. (2838)
#59	54 or 55 or 56 or 57 or 58 (5067)
#60	(animal or animals or rat or rats or mouse or mice or hamster or hamsters or dog or dogs or cat or cats or bovine or sheep or ovine or pig or pigs).ab,ti,id,de. (364348)
#61	editorial.dt. (44230)
#62	letter.dt. (23631)
#63	dissertation abstract.pt. (514311)

#64	60 or 61 or 62 or 63 (923578)
#65	53 not (59 or 64) (31941)
#66	36 and 65 (319)
#67	impact analys#s.mp. (250)
#68	impact assessment*.mp. (643)
#69	social return* on investment*.mp. (44)
#70	SROI.ti,ab. (34)
#71	beneficiary assessment*.mp. (2)
#72	67 or 68 or 69 or 70 or 71 (933)
#73	36 and 72 (12)
#74	66 or 73 (329)
#75	limit 74 to yr="2010 -Current" (206)
#76	limit 75 to (english or german) (191)
Total: 191 hits	

Search strategy for Web of Science database

Search name: COPMI economics	
Search Date: 27/05/201	
#1	TI=((child* OR adolesc* OR son OR sons OR daughter*) NEAR/3 ((parent* OR mother* OR father* OR family OR families OR care*giver* OR care-giver* OR carer*) NEAR/3 (mental* OR psychiatr*))) Indexes=SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC Timespan=All years
#2	AB=((child* OR adolesc* OR son OR sons OR daughter*) NEAR/3 ((parent* OR mother* OR father* OR family OR families OR care*giver* OR care-giver* OR carer*) NEAR/3 (mental* OR psychiatr*))) Indexes=SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC Timespan=All years
#3	ALL=(COPMI) Indexes=SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC Timespan=All years
#4	#3 OR #2 OR #1 Indexes=SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC Timespan=All years
#5	#3 OR #2 OR #1 Refined by: WEB OF SCIENCE CATEGORIES: (ECONOMICS OR PUBLIC ENVIRONMENTAL OCCUPATIONAL HEALTH) Indexes=SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC Timespan=All years
#6	TOPIC: ("parents with mental illness") Indexes=BKCI-S, ESCI, SSCI, BKCI-SSH, SCI-EXPANDED, IC, A&HCI, CPCI-SSH, CPCI-S, CCR-EXPANDED Timespan=All years
#7	#6 OR #5 Indexes=SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC Timespan=All years
#8	#6 OR #5 Refined by: PUBLICATION YEARS: (2021 OR 2020 OR 2019 OR 2018 OR 2017 OR 2016 OR 2015 OR 2014 OR 2013 OR 2012 OR 2011 OR 2010) Indexes=SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC Timespan=All years
#9	#6 OR #5 Refined by: PUBLICATION YEARS: (2021 OR 2020 OR 2019 OR 2018 OR 2017 OR 2016 OR 2015 OR 2014 OR 2013 OR 2012 OR 2011 OR 2010) AND LANGUAGES: (ENGLISH OR GERMAN) Indexes=SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC Timespan=All years
Total: 410 hits	

Search strategies for CINAHL and EconLit

Search strategies upon request.

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Austrian Institute for
Health Technology Assessment
GmbH