

Costs and Benefits in Education

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Costs and Benefits in Education

Addressing Methodological Challenges
and Broadening the Societal Perspective
in Mental Health Economics

Irina Pokhilenko

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COSTS AND BENEFITS IN EDUCATION

Addressing Methodological Challenges and Broadening the Societal
Perspective in Mental Health Economics

DISSERTATION

to obtain the degree of Doctor at the Maastricht University,

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Prof.dr. Pamela Habibović

in accordance with the decision of the Board of Deans

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by

Irina Pokhilenko

Supervisors:

Prof. dr. S.M.A.A. Evers

Prof. dr. A.T.G. Paulus

Co-supervisor:

Dr. R.M.W.A. Drost

Assessment committee:

Prof. dr. H.J.M. Majoie (chair)

Prof. dr. W.B.F. Brouwer, Erasmus University Rotterdam

Dr. L. Jackson, University of Birmingham, UK

Prof. dr. M.A. Joore

Prof. dr. T. Schils

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Chapter 1. General Introduction

Mental problems are associated with a significant proportion of health expenditures,¹ but also costs in other sectors, including the education sector.² At the same time, interventions to prevent or treat mental problems can lead to benefits in the education sector. However, health economic evaluations rarely account for costs and benefits in the education sector.^{3,4} This dissertation studies the intersectoral costs and benefits related to mental health in the education sector and proposes evidence-based methods that have the potential to help capture these in health economics research. This introductory chapter provides the general background of the research presented in this dissertation. It describes the basic principles of the scientific discipline of health economics, the relationship between the mental health domain and the education sector, and provides an outline of currently available methods for capturing this relationship and their respective limitations. Furthermore, this chapter highlights the need for developing standardized evidence-based methods for including education-related costs and benefits in health economics studies. The chapter concludes with a description of the aims of this dissertation and its outline.

Terminology in this dissertation

In this dissertation, we use different terms when describing the economic impact of mental health on societal sectors, in particular the education sector. The concept of intersectoral costs and benefits, defined as ‘the costs and benefits for all non-healthcare sectors of society, attributable to the implementation of a health intervention’, is a term in health economics literature.² It is used to refer to costs and benefits of health interventions that occur in various sectors of society. It is important to mention the terminological variability in relation to this concept. Some studies use such terms as ‘multi-sectoral’, ‘cross-sectoral’, and ‘indirect’ costs when referring to intersectoral costs and benefits.

The central concept of this dissertation is the concept of intersectoral costs and benefits in the education sector. In this dissertation, the education sector is defined as one of the key social sectors aimed at facilitating the process of learning and acquisition of knowledge. It encompasses both core goods and services, i.e. expenditures related to instruction and education, and ancillary services such as provision of meals, transport and housing.⁵ Furthermore, in this dissertation, we consider not only the costs and benefits related to the use of education services, but also costs and benefits related to educational outcomes such as productivity at school and educational attainment.

Economic impact of mental conditions

Worldwide, health expenditures continue to rise at alarming rates, due to ageing societies, the spread of non-communicable diseases, and the development of expensive new treatments and interventions. In fact, health spending has been growing faster than the global economy – at an average of 3.9% versus 3% over the past 20 years.¹ Mental health accounts for up to 10% of global disease burden, which makes mental health conditions one of the most significant contributors to the economic impact of ill health on societies worldwide.^{6,7} People with mental health conditions have an increasing need for healthcare services.^{8,9} On the one hand, this can be attributed to higher utilization of mental health services, such as contacts with a psychotherapist or the utilization of inpatient mental health care.¹⁰ On the other hand, the

presence of mental health conditions requiring care is often linked to physical health conditions. For example, people diagnosed with depression are often at higher risk of developing cardiovascular disease and vice versa.¹¹ Therefore, the economic impact of mental ill health is broader than in (mental) health care alone.

In fact, mental problems can have an extensive effect on all areas of a person's life. People with mental problems often have a diminished ability to participate in education or work, and to enjoy leisure time.¹²⁻¹⁴ Children with mental disorders tend to have lower school attendance and lower school attainment compared to their healthy peers.^{15,16} Similarly, adults with mental disorders are likely to be less productive at work due to absenteeism, presenteeism and early retirement.¹⁷⁻¹⁹ Laszewska et al. (2020) reported that the costs of lost productivity were 2.5 times higher for people with mental disorders compared to people without mental disorders.²⁰ For particular disorders such as depression, lost productivity can account for the highest proportion of the economic burden. For example, the study by Sobocki and colleagues (2007), conducted among the adult population in Sweden, estimated that up to 86% of the economic burden of depression was due to lost productivity.¹⁷ Higher utilization of health care is also associated with personal expenses for travel to places where services are provided, and with loss of leisure time.¹⁰ Furthermore, the impact of mental conditions often goes beyond the affected individuals to affect the well-being of their families and close ones.^{21,22}

People with severe mental conditions often require additional support to function in society, in the form of assisted housing²³ and supported employment.²⁴ In addition, people with certain mental conditions (e.g. schizophrenia) are more likely to be involved with safety and justice services either as a victim or a perpetrator of a crime, which can lead to high costs in the criminal justice system.^{25,26} This was demonstrated in several previous studies including Healy and colleagues (1998), who found that up to 80% of the societal costs of drug dependency could be associated with costs in the criminal justice system.²⁷

Mental health and education

Apart from the sectors that are affected by changes in the health of the adult population, mental conditions also contribute to costs in the education sector; these costs are particularly relevant in the context of interventions targeting children and adolescents.²⁸ Mental problems often begin to develop in childhood or adolescence and coincide with the critical period of developing educational trajectories.²⁹ In comparison with their healthy peers, students with mental problems tend to be absent and/or drop out of school more often, and hence are more likely to have lower educational attainment.^{15,16} Research shows that supporting individuals with mental problems can be far more costly than supporting their peers without these problems, due to the use of additional specialist education services.^{30,31} This also suggests that students with mental problems are less likely to succeed in the labor market, are more likely to have lower income and a lower socio-economic status, leading to loss of societal welfare.³²

The economic impact of mental problems on the education sector is likely to be particularly evident in the context of common mental conditions among children and adolescents (e.g. attention deficit hyperactivity disorder). Several cost-of-illness studies of mental conditions in children and adolescents demonstrated that the proportion of education costs in relation to

the total disease burden can be significant and amount to over half of the total disease burden for this group.^{33,34} This can be attributed to the increased use of education services (e.g. extra lessons), the need for special education services, which are more costly in comparison with regular education, and absenteeism, among other consequences. For example, a cost-of-illness study conducted by Le and colleagues (2014) in the Netherlands found that costs in the education sector (i.e. costs incurred due to the additional use of education services) could account for up to 62% of the costs associated with attention-deficit hyperactivity disorder.³³ Despite this knowledge about the economic burden of mental conditions on the educational sector, the integration of education costs and benefits in the evaluation of health interventions remains scarce.^{3,4} The key to changing this is to develop health economic tools to further enable researchers and better inform decision makers.

Health economics

As the increasing need for health care outweighs available resources, evidence-based decisions and policies related to health and health care become increasingly important. Health economics is a science of scarcity in the context of health(care) that studies, among other factors, the value of new health technologies in relation to their effectiveness. The emergence of this discipline is attributed to a seminal piece by Kenneth Arrow (1963) that outlines the distinct characteristics of healthcare markets, such as the derived demand for health care and uncertainties regarding the effectiveness of treatment.³⁵ Since then health economists have developed methods and a large body of knowledge to help policymakers with the efficient allocation of resources in the healthcare sector. The use of health economics evidence in policy decision-making became more important in the 90s, when it became a mandatory requirement for the pricing and reimbursement of pharmaceuticals by government bodies and health insurers (e.g. in Australia and Canada).³⁶ Subsequently, this requirement has been extended to other types of health and medical technology, including medical devices and public health interventions.^{37,38}

One of the most commonly used tools in health economics is an economic evaluation. In an economic evaluation, two or more alternative interventions are compared in terms of costs and effects. For example, the study by Wansink and colleagues (2016) compared preventive basic care management to care as usual in terms of their respective costs and outcomes in the population of children of parents with a mental illness.³⁹ The evidence generated by these studies helps policymakers select and invest in the most attractive alternative, which will generate the most value for money (i.e. better health outcomes, savings). In case of the study by Wansink and colleagues (2016), the results suggested that preventive basic care management was more effective but also more costly, with cost-effectiveness improving when a societal perspective was adopted. Other health economics studies such as cost-of-illness studies and budget impact analysis also help portray the economic impact of specific disorders or new interventions on society.

Choice of perspective in health economics

Health economics studies can adopt different perspectives. The choice of perspective determines what costs and effects of a health intervention under consideration are included in

the analysis.⁴⁰ In studies conducted from a healthcare system perspective, only healthcare costs associated with an intervention (e.g. visits to general practitioner, hospital stay) and health outcomes are considered. In a payer perspective, costs and benefits relevant to the payer, for example, the government, are considered. In studies conducted from a societal perspective, which is arguably the broadest perspective, all costs and benefits associated with an intervention are included regardless of who experiences them. In addition to costs and benefits relevant to the healthcare system and/or the payer, productivity losses (i.e. costs associated with diminished or lost productivity at work due to being ill) and patient and family costs and benefits (e.g. out of pocket expenses, informal care) are the most common components of health economics studies conducted from a societal perspective.^{3,4} However, costs and benefits in other sectors (e.g. education, criminal justice, housing) can be relevant as well.³

The choice of perspective has implications for selecting the appropriate methodology for conducting a study, particularly with regard to data collection. In health economics studies, robust data is essential for producing reliable evidence. Health economics data are most often collected either from administrative data sources (e.g. hospital registries) or through patients' self-reports. Administrative data are considered to be more accurate and detailed in comparison with self-reported data. However, administrative data sources are usually limited to a particular type of data and rarely provide a broad overview of costs.⁴¹ For example, hospital registries contain only data on patients' use of hospital services, but no information on the use of social care services, informal care, or out-of-pocket expenses. Therefore, self-reported data is preferred when the analysis takes a broader perspective (e.g. societal perspective), as self-reported data allows for collecting information on broader use of resources.^{41,42}

The choice of perspective also has implications for study results and their interpretation. Economic evaluations are usually commissioned by a national health authority who is also responsible for the health system budget. Hence, a budget holder perspective, i.e. a healthcare or a health system perspective, is often chosen for the pragmatic reason of informing the decision maker while taking into account the context in which they operate (e.g. a limited healthcare budget).⁴³ However, from a theoretical point of view, a societal perspective is preferred over more narrow healthcare or payer perspectives. According to the theory of welfarism, which lies at the foundation of health economics, the goal of health interventions is to maximize social welfare.⁴⁴ Therefore, one needs to consider the broader impact of an intervention on society to evaluate its contribution to social welfare. Furthermore, from a practical point of view, health affects all aspects of one's life and is dependent on an array of factors including the environment, demographics, education, politics, and many others. Investments in health can generate savings in other sectors and vice versa. To demonstrate this in an economic evaluation, all relevant costs and/or benefits of health interventions need to be considered. For example, the study by Lavelle and colleagues (2019)⁴⁵ demonstrates that including the family spillover health effects and costs in economic evaluations of childhood illnesses can have a significant influence on the study results, often resulting in a more favorable conclusion. Therefore, a societal perspective is preferred for informing societal

decision-making in relation to health.⁴⁶ The growing importance of adopting a broader perspective in health economics also corresponds with the increasing interest in broader research and policy approaches such as the whole systems approach and Health in All Policies that advocate for taking into account the health implications of all policy decisions on every level of policy-making.^{47,48}

Nevertheless, there is currently no universal guidance regarding which cost categories could be relevant for inclusion. Researchers commonly rely on national pharmacoeconomic or costing guidelines such as the guidelines for conducting economic evaluation by the National Health Care Institute in the Netherlands⁴⁹ or the guidance for the appraisal of health technologies by the National Institute for Health and Care Excellence in the United Kingdom.⁵⁰ These guidelines prescribe the predominant perspective of the analysis and outline which costs need to be considered in the reference case, i.e. the primary analysis, and, if applicable, which costs could be considered in secondary analyses. However, there is a lot of variation between these guidelines. Most of them recommend adopting a healthcare perspective for the primary analysis, while a few recommend adopting a broader perspective and including costs in other sectors in addition to the costs in the healthcare sector.⁴ This leads to a lot of variation in the conceptualization of a societal perspective, in the methods applied in conducting health economics studies and hence in the outcomes of these studies; this variation ultimately limits the comparability and transferability of the study results. Furthermore, costing guidelines were originally developed to support economic evaluations of pharmaceuticals and somatic (health) care interventions using a more narrow healthcare perspective. Economic evaluations of more complex interventions, such as public health and mental health interventions, require an adapted analysis framework to account for the complex interrelations of these interventions within their context and their broad intersectoral impact.⁵¹

The Second Panel of Cost-effectiveness in the United States has proposed an impact inventory (Figure 1) that can be used to identify potentially relevant health and non-health related costs and consequences when appraising an intervention.⁵² The impact inventory is designed to help evaluate the broader effects of interventions in an explicit and transparent manner. In addition to costs and consequences in the healthcare sector, it includes costs and consequences in other sectors such as productivity, consumption, social services, criminal justice, education, housing, and the environment as well as any other potentially relevant sectors. However, to date it has not been widely used.⁴ There are also other studies exploring the conceptualization of broader costs and benefits in health economics. The handbook for conducting economic evaluations by Drummond et al. (2015) outlines cost categories including costs in non-healthcare sectors that could be relevant for consideration, depending on the nature of the intervention being studied.⁴⁰ The classification scheme by Drost et al. (2013) incorporates a more detailed overview of the intersectoral costs and benefits associated with mental health interventions in the following domains: education, labour and social security, household and leisure, the criminal justice system, and the individual and family.²

Sector	Type of Impact (list category within each sector with unit of measure if relevant) ^a	Included in This Reference Case Analysis From...Perspective?		Notes on Sources of Evidence
		Health Care Sector	Societal	
Formal Health Care Sector				
Health	Health outcomes (effects)			
	Longevity effects	<input type="checkbox"/>	<input type="checkbox"/>	
	Health-related quality-of-life effects	<input type="checkbox"/>	<input type="checkbox"/>	
	Other health effects (eg, adverse events and secondary transmissions of infections)	<input type="checkbox"/>	<input type="checkbox"/>	
	Medical costs			
	Paid for by third-party payers	<input type="checkbox"/>	<input type="checkbox"/>	
	Paid for by patients out-of-pocket	<input type="checkbox"/>	<input type="checkbox"/>	
Future related medical costs (payers and patients)	<input type="checkbox"/>	<input type="checkbox"/>		
Future unrelated medical costs (payers and patients)	<input type="checkbox"/>	<input type="checkbox"/>		
Informal Health Care Sector				
Health	Patient-time costs	NA	<input type="checkbox"/>	
	Unpaid caregiver-time costs	NA	<input type="checkbox"/>	
	Transportation costs	NA	<input type="checkbox"/>	
Non-Health Care Sectors (with examples of possible items)				
Productivity	Labor market earnings lost	NA	<input type="checkbox"/>	
	Cost of unpaid lost productivity due to illness	NA	<input type="checkbox"/>	
	Cost of uncompensated household production ^b	NA	<input type="checkbox"/>	
Consumption	Future consumption unrelated to health	NA	<input type="checkbox"/>	
Social Services	Cost of social services as part of intervention	NA	<input type="checkbox"/>	
Legal or Criminal Justice	Number of crimes related to intervention	NA	<input type="checkbox"/>	
	Cost of crimes related to intervention	NA	<input type="checkbox"/>	
Education	Impact of intervention on educational achievement of population	NA	<input type="checkbox"/>	
Housing	Cost of intervention on home improvements (eg, removing lead paint)	NA	<input type="checkbox"/>	
Environment	Production of toxic waste pollution by intervention	NA	<input type="checkbox"/>	
Other (specify)	Other impacts	NA	<input type="checkbox"/>	

Figure 1. Impact Inventory Template (Sanders et al. 2016)

Previous research on education costs and benefits in health economics

Given that health is intertwined with many aspects of a person's life, health interventions can lead to significant costs and/or benefits (i.e. cost savings, cost offsets) in sectors outside the healthcare sector, including the education sector. As mentioned previously, education costs and benefits associated with health interventions are particularly relevant for inclusion in studies focusing on the population of children and adolescents. Several economic evaluations and cost-of-illness studies incorporated costs and/or benefits in the education sector;^{33,34,53-55} however, they remain scarce.^{3,4} This can be attributed to various factors including the

predominance of a narrow perspective. Only a limited number of national pharmacoeconomic guidelines suggest including education costs and benefits in health economic analyses if these are relevant to the context of the study.^{49,50,52,56} Furthermore, proper guidance on the inclusion of these costs and benefits is lacking. Compared to the amount of research done on the inclusion of costs and benefits in the healthcare sector, little attention has been paid to exploring the methodology for including intersectoral education costs and benefits. Nevertheless, several previous studies have investigated the methods for their identification, measurement, valuation, and ultimately inclusion in the analyses.

Drost et al. (2013)² identified costs and benefits in the education sector based on a review of peer-reviewed literature and expert consultation. The list includes fourteen education costs and benefits that can be relevant for inclusion in economic evaluations of mental health interventions (Figure 2). Education costs and benefits on this list are categorized into those that occur during pre-school (e.g. change in school readiness) and those that occur while students attend school (e.g. low school performance). It is important to note that the list presented in this study is a collection of mixed terms including services (e.g. special education services) and outcomes for the affected individuals (e.g. low school attainment).

Students

Education

Pre-school (E-p)

- Change in school readiness
- Problems with school entry

School (E-s)

- Low school adaption/competence
- Low school participation/engagement
- Low school attainment/productivity/performance (absenteeism/presenteeism)
 - due to morbidity
 - due to death of a sick relative
- Cognitive deficits (I&F-p)
 - e.g. learning disabilities
- Failing to meet school obligations/low educational outcomes/school failure
 - leading to grade retention
- Disrupted school experience
- Teacher-student conflicts (I&F-s)
- School dropout/pre-mature leave
- Special education services
- Home education/residential programs
- Risk of dropping out of high school due to morbidity earlier in time*
- Decreased intellectual and academic performance due to morbidity earlier in time
- performance due to morbidity earlier in time

Figure 2. Education costs and benefits of mental health interventions
(adapted from Drost et al. (2013))

Mayer et al. (2017)⁵⁷ conducted a systematic literature review of health-related resource-use measurement instruments that included education costs and benefits. Twenty-one resource-use measurement instruments were found to have included education costs and benefits, with the most common items being “absenteeism from school”, “tutoring”, “classroom assistance”, “special school/boarding school”, and “school/social functioning”.

Drost et al. (2017)⁵⁸ proposed four methods for the valuation of education costs and benefits, and, based on this, developed a costing manual for the selected items for the Netherlands. In addition, Andronis et al. (2019)⁵⁹ reviewed available evidence on the valuation of the forgone childhood time, including the valuation of lost school productivity.

Although the abovementioned literature offers some guidance for the inclusion of education costs and benefits in health economics studies, information on the relative importance of integrating these cost and benefits is limited. Furthermore, in comparison with the amount of research done on the proper identification, measurement and valuation of costs and benefits in other sectors (e.g. health care, social care, productivity, and informal care), research on the proper inclusion of intersectoral education costs and benefits is scarce. There is a lack of evidence-based standardized methods for including these costs and benefits in economic evaluations, in particular those that could support multi-national studies. This hampers researchers in producing reliable, comparable and transferable economic evidence to support optimal policy-making within and across countries, leading to inefficiencies in resource allocation. The importance of further methodological research in this domain is also highlighted by the increasing number of economic evaluations conducted in the population of children and adolescents, for which the inclusion of education costs and benefits is likely to be relevant.⁶⁰

Context of this dissertation

The research presented in this dissertation was conducted alongside the ProgrammE in Costing, resource use measurement and outcome valuation for Use in multi-sectoral National and International health economic evaluations (PECUNIA) project funded by the European Union’s (EU) Horizon 2020 research and innovation programme.⁶¹ The main objectives of PECUNIA were to tackle the healthcare challenges of an ever-growing and rapidly ageing population in the EU by developing new multi-sectoral, standardized, harmonized and validated methods and tools for the assessment of costs and outcomes in European healthcare systems. In line with the PECUNIA’s focus on the methods for conducting multi-sectoral economic evaluations, this dissertation presents recent methodological developments to facilitate the inclusion of education costs and benefits in health economics research.

Objectives of the dissertation

In light of the abovementioned knowledge gaps, the objective of this dissertation is two-fold. The first aim is to investigate the available methods for including intersectoral education costs and benefits in health economics research and the attitude among health economists towards doing this. The second aim is to propose new standardized, internationally applicable identification, definition, measurement and valuation methods that can enable the inclusion

of education costs and benefits in health economics studies in a transparent way and enhance the comparability and transferability of the results within and across countries.

Outline of the dissertation

Chapter 2 explores the existing methods for including intersectoral education costs and benefits in health economics studies. By reviewing economic evaluations and cost-of-illness studies set in the psychosocial domain in the child and adolescent population, an overview of the applied identification, measurement and valuation methods is presented. Furthermore, the proportion of the education costs and benefits to the total costs is investigated. In Chapter 3, an overview of education costs and benefits is developed based on the review of peer-reviewed and grey literature and on expert opinions. Using the overview of education costs and benefits presented in Chapter 3, Chapter 4 investigates the relative importance of the education and criminal justice costs and benefits in the context of mental and behavioral disorders. Health economists were asked to rank the attributes in a best-worst scaling experiment. Furthermore, the experts were asked whether they thought it was relevant to include education costs and benefits in economic evaluations in this domain and whether they previously included education costs and benefits in their research. Chapter 5 describes the development process of a standardized multi-sectoral resource-use measurement instrument to enable the measurement of broader costs, including education costs, in a more comparable way. In Chapter 6, a standardized valuation method is applied to calculate and compare the unit costs of the selected education services in five European countries.

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Chapter 2. Do Costs in the Education Sector Matter? A Systematic Literature Review of the Economic Impact of Psychosocial Problems on the Education Sector

Chapter 2 draws upon:

Pokhilenko, I., Janssen, L. M., Evers, S. M., Drost, R. M., Schnitzler, L., & Paulus, A. T. 2021.
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impact of psychosocial problems on the education sector. *PharmacoEconomics*, 39, 889-900.
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Abstract

Background. Psychosocial (e.g., anxiety or behavior) problems lead to costs not only in the healthcare sector but also in education and other sectors. As psychosocial problems develop during the critical period of establishing educational trajectories, education costs are particularly relevant in the context of psychosocial problems among children and adolescents.

Objectives. This study aimed to gain insights into the methods used for the inclusion of education costs in health economics studies and into the proportion of the education costs in relation to the total costs associated with a condition or an intervention.

Methods. We systematically searched the PubMed, Embase, SSCI, CINAHL, PsycINFO, ERIC, and Econlit databases in August 2019 for economic evaluations of mental health, psychosocial and educational interventions, and cost-of-illness studies of mental, behavioral, and neurodevelopmental disorders conducted from a societal perspective in populations of children and adolescents. An additional search was conducted in February 2021 to update the review.

Results. In total, 49 articles were included in the analysis. The most common cost items were special education, school absenteeism, and various educational professionals (educational psychologist). A variety of methods were employed for the identification, measurement, and/or valuation of education costs. The proportion of education costs to the total costs of condition/intervention ranged from 0 to 67%, with the mean being 18.5%.

Discussion. Since education costs can constitute a significant proportion of the total costs of an intervention or condition, including them in health economics studies might be important in informing optimal resource allocation decisions. Although various methods are available for including education costs in health economics studies, further research is needed to develop evidence-based methods for producing comparable estimates.

Introduction

Individuals affected by psychosocial problems have not only an increased need for health care; their ability to participate in education and work or enjoy leisure time may also be diminished. Hence, the economic impact of psychosocial problems is cross-sectoral and is not limited to the healthcare sector.¹ In fact, a large proportion of the costs associated with psychosocial problems occur in sectors other than healthcare, including education, employment and productivity, informal care, and criminal justice.^{1,2} In the absence of a commonly accepted definition, for this study psychosocial problems were defined as a range of psychological problems (e.g. learning difficulties, anxiety, behavioral issues) and social problems (e.g. educational or family difficulties) that affect one's "functioning in daily life, his or her environment and/or life events".³ As psychosocial problems often begin to develop in childhood or adolescence, their onset might occur during the period when individuals establish their educational trajectories.⁴ Therefore, the economic impact of psychosocial problems on the education sector is likely to be particularly evident in the context of common conditions among children and adolescents (e.g. autism and attention deficit hyperactivity disorder). Research shows that supporting individuals with psychosocial problems can be far more costly in comparison with their peers without these problems.^{5,6} Psychosocial problems among children and adolescents can lead to diminished school productivity and school performance, increased need for additional support at school, early dropout, and potentially reduced educational attainment, all of which are associated with substantial overall societal losses and excessive costs for the education sector in particular.^{2,5,7,8} Several health economic studies in the psychosocial domain also demonstrate that education costs can constitute a large proportion of the total costs and even be the most costly cost category.^{9,10} Therefore, including education costs in health economics research studies could be crucial to ensuring optimal resource allocation decision-making.¹¹

Despite recent methodological developments in identifying, measuring and valuing education costs,¹²⁻¹⁶ their inclusion in costing research remains limited.¹⁷ This could be attributed to several reasons. First, a narrow perspective is still predominant in the majority of current pharmacoeconomic guidelines, of which many neglect costs other than healthcare and productivity costs.¹⁸ Second, there is limited explicit methodological guidance on how to collect, measure and value education costs in health economics studies. The current methods for the identification, measurement and valuation of the education costs in health economics focus only on a limited number of costs and do not allow for producing valid and internationally comparable estimates. Third, the evidence regarding the impact of education costs on the results of health economics studies is lacking. While several studies demonstrate that the proportion of education costs to total costs can be significant,^{9,10} a systematic synthesis of the existing evidence could provide a sound rationale for including education costs in health economics research studies in the psychosocial domain.

Given the above mentioned research gaps, a systematic review of health economics studies, i.e. economic evaluations and cost-of-illness studies, in the psychosocial domain was conducted to fulfill the aims of this research. As will be discussed in more detail in the methods section, potentially relevant studies include economic evaluations of psychosocial

interventions and cost-of-illness studies of mental health, behavioral and neurodevelopmental disorders. First, this study aimed to gain insight into whether the studies in the psychosocial domain include education costs, and, if so, what methods they used to identify, measure and value education costs. This would provide an overview of the available methods for including education costs in health economics studies and highlight current knowledge gaps, information that can direct further research in this field. Second, this study aimed to estimate the proportion of the total costs that education costs account for, to provide justification for including education costs in health economics studies.

Methods

This study adopted a systematic review methodology standardized in health economics, based on the methods described by van Mastrigt et al. (2016).¹⁹ Reporting of the results was based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement.²⁰

Data collection

To identify health economics research studies that included costs in the education sector, seven electronic databases were searched in August 2019, including PubMed, Embase (Ovid), SSCI (Web of Science), CINAHL (EBSCO), PsycINFO (EBSCO), ERIC (EBSCO), and Econlit (EBSCO). The selection of databases and the search string were developed and consolidated by the research team in collaboration with an information specialist. An example of the search string in PubMed is available in Supplementary File 1. An additional search using the same approach was carried out in February 2021 and covered the period between August 2019 and February 2021.

In- and exclusion criteria of the identified studies were based on the PICOS framework.²¹ They included studies with a population of children and adolescents aged 0-25 years (P), investigating psychosocial and educational interventions (I), regardless of the comparator (C) or the outcomes (O), relating societal costs, and designed either as a full economic evaluation or cost-of-illness study (S). Psychosocial interventions were defined as “interpersonal or informational activities, techniques, or strategies that target biological, behavioral, cognitive, emotional, interpersonal, social, or environmental factors with the aim of improving health functioning and well-being”.²² Both economic evaluations and cost-of-illness studies were considered for inclusion only if costs in the education sector were included as intersectoral costs associated with a condition or an intervention. Studies that included costs in the education sector as part of the intervention costs (e.g. training of professionals, materials) were considered beyond the scope of the current review and excluded, as there is research that provides detailed methodological guidance on how to include intervention costs in economic evaluations.²³ The relevance of cost-of-illness studies was determined based on the International Classification of Diseases 11th revision; the relevant conditions were mental health, behavioral and neurodevelopmental disorders.²⁴ Studies were excluded when the abstract and/or the full text was not available in the English language or could not be accessed.

Identification and screening of relevant studies

All hits were imported in Endnote X8.2 and de-duplicated using the method determined by Bramer et al. (2016).²⁵ One reviewer (IP) performed title and abstract screening. The second reviewer (LJ) screened 10% of randomly assigned titles and abstracts. A similar procedure was employed for the full text screening. Differences in inclusion were resolved by discussion between the two reviewers. The ultimate inclusion of studies based on full text screening was determined in the discussion between the two reviewers.

Data extraction and analysis

The number of studies that were excluded during full-text screening because they did not include education costs and the number of studies that complied with the inclusion criteria were used to calculate the proportion of the studies that included education costs. Data extraction for the studies that complied with the inclusion criteria was conducted by multiple reviewers independently. One reviewer extracted data from all studies (IP); two reviewers (RD, LS) extracted data from 50% of the included studies each. The data on the general characteristics of the selected articles and the methods used to identify, measure and value education costs were extracted using a pre-defined template adapted from Wijnen et al. (2016),²⁶ which was piloted on 10% of the included studies by two reviewers (IP & LJ) (Supplementary File 2).

The reported unit costs were recalculated to Euros (2021) using an online inflation tool²⁷ and a currency converter²⁸ and presented in ranges per reported measurement unit (e.g. per day, per year). When the original year for reported unit costs was not provided, the year of the article's publication was used for the recalculation. For studies that included costs in the education sector and reported the breakdown of total costs and/or incremental cost-effectiveness ratio (ICER) calculation, the percentage of total costs that was accounted for by costs in the education sector was determined. When multiple groups (i.e. treatment arms) were compared in an article, the percentage of education costs was calculated separately for each group. The overall proportion of the education costs was calculated as the average of every estimate. In addition, this proportion was also calculated by averaging the mean estimates per article to avoid bias. The percentages of education costs for each condition were summarized and presented separately. When articles reported the inclusion of costs in the education sector but did not provide the breakdown of total costs, the authors of these articles were contacted for further information.

Results

The database search conducted in August 2019 resulted in 10,236 hits. After de-duplication, the titles and abstracts of 6,248 articles were screened and 5,937 of them were excluded. The full texts of 311 records were assessed for eligibility, resulting in 270 records being excluded. The additional search in February 2021 resulted in 1,705 hits. After de-duplication, the titles and abstracts of 1,048 articles were screened and 995 of them were excluded. The full texts of 53 records were assessed for eligibility, resulting in 45 records being excluded. In total, forty-one records identified in the search in August 2019 and eight records identified in the search

in February 2021 fulfilled the inclusion criteria and were included in the analysis. The process of the literature search and selection is shown in the PRISMA flowchart (Figure 1).

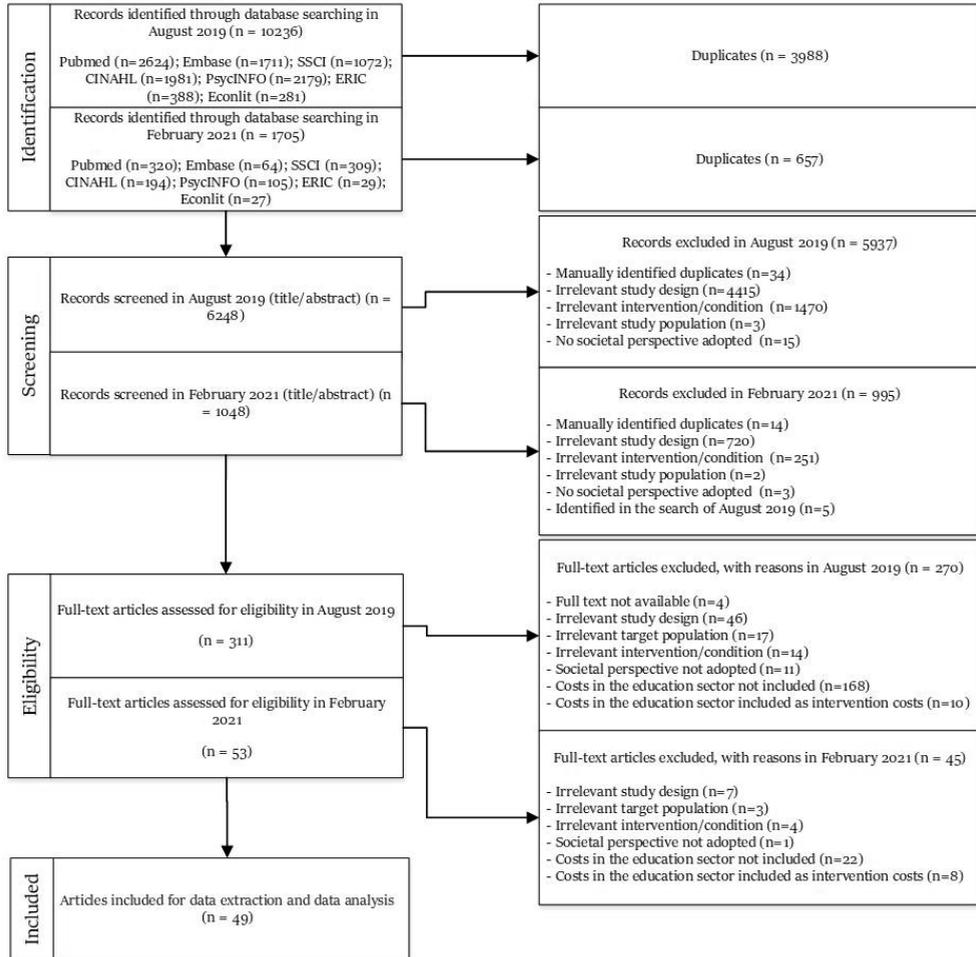


Figure 1. PRISMA flowchart of the literature search and selection

General study characteristics

Forty-nine studies fulfilled the inclusion criteria and were included for data extraction and data analysis^{9,10,29-75}. The majority of the studies (n=16, 33%) were conducted in the Netherlands, the United States (n=8, 16%), and Canada (n=7, 14%), followed by the United Kingdom (n=5, 10%), Sweden (n=5, 10%), Germany (n=2, 4%), Australia (n=1, 2%), Spain (n=1, 2%), and Ireland (n=1, 2%). Furthermore, two studies focused on multiple countries^{36,45} and one study did not focus on any particular country.⁵⁵ Concerning the study design, over a half of the studies were cost-of-illness studies (n=26); the remaining studies were either trial-

based (n=12) or model-based (n=8) economic evaluations or a combination of both (n=1).⁵¹ One study described an economic evaluation study using a quasi-experimental design.⁷⁴ One study combined a cost-of-illness study with a model-based economic evaluation.³² The majority of the studies focused on mental and neurodevelopmental disorders among children and adolescents, including autism spectrum disorder (n=10), attention deficit hyperactivity disorder (n=8), and anxiety disorders (n=6).

Methods for including education costs

Identification

Ninety-two education cost items were extracted from the 49 studies, and clustered based on similar descriptions (Supplementary File 3). The most commonly identified education cost items were special education (n=22), school absenteeism (n=17), different types of professionals in the education sector (n=17), and additional educational support services such as tutoring and homework help (n=8). Two studies reported using the classification scheme by Drost et al. (2013)¹⁴ to identify relevant education costs.^{41,73} In five studies, relevant cost categories were identified based on the literature search.^{9,45,52,55,64} In one study cost selection was based on the data included in the administrative records.⁵¹ In addition, the authors of five studies justified their selection of cost categories as including all potentially relevant costs based on the definition of a societal perspective,^{30,33,38,47,71} and the education costs in particular as “typical components of direct non-medical costs”,⁴³ “important cost for many children with autism”,⁴⁷ “important resource units”.⁷² The majority of the studies (70%) did not report any specific methods used for identifying the education cost items.

Measurement

For measuring education costs, 27 studies used self-reported data, thirteen studies derived data from the literature (i.e. other studies), six studies used other secondary sources (e.g. national-level statistics data, government reports, administrative records), two studies used a combination of primary and secondary data,^{45,74} and one study used literature combined with expert opinion.⁶⁴ Figure 2 illustrates the frequency of use of measurement methods per cost item. For example, special education was measured most often by using literature (n=8) or self-reported data (n=8), followed by a combination of methods (n=4) and other secondary sources (n=2). The figure shows that the majority of education cost items (n=65) were measured by using self-reported data. It is important to note that in Figures 2 and 3 the extracted cost items are presented based on the clustering as described above.

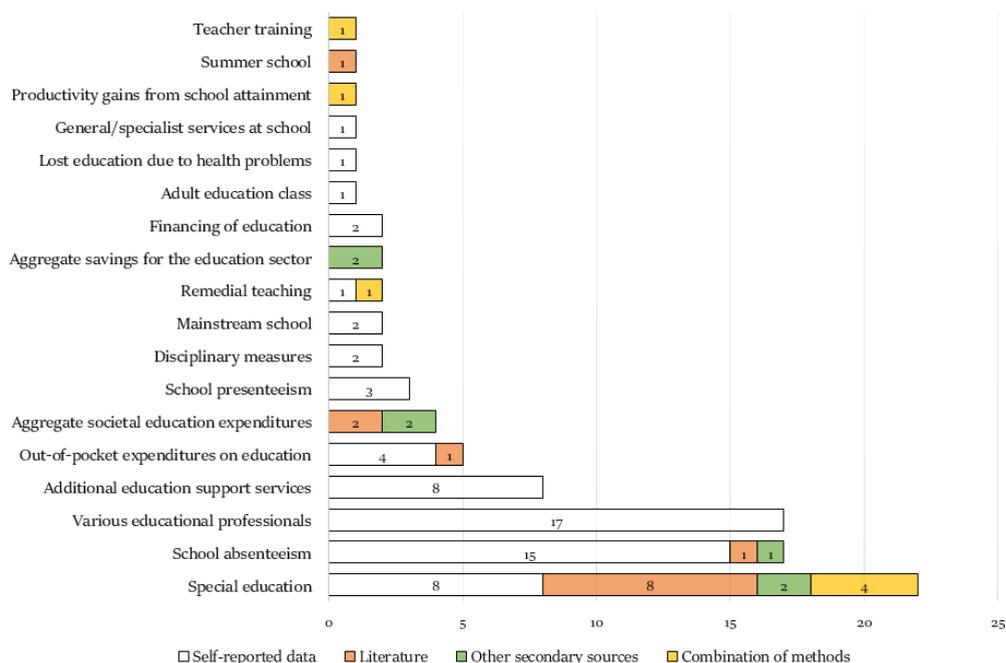


Figure 2. Methods used for the measurement of the education costs

The majority of studies collected self-reported data from parents or other primary caregivers (n=21); fewer studies collected self-reported data from children or adolescents alone or in addition to caregivers (n=5), study investigators (n=1) or government officials (n=1). The most common methods for collecting self-reported data were questionnaires (n=21), followed by cost diaries (n=5) and case report forms (n=1). The resource-use measurement questionnaires that were used included (modified) Treatment Inventory of Costs in Patients with psychiatric disorders (TiC-P) (n=6), modified CSRI (n=1), Child and Adolescent Service Use Schedule (CASUS) (n=2), Carer Service Use Schedule (CARER-SUS) (n=1), Expenditures for Health and Social Service Utilization Questionnaire (n=1), Child and Adolescent Services Assessment (n=1), Studying the Costs of Parental Expenditures (SCOPE) (n=1), or other study-specific resource-use measurement instruments (n=5). Nine articles did not report what specific resource-use measurement instruments were used for collecting self-reported data.

Valuation

The unit costs for the extracted education cost items (n=92) were derived from various secondary data sources (n=65, 70%), calculated by the authors using secondary data (n=12, 13%), taken at face value (e.g. out of pocket expenses) (n=5, 5%) or calculated based on the market price of a given service (n=1, 1%). Nine items (10%) were not valued or were included as either intangible costs (i.e. they were measured but not valued) or as the impact on a caregivers' costs (e.g. lost productivity, time or money). The secondary sources of unit costs included official public sources such as national level public data (e.g. government reports) (n=34), costing guidelines (n=12), literature (n=11), or a combination of literature and public

sources (n=1). For seven items, the sources of unit costs were not reported. For the 30 extracted items, the unit costs were not reported.

Figure 3 illustrates the frequency of use of different methods for the valuation of education cost items. For example, the unit cost of school absenteeism was most frequently developed by the authors (n=8), extracted from secondary national sources (n=4), in three studies school absenteeism was included it as an intangible cost, and two studies used costing guidelines. Supplementary File 3 provides a detailed overview of the measurement and valuation methods applied to measure and value the extracted items.

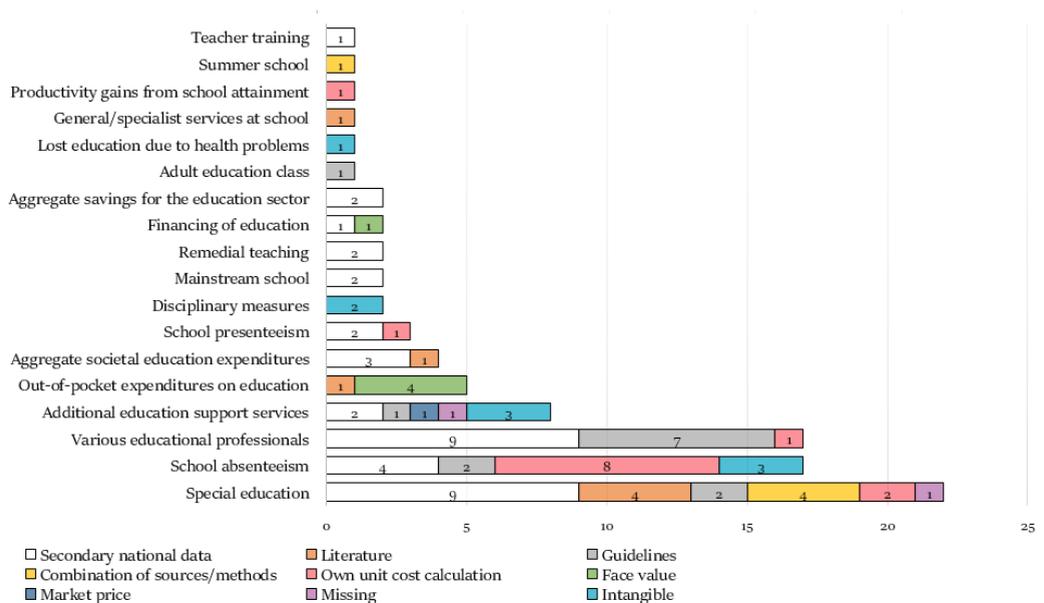


Figure 3. Methods used for the valuation of the education costs

The reported unit costs of the education cost items are presented in Table 1 in Euros (2021) per measurement unit. The items for which the unit of measurement was not reported are presented in separate rows.

Table 1. Unit costs of the education cost items

Item	Measurement unit	Range (Euros 2021)	Sources
Special education	Per day	15.4 - 15.6	64,70
	Per month	449.8	69
	Per year	588.1 - 56,534.5	10,42,45,47,56,58,59,71
	Average lifetime cost	7,949.1 - 13,852.8	32,57
	Not reported	677 - 1,771.32	62
School absenteeism	Per hour	5.3 - 11.4	33,34,65,66,71
	Per day	28.6 - 68	38,48,53,54,63
School presenteeism	Per day	61.6 - 68	48,53,63
Remedial teaching	Per visit	63.6	64
	Per hour	63.1	69
Aggregate societal expenditures for education	Per year	5,599.3 - 6,981.7	9,55
Study/homework help	Per hour	44.1	48
Tutoring help	Per hour	48.7	53
Educational psychologist	Per hour	48.5	62
	Not reported	51	38
Attendance officer	Per hour	47	41,73
Teacher training	Per visit	83.4	64
General/specialist services at school	Not reported	25.3	40
Education welfare officer	Not reported	27.6	38
Family liaison officer (school)	Per hour of client work	67.5	38
School counsellor	Per hour	14.9	30

Item	Measurement unit	Range (Euros 2021)	Sources
Special needs assistant	Per hour	18.6	62
Teacher	Not reported	48.2	38
Parental out-of-pocket expenses related to child's education	Annual per patient and total costs	64.3	9
Financing of education	Per year	13,846.7 or 27693.4 (depending on the disability level)	44
	Not reported	193 - 1,094	62
Aggregate savings for the education sector	Per year per child	8,185.40	61
Productivity gains from school attainment	Per high school diploma	171,133 - 23,7784.9	74

Contribution of the education costs to the total costs

Thirty-one out of the 49 included studies contained sufficient detailed information to calculate the proportion of the education costs. Contact with the authors did not allow for collecting any additional information either due to non-response or lack of time to provide further information. Because some studies included multiple groups or treatment arms, the number of estimates (n=72) is higher than the number of studies (n=31). The average proportion of the education costs based on the average of every estimate was 18.48% (n=72), ranging from 0% to 78.5%. A slightly higher proportion of 20.37% was estimated based on the average of the means per study (n=31). Figure 4 illustrates the percentage of the education costs to total costs. The majority of the estimates (64%, n=46) lie below the mean value of 18.48%. Approximately a quarter of the estimates (22%, n=16) are within the range between the mean and 40%. Only 13% (n=10) of the estimates are above 40%, in the range of 44.3-78.5%. The highest estimates (above 50% of the total costs) were reported in six studies; one of them is an economic evaluation of an exposure-based cognitive behavioral therapy for adolescents diagnosed with irritable bowel syndrome;⁶³ five of them are cost-of-illness studies of autism spectrum disorder, intellectual disability, attention deficit hyperactivity disorder, and mental problems in general in the population of children and adolescents.^{9,10,36,44,52} The summary of the studies is provided in Supplementary File 4.

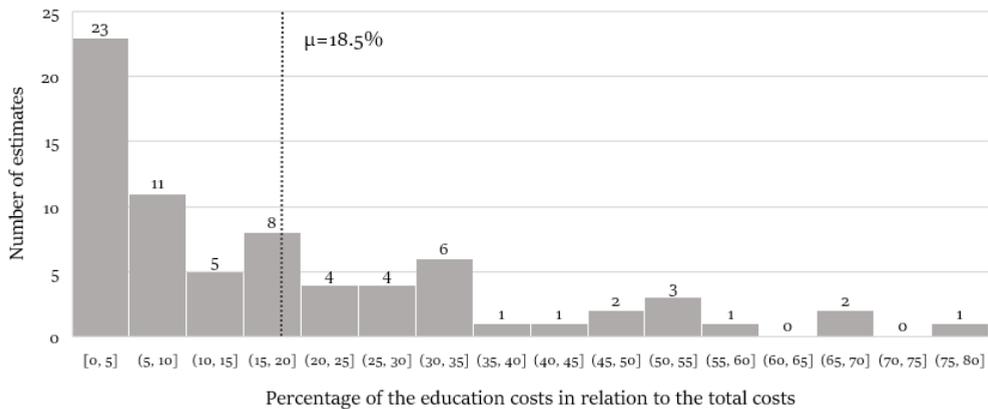


Figure 4. Relative proportion of the education costs in relation to the total costs (n=72)

The largest number of estimates was provided for the studies that investigated autism spectrum disorder (n=18), anxiety disorders (n=9), and attention deficit hyperactivity disorder/attention deficit disorder (n=8). The highest proportion of education costs was observed for intellectual disability disorder (67%); however, this is based on a single study.⁴⁴ Studies that focused on autism spectrum disorder and attention deficit hyperactivity disorder/attention deficit disorder accounted for the second and third highest proportion of education costs, 30% and 24%, respectively.

Discussion

Psychosocial problems have a far-reaching impact on all societal sectors, including the education sector.² The economic impact of psychosocial problems on the education sector is especially relevant for children and adolescents.⁵ Nevertheless, little is known about including costs from the education sector in health economics studies in the psychosocial domain. The aim of this study was to investigate whether health economics studies in the psychosocial domain that focus on the child and adolescent population include costs from the education sector, and, for those studies that do, what methods the authors used to identify, measure and value these costs. In addition, for the studies that included education costs, this study aimed to estimate the proportion of education costs to total costs.

The findings of this study revealed that only approximately 20% of the health economics studies relating to the psychosocial domain included education costs. Regarding the country of origin of the included studies, the consideration of education costs is explicitly recommended only in the Dutch, Canadian and Irish pharmacoeconomic guidelines.⁷⁶⁻⁷⁸ However, in the absence of evidence-based guidelines in other countries regarding including education costs in health economics studies, it is difficult to assess whether the education costs should have been included in each particular study. For example, the guidelines of the National Institute of Health and Care Excellence in the UK suggest considering broader costs if these are substantially influenced by an intervention, but do not provide any further guidance in terms of the types of costs to be included and how to determine their relevance.⁷⁹ This means that the decision (not) to include education costs oftentimes depends on the researchers, who might

not be aware of the relevance of the education costs and might be constrained by a lack of resources, the requirements of the national pharmacoeconomic guidelines, and other factors. In addition, the decision to include education costs might also lead to the need for additional resources (e.g. extended questionnaires, more time for data analysis), which are often scarce.

The education cost items identified in this review comprise services, professionals, lost productivity (i.e. absenteeism and presenteeism), and the financial expenses on education. The list also includes disciplinary measures (i.e. detention, expulsion, etc.) which were relevant in the context of attention deficit hyperactivity disorder, although these were not valued. In comparison with the existing classifications,^{14,16} this study identified more services and educational professionals. However, intangible consequences, such as reduced educational attainment or problems with school entry, were scarcely captured in the reviewed studies. It is important to note that the costs associated with reduced school attainment become more apparent in the longer term, which makes it more challenging to capture these costs in studies with shorter time horizons of one or two years. In addition, the most frequently included education cost items, “special education”, “absenteeism from school” and “additional educational support (e.g. tutoring, homework help)”, correspond with the findings of the recent review of health-related resource-use measurement instruments by Mayer et al. (2017).¹³ However, less tangible resource-use items such as “school functioning” and “social functioning” were also not included in the reviewed studies. This might be attributed to the limited time horizon of the studies as well as the difficulty of valuing these costs. Furthermore, one can argue that these costs can also be captured by quality of life measurement instruments. This highlights the relevance of the issue of double counting, which researchers need to be aware of.

A variety of methods were employed for measuring and valuating the education costs. Self-reported data was used more frequently than secondary data sources. Furthermore, the studies that were based on self-reported data included more cost items in comparison with the studies that relied on secondary sources, which often provide pooled estimates. Resource-use measurement questionnaires were preferred over other self-reported data collection methods, with TiC-P being the most commonly used. There was considerable variation between the estimates for comparable items, which could be attributed to the selection of the measurement and valuation methods, among other factors. In addition, the content of the items was not always clearly based on the name and/or the semantic description of the item, which further limits comparability. For example, special education was the most commonly included education cost in the reviewed studies. However, it was not always clear how the authors defined special education and, based on the definition, what type of service was measured and valued. Above all, the findings suggest that the variability in the methodology used to include education costs and the general lack of transparent reporting limits meaningful comparison between the studies. This also indicates the need for developing evidence-based harmonized methods and instruments.

The proportion of education costs in the reviewed studies varied from 0 to 78.5%. The highest proportion (above 50% of the total costs) was observed in studies of autism spectrum disorder, intellectual disability, attention deficit hyperactivity disorder, and mental problems in general

in the population of children and adolescents. It is important to note that the proportion of the education costs in the reviewed studies might have been influenced by the choice of study design, as well as by the number of cost categories and the number of the education cost items included in each particular study. Nevertheless, this finding suggests that education costs can constitute a significant proportion of the total costs and thus are likely to influence the results of the study. This also provides a rationale for considering including education costs in health economics studies in the psychosocial domain to ensure optimal resource allocation in society.

Methodological reflections

To the best of our knowledge, this is the first study that systematically reviewed health economics literature in relation to the inclusion of education costs. Broad inclusion criteria were employed to incorporate studies with various designs. Furthermore, a detailed overview of possibly relevant cost items and currently available methods for including education costs in health economics studies was presented. Nevertheless, the study is also subject to several limitations. First, the context of this study was limited to the psychosocial domain, while education costs can also be relevant for other disease areas (e.g. infectious diseases).¹⁷ Using the methods of the current study to investigate economic evaluations and cost-of-illness studies in other contexts could provide a more complete overview of the available methods for including education costs. Second, the inclusion criteria for this study were fairly broad, which allowed for collecting more data, but did not allow for making meaningful comparison between the studies that included education costs with the studies that did not, due to heterogeneity. Third, the involvement of a second reviewer in the screening and data extraction process was limited to a random sample of 10% of the studies at each stage. Fourth, the findings of the current study depended on the quality of reporting in the included articles, in particular regarding transparency and the level of detail when reporting the methods that were used for identification, measurement (e.g. measurement units, resource-use measurement instrument used) and valuation (e.g. unit costs and their sources) of the education costs. Furthermore, the lack of details in several studies did not allow for the calculation of the proportion of the education costs. Although several attempts were made to contact the authors of the studies, they were not able to provide additional information, either due to lack of time or non-response.

It is also important to note that the definition of the education sector and what falls under it might not always be clear. The reviewed studies included several cost items which might not necessarily fall within the education sector. For example, government subsidy on education might fall within the government sector, as it does in the Canadian costing guidelines.⁷⁷ Another example is out of pocket expenses on education, which might in fact fall within the category of patient and family costs, as these costs are incurred by the household. In addition, training of professionals (e.g. nurses) who will be delivering an intervention might also be an example of an education cost. In this study, we included training of professionals as part of intervention costs, although if the intervention was set in the education sector, it was considered an education cost, while if the intervention was set in another sector (e.g. healthcare sector), it was not considered an education cost. While this could be considered a limitation, the literature provides detailed methodological guidance on how to include

intervention costs in economic evaluations.²³ Therefore, further investigation of these studies was beyond the scope of the current article.

Implications for further research

The findings of this study demonstrate that incorporating education costs in health economic studies can be hindered due to the lack of methodological guidance and difficulties of collecting reliable comparable data to estimate these costs. These challenges dictate the need for further research in this direction. First, the relevance of including education costs depends on various factors such as target population, condition and/or intervention of interest, national setting, among others. Further research could focus on investigating the relevance of specific education cost items in relation to these factors - for example, which costs are most relevant for the child population with attention deficit hyperactivity disorder. Second, the use of standardized terminology might help facilitate the comprehensibility and comparability of the cost items. One example of a classification system that can facilitate the comparability of the services is the standard taxonomy for description, mapping and comparison of services for Long-Term Care (DESDE-LTC).⁸⁰ It has been widely used in health services research to enable comparison of services across settings.⁸¹ Application of this system to other sectors, including the education sector, might help facilitate the comparability of the services included in future health economics studies. Third, reduced educational attainment due to psychosocial problems in childhood and adolescence can influence one's labor market outcomes,⁵ which might be especially notable for the individuals whose educational needs have not been met. Accounting for this relationship would be particularly relevant for the studies that adopt a long-term timeframe and would require further research to develop appropriate methods for capturing this effect in health economic evaluations and cost-of-illness studies. It is also important to consider the potential issue of double counting that might arise when as costs associated with educational attainment and consequences on the labor market would be interlinked. Fourth, future research is needed to investigate the appropriate methods for the valuation of school productivity, since school productivity is conceptually different from work-related productivity.

Conclusion

Education costs can constitute a large proportion of the total costs in health economics studies set in the psychosocial domain, especially in the population of children and adolescents. Therefore, inclusion of education costs, if those are substantially affected given the study context, could be crucial for informing optimal resource allocation decisions. The findings of this study demonstrate that although methods for the identification, measurement, and valuation of the education costs are available, the lack of methodological guidance regarding the proper use of the methods leads to considerable variation and limits the comparability of the studies. This indicates the need for further research in this direction, especially with respect to the development of evidence-based methodological recommendations for the inclusion of the education costs in health economics studies. By demonstrating the broader impact of psychosocial conditions and interventions to treat them on the education sector, health economics research can play a crucial role in optimizing the use of limited societal resources. Further research in this direction also has the potential to alleviate the urgent

problem of underfunding of the mental health domain by attracting additional (cross-sectoral) resources.

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Supplementary Files

Supplementary File 1. Example of a search strategy in PubMed

PubMed		
"Infant"[Mesh] OR "Child"[Mesh] OR "Adolescent"[Mesh] OR "Young Adult"[Mesh] OR infan*[tiab] OR baby[tiab] OR babies[tiab] OR newborn*[tiab] OR child*[tiab] OR pupil*[tiab] OR teen*[tiab] OR adolescen*[tiab] OR young adult*[tiab] OR pediatric*[tiab] OR paediatric*[tiab] OR pube*[tiab] OR juvenil*[tiab] OR youngster*[tiab] OR youth*[tiab] OR kid[tiab] OR kids[tiab] OR prepube*[tiab] OR preadolescen*[tiab] OR junior high*[tiab] OR young people*[tiab] OR minors[tiab] OR preschool*[tiab] OR pre-school*[tiab] OR kindergart*[tiab] OR playgroup*[tiab] OR play- group*[tiab] OR playschool*[tiab] OR schoolgirl*[tiab] OR schoolboy*[tiab] OR "Schools"[Mesh:NoExp] OR School[tiab] OR Schools[tiab] OR highschool*[tiab]	"Costs and Cost Analysis"[Mesh:noexp] OR "Cost- Benefit Analysis"[Mesh] OR "Cost of Illness"[Mesh] OR cost[tiab] OR costs[tiab] OR economic evaluation[tiab] OR Economics[Subheading] OR health expenditure*[tiab] OR "health care costs"[MeSH:noexp] OR benefits[tiab] OR burden[tiab]	Societal[tiab]

Supplementary File 2. Template for data extraction

Author		
Year		
Title		
Country		
Condition		
Type of study (cost-of-illness, economic evaluation)		
Intervention (if applicable)		
Control (if applicable)		
Analytic approach (model/trial)		
Population		
Study perspective		
Cost categories		
Identification - included cost item(s)		
Identification - method		
Measurement - method		
Measurement - instrument		
Valuation - method		
Total costs		
Education costs		
Proportion of the education costs to the total costs		

Supplementary File 3. Summary of measurement and valuation methods per extracted item

Item	Number of studies	First author (year), country	Measurement method (instrument)	Unit cost (currency, year)	Unit	Source of unit cost
Special education	22	Barrett (2011), UK	Self-reported questionnaire (CA-SUS)	Not reported	Not reported	National-level public data obtained via personal communication with government departments and from national surveys
		Beaulieu 2019	Literature	18308 (CAD, 2014)	Lifetime costs	Literature and government report
		Buescher (2014), UK/US	Literature	Not reported	Not reported	Literature and government reports
		Byford (2015), UK	Self-reported questionnaire (CA-SUS (modified), CARER-SUS (modified))	Not reported	Not reported	National-level public data obtained from government departments for school costs
		Ericson (2017), Sweden	Literature (prevalence-based)	53745 (EUR, 2014)	Per year	National-level public data obtained from government sources (Swedish National Agency for Education)
		Ganz (2007), US	Literature and government report (prevalence-based)	Not reported	Not reported	Literature and government report
		Greenmyer (2018), US/Canada/Sweden/New Zealand	Literature (pooled estimate)	7179 (USD, 2017)	Per year	Literature (pooled estimate)

Item	Number of studies	First author (year), country	Measurement method (instrument)	Unit cost (currency, year)	Unit	Source of unit cost
		Honeycutt (2004), US	National-level government data (Special Education Expenditure Project for the 1999–2000 school year)	Not reported	Not reported	Government report (Special Education Expenditure Project for the 1999–2000 school year)
		Jarbrink (2001), UK	Literature and national-level data (prevalence-based)	10778 (GBP, 1997-1998) for children 5-19 with autism and additional learning disability 7216 (GBP, 1997-1998) for children with high functioning autism 323 (GBP, 1997-1998) for adults with autism and additional learning disability	Per year	Own calculation based on literature and national-level data obtained via personal communication
		Jarbrink (2007), Sweden	Self-reported questionnaire (not provided, based on CSRI and tested in a comparable sample)	26263 (EUR, 2005) range 24347 – 163147 depending on the type of schooling	Per year	Own unit cost calculation based on the data provided by service providers in the municipality

Item	Number of studies	First author (year), country	Measurement method (instrument)	Unit cost (currency, year)	Unit	Source of unit cost
		Penner (2015), Canada	Literature	14057 (CAD, 2013) for semi-dependent individuals 31629 (CAD, 2013) for dependent individuals	Per year	Government report (Zegarac et al. 2008)
		Peterson (2018), US	National-level government data	8393 (USD, 2013)	Average lifetime cost per victim	Literature
		Piccininni (2017), Canada	Literature	14057 (CAD, 2013) for semi-dependent individuals 31629 (CAD, 2013) for dependent individuals	Per year	Government report (Zegarac et al. 2008)
		Popova (2016), Canada	Data collected via publicly available sources and a study-specific questionnaire (not provided) from Ministries of Education of each Canadian province	Range 5564-9220 (CAD, 2013) depending on the province	Average cost per child per year	Provincial data
		Roddy (2019), Ireland	Self-reported questionnaire (The Economic Costs for Families Raising a Child with an Autism Spectrum Disorder)	Range 670 - 1753.33 depending on the type of schooling (EUR)	Not reported	National-level data obtain via personal communication with government officials (Special Education Section in the department of Education and Skills)

Item	Number of studies	First author (year), country	Measurement method (instrument)	Unit cost (currency, year)	Unit	Source of unit cost
		Schawo (2015), Netherlands	Literature and expert opinion	13.63 (EUR, 2014)	Additional costs per day	National-level public data obtained from government sources (Dutch Ministry of Education, Culture and Science annual report)
		Steensel (2013), Netherlands	Self-reported questionnaire (not provided)	4728.53 (EUR, 2010)	Per year	Costing guidelines (Dutch Guidelines for Cost-Research)
		Thanh (2009), Canada	Literature	Not reported	Per year	Literature
		Thurston (2011), Canada	Self-reported questionnaire (inventory developed initially by Browne et al. (1990), currently updated to be the Expenditures for Health and Social Service Utilization Questionnaire (Browne et al. 2006))	Not reported	Not reported	Not reported
		Van Der Kolk (2015), Netherlands	Self-reported questionnaire (TiC-P (both child and adult versions, modified))	392 (EUR, 2012)	Per month (in addition to costs for regular education)	National-level public data obtained from government sources (Dutch Ministry of Education, Culture and Science annual report)
		Van Der Schans (2015), Netherlands	Literature	12.05 (EUR, 2005)	Per day	Literature (Faber et al. 2008)

Item	Number of studies	First author (year), country	Measurement method (instrument)	Unit cost (currency, year)	Unit	Source of unit cost
School absenteeism	17	Wansink (2016), Netherlands	Self-reported study-specific questionnaire (Dutch Services and Support Questionnaire (based on TiC-P))	Not reported	Not reported	Costing guidelines (Dutch manual for intersectoral costs and benefits of (preventive interventions))
		Bodden (2008)a, Netherlands	Self-reported data collected using a prospective cost diary (not provided)	4.37 (EUR, 2003)	Per hour	Own calculation based on the national-level data (National Institute of Budget information)
		Bodden (2008)b, Netherlands	Self-reported data collected using a prospective cost diary (not provided)	4.37 (EUR, 2003)	Per hour	Own calculation based on the national-level data (National Institute of Budget information)
		Bodden (2018), Netherlands	Self-reported data collected using a retrospective cost questionnaire (not provided)	Not reported	Not reported	Own calculation based on the national-level data (National Institute of Budget information)
		Creswell (2019), UK	Self-reported data collected using a parent diary (not provided)	25-55 (GBP, 2011-2012)	Per day	Government report (National per pupil medians Income and Expenditure data 2011-2012)
		Day 2020	Self-reported questionnaire (CSRI (modified))	Not valued		
		Drost (2016), Netherlands	Self-reported web-based questionnaire (not provided)	Not reported	Not reported	Costing guidelines (Dutch manual for intersectoral costs and benefits of (preventive interventions))

Item	Number of studies	First author (year), country	Measurement method (instrument)	Unit cost (currency, year)	Unit	Source of unit cost
		Hakkaart van Rooijen (2007), Netherlands	Self-reported questionnaire (TiC-P)	Not valued		
		Jolstedt (2018), Sweden	Self-reported questionnaire (TiC-P)	61.2 (EUR, 2016)	Per school day	National-level public data obtained from government sources (National Agency for Education)
		Leijdesdorff 2020	Self-reported study-specific questionnaire (provided)	Not reported	Day	Own unit cost calculation based on national-level data (Statistics Netherlands)
		Lenhard (2017), Sweden	Self-reported questionnaire (adapted version of parent-rated TiC-P)	66.85 (USD, 2016)	Per child per day	National-level public data obtained from government sources (Swedish National Agency for Education)
		Oosterhoff (2020), Netherlands	Literature	27 (EUR, 2018)	School day	Costing guidelines (Dutch manual for intersectoral costs and benefits of (preventive) interventions)
		Reynolds (2011), US	Administrative data from public records	Not reported	Average longterm costs	National-level public data obtained from government sources (National Institution of Justice)
		Sampaio 2019	Self-reported questionnaire (TiC-P (modified))	73.77 (USD, 2016)	Day	Own calculation based on the national-level data (The Swedish National Agency for Education)
		Simon (2012), Netherlands	Self-reported data collected using standardized cost diaries (not provided)	4.8 (EUR, 2008)	Per hour	Own calculation based on the national-level data (National Institute of Budget Information)

Item	Number of studies	First author (year), country	Measurement method (instrument)	Unit cost (currency, year)	Unit	Source of unit cost
		Simon (2013), Netherlands	Self-reported data collected using prospective cost-diaries (not provided)	4.8 (EUR, 2008)	Per hour	Own calculation based on the national-level data (National Institute of Budget Information)
		Steenel (2013), Netherlands	Self-reported questionnaire (not provided)	4.88 (EUR, 2010) for regular education	Per hour	Own unit cost calculation based on national-level data obtained from government sources (CPB Netherlands Bureau for Economics Policy Analysis)
			Self-reported questionnaire (not provided)	9.75 (EUR, 2010) for special education	Per hour	Own unit cost calculation based on national-level data obtained from government sources (CPB Netherlands Bureau for Economics Policy Analysis)
		Van Der Kolk (2015), Netherlands	Self-reported questionnaire (TiC-P (both child and adult versions, modified))	Not valued		
Educational professionals						
Educational psychologist	4	Barrett (2011), UK	Self-reported questionnaire (CA-SUS (modified))	Not reported	Not reported	Costing guidelines
		Byford (2015), UK	Self-reported questionnaire (CA-SUS (modified), CARER-SUS (modified))	Not reported	Not reported	National-level public data obtained from government departments for school costs

Item	Number of studies	First author (year), country	Measurement method (instrument)	Unit cost (currency, year)	Unit	Source of unit cost
		Creswell (2019), UK	Self-reported data collected using parent diaries (not provided)	37.29 (GBP, 2011-2012)	Not reported	Own unit cost calculation based on the national-level data (Local Government Earnings Survey 2011/12)
		Roddy (2019), Ireland	Self-reported questionnaire (The Economic Costs for Families Raising a Child with an Autism Spectrum Disorder)	47.97 (EUR)	Per hour	National-level data obtain via personal communication with government officials (Special Education Section in the department of Education and Skills)
Attendance officer	2	Drost (2016), Netherlands	Self-reported web-based questionnaire (not provided)	40.61 (EUR, 2012)	Per hour	Costing guidelines (Dutch manual for intersectoral costs and benefits of (preventive interventions))
		Wansink (2016), Netherlands	Self-reported study-specific questionnaire (Dutch Services and Support Questionnaire (based on TiC-P))	40.61 (EUR, 2012)	Per hour	Costing guidelines (Dutch manual for intersectoral costs and benefits of (preventive interventions))
Special education needs coordinator	2	Barrett (2011), UK	Self-reported questionnaire (CA-SUS (modified))	Not reported	Not reported	Costing guidelines
		Byford (2015), UK	Self-reported questionnaire (CA-SUS (modified), CARER-SUS (modified))	Not reported	Not reported	National-level public data obtained via personal communication with government departments and from national surveys

Item	Number of studies	First author (year), country	Measurement method (instrument)	Unit cost (currency, year)	Unit	Source of unit cost
Education welfare officer	1	Creswell (2019), UK	Self-reported data collected using parent diaries (not provided)	20.44 (GBP, 2011-2012)	Not reported	National-level data (Local Government Earnings Survey 2011/12)
Family liaison officer (school)	1	Creswell (2019), UK	Self-reported data collected using parent diaries (not provided)	49 (GBP, 2011-2012)	Per hour of client work	Costing guidelines (Unit Costs of Health and Social Care 2012)
Home tutor	1	Byford (2015), UK	Self-reported questionnaire (CA-SUS (modified), CARER-SUS (modified))	Not reported	Not reported	National-level public data obtained from government departments for school costs
Interne special education teacher	1	Wansink (2016), Netherlands	Self-reported study-specific questionnaire (Dutch Services and Support Questionnaire (based on TiC-P))	Not reported	Not reported	Costing guidelines (Dutch manual for intersectoral costs and benefits of (preventive) interventions)
Portage worker	1	Byford (2015), UK	Self-reported questionnaire (CA-SUS (modified), CARER-SUS (modified))	Not reported	Not reported	National-level public data obtained from government departments for school costs
Remedial teacher	1	Vermeulen (2017), Netherlands	Standardized self-reported questionnaire (TiC-P (modified))	Not reported	Not reported	Dutch costing guidelines

Item	Number of studies	First author (year), country	Measurement method (instrument)	Unit cost (currency, year)	Unit	Source of unit cost
School counsellor	1	Arora (2020), Australia	Self-reported retrospective questionnaire (not provided)	21.77 (AUD, 2018-2019)	Per hour	National-level data obtained from government sources
Special needs assistant	1	Roddy (2019), Ireland	Self-reported questionnaire (The Economic Costs for Families Raising a Child with an Autism Spectrum Disorder)	18.4 (EUR)	Per hour	National-level data obtain via personal communication with government officials (Special Education Section in the department of Education and Skills)
Teacher	1	Creswell (2019), UK	Self-reported data collected using a parent diary (not provided)	35.41 (GBP, 2011-2012)	Not reported	National-level data obtained from government sources (School Workforce in England, November 2011)
Additional education support services						
Study/home work help	1	Jolstedt (2018), Sweden	Self-reported questionnaire (TiC-P)	47.5 (USD, 2016)	Per hour	Average market price
Tutoring help	1	Lenhard (2017), Sweden	Self-reported questionnaire (adapted version of parent-rated TiC-P)	53.06 (USD, 2016)	Per hour	National-level data obtained from government sources (Sweden's Municipalities and Counties)
Adapted school curriculum	1	Quintero (2018), Spain	Case report form	Included as intangible costs		
Reinforcement/tutoring lessons	1	Quintero (2018), Spain	Case report form	Included as intangible costs		

Item	Number of studies	First author (year), country	Measurement method (instrument)	Unit cost (currency, year)	Unit	Source of unit cost
Additional services to support education (special transport to school, special equipment, special training devices)	1	Roddy (2019), Ireland	Self-reported questionnaire (The Economic Costs for Families Raising a Child with an Autism Spectrum Disorder)	Not reported	Not reported	National-level data obtain via personal communication with government officials (Special Education Section in the department of Education and Skills)
Other special education supports	1	Thurston (2011), Canada	Self-reported questionnaire (inventory developed initially by Browne et al. (1990), currently updated to be the Expenditures for Health and Social Service Utilization Questionnaire (Browne et al. 2006))	Not reported	Not reported	Not reported
Specialized education services	1	Wansink (2016), Netherlands	Self-reported study-specific questionnaire (Dutch Services and Support Questionnaire (based on TiC-P))	Not reported	Not reported	Costing guidelines (Dutch manual for intersectoral costs and benefits of (preventive interventions))

Item	Number of studies	First author (year), country	Measurement method (instrument)	Unit cost (currency, year)	Unit	Source of unit cost
Additional services required for child's academic achievement	1	Zhao (2019), US	Self-reported study-specific questionnaire (The Impact Questionnaire)	Valued as the impact on caregivers' costs		
		Parental out-of-pocket expenses related to child's education				
Parental out-of-pocket expenses related to child's education	5	Genereaux (2015), Canada	Self-reported web-based survey (SCOPE (modified))	n/a	Per year	Costs taken at face value as reported by the respondents
		Le (2014), Netherlands	Literature	56 (EUR, 2012)	Annual per patient and total costs	Literature
		Roddy (2019), Ireland	Self-reported questionnaire (The Economic Costs for Families Raising a Child with an Autism Spectrum Disorder)	n/a	Cost per type of expense	Costs taken at face value as reported by the respondents
		Weimer 2019	Self-reported data collected in the interview with parents (instrument not provided)	n/a	n/a	Costs taken at face value as reported by the respondents
		Zhao (2019), US	Self-reported study-specific questionnaire (The Impact Questionnaire)	n/a	Approximate cost per academic tool	Costs taken at face value as reported by the respondents

Item	Number of studies	First author (year), country	Measurement method (instrument)	Unit cost (currency, year)	Unit	Source of unit cost
Aggregate societal expenditures of education	4	Konnopka (2007), Germany	National-level data (German statistical office)	Not reported	Not reported	National-level data (German statistical office)
		Konnopka (2009), Germany	National-level data (German statistical office)	Not reported	Not reported	National-level data (German statistical office)
		Le (2014), Netherlands	Literature	6085 (EUR, 2012)	Annual per patient and total costs	Literature
		Pelham (2007), n/a	Literature	4900 (USD, 2005)	Per year	National-level data obtained from government sources (US Department of Education)
School presentation	3	Jolstedt (2018), Sweden	Self-reported questionnaire (TiC-P)	61.2 (EUR, 2016)	Per school day	National-level data obtained from government sources (National Agency for Education)
		Lenhard (2017), Sweden	Self-reported questionnaire (adapted version of parent-rated TiC-P)	66.85 (USD, 2016)	Per child per day	National-level data obtained from government sources (National Agency for Education)
		Sampaio 2019	Self-reported questionnaire (TiC-P (modified))	73.77 (USD, 2016) multiplied by a coefficient	Per day	Own estimate based on publicly available data and literature
Disciplinary measures at school	2	Quintero (2018), Spain	Case report form	Included as intangible costs		
		Zhao (2019), US	Self-reported study-specific questionnaire (The Impact Questionnaire)	Valued as the impact on caregivers' costs		

Item	Number of studies	First author (year), country	Measurement method (instrument)	Unit cost (currency, year)	Unit	Source of unit cost
Mainstream school	2	Barrett (2011), UK	Self-reported questionnaire (CA-SUS (modified))	Not reported	Not reported	National-level public data obtained via personal communication with government departments and from national surveys
		Byford (2015), UK	Self-reported questionnaire (CA-SUS (modified), CARER-SUS (modified))	Not reported	Not reported	National-level public data obtained from government departments for school costs
Remedial teaching	2	Schawo (2015), Netherlands	Literature and expert opinion	58.49 (EUR, 2014)	Per visit	Standard prices obtained via website of the national association of remedial teachers
		Van Der Kolk (2015), Netherlands	Self-reported questionnaire (TiC-P (both child and adult versions, modified))	55 (EUR, 2012)	Per hour	Based on the average price per hour obtained from the website of the national association of remedial teachers
Financing of education	2	Genereaux (2015), Canada	Self-reported web-based survey (SCOPE (modified))	18,300 (CAD, 2014) or 36,600 (CAD, 2014) depending on the disability level	Per year	Costs taken at face value as reported by the respondents

Item	Number of studies	First author (year), country	Measurement method (instrument)	Unit cost (currency, year)	Unit	Source of unit cost
		Roddy (2019), Ireland	Self-reported questionnaire (The Economic Costs for Families Raising a Child with an Autism Spectrum Disorder)	840 (EUR) for primary school 191 (EUR) for post-primary school 1083.33 (EUR) for one time cost for ASD unit set-up	Not reported	National-level data obtained via personal communication with government officials (Special Education Section in the department of Education and Skills)
Aggregate savings for the education sector	2	Kuklinski (2020), US	National-level public data	Not reported	Lifetime costs averted	Washington State Institute for Public Policy model and software tool
		Reynolds (2011), US	Administrative data from public records	7596 (USD, 2007)	Per year per child	National-level data obtained from government sources (Illinois State Board of Education, 1997)
Adult education class	1	Day 2020	Self-reported questionnaire (CSRI (modified))	Not reported	Per hour per team member to NHS Community mental health team for adults with mental health problems	Costing guidelines

Item	Number of studies	First author (year), country	Measurement method (instrument)	Unit cost (currency, year)	Unit	Source of unit cost
Lost education owing to health problems (not valued)	1	Day 2020	Self-reported questionnaire (CSRI (modified))	Not valued		
General/specialist services at school	1	Domino (2008), US	Self-reported questionnaire (Child and Adolescent Services Assessment)	20.69 (USD, 2003)	Not reported	Literature
Productivity gains from school attainment	1	Weimer 2019	Self-reported data collected in the interview with parents (instrument not provided), administrative data from school records	estimated shadow price for a high school diploma of \$264 thousand (2017) (SD = \$56.5 thousand) Excluding externalities, the shadow price is \$190 thousand (2017) (SD = \$41.3 thousand)	Per high school diploma	Shadow price based on publicly available data and own estimations Washington State Institute for Public Policy 2014 Current Population Survey
Summer school	1	Buescher (2014), UK/US	Literature	Not reported	Not reported	Literature and government reports
Teacher training	1	Schawo (2015), Netherlands	Literature and expert opinion	76.05 (EUR, 2014)	Per visit	Government report (Tariffs AWEZ-institutions 2005)

Supplementary File 4. Summary of the general characteristics of the included studies

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Arora (2020), Australia	Intellectual disability	COI	Children aged 2-10 (mean age 5.6) with a diagnosis of ID or developmental delay	n/a	Societal	Costs to government, individual/family, and society including child's and caregiver's healthcare service utilisation, the child's pharmaceutical use, caregiver productivity losses, and time spent by the caregiver providing care because of the child's disability	School counsellor	Insufficient information
Barrett (2012), The United Kingdom	Autism	COI	152 children with autism, average age 44.8 months (SD=7.9, range=24-60 months)	n/a	Service and wider societal costs	Accommodation, hospital based health services, community health and social services, medication, voluntary sector services, education and childcare, out-of-pocket expenses, productivity losses	Educational psychologist, special educational needs coordinator, mainstream school, specialist school	Insufficient information

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Beaulieu (2019), Canada	Pediatric abusive head trauma	EE+ COI	Children aged 0-24 months old with a definite diagnosis of abusive head trauma	Pediatric abusive head trauma prevention intervention vs care as usual before the implementation	Societal	Intervention costs, mortality, health care, social care, criminal justice, education, productivity, individuals	Special education	2.6
Bodden (2008) ^a , The Netherlands	Anxiety disorders	COI	74 girls (63%) and 44 boys (37%), range 8-17 years (M=12.4, SD=2.6) with a primary anxiety disorder (except for obsessive-compulsive and posttraumatic stress disorder)	n/a	Societal	health care costs, patient and family costs and costs in other sectors (productivity, education)	School absence	16.6

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Bodden (2008) ^b , The Netherlands	Anxiety disorders	EE	72 girls (62%) and 44 boys (38%), aged 8–17 years (M= 12.3, SD = 2.6) with primary anxiety disorder (except for obsessive-compulsive disorder and post-traumatic stress disorder)	12-session individual cognitive behavioral therapy (intervention) vs family cognitive behavioral therapy (control)	Societal	direct healthcare costs, direct non-healthcare costs, indirect costs and out-of-pocket costs	School absence	Intervention : 1.7 Control: 14.2
Bodden (2018), The Netherlands	Depression	COI	49 girls (87.5%) and 7 boys (12.5%) with a age range of 12 to 21 years (M=16.2, SD= 2.07), a primary diagnosis of Major Depressive Disorder or Dysthymic disorder	n/a	Societal	Direct health care, direct non health care (professional and informal care), indirect (parents' productivity, school absence, costs of substance abuse), out of pocket expenses	School absence	10.17

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Buescher (2014), The United Kingdom/The United States	Autism spectrum disorder	COI	Based on accepted prevalence estimates from each country. UK: 10% of children aged 0-3 years with ASDs have their condition diagnosed and receive some type of service in response (0.1% of all children in this age group), and 1.1% of children of all other ages. US: assumed 1.1% of children and adults had ASDs and were treated	n/a	Societal	Accommodation, medical services, non-medical services (special education, treatment for autism spectrum disorder-related needs (in the United Kingdom), child care, special programs, after-school care, day care, weekend programs, summer school, overnight and other respite, travel to medical appointments, home care modifications, and damage replacement), and out-of-pocket payments by families	Special education, summer school	UK 2-3 with ID: 24.63 UK 4-11 with ID: 34.57 UK 12-17 with ID: 65.24 UK 2-3 wo ID: 37.34 UK 4-17 wo ID: 47.05 US 0-5 with ID: 58.33 US 6-17 with ID: 32.63 US 0-5 wo ID: 49.71 US 6-17 wo ID: 26.78 UK adults with ID: 3.04 UK adults wo ID: 6.9

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Byford (2015), The United Kingdom	Autism	EE	Families with a child aged 2 years to 4 years 11 months; n=74 (intervention), n=69 (control), mean age = 4 (2-5)	Pre-school autism communication trial intervention vs treatment as usual	Service, societal	health, social and education services, parental out of pocket expenditure, productivity losses and informal care	Educational psychologist, special education needs coordinator, portage worker, mainstream school, specialist school, home tutor	Insufficient information

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Creswell (2019), The United Kingdom	Anxiety disorders	EE	211 children, 85% White British, aged 7–12 years (mean age = 10.22, SD = 1.58), with a current anxiety disorder, together with their mothers, who also had a current anxiety disorder	(a) child cognitive behaviour therapy plus control interventions to balance for therapist time (CCBT+Con); (b) CCBT plus cognitive behaviour therapy for maternal anxiety disorder (CCBT+MCBT); or (c) CCBT plus treatment focused on the mother–child interaction (CCBT+MCI)	NHS, societal	Intervention costs, school absence, time off work and lost leisure time (for mothers), use of non-NHS services (e.g. educational services) and personal costs of medications	School absence, use of educational services (education welfare officer, educational psychologist, family liaison officer, teacher)	Could only be calculated for school absence: CCBT+MCB T: 5.42 CCBT+MCI: 4.09 CCBT(+Con): 6.02

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Day (2020), the United Kingdom	Mental health problems	EE	48 children aged with the mean age of 7.8 (2.2) experiencing emotional and/or behavioural difficulties and their parent(s)	16-week psychoeducational program on parenting "Helping Families Programme - Modified" vs standard care augmented by a single psychoeducational session	National Health Service/ Personal Social Services and societal	Health care, social care, education, criminal justice, benefits, intervention	Adult education class, lost education owing to health problems (not valued), days off school (not valued)	Intervention group: 2 time point - 3.7; 3 time point - 0.2; Control group: 2 time point - 0.9; 3 time point - 3.7
Domino (2008), The United States of America	Depression	EE	The sample (N=369) included adolescents ages 12-18 (mean=14.6 years) and was 57% female with a primary DSM-IV diagnosis of major depressive disorder	Fluoxetine alone, CBT alone, combination therapy, or medical management with pill placebo	Societal	Medication, intervention, inpatient hospital use, education services, time and travel costs for caregivers	General/specialist services at school (e.g. school-based counselling)	Insufficient information

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Drost (2016), The Netherlands	Substance abuse	EE	Dutch adolescents (n = 2493, aged 15-19 years, mean age 16.3 (1.2)) attending school	Game on alcohol awareness vs care as usual	Healthcare, societal	Intervention costs, health care costs, intersectoral costs (education, criminal justice, labour and social security, household and leisure), costs of substance use	School absenteeism, contacts with an attendance officer	Intervention : 15.31 Comparator: 26.3

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Ericson (2017), Sweden	Fetal alcohol spectrum disorder	COI	The proportion of individuals with the secondary disabilities with a verified diagnosis of FAS. The calculations are based on individuals up to the age of 64 years, and are divided by age into children (0-17 years) and adults (18-64 years). The FAS group (n = 79; mean age 32 years) was composed of individuals who were all diagnosed with FAS, when they were infants or children. An additional comparison group (n = 3160) matched by age, gender, and place of birth was also included.	n/a	Societal	Direct (societal support - housing, special education, costs of psychiatric disorders and substance abuse), Indirect (productivity, informal care)	Special education school (including school transport and travel expense compensation) Mentioned, but not included due to insufficient information: special needs teacher and personal assistance	17.87 (for children with fetal alcohol spectrum disorder 0-17) 0 (for adults with fetal alcohol spectrum disorder 18-64) 15.88 (for children with fetal alcohol spectrum disorder 0-17) 0 (for adults from the comparison group 18-64)

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Ganz (2007), The United States	Autism	COI	US population with autism based on the prevalence data	n/a	Societal	Health and social care, education, productivity	Special education	4.76
Generaux (2015), Canada	Intellectual disability disorder	COI	Convenience sample of 206 British Columbian parents of children aged 1 through 18 years, referred to Children's & Women's Health Centre of British Columbia with confirmed IDD of unknown cause. There were 33 mild, 30 moderate, and 17 severe/profound diagnoses with mean ages of 6.7, 6.3, and 7.3 years, for each severity level, respectively.	n/a	Parental, societal	Government benefits, non-hospital therapies, child care, family (time lost, travel, income loss, out of pocket expenses)	School subsidy (educational supplements for special needs children), parental expenses on education (school, preschool, and respite-related expenses)	66.72

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Greenmyer (2018), The United States/Canada/Sweden/New Zealand	Fetal alcohol spectrum disorder	COI	Children and adults	n/a	Economic impact for individual, caregivers and society	Health care, residential care, special education, criminal justice system, productivity losses, and intangible losses	Special education	All ages: 30.16 Children: 19.94
Hakkaart van Rooijen (2007), The Netherlands	Attention deficit hyperactivity disorder	COI	Group of 70 children who were being treated by a paediatrician for ADHD, 60 children with no behaviour problems, and 35 children with behaviour problems ADHD: 10.5 (2.7) Behavioral problems: 9.3 (1.0) No behavior problems: 7.8 (1.0)	n/a	Societal	Direct (health care, out of pocket) and indirect (productivity) costs, school absenteeism, failure to participate in sports	Health-related school absenteeism	Education costs were not valued

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Honeycutt (2004), The United States	Mental retardation	COI	Hypothetical cohort of persons born in 2000 with mental retardation, cerebral palsy, hearing loss or vision impairment	n/a	Societal	Direct medical and non-medical (home/automobile modifications, special education), indirect (productivity)	Special education	42%-82% of total direct costs (reported by the authors, not included in the synthesis)
Jarbrink (2007), Sweden	Autism spectrum disorder	COI	children aged 1-18 years who had previously been identified as having autistic spectrum disorder and living in Härryda municipality; 26 of 33 were boys, the average age being 11.6 years with a range of 4 to 18 years	n/a	Societal	Health care, community support, schooling, informal care, out of pocket expenses	Schooling (including personal assistance within school, school transport, aid and special diet) of different types (special training school, residential school or school at a treatment home)	50.63

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Jarbrink (2001), The United Kingdom	Autism	COI	UK population with an assumed prevalence of autism of 5 per 10,000	n/a	Societal	Health and social services, living support, voluntary support, special education, medication, sheltered work, day care provision, productivity losses for people with autism, family members' time costs (the time costs for people with autism was not included), family expenses	Special education	6.7
Jolstedt (2018), Sweden	Anxiety disorders	EE	Children 8-12 (mean age=9.95, SD=1.39)	Therapist-supported and parent-assisted internet-delivered cognitive behaviour therapy vs internet-delivered child-directed play	Societal	Intervention, healthcare, education (supportive resources and productivity), dietary supplements, parents' productivity	Child's absence from school, presenteeism when feeling ill, study / homework help	Insufficient information

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Konnopka (2007), Germany	Alcohol consumption	COI	German population, n=8139, aged 18-59	n/a	Societal	Direct medical (health care and out of pocket expenses) and non-medical (rescue services, administration, prevention, research, education, and investments), indirect (productivity, early retirement and mortality)	Education expenditures	Insufficient information
Konnopka (2009), Germany	Alcohol consumption	COI	German population from 15 years old onwards	n/a	Societal	Direct medical and non-medical (preventative care, paramedics, transportation to hospital, administration, research, education, medical infrastructure, other institutions, and associated private household costs) and indirect (productivity, early retirement, and mortality)	Education expenditure	Insufficient information

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Kuklinski (2020), The United States	Child abuse and neglect	EE	Children up to 5 years old	"Promoting First Relationships" - 10-week home-visiting intervention for caregivers of children from birth to age 5 years that aims to increase caregiver sensitivity and responsiveness to children's behavioral and emotional needs vs provision of some support to vulnerable control arm families, offering three resource and referral calls	Societal	Intervention costs, monetized intervention impacts (health care, social care, education, crime, productivity, intangible costs for individuals (pain, suffering, psychological distress, diminished quality of life), premature mortality)	Aggregate savings for the education sector (reduced special education placement and grade retention)	Scenario 2&3 60% - 2 80% - 2.1 100% - 2.2

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Le (2014), The Netherlands	Attention deficit hyperactivity disorder	COI	Children and adolescents (7-17) with ADHD in the Netherlands	n/a	Societal	Health care, productivity, education, social services	Costs to parents for extra lessons, education system costs (special school needs, time commitment from teachers, social workers, counsellors, etc. and services of educational psychologists and school/doctors)	52.34 (range 42.4 - 62.28)
Leijdesdorff (2020), The Netherlands	Mental problems	COI	Youth 12-25 years old (mean age 20.5) with problems around mental health and well-being	n/a	Societal	Health care, education	Number of days skipped school	78.5

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Lenhard (2017), Sweden	Obsessive compulsive disorder	EE	12 to 17 years of age (n=67, 46% girls, mean age 14.60 (1.71))	web-based, therapist-guided and parent-assisted CBT intervention with treatment components in line with clinical expert guidelines for OCD treatment vs waitlist	Healthcare, societal	Intervention, healthcare (healthcare use and medicines), education (absenteeism, presenteeism, tutoring), personal expenses	Private tutoring, absenteeism from school, academic productivity loss when being at school despite not feeling well	Insufficient information

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Oosterhoff (2020), The Netherlands	Health promotion	EE	Children 4-12 years old	"Healthy Primary School of the Future" and "Physical Activity School" (PAS) - health promotion initiatives at primary schools in the Netherlands vs regular school curriculum	Societal	Intervention, healthcare, education, productivity	School absenteeism	Regular school curriculum 0.7 HPSF 0.7 PAS 0.7
Pelham (2007)	ADD/ADHD	COI	Samples of children and adolescents with ADD/ADHD	n/a	Societal	Healthcare providers and insurers, education, other non-health systems in the public sector (juvenile justice), and other members of society (victims of crime, children in the same classroom, taxpayers)	Aggregate education costs including special education services, school counselling, (grade) retention, disciplinary referrals	32

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Penner (2015), Canada	Autism	EE	Toddlers from 2 y/o modelled until they are 65 y/o	<ol style="list-style-type: none"> 1. Intensive Early Start Denver Model - ESDM (ESDM-I) 2. Pre-diagnosis parent-delivered ESDM (ESDM-PD) 3. Care as usual 	Provincial government, societal	Intervention costs, provincial costs (health and social care, education), productivity losses, caregiver costs	Special education	Insufficient information
Peterson (2018), The United States	Child abuse and neglect	EE	Current US population	<ol style="list-style-type: none"> 1. Child-Parent Centers 2. Nurse-Family Partnership 3. No program 	Payer (government), societal	Program costs, cost of parents' time to attend program sessions and lifetime work productivity gains, costs averted (special education, health care, CJ, lost productivity)	Special education costs	3.77 for each of the three scenarios

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Piccininni (2017), Canada	Autism spectrum disorder	EE	The mean starting ages were determined to be 5.24 years for the CWT scenario, 3.89 years for the RWT scenario, and 2.71 years for the EWT scenario	Reduced wait time Eliminated wait time Current waiting time	Provincial government, societal	Public funding for education, health, and social services, costs to families, including costs associated with caregiving and lost productivity	Special education	Insufficient information
Popova (2016), Canada	Fetal alcohol spectrum disorder	COI	Canadian population	n/a	Societal	health care, law enforcement, children and youth in care, special education, supportive housing, long-term care, prevention and research, productivity losses of individuals with FASD due to their increased morbidity and premature mortality	Special education	3.26 (lower estimate: 4.21, upper estimate: 2.31)
Quintero (2018), Spain	Attention deficit hyperactivity disorder	COI	321 children with ADHD, mean age 12.7 (2.9, 6.3-18)	n/a	Healthcare system, societal	Direct medical, direct non-medical (transport), indirect (patient and family costs, productivity)	Adapted school curriculum, reinforcement/tutoring lessons, expulsion, grade repetition	Education costs were not valued

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Reynolds (2011), The United States	Well-being of less privileged population groups	EE	1,373 participants with known educational attainment by August 31, 2005 (average age of 25.5 years)	Child-Parent Center that provides services up to age 9 for economically disadvantaged children vs usual early intervention	Societal and public benefits	Savings associated with education, criminal justice, maltreatment, averted tangible and intangible expenditures to crime victims, reductions in health and substance abuse treatment, and increases in projected earnings and tax revenues as a result of higher educational attainment	Savings in school remedial services (grade retention, special education), missed school days (as part of victims' tangible and intangible costs)	Preschool program: 7 School-age program: 29.1 Extended program: 13.89 (benefits)

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Roddy (2019), Ireland	Autism spectrum disorder	COI	195 parents of 222 children aged between 2 and 18 years of age with a clinically diagnosed autism spectrum disorder	n/a	Societal	Direct costs (health service utilization, medication costs, educational resources related to autism spectrum disorder), parental out-of-pocket expenditures (living costs, health and social care use, education, travel, training/support, autism assistance dogs, and other), loss of parental earnings, informal care	Schooling, additional services to support education (special transport to school, special equipment, special training devices), special needs assistant, educational psychologist, special needs financing, parents' out of pocket expenses on education	30
Sampaio (2019), Sweden	Irritable bowel syndrome	EE	101 adolescents aged 13-17 with the diagnosis of irritable bowel syndrome	Exposure-based internet cognitive behavioral therapy vs waitlist	Societal	Health and healthcare use, medication, productivity losses for parents and children, intervention costs	Productivity losses associated with absenteeism and reduced efficiency at school by adolescents	Intervention group Follow-up: 44-3 Control group Follow-up: 54-5

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Schawo (2015), The Netherlands	Attention deficit hyperactivity disorder	EE	Patients (with suboptimal response to IR) enter a model at 6 years	MPH osmotic-release oral system vs short-acting MPH immediate-release	Societal	Direct medical and nonmedical costs as well as spillover effects on caregivers Patient: medication, consultation, special intervention, special education Caregiver: medical costs, production losses	Special education, costs for teacher training, remedial education	Insufficient information
Simon (2012), The Netherlands	Anxiety disorders	EE	Parents and children (average age of 10) who scored in top-15% on the Screen for Child Anxiety Related Emotional Disorders-71	Child-focused intervention Parent-focused intervention No intervention	Societal	direct healthcare, direct non-healthcare (child care, informal care, professional help at home), indirect (parental productivity, child's school absence), and out-of-pocket costs (medication, transportation, alternative care)	Child's school absence	CI: 7.87 PI: 9.9 NI: 11.11

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Simon (2013), The Netherlands	Anxiety disorders	EE	primary schools in the Netherlands (age 8-12), high- (mean age 9.82, SD=1.19) and median-anxious (mean age 10.31, SD=1.15) children were selected	1. applying the screening procedure and offering the child-focused intervention 2. applying the screening procedure and offering the parent-focused intervention 3. applying the screening procedure and differentially offering the parent- or child-focused intervention, depending on parental anxiety 4. doing nothing	Societal	Direct health care and non-healthcare costs, indirect costs and out of pocket costs	School absence	Insufficient information

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Steensel (2013), The Netherlands	Autism spectrum disorder, (comorbid) anxiety disorder	COI	194 children aged 7-18 years	n/a	Societal	Health care and non health care costs (social care, education, productivity, out of pocket, informal care, time)	Special education/extra help at school, school absence (regular education, special education)	ASD: 13 AD: 17.57 CON: 30.74
Thahn (2009), Canada	Fetal alcohol spectrum disorder	COI	Children born with FASD	n/a	Societal	medical, education, social services, direct costs to the patient/family, productivity losses, and externalizing behaviors	Special education	33 (as reported by the authors, was not included in the synthesis)

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Thurston (2011), Canada	Psychiatric distress	COI	The average child age at interview was 7.94 years with 67% of the sample being male. The average guardian was 40 years.	n/a	Not specified	Health and social care, education, out of pocket expenses	Special education services, other special education supports	K10 (measure of psychiatric distress) equal or above 20: 0.03 (for guardian), 5.7 (for child) K10 below 20: 0 (for guardian), 5.9 (for child)
van der Kolk (2015), The Netherlands	Attention deficit hyperactivity disorder	COI	618 questionnaires of children (11.8 (8–18)) and 590 questionnaires of parents (43.2 (30–63))	n/a	Societal	Health and social care, education, productivity, justice, family costs	Remedial teaching at school, special education (primary and secondary), absence from school	18.14

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
van der Schans (2015), The Netherlands	Attention deficit hyperactivity disorder	EE	patients who were sub-optimal responders to IR-MPH treatment due to adherence problems with the multiple doses short-acting regimen (3-5 hours)	use of ER-MPH in patients who were sub-optimally treated with IR-MPH (switching to ER-MPH) compared to sub-optimally treated patients with ADHD remaining on IR-MPH	Societal	Direct (consultation, intervention, medication, and special education costs) and indirect (productivity)	Special education costs	IR-MPH: 24.73 MPH-OROS: 21.32 Medikinit CR/ Equasym XL: 22.58
Vermeulen (2017), The Netherlands	Antisocial problems	EE	Fifty-one adolescents were included in the MST group (16 (1.36)) and 65 in the TAU group (16 (1.17)) and parents	Multisystemic therapy vs treatment as usual	Societal	Direct (health and social care, education, out of pockets, justice) and indirect (productivity, travelling)	Remedial teacher	Insufficient information

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Wansink (2016), The Netherlands	Mental illness	EE	Ninety-nine families were included and randomly allocated to either PBCM (n = 49) or to the control condition (n = 50). Of the 49 families allocated to PBCM, 38 (77 %) actually did receive the intervention.	Preventive basic care management a preventive program targeting threats to parenting quality vs care as usual	Health care, social care and societal	intervention costs, healthcare costs, childcare costs, and other inter-sectoral costs (education, criminal justice, and services for debt restructuring)	Special education, specialized educational services, attendance officer, interne special education teacher	Intervention : 6.25 Control: 7.34

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Weimer (2019), The United States of America	Substance use disorders	EE	294 adolescent students who received substance use treatment with the mean age of 16.3	Recovery high school attendance after receiving treatment for substance use disorder (schools that provide educational and therapeutic support to students with treated substance use disorders) vs regular school attendance after receiving treatment for substance use disorder	Societal	Intervention costs (incremental costs of recovery high schools), parental expenditures on healthcare and education, parental productivity losses, productivity gains from school attainment (included as benefit), crime reduction (included as benefit), healthcare costs avoided (included as benefit)	Parental out-of-pocket expenses on education, productivity gains from school attainment	Insufficient information

First author, year, country	Condition	Design	Population	Intervention/comparator	Perspective	Cost categories	Education costs	% of the education costs
Zhao (2019), The United States	Attention deficit hyperactivity disorder	COI	Two cohorts of participants were recruited at age four to six. A total of 56 parents in the ADHD group and 30 parents in the control group completed the current study.	n/a	Not specified	Direct: education, criminal justice, out of pocket, Indirect: productivity, income loss, out of pocket expenses	Expenses related to academic support (e.g. purchases of educational software, tutoring, extra books), additional services required for child's academic achievement, discipline problems at school (suspension, detention, expulsion)	With ADHD: 4.6 Comparison group: 18.05

Chapter 3. Exploring the Identification, Validation and Categorization of Costs and Benefits of Education in Mental Health: The PECUNIA Project

Chapter 3 draws upon:

Pokhilenko, I., Janssen, L.M., Evers, S.M., Drost, R.M., Simon, J., König, H.H., Brodzky, V., Salvador-Carulla, L., Park, A.L., Hollingworth, W.W. and Paulus, A.T., 2020. Exploring the identification, validation, and categorization of costs and benefits of education in mental health: The PECUNIA project. *International Journal of Technology Assessment in Health Care*, 36(4), 325-331. <http://dx.doi.org/10.1017/S0266462320000203>

Abstract

Background. Mental health problems can lead to costs and benefits in other sectors (e.g., in the education sector) in addition to the healthcare sector. These related costs and benefits are known as intersectoral costs and benefits (ICBs). Although some ICBs within the education sector have been identified previously, little is known about their extensiveness and transferability, which is crucial for their inclusion in health economics research.

Objectives. The aim of this study was to identify ICBs in the education sector, to validate the list of ICBs in a broader European context, and to categorize the ICBs using mental health as a case study.

Methods. Previously identified ICBs in the education sector were used as a basis for this study. Additional ICBs were extracted from peer-reviewed literature in PubMed and grey literature from six European countries. A comprehensive list with unique items was developed based on the identified ICBs. The list was validated by surveying an international group of educational experts. The survey results were used to finalize the list, which was categorized according to the care atom.

Results. Additional ICBs in the education sector were retrieved from ninety-six sources. Fourteen experts from six European countries assessed the list for completeness, clarity and relevance. The final list contained twenty-four ICBs categorized into input, throughput and output.

Conclusion. By providing a comprehensive list of ICBs in the education sector, this study laid further foundations for the inclusion of important societal costs in health economics research in the broader European context.

Introduction

Mental and psychosocial problems have a broad societal impact on sectors outside health care.¹ As the age of onset of mental problems often coincides with the critical period of establishing educational trajectories,² the economic impact of these problems on the education sector can be particularly substantial.³ Several cost of illness studies have shown that costs in the education sector can constitute a considerable part of the total societal costs of mental and psychosocial problems.^{4,5} Therefore, the interventions through which children and adolescents with mental and psychosocial problems are targeted are also likely to impact on the resource-use in the education sector. Such costs and benefits that are associated with health interventions but are incurred outside the healthcare sector are known as intersectoral costs and benefits (ICBs). Inclusion of ICBs could be particularly important for economic evaluations conducted from a societal perspective, in order to determine all relevant costs and outcomes associated with an intervention taking place in a broader societal context.⁶

Among other factors, the lack of validated methodological guidance in the health economic literature has resulted in little emphasis being placed on taking ICBs in the education sector into account in health economics research, in comparison with other sectors, such as informal care or productivity, for which methods and tools are widely available. This is the case, even though in the context of mental health and psychosocial interventions for children and adolescents, education ICBs can be particularly significant.^{1,3-5,7} Overlooking these ICBs in economic evaluations can have serious implications for the validity of the results, particularly in the context of interventions aimed at youngsters, as their health status has a more significant impact on the resource-use in the education sector, in comparison with older population groups. Furthermore, mental health related ICBs of younger people in the education sector can even outweigh their resource-use in the healthcare sector.^{7,8}

Although research into ICBs is limited, the importance of including ICBs in the education sector in health economics research is highlighted in the literature⁹ and in several national pharmaco-economic guidelines.^{10,11} Furthermore, Drost et al.¹² developed an overview of ICBs associated with mental health interventions and distinguished five non-healthcare sectors, including the education sector, in which ICBs can be incurred. This scheme could be useful for the identification of relevant ICBs, as the first step of the costing process. In addition, several methods for the measurement and valuation of ICBs in the education sector are available.^{13,14} Nevertheless, current methods focus only on a limited number of education ICBs. Similarly, in the few health economic evaluation studies that did incorporate ICBs in the education sector, the number of items was also limited.^{8,15} In addition, while the scheme by Drost et al.¹² is intended for international use, its transferability has not been properly tested in a broader international context. This is of great importance for the ICBs in the education sector in particular, as, although valid transferable tools for the identification, measurement and valuation of ICBs in this sector are needed, they are scarcely available. Furthermore, proper classification of ICBs in the education sector is still lacking, while this could provide additional value for the development of a consistent harmonized approach for the inclusion of ICBs in health economics research and foster comparability between studies.

To address these knowledge gaps and to make the first steps towards the development of validated tools for including ICBs in the education sector in health economics studies, the objective of this study was threefold. First, this study aimed to further identify education ICBs attributed to mental health interventions. Second, this study aimed to validate the list of the identified ICBs in a broader European context. Third, this study aimed to categorize the identified ICBs as the first step towards the development of a formal classification.

Methods

Context: The PECUNIA Project

This study was conducted alongside the ProgrammE in Costing, resource-use measurement and outcome valuation for Use in multi-sectoral National and International health economic evaluations (PECUNIA) project, which aims to tackle the healthcare challenges of an ever-growing and rapidly ageing population in the EU by developing new standardised, harmonised and validated methods and tools for the assessment of costs and outcomes in European healthcare systems.¹⁶ The PECUNIA consortium is a network of ten institutions in six European countries (Austria, Germany, Hungary, Spain, the Netherlands, and the United Kingdom (UK)) with relevant complementary methodological expertise. Three mental disorders (depression, schizophrenia, and post-traumatic stress disorder (PTSD)) with high disease, societal and economic burdens were selected as exemplars for service identification, to address methodological issues and to contribute to major public health challenges with highly needed applied evidence. Methodological choices for this study were made in collaboration with the international health economics and health technology assessment experts within the PECUNIA consortium. For this study, the education sector was defined as encompassing all education services provided to primary and secondary level students with (the risk of developing) mental or psychosocial problems.

Identification

Drost et al.¹² identified fourteen ICBs in the education sector attributed to mental health (preventive) interventions, which were used as a basis for this study. To identify additional ICBs, a search of peer-reviewed and grey literature was conducted in the autumn of 2019. The methodology of the peer-reviewed literature search for this study was similar to the one used by Drost et al.¹². The search was conducted in PubMed; the search strategy is available on request to the corresponding author. Based on the number of hits (13,137) and the scope of the PECUNIA project, the search was not extended to additional databases and was further limited to the articles pertaining to depression, PTSD, and schizophrenia. To complement the search with potentially relevant national sources and to increase the transferability of the results, grey literature sources (e.g. ministry reports, national (costing) guidelines) were reviewed by co-authors in PECUNIA partner countries. The full texts of peer-reviewed and grey literature sources were reviewed and the studies that did not contain education ICBs were excluded. The literature search generated a pool of ICBs, which were compiled in a list. Similar or duplicated items were clustered under one heading using the classification scheme of Drost et al.¹² as a reference. Items that referred to healthcare services provided in a school setting (e.g. a school nurse) and country-specific mental health promotion programs were excluded. As a result, an expanded draft list of ICBs in the education sector was developed.

Validation

To validate the transferability of the list of ICBs in the international context, an expert survey was conducted (Supplementary File 1). The aim of the survey was to assess the clarity and relevance of the identified ICBs, and to assess the completeness of the draft list. The survey was conducted with an international group of education experts in the six PECUNIA countries between November 2018 and January 2019. Experts could be either researchers or professionals in the field of education, i.e. with knowledge of and/or experience in the education sector. The survey was administered via email in the form of an Excel file. The file contained instructions for completing the survey, survey questions, and the list of ICBs. The ICBs were accompanied by short descriptive definitions, which were developed by the co-authors with the aim of clarifying the meaning of the items to the experts. The email also contained a consent form, in which the experts could give their permission to be acknowledged on the project website and in any publications resulting from the survey. The estimated time to complete the survey was thirty minutes. To analyse the responses regarding the clarity and relevance of the ICBs, the answers were summarised to indicate the number of positive, negative, neither positive nor negative (which were regarded as unclear), and missing responses. To assess the completeness of the list, the items suggested by the experts were listed alongside the draft list of ICBs and clustered with previously identified items. The feedback provided by the experts was used to finalize the list of ICBs. It is important to note that due to the exploratory nature of the study, the aim of the expert survey was to gain insight into the experts' opinions about the list of identified ICBs. Hence, the responses were treated as qualitative rather than quantitative data. The survey results were used to finalise the list of ICBs in project team discussions.

To optimize the final list of ICBs via group discussions with relevant health economic experts, it was presented and discussed at multiple international conferences, including the Fourteenth Workshop on Costs and Assessment in Psychiatry (Venice, Italy), the 11th edition of the Lowlands Health Economic Study Group conference (Almen, The Netherlands), the Health Technology Assessment international 2019 Annual Meeting (Cologne, Germany), and the International Health Economic Association 2019 Congress (Basel, Switzerland).

Categorization

Within the PECUNIA project, a conceptual framework for semantic standardization, the PECUNIA care atom, was developed to categorize resource-use items, including the ICBs in the education sector.¹⁷ The draft list was made up of mixed concepts and terms (e.g. services, interventions, outcomes) that needed to be defined and organised in conceptually harmonized clusters with the aim of facilitating the comparability of health systems analyses while taking their complexity into account. The PECUNIA care atom describes the minimum units of analysis of the three phases of the care delivery process: input, throughput, and output. Inputs encompass the resources introduced in a mental healthcare system including services, staff, equipment, consumables and facilities. Throughputs refer to the resource utilization by consumers and include activities resulting from the delivery of services (e.g. interventions). Outputs refer to the consequences of using care resources such as changes in functioning, morbidity and mortality associated with mental health and psychosocial problems. For the

current study, the final list of ICBs in the education sector was categorized according to this framework.

Results

Identification

The search in PubMed yielded 13,137 hits. After title and abstract screening, 12,941 records were excluded. Full texts of the remaining 196 studies were reviewed and 113 studies were excluded because they did not contain ICBs in the education sector. In addition, thirteen out of ninety-eight reviewed international grey literature sources were included. Ultimately, ICBs in the education sector were extracted from ninety-six peer-reviewed and grey literature sources. The list of peer-reviewed sources is available in Supplementary File 2. Out of the eighty-three selected peer-reviewed sources, twenty focused on mental health in general, thirty-three on depression, five on PTSD, nineteen on schizophrenia, four on depression and anxiety, one on PTSD and depression, and one on schizophrenia and depression. Out of the thirteen grey literature sources, four sources were websites of public organizations, three sources were national guidelines, three sources were ministry reports, one source was an unpublished article, one source was a Master's thesis dissertation, and one source was a piece of national legislation. The literature search is outlined in Figure 1.

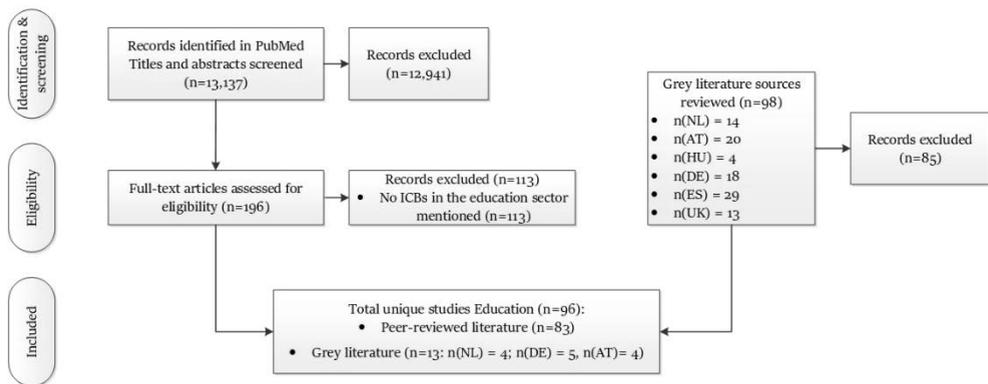


Figure 1. Flowchart of the literature search and the development of the list of intersectoral costs and benefits (ICBs); abbreviations: the Netherlands (NL), Austria (AT), Hungary (HU), Germany (DE), Spain (ES), the United Kingdom (UK)

The ICBs retrieved from the literature were clustered in a comprehensive draft list with thirty-eight unique ICBs (Supplementary File 3).

Validation by Experts

Out of thirty-seven experts invited to participate, fourteen experts (38%) completed the survey. Each participating country recruited at least one expert. The highest number of responses (n=4) was obtained in Austria, while the lowest number of experts (n=1) was recruited in Hungary and Germany. Information on the experts' background and the number of responses per question is presented in Supplementary File 4.

Fourteen experts commented on the clarity of the ICBs. Only few missing responses were registered. All items were found to be clear by at least eight experts. Twenty-one items were found clear by at least twelve experts. Seventeen items were found unclear by three of four experts. The least clear items were “learning therapy”, “student counselling”, and “low school attainment/productivity/performance”. Four experts mentioned that the difference between “learning therapy” and “special needs diagnostics” was ambiguous, while it was also unclear what “student counselling” entails and why the item “low school attainment/productivity/performance” contained multiple terms under one heading.

Although fourteen experts commented on the relevance of the ICBs, the responses of one expert were excluded as his/her responses to all but one item in this section of the survey were missing. Apart from this, few missing responses were registered. The experts found most items relevant. Thirty-one items were found relevant by twelve or more experts. Over half of the experts found the items “night school” and “attendance officer” the least relevant. The item “night school” was found to be less relevant because it is applicable to adults rather than to younger students. The item “attendance officer” was deemed irrelevant due to cross-country differences. According to the Austrian and German experts, absenteeism is dealt with by regular schools, and there are no special officers responsible for this task.

Eleven experts commented on the completeness of the list. Five experts found the list of ICBs to be complete. Six experts suggested fifteen additional items. The majority of the suggested items had either a degree of overlap with the previously identified ICBs (e.g. “additional lessons” or measures to improve inclusivity in regular classrooms) or could not be allocated to the education sector (e.g. “clinic schools” or “joint work between teaching and health professionals”). Nevertheless, one unique relevant item “support and training services for teachers” was added to the final list based on the suggestions of the experts.

Finalization and Categorization of the List

In addition to “yes”/“no”/“I don’t know” answers, the experts provided qualitative comments regarding the clarity and relevance of each item. For example, it was emphasized that “*special education services can also be provided in a mainstream environment in the form of additional support*” and not only in a special education school. Another expert mentioned that “*social reintegration is not necessarily related to the school. Maybe call it school reintegration?*” In addition, for some ICBs, the experts mentioned alternative naming used in their country, such as “special need financing” in the UK instead of “student-related financing”. The comments were taken into account when finalizing the list of ICBs. The final list contained twenty-four ICBs, in comparison with thirty-eight ICBs in the draft list. Four items were excluded (“attendance officer”, “night school”, “indirect effect of premature school leave/dropout”, “school-based health promotion interventions”). One item was added based on the experts’ comments (“support and training services for teachers”). The overview of the experts’ comments and more details about the process of transforming the draft list of ICBs into the final list are presented in Supplementary File 3. Nine items were categorized as inputs, one item was categorized as throughput, and fourteen items were categorized as outputs according to the PECUNIA care atom. The final list of ICBs accompanied by short descriptions is presented in Table 1.

Table 1. Categorization of intersectoral costs and benefits in the education sector according to the care atom; ¹ICBs extracted from the scheme by Drost et al. (2013)(12), ²ICBs extracted from peer-reviewed literature, ³ICBs extracted from grey literature, ⁴ICBs added based on the expert survey; inputs - resources introduced in a mental healthcare system (e.g. services, staff, equipment, consumables, and facilities), throughputs - resource utilization and activities necessary for delivery of services (e.g. interventions), outputs - changes in functioning, morbidity and mortality associated with mental health and psychosocial problems (e.g. impaired functioning)

Intersectoral costs and benefits (ICBs) in the education sector	Description	Category
Special education school ¹	School for students who cannot be supported in regular school classes concerning personal development and achievements	Input
Additional education services/assistance in a regular school ¹	Education services provided to students with learning difficulties in a regular school in addition to regular curriculum (e.g. tutoring, extra time to complete a task)	Input
Home education ¹	Schooling provided in the home environment	Throughput
Educational therapy ³	Form of therapy used to treat individuals with learning differences, disabilities, and challenges	Input
Special needs diagnostics ³	Form of diagnostics to detect potential need for special education	Input
Student counselling ³	Counselling services for students who experience difficulties at school	Input
Counselling of legal guardians ³	Counseling of legal guardians concerning development, support and abilities of their child	Input
Student transport to education facility ³	Services aimed at transporting students to the education facility	Input
Student-related financing ³	Financial support for schools meant for students who experience learning disabilities for providing additional guidance and adapted lesson materials	Input
Training and support services for teachers ⁴	Training and support services for teachers who deliver education to students with mental problems	Input
Reduced school readiness ¹	Due to mental problems, student enters school less ready to engage in and benefit from early learning experiences that best promote the child's success	Output
Problems with school entry ¹	Delayed or fraught school entry due to child's mental problems	Output

Intersectoral costs and benefits (ICBs) in the education sector	Description	Category
Learning disabilities ¹	Impaired learning developing e.g. related to reading, spelling and/or calculating	Output
Reduced school adaptation ¹	Impaired adaptation to a school environment	Output
Reduced school competence ¹	Impaired academic capability due to mental problems	Output
Reduced school participation ¹	Impaired involvement in school activities	Output
Reduced school engagement ¹	Impaired academic achievement due to mental problems	Output
Reduced school attainment ¹ as a result of: <ul style="list-style-type: none"> - School drop out/leaving prematurely¹ - Refusal of admission³ - Exemption from compulsory education³ - Suspension³ - Change in the educational level³ 	Reduced level of completed education due to mental problems	Output
Reduced school productivity ¹	Presenteeism due to student's mental problems	Output
Reduced school performance ¹	Impaired academic achievement due to mental problems	Output
Grade retention ¹	Repetition of a school year in case a student is not entitled to advance to the next year	Output
Negative school experiences ¹ incl.: <ul style="list-style-type: none"> - negative peer relations (conflicts between students², conflicts between students and teachers¹) - discrimination² - negative feelings about school² 	Student's negative experiences at school due to e.g. conflicts with peers, discrimination or school dislike	Output
School (re-) integration ³	Measures taken by the school to promote integration after a students' time of absence due to illness or hospital stay	Output

Intersectoral costs and benefits (ICBs) in the education sector	Description	Category
Disruptive school behavior ²	Student's school behavior that disrupts learning process in class	Output

Discussion

This study aimed to identify relevant ICBs in the education sector, to validate the list of ICBs in a broader European context, and to categorize the ICBs using mental health as a case study. Based on these aims, a comprehensive list of ICBs in the education sector was developed that aims to support and to lay further foundations for the inclusion of important societal costs in health economics research. Although the case of mental health was used to develop the list, it is meant to be generic and applicable to other disease areas. Furthermore, as the list is based on the international literature and has been validated by an international group of experts, it can be used by researchers to select relevant cost items for health economic studies at both national and international levels in a broader European context.

Taking into account the far-reaching effect of health interventions on society is recognized to be a crucial aspect of the overall methodological quality of economic evaluations,¹⁸ as well as an important factor in national decision making.¹⁹ In line with previous research, the current study demonstrates that mental health problems have an (economic) effect on a wide range of sectors in society, including the education sector.^{3,15} This implies that ICBs in the education sector could constitute a large proportion of total disease costs for certain disease areas, including mental health, and, if relevant to the context of the study, should be included in economic evaluations. In comparison with the scheme of ICBs by Drost et al.¹² the current overview has been supplemented by the findings from the international grey literature, and its face validity has been assessed by education experts from within a broader European context. Furthermore, the results of this study complement the existing national pharmacoeconomic guidelines that recommend considering ICBs in the education sector in health economics evaluations.^{10,11}

The categorization of ICBs based on the PECUNIA care atom presents an important step towards the development of a formal ontology of ICBs in the education sector.²⁰ Such an ontology will subsequently serve as a basis for the development of standardized measurement and costing tools for the inclusion of these ICBs in health economics research, and ultimately contribute to improving the comparability of health economic analyses. The categorization also demonstrates the heterogeneity of the identified ICBs and implies that for each category of ICBs different measurement and costing approaches might be needed. While the methods for the measurement and valuation of input and throughput ICBs based on the quantities of resource-use and on the cost-price are relatively straightforward, quantifying ICBs categorized as output can be more complicated. Drost et al.¹⁴ developed methods for valuing ICBs, in particular those pertaining to changes in the level of economic losses and in the use of services. Nevertheless, for the valuation of the ICBs concerning other effects of mental health problems in the context of the education sector, alternative methods, for example, methods for eliciting

stated preferences,²¹ need to be considered. In addition, some ICBs categorized as output can be measured not only as a cost item but can also be incorporated in the health-related quality of life. This has implications for double counting in economic evaluations and researchers need to be aware of this potential bias.

As mentioned previously, mental and psychosocial problems can affect multiple non-health care sectors besides education. However, the boundaries of the sectors are not always clear. One of the ICBs included in the draft list, “the indirect effects of premature school leaving”, refers to the effect of lower school attainment due to a mental health problem on an individual’s success in later life (e.g. lower income, fewer job opportunities). While this item is linked to the education sector, it belongs to the labour sector, as it illustrates the reduced capacity of the individual on the job market. Similar issues were encountered during the literature search. ICBs in other sectors such as health care (e.g. school nurse/physician, education for hospitalized children) and patient and family (e.g. parental stress) were initially extracted from the literature, but subsequently not included in the draft and final lists. There might be several ways to define the intersectoral boundaries. Allocation of ICBs categorized as input (i.e. services or professionals) to a specific sector could be based on the primary aim of the service, while for throughput ICBs (i.e. interventions or activities), allocation could be based on the source of financing. Furthermore, the complex interrelatedness of the sectors indicates the need for a broader approach to conducting health economics research, looking not only at changes in resource-use on the individual level, but also taking into account the environment (e.g. family as a unit of analysis).

Methodological reflection

This study provides a comprehensive overview of ICBs in the education sector based on the extensive search of peer-reviewed and international grey literature as well as the survey of an international group of experts. While peer-reviewed literature accentuated the relevance of previously identified ICBs, grey literature was of added value, particularly in identifying additional relevant ICBs in the education sector and in developing a more comprehensive list in comparison with the previous overview.¹² Furthermore, the survey with the international experts provided insight into the clarity and relevance of the identified ICBs, and into the transferability of the list in a broader European context. Nevertheless, the current study is largely exploratory in nature and is to be viewed in the context of several limitations. First, the expert survey to validate the list of ICBs was challenging. While recruiting experts via phone proved to be a more effective recruitment strategy in comparison with recruitment via email, the overall response rate was low (38%). Second, the list of ICBs was partially based on the peer-reviewed literature pertaining to depression, PTSD and schizophrenia due to the focus of PECUNIA project. This, while other mental disorders (e.g. autism, attention deficit hyperactivity disorder) might also be relevant given the study context. Nevertheless, because other sources were not limited to specific mental disorders, it is assumed that the list developed in this study contains is comprehensive and contains all relevant ICBs in the education sector. Third, although the list of ICBs developed in this study is meant to be generic and applicable to multiple disease areas, it is derived from the mental health literature. Additional literature or qualitative research might be needed to select and prioritize the education ICBs with respect

to a specific context. Fourth, the identified ICBs are mainly applicable in the context of primary and secondary education, as these are generally compulsory. Some education ICBs might also be applicable to students with mental problems in higher education, although for the older population consequences in the labour sector might be more relevant.

Implications for further research

A broader intersectoral approach to conducting economic evaluations is a new development in the field of health economics, and highlights the need for further work in this direction. First, the impact of the education ICBs on the study outcomes should be further explored. This would potentially demonstrate that ICBs can constitute a large proportion of the total costs and could provide a sound argument for their inclusion in health economics research. Second, to improve the transferability of the results, the ICBs identified in this study need to be further defined and classified to demonstrate their usability, reliability, shareability, portability and interoperability for being transformed into a formal ontology that can be used across different countries and databases.²² One example of such an ontology is DESDE-LTC, the standard taxonomy for description, mapping, and comparison of services for long-term care,²³ which was selected to classify services identified within the PECUNIA project. DESDE-LTC has been used extensively for assessing the classification of care provision, including specific education services targeting students with mental problems.²⁴ Third, this study forms a basis for the development of standardized measurement and costing tools, which will also be done within the scope of the PECUNIA project. Fourth, while the current list provides a comprehensive overview of ICBs in the education sector, it is also important to investigate which ICBs are the most important to be included in economic evaluations, which could be done by means of best-worst scaling, among other methods.

Policy implications

The findings indicate that the impact of mental and psychosocial problems on the education sector is indisputable. As current pharmaco-economic guidelines are shifting towards recommending the adoption of a societal perspective, it is of increasing importance to consider ICBs in the education sector, as these costs might be particularly substantial in the context of certain disease areas and population groups.^{7,13} Furthermore, by highlighting the far-reaching economic impact of mental health interventions on the education sector and on society in general, this study is in line with the policy approaches that stress the importance of intersectoral collaboration,²⁵ which is particularly relevant given the severe underfunding on mental health.¹ Within the scope of the PECUNIA project, this study will ultimately contribute to the development of efficient, evidence-based collaborative care models and intersectoral funding arrangements that are crucial for improving chronic and mental health care in healthcare systems in a broader European context.

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Supplementary files

Supplementary File 1. Questions of the expert survey

Question	Answer options
Q1. Please comment on the naming and the short definition of the item. Based on the name and the short description, is it clear to you what the item entails?	Yes/No (if no, please suggest an alternative name/definition that you believe is more clear)
Q2. Do you think that this item is applicable to students with mental disorders?	Yes/No (if no, please explain)
Q3. Do you think the list of items is complete?	Yes/No (if no, please mention additional items that you think are/can be applicable)

Supplementary File 2. Peer-reviewed sources included in the literature review (in alphabetical order)

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Supplementary File 3. Transformation of the draft list of ICBs in the education sector based on the comments of the experts

Draft list of ICBs in the education sector	Sources	Summary of experts' comments	Final list of ICBs in the education sector
Special education services	Drost et al (2013)	Special education can be provided in regular schools/classrooms but also in special education schools	Special education school Additional education services/assistance in a regular school
Learning therapy	Grey literature Austrian online portal for career planning: https://www.bic.at/berufsinformation.php?brfid=2195 German guideline : Leitfaden zur Feststellung sonderpädagogischen Förderbedarfs an Berliner Schulen (2015) https://www.berlin.de/sen/bildung/schule/.../leitfaden_foerderbedarf-2017_nov15.pdf	Confused with learning diagnosis, must be clearly defined Learning support when it is organised in school and learning therapy when outside school	Educational therapy
Home education	Drost et al (2013)	Not provided only by parents, could also be by teachers/tutors	Home education
School-based health promotion interventions	Peer-reviewed literature Bowman, S., McKinstry, C., & McGorry, P. (2017). Youth mental ill health and secondary school completion in Australia: time to act. <i>Early intervention in psychiatry</i> , 11(4), 277-289. Deady, M., Choi, I., Calvo, R. A., Glozier, N., Christensen, H., & Harvey, S. B. (2017). eHealth interventions for the prevention of depression and anxiety in the general population: a systematic review and meta-analysis. <i>BMC psychiatry</i> , 17(1), 310.		Excluded as it belongs to the health care sector rather than the education sector

	<p>Kuyken, W., Nuthall, E., Byford, S., Crane, C., Dalgleish, T., Ford, T., ... & Williams, J. M. G. (2017). The effectiveness and cost-effectiveness of a mindfulness training programme in schools compared with normal school provision (MYRIAD): study protocol for a randomised controlled trial. <i>Trials</i>, 18(1), 194.</p> <p>Law, W. C., McClanahan, R., & Weismuller, P. C. (2017). Depression Screening in the School Setting: Identification of the Depressed Adolescent. <i>NASN school nurse</i>, 32(6), 364-370.</p> <p>Lyon, A. R., Maras, M. A., Pate, C. M., Igusa, T., & Vander Stoep, A. (2016). Modeling the impact of school-based universal depression screening on additional service capacity needs: A system dynamics approach. <i>Administration and Policy in Mental Health and Mental Health Services Research</i>, 43(2), 168-188.</p> <p>Ball, A., Rittner, B., Chen, Y. L., & Maguin, E. (2018). Impact of individualized education plans on academic success of youth with early onset schizophrenia. <i>Journal of evidence-informed social work</i>, 15(5), 534-549.</p>		
Liaison teacher	<p>Grey literature</p> <p>German federal ministry website: Behörde für Schule und Berufsbildung Hamburg, Information Bildungs- und Beratungszentrum Pädagogik bei Krankheit und Autismus. https://www.hamburg.de/bsb/bbz-einfuehrung/</p>	Suggested alternative namings “therapeutic pedagogy teacher” or “mentor”	Included in "additional education services/assistance in a regular school" based on re-clustering
Compensation for disadvantages	<p>Grey literature</p> <p>Website private school counselling institution in Lower Austria: Helpdesk für Lehrer/innen verhaltensauffälliger Schüler/innen, http://helv.ph-noe.ac.at/index.php?id=22&tx_ttnews%05Btt_news%05D=625&cHash=1f437c496aa4d2d9c8904537a4f460b5</p> <p>German Federal Ministry website: Behörde für Schule und Berufsbildung Hamburg,</p>	Special support facilities/differentiation in classroom	Included in "additional education services/assistance in a regular school" based on re-clustering

	<p>Information Bildungs- und Beratungszentrum Pädagogik bei Krankheit und Autismus. https://www.hamburg.de/bsb/bbz-einfuehrung/</p>		
Special needs diagnostics	<p>Grey literature German guideline : Leitfaden zur Feststellung sonderpädagogischen Förderbedarfs an Berliner Schulen (2015), https://www.berlin.de/sen/bildung/schule/.../leitfaden_foerderbedarf-2017_nov15.pdf</p>	<p>In the UK referred to as Special Education Needs assessment (SEN) or Education, Health and Care (EHC) assessment Difference with “learning therapy“ is not clear</p>	Special needs diagnostics
Counseling of legal guardians	<p>Grey literature Austrian guideline of the Ministry of education: Bundesministerium für Bildung, Mobbing an Schulen, 2017, https://www.saferinternet.at/fileadmin/cat egorized/Materialien/Mobbing_an_Schulen _Leitfaden_BMB.pdf</p> <p>German guideline : Leitfaden zur Feststellung sonderpädagogischen Förderbedarfs an Berliner Schulen (2015), https://www.berlin.de/sen/bildung/schule/.../leitfaden_foerderbedarf-2017_nov15.pdf</p>	No comments were given	Counselling of legal guardians
Student counselling	<p>Grey literature Austrian guideline of the Ministry of education: Bundesministerium für Bildung, Mobbing an Schulen, 2017, https://www.saferinternet.at/fileadmin/cat egorized/Materialien/Mobbing_an_Schulen _Leitfaden_BMB.pdf</p> <p>German national guideline :</p>	Can also take place during school, and not only for students with performance difficulties	Student counselling

	<p>Leitfaden zur Feststellung sonderpädagogischen Förderbedarfs an Berliner Schulen (2015), https://www.berlin.de/sen/bildung/schule/.../leitfaden_foerderbedarf-2017_nov15.pdf</p> <p>Dutch costing manual: Drost, R. M., Paulus, A. T. G., Ruwaard, D., & Evers, S. M. A. A. (2014). Handleiding intersectorale kosten en baten van (preventieve) interventies: Classificatie, identificatie en kostprijzen. Maastricht University, Department of Health Services Research.</p>		
Temporary study group	<p>Grey literature German unpublished article: Senatsverwaltung für Gesundheit, Soziales und Verbraucherschutz Berlin & Senatsverwaltung für Bildung, Jugend und Sport Berlin (2003) Kooperation von Kinder- und Jugendpsychiatrie, Jugendhilfe und Schule. https://www.efb-berlin.de/wp-content/uploads/pdf_fachleute/kooperation_psychiatrie_jugendhilfe_schule.pdf</p>	Might also be not temporary, study support group, disability learning group	Included in "additional education services/assistance in a regular school" based on re-clustering
Social and educational therapy boarding school	<p>Grey literature German school website: http://www.weisse-villa-harz.de/</p>	Special needs boarding school, residential school therapeutic boarding school	Special education school
Night school	<p>Grey literature German school website: http://hasa.grone-netzwerk.de/?gclid=EAIaIQobChMIhqGJ1cXJ3QIVDud3Ch3_sAYGEAAyAAEgIXofD_BwE</p>	<p>Could also take place in the evening</p> <p>Only for adults, while the focus of this study is on younger students</p>	Excluded, as this facility is meant for adults rather than children and adolescents
Attendance officer	<p>Grey literature Dutch costing manual: Drost, R. M., Paulus, A. T. G., Ruwaard, D., & Evers, S. M. A. A. (2014). <i>Handleiding intersectorale kosten en baten van (preventieve) interventies: Classificatie, identificatie en kostprijzen.</i></p>	Dealing with absenteeism is a regular task of schools, there are no special	Excluded

	Maastricht University, Department of Health Services Research.	officers for this task Does not exist in some countries	
Student transport to special education facility	Grey literature Dutch costing manual: Drost, R. M., Paulus, A. T. G., Ruwaard, D., & Evers, S. M. A. A. (2014). <i>Handleiding intersectorale kosten en baten van (preventieve) interventies: Classificatie, identificatie en kostprijzen</i> . Maastricht University, Department of Health Services Research.	Should not be limited to travel to special education facilities, as transport can be provided to mainstream schools and is not necessarily only available for those with SEND.	Student transport to special education facility
Student-related financing	Grey literature Dutch costing manual: Drost, R. M., Paulus, A. T. G., Ruwaard, D., & Evers, S. M. A. A. (2014). <i>Handleiding intersectorale kosten en baten van (preventieve) interventies: Classificatie, identificatie en kostprijzen</i> . Maastricht University, Department of Health Services Research.	Special needs financing/premium, but might be related to regular school financing	Student-related financing
Change in school readiness	Drost et al (2013)	Rather reduced	Reduced school readiness
Problems with school entry	Drost et al (2013)	Rarely occurs	Problems with school entry
Learning disabilities	Drost et al (2013)	Impaired development of learning skills	Learning disabilities
Cognitive deficits	Drost et al (2013)	Fall under learning disability	Included in "learning disabilities" based on re-clustering
Low school adaptation/competence	Drost et al (2013)	"Low" → "reduced" "Adaptation" and	Reduced school adaptation, Reduced

		“competence” should not be grouped together	school competence based on re-clustering
Low school participation /engagement	Drost et al (2013)	“Participation“ and “engagement” should not be grouped together Can also be low school motivation or classroom participation	Reduced school participation, Reduced school engagement based on re-clustering
Low school attainment/p roductivity/ performance	Drost et al (2013)	“Attainment”, “productivity” and “performance” are different terms	Reduced school attainment, Reduced school productivity, Reduced school performance based on re-clustering
Grade retention	Drost et al (2013)	Suggest 're-sit of the school year' Rare	Grade retention
Disrupted school experience	Drost et al (2013)	Very broad term	Negative school experiences
Teacher-student conflicts	Drost et al (2013)	No comments were given	Included in "negative school experiences" based on re-clustering
School droupout/pr e-mature leave	Drost et al (2013)	Leaving school without qualifications	Included in "reduced school attainment"

			based on re-clustering
Indirect effect of premature school leave/drop-out	Grey literature Dutch societal cost-benefit analysis in the education sector: KBA onderwijs en jeugdwerkloosheid, https://www.cpb.nl/sites/default/files/publicaties/download/cpb-notitie-maatschappelijke-kosten-baten-analyses-het-onderwijs.pdf	Can also include positive effects	Excluded as this ICB occurs in the labour sector
(Social) reintegration	Grey literature German federal ministry website: Behörde für Schule und berufsbildung Hamburg, Information Bildungs- und Beratungszentrum Pädagogik bei Krankheit und Autismus. https://www.hamburg.de/bsb/bbz-einfuehrung/	Might be too broad, better call it school re-integration	School (re-)integration
Inclusion	Grey literature German federal ministry website: Behörde für Schule und berufsbildung Hamburg, Information Bildungs- und Beratungszentrum Pädagogik bei Krankheit und Autismus. https://www.hamburg.de/bsb/bbz-einfuehrung/	Unsure whether "inclusion" as a policy is limited to students with social disabilities	Excluded as it is an antonym for discrimination
Refusal of admission	Grey literature Dutch costing manual: Drost, R. M., Paulus, A. T. G., Ruwaard, D., & Evers, S. M. A. A. (2014). Handleiding intersectorale kosten en baten van (preventieve) interventies: Classificatie, identificatie en kostprijzen. Maastricht University, Department of Health Services Research.	Not permitted in the UK and Germany	Included in "reduced school attainment" based on re-clustering
Change in educational level	Grey literature Dutch costing manual: Drost, R. M., Paulus, A. T. G., Ruwaard, D., & Evers, S. M. A. A. (2014). Handleiding intersectorale kosten en baten van (preventieve) interventies: Classificatie, identificatie en kostprijzen. Maastricht University, Department of Health Services Research.	"Fallback" Can also be upwards	Included in "reduced school attainment" based on re-clustering
Exemption from compulsory education	Grey literature Dutch costing manual: Drost, R. M., Paulus, A. T. G., Ruwaard, D., & Evers, S. M. A. A. (2014). Handleiding intersectorale kosten en baten van (preventieve) interventies:	Not possible when child is obliged by law to go to school	Included in "reduced school attainment"

	Classificatie, identificatie en kostprijzen. Maastricht University, Department of Health Services Research.		based on re-clustering
Talent development	Grey literature Societal cost-benefit analysis of an educational concept "Broad School" (NL: Brede School): MKBA Brede School, http://www.lpb.nl/wp-content/uploads/2016/03/MKBA-Brede-school-2007.pdf	Add "impaired" Perhaps skills/competence development would be better Needs to be clarified	Included in "reduced school competence" based on re-clustering
Discrimination	Peer-reviewed literature Bowman, S., McKinstry, C., & McGorry, P. (2017). Youth mental ill health and secondary school completion in Australia: time to act. <i>Early intervention in psychiatry</i> , 11(4), 277-289.	No comments were given	Included in "negative school experiences" based on re-clustering
Peer relations	Peer-reviewed literature Kuyken, W., Nuthall, E., Byford, S., Crane, C., Dalgleish, T., Ford, T., ... & Williams, J. M. G. (2017). The effectiveness and cost-effectiveness of a mindfulness training programme in schools compared with normal school provision (MYRIAD): study protocol for a randomised controlled trial. <i>Trials</i> , 18(1), 194. Dodge, K. A., Bierman, K. L., Coie, J. D., Greenberg, M. T., Lochman, J. E., McMahon, R. J., ... & Conduct Problems Prevention Research Group. (2014). Impact of early intervention on psychopathology, crime, and well-being at age 25. <i>American journal of psychiatry</i> , 172(1), 59-70. Campbell, R., & Riggs, S. A. (2015). The role of psychological symptomatology and social support in the academic adjustment of previously deployed student veterans. <i>Journal of American College Health</i> , 63(7), 473-481. Bowman, S., McKinstry, C., & McGorry, P. (2017). Youth mental ill health and secondary school completion in Australia: time to act. <i>Early intervention in psychiatry</i> , 11(4), 277-289. Betancourt, T. S., McBain, R., Newnham, E. A., Akinsulure-Smith, A. M., Brennan, R. T., Weisz, J. R., & Hansen, N. B. (2014). A behavioral intervention for war-affected	Include the word negative in the title, "negative peer relations"	Included in "negative school experiences" based on re-clustering

	<p>youth in Sierra Leone: a randomized controlled trial. <i>Journal of the American Academy of Child & Adolescent Psychiatry</i>, 53(12), 1288-1297.</p> <p>Beecham, J. (2014). Annual research review: Child and adolescent mental health interventions: A review of progress in economic studies across different disorders. <i>Journal of Child Psychology and Psychiatry</i>, 55(6), 714-732.</p> <p>Ball, A., Rittner, B., Chen, Y. L., & Maguin, E. (2018). Impact of individualized education plans on academic success of youth with early onset schizophrenia. <i>Journal of evidence-informed social work</i>, 15(5), 534-549.</p>		
Suspension	<p>Grey literature</p> <p>Austrian law</p> <p>Bundesgesetz über die Ordnung von Unterricht und Erziehung in den im Schulorganisationsgesetz geregelten Schulen (Schulunterrichtsgesetz - SchUG), § 49</p> <p>https://www.ris.bka.gv.at/GeltendeFassung.wxe?Abfrage=Bundesnormen&Gesetzesnummer=10009600</p> <p>Ducth unpublished MSc thesis</p>	Temporary/permanent exclusion	Included in "reduced school attainment" based on re-clustering
Negative feelings about school	<p>Peer-reviewed literature</p> <p>Hawkrigg, S., & Payne, D. N. (2014). Prolonged school non-attendance in adolescence: a practical approach. <i>Archives of disease in childhood</i>, 99(10), 954-957.</p>	Negative feelings are about more than just dislike	Included in "negative school experiences" based on re-clustering
Classroom behaviour	<p>Peer-reviewed literature</p> <p>Betancourt, T. S., McBain, R., Newnham, E. A., Akinsulure-Smith, A. M., Brennan, R. T., Weisz, J. R., & Hansen, N. B. (2014). A behavioral intervention for war-affected youth in Sierra Leone: a randomized controlled trial. <i>Journal of the American Academy of Child & Adolescent Psychiatry</i>, 53(12), 1288-1297.</p> <p>Bowman, S., McKinstry, C., & McGorry, P. (2017). Youth mental ill health and secondary school completion in Australia: time to act. <i>Early intervention in psychiatry</i>, 11(4), 277-289.</p>	Relates to peer relations Not clear who it is referred to a student or a class as a whole	Disruptive school behavior

Supplementary File 4. Descriptive information about the expert survey

Country	Number of experts	Professional background		Number of responses per question (Q)		
		Researcher	Educational expert	Q1	Q2	Q3
Austria	4		4	4	4	4
Germany	1	Unknown		1	1	1
Hungary	1	1		1	1	0
Spain	3		3	3	3	3
Netherlands	3	3		3	3	2
UK	2	2		2	1	1
Total	14	6	7	14	13	11

Chapter 4. The Relative Importance of Education and Criminal Justice Costs and Benefits in Economic Evaluations: A Best-Worst Scaling Experiment

Chapter 4 draws upon:

Pokhilenko, I., Janssen, L. M., Hiligsmann, M., Evers, S. M., Drost, R. M., Paulus, A. T., & Bremmers, L. G. 2021. The relative importance of education and criminal justice costs and benefits in economic evaluations: a best-worst scaling experiment. *PharmacoEconomics*, 39(1), 99-108. <http://dx.doi.org/10.1007/s40273-020-00966-8>

Abstract

Objectives. Mental and behavioral disorders (MBDs) and interventions targeting MBDs lead to costs and cost savings in the healthcare sector, but also in other sectors. The latter are referred to as intersectoral costs and benefits (ICBs). Interventions targeting MBDs often lead to ICBs in the education and criminal justice sectors, yet these are rarely included in economic evaluations. This study aimed to investigate the attitudes held by health economists and health technology assessment experts towards education and criminal justice ICBs in economic evaluations, and to quantify the relative importance of these ICBs in the context of MBDs.

Methods. An online survey containing open-ended questions and two best-worst scaling object case studies was conducted, in order to prioritize a list of twenty education ICBs and twenty criminal justice ICBs. Mean relative importance scores for each ICB were generated using Hierarchical Bayes analysis.

Results. Thirty-nine experts completed the survey. The majority of the respondents (68%) reported that ICBs were relevant, but only a few (32%) included them in economic evaluations. The most important education ICBs were “special education school attendance”, “absenteeism from school”, and “reduced school attainment”. The most important criminal justice ICBs were “decreased chance of committing a crime as a consequence/effect of mental health programs/interventions”, “jail and prison expenditures”, and “long-term pain and suffering of victims/victimization”.

Conclusions. This study identified the most important education and criminal justice ICBs for economic evaluations of interventions targeting MBDs and suggests that it could be relevant to include these ICBs in economic evaluations.

Introduction

Mental and behavioural disorders (MBDs) are estimated to affect more than one in six people during their lifetime, making it one of the top public health challenges worldwide.^{1,2} These disorders are associated with impaired capabilities in personal, familial and social functional areas and may adversely affect one's ability to actively participate in society.³ Up to 50% of the economic burden of MBDs is reflected in indirect costs to the labor market, due to the lower productivity associated with MBDs and overall impact on economic growth.^{1,4} Furthermore, MBDs are associated with reduced educational attainment⁵ and increased likelihood of contact with the justice system.⁶ The cost of supporting individuals with MBDs can be far higher than for their peers, as they often require additional support to achieve the same educational outcomes and increasingly engage in risky behavior and criminal activity.⁷ This is further supported by the evidence from several cost of illness studies of MBDs, which demonstrate that the costs of these disorders that fall on the education and criminal justice sectors can be significant.^{8,9} Hence, effective health interventions for MBDs can result in cost savings, i.e. benefits, in these sectors.

Costs and benefits attributable to the implementation of health interventions but that occur in sectors outside the healthcare sector are referred to as intersectoral costs and benefits (ICBs). Within this concept, benefits can be best described as prevented costs, i.e. cost savings that can be associated with lower resource-use, improved outcomes and/or averted consequences. From a welfarist theoretical framework, which focuses on maximizing societal well-being and underlies the definition of a societal perspective, all costs and benefits engendered by an intervention should be included in economic evaluations regardless of on whom they fall.¹⁰ Therefore, considering that interventions targeting MBDs are expected to generate ICBs in the education and criminal justice sectors,¹¹ it is necessary to account for them when conducting economic evaluations. Failure to include relevant ICBs in an economic evaluation might lead to suboptimal and misinformed decision-making.¹²

The inclusion of education and criminal justice ICBs in economic evaluations is supported by several national pharmaco-economic guidelines.¹³⁻¹⁵ Furthermore, several studies investigated the identification, measurement and valuation of education and criminal justice ICBs in health economics research.¹⁶⁻¹⁸ Nevertheless, to date, few economic evaluations incorporate these ICBs, even though they might be relevant to the study context.^{19,20} This can be due not only to the scarcity of validated methods and tools to measure and value education and criminal justice ICBs, but also to the lack of resources to collect data, the lack of knowledge about the relevance of ICBs and which ICBs are the most important to include in economic evaluations. Furthermore, national policy guidelines do not always recommend adopting a societal perspective.

Although an overview of relevant education and criminal justice ICBs does exist,¹⁶ it does not provide guidance in terms of which ICBs are the most important to include in economic evaluations. Mayer and colleagues (2017)¹⁸ developed a list of the most common education and criminal justice ICBs, based on the existing health-related resource-use measurement instruments. While this list could help to gain insight into the importance of these ICBs, it does not provide a ranking. To date, no research has focused on investigating the relative

importance of education and criminal justice ICBs in economic evaluations. Given that this knowledge would be particularly valuable for health economists and Health Technology Assessment (HTA) experts conducting economic evaluations in the MBD domain and for researchers undertaking further methodological research in the field of ICBs, the aim of this study was to assess the relative importance of the education and criminal justice ICBs that are of importance for economic evaluations in the disease area of MBDs among health economists and HTA experts, using best-worst scaling (BWS). Furthermore, this study aimed to gain insight into the experts' attitudes towards and experiences with including ICBs in economic evaluations.

Methods

Best-worst scaling

BWS is a survey method for assessing an individual's priorities or preferences for a set of attributes.²¹ A BWS object case, a type of BWS survey method, identifies the relative values associated with each of the attributes on a master attribute list.

Two BWS object case surveys (one for education and one for criminal justice) were conducted to obtain experts' preferences with regard to the most important education and criminal justice ICBs for inclusion in economic evaluations conducted from a societal perspective in the disease area of MBDs. The Checklist for Conjoint Analysis Applications in Health from the International Society for Pharmacoeconomics and Outcomes Research (ISPOR) was followed to design and conduct the BWS survey and to report the results of the study.²²

Identification of the attributes

Prior to the BWS object case surveys, a mutually exclusive attribute list was generated using a two-stage procedure. A mutually exclusive attribute list is essential for obtaining valid outcomes in BWS analysis.²³ First, potential attributes were extracted by one researcher (LB) from earlier research on ICBs^{16,18} and resource-use measurement instruments, via a hand search of the Database of Instruments for Resource Use Measurement (DIRUM).²⁴ Second, the attributes were compiled and clustered based on similarity using the classification of Drost et al. (2013)¹⁶ as a reference. Third, the list of attributes was defined and validated in a working group of five co-authors with expertise in the field of ICBs (IP, LJ, RD, AP, LB) to ensure completeness and mutual exclusivity. This resulted in the final list of twenty education ICBs and twenty criminal justice ICBs (i.e. attributes) to be assessed in the BWS survey (Supplementary File 1). It is important to note that every attribute could be viewed both as a cost and as a benefit (i.e. cost saving or improved outcome).

Survey respondents

Health economists and HTA experts were chosen as the target group for this study because they are involved in the selection of relevant costs and benefits for the inclusion in economic evaluations. Convenience and snowball sampling methods were used to recruit respondents. Experts were screened for inclusion based on a question integrated in the survey, which assessed whether they had work experience in the field of health economics in general and experience with conducting health economic evaluations. Respondents who reported having

no work experience in the field of health economics were redirected to the end of the survey. Respondents with experience in health economics but with no specific experience in economic evaluations were assumed to have sufficient knowledge to participate in the study. Potential respondents were approached at several health economics and HTA conferences between May and November 2019. In addition, targeted e-mails were sent to potentially eligible experts. Respondents were asked to invite and/or provide contact information of colleagues who would be interested in completing the survey. Social media promotion was also employed. The study protocol was reviewed and approved by the Medical Ethics Committee of the Academic Hospital Maastricht and Maastricht University, the Netherlands (Approval Number: FHML-REC/2019/030). All respondents provided informed consent prior to completing the survey and they were free to stop participation at any moment.

BWS survey

The experimental design of the BWS object case surveys was developed using Sawtooth Software's SSI Web platform, resulting in fractional and efficient designs, which are characterized by orthogonality, minimal overlap, positional balance, connectivity, and stability. To improve statistical efficiency, two different versions of the questionnaire were generated per object case survey, i.e. two versions for education and two versions for criminal justice. Each version contained twelve education and twelve criminal justice choice sets with five attributes per choice set. Each attribute was presented twelve times, was combined at least once with every other cost and benefit and appeared two to four times in each position in the choice set. Definitions were provided alongside each choice set as recommended by good research practice guidelines.²² For each choice set, respondents were asked to select which cost and benefit was the most and least important to include in economic evaluations in the disease area of MBDS. An example choice set is presented in Figure 1.

Best-worst scaling survey: Criminal justice sector		
Please identify which cost and benefit in the criminal justice sector you believe is the MOST important and which is the LEAST important to include in economic evaluations conducted in the disease area of mental and behavioral disorders		
Trade-off scenario 1		
Most important		Least important
x	Jail and prison expenditures	
	Short-term pain and suffering of others	
	Property loss of offender	x
	Probation/Parole (including electronic monitoring)	
	Forensic services	

Figure 1. Example of a completed criminal justice choice set from the best-worst scaling survey

The online survey was designed in the form of a self-administered questionnaire using Qualtrics®.²⁵ Respondents were randomly allocated to one of the two versions of the education ICBs object case surveys and one of the two versions of the criminal justice ICBs

object case surveys. In addition to the choice sets, respondents were asked to answer questions about their professional characteristics.

The survey also contained six questions to investigate experts' attitudes towards and experiences with including ICBs in economic evaluations. The experts were asked to indicate whether they thought including education and criminal justice ICBs in economic evaluations could be relevant and to motivate their response. The experts were also asked whether they had previously included education and/or criminal justice ICBs in economic evaluations. At the end of each BWS object case study, respondents had the opportunity to report any additional education and criminal justice ICBs that they thought were missing from the master attribute list. Furthermore, respondents were asked to rate the difficulty of the choice tasks using a Likert scale (0 = easy to 10 = difficult). The questionnaire was piloted among health economists (n=2), a researcher (n=1), and health economics students (n=2) from Maastricht University. The estimated time to complete the questionnaire was 15 minutes. The paper-pencil version of the questionnaire is available in Supplementary File 2.

Data analysis

First, descriptive statistics were used to generate the respondents' professional characteristics, whether they found ICBs relevant and whether they had previously included ICBs in economic evaluations. Second, a Fischer's exact test was employed to test the association between the respondents' professional characteristics and whether they found ICBs relevant and had previously included them in economic evaluations. Two separate subgroup analyses were performed: those respondents with the number of years of experience in health economics below the mean were compared with the respondents with the number of years of experience equal to or above the mean; respondents employed in the Netherlands were compared with respondents employed elsewhere. In addition, respondents' open-ended answers concerning the relevance of and previous experiences with including ICBs in economic evaluations were summarized to identify common themes. SPSS for Windows V.25 was used to perform statistical analysis; $p < 0.025$ was considered statistically significant.

Third, Sawtooth SSI Web version 8.2.0 was used to perform a hierarchical Bayes estimation and to calculate the mean relative importance score (RIS) for each attribute. Rescaled scores were estimated based on the raw coefficient of the preference function. These scores represent the probability that the respondent chooses a selected attribute over other attributes. The RISs for each individual cost and benefit add up to 100, with a higher score indicating a higher relative importance for that attribute.²⁶ An individual fit statistic was employed to examine the quality of the responses. Responses with an individual fit statistic lower than 0.25, indicating that the answers were likely to be provided at random,²⁷ were excluded from the analysis.

Fourth, the RISs of education and criminal justice object case studies were calculated separately for the groups of respondents, based on their years of experience in health economics (i.e. below and above the mean) and country of employment (i.e. experts employed in the Netherlands vs experts employed elsewhere).

Results

Descriptive statistics

The survey was accessed 70 times between May 2019 and October 2019. In total, 39 health economists and HTA experts completed the questionnaire (response rate of 56%). Over half of the respondents (n=23, 59%) reported being employed in the Netherlands. The respondents had on average 7.1 years (SD=5.3) of experience in the field of health economics, and the majority (85%) reported having conducted economic evaluations prior to completing the survey. The descriptive statistics of the respondents are presented in Table 1.

Table 1. Descriptive statistics of the survey respondents

Country of employment (n=39*)	Netherlands	23
	Australia	3
	Germany	3
	United Kingdom	3
	Belgium	2
	Austria	1
	Colombia	1
	Hungary	1
	Spain	1
	United Kingdom/Canada	1
Years of experience in health economics		Mean (SD)
		7.1 (5.3)
Conducted economic evaluations		N (%)
	Yes	33 (85)
	No	6 (15)
Relevance of including criminal justice costs and benefits in economic evaluations according to the respondents		N (%)**
	Relevant	29 (88)
	Not relevant	4 (12)
Relevance of including education costs and benefits in economic evaluations according to the respondents		N (%)**
	Relevant	31 (94)
	Not relevant	2 (6)
Previously included criminal justice costs and benefits in economic evaluations		N (%)**
	Yes	9 (27)
	No	24 (73)
Previously included education costs and benefits in economic evaluations		N (%)**
	Yes	10 (30)
	No	23 (70)

*39 individuals completed the questionnaire for criminal justice, 35 completed the questionnaire for education

**This question was displayed only to the individuals who reported to have had previously conducted economic evaluations (n=33)

Thirty-nine respondents completed the BWS object survey for criminal justice and 35 out of these 39 completed the BWS object case survey for education. The overall fit statistic was 0.47 (SD=0.09) for the criminal justice object case survey and 0.45 (SD=0.07) for the education object case survey. Three responses had an individual fit statistic below 0.25 (n=2 for the education survey, n=1 for both the education and criminal justice surveys) and were excluded from the BWS analysis. The average difficulty of the questionnaire was 7 (SD=1.8) on a 10-point Likert scale (1 – “very easy”, 10 – “very difficult”).

Attitudes and experiences of including ICBs in economic evaluations

Criminal justice and education ICBs were found relevant for inclusion in economic evaluations by 29 and 31 respondents, respectively (Table 1). The majority of the respondents who found ICBs relevant had never included them in economic evaluations (68%) (Supplementary File 3). No significant differences were found between the subgroups of the respondents based on their years of experience in health economics and their country of employment in relation to their opinions regarding the relevance of the ICBs and their experience with including them in economic evaluations (Supplementary File 4).

Thirty-four experts answered the open-ended questions regarding the general relevance of education and criminal justice ICBs for economic evaluations. The experts agreed that the relevance of these ICBs is dependent on the intervention/disease, the target group and the budget holder. Several arguments in favor of including these ICBs in economic evaluations conducted from a societal perspective were mentioned. First, the definition of the societal perspective implies the inclusion of all relevant costs and benefits associated with the intervention. Second, education and criminal justice ICBs can constitute a significant proportion of the total costs and influence the results; hence, by not incorporating them in the study, the risk of bias is increased. Third, including these ICBs can demonstrate the wider impact of interventions. The reasons for not including education and criminal justice ICBs in economic evaluations were the irrelevance of these costs and benefits to the budget holder, the difficulty of obtaining information, the difficulty of quantifying these ICBs, and the difficulty of defining the boundaries of a broader (societal) perspective. Furthermore, one expert mentioned that while it is important to quantify the impact of diseases/health interventions on the education sector, the impact of educational decisions on the healthcare sector also ought to be quantified.

BWS analysis – education ICBs

Figure 2 illustrates the RISs and the confidence intervals of the education attributes. The most important education ICBs were “special education school attendance” (RIS=11.21), “absenteeism from school” (RIS=10.08), “reduced school attainment” (RIS=9.43), “additional education services provided at a regular school outside operating hours” (RIS=7.49), and “reduced school performance” (RIS=7.44). The five least important education ICBs were “student transport to school” (RIS=3), “reduced school engagement” (RIS=2.8), “reduced school adaption” (RIS=2.54), “attendance officer” (RIS=1.6), and “home education/homeschooling” (RIS=0.91). The scores and the confidence intervals of the education attributes are presented in Supplementary File 5.

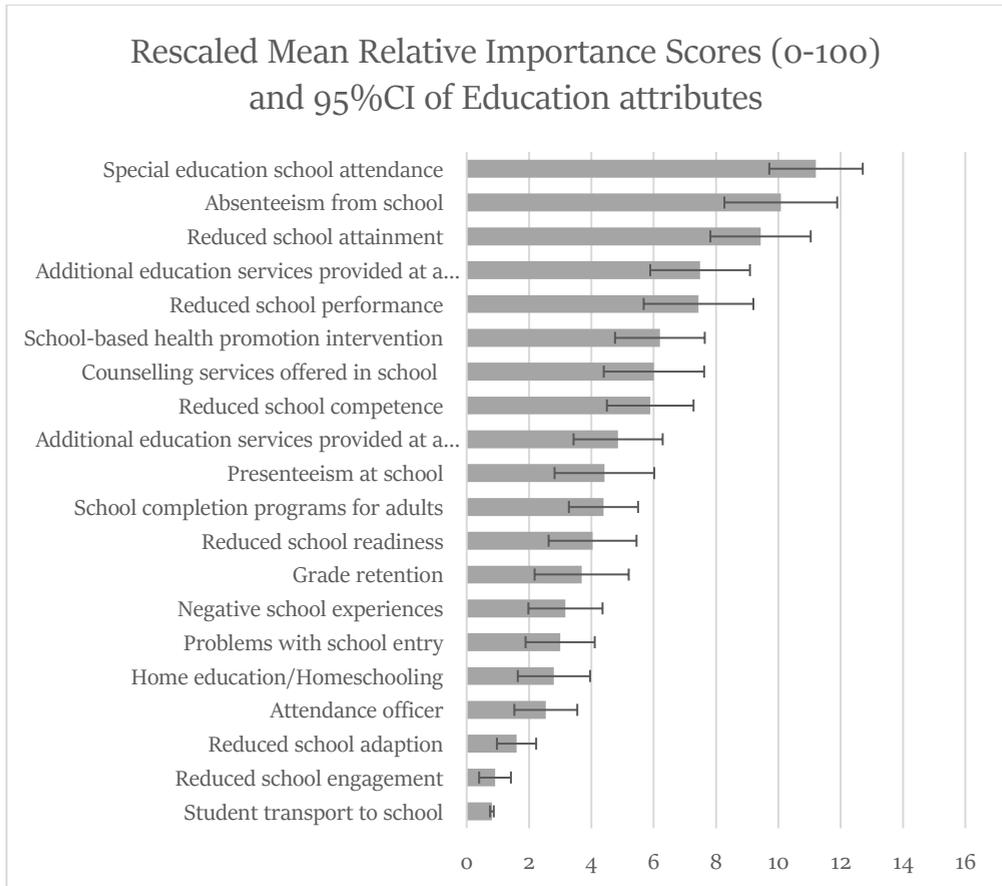


Figure 2. Relative importance scores of the education attributes (n=32)

BWS analysis – criminal justice ICBs

Figure 3 illustrates the RISs and the confidence intervals of the criminal justice attributes. The most important criminal justice ICBs were “decreased chance of committing a crime as a consequence/effect of mental health programs/interventions” (RIS=12.94), “jail and prison expenditures” (RIS=10.88), “long-term pain and suffering of victims/victimization” (RIS=9.94), “short-term pain and suffering of victims/victimization” (RIS=8.59), and “long-term pain and suffering of others” (RIS=7.21). The five least important criminal justice ICBs were “illegal untaxed income of the offender” (RIS=2.73), “property loss of offender” (RIS=2.3), “lost freedom of the offender” (RIS=1.1), “fire and rescue services” (RIS=0.86), and “forensic services” (RIS=0.78). The scores and the confidence intervals are presented in Supplementary File 5.

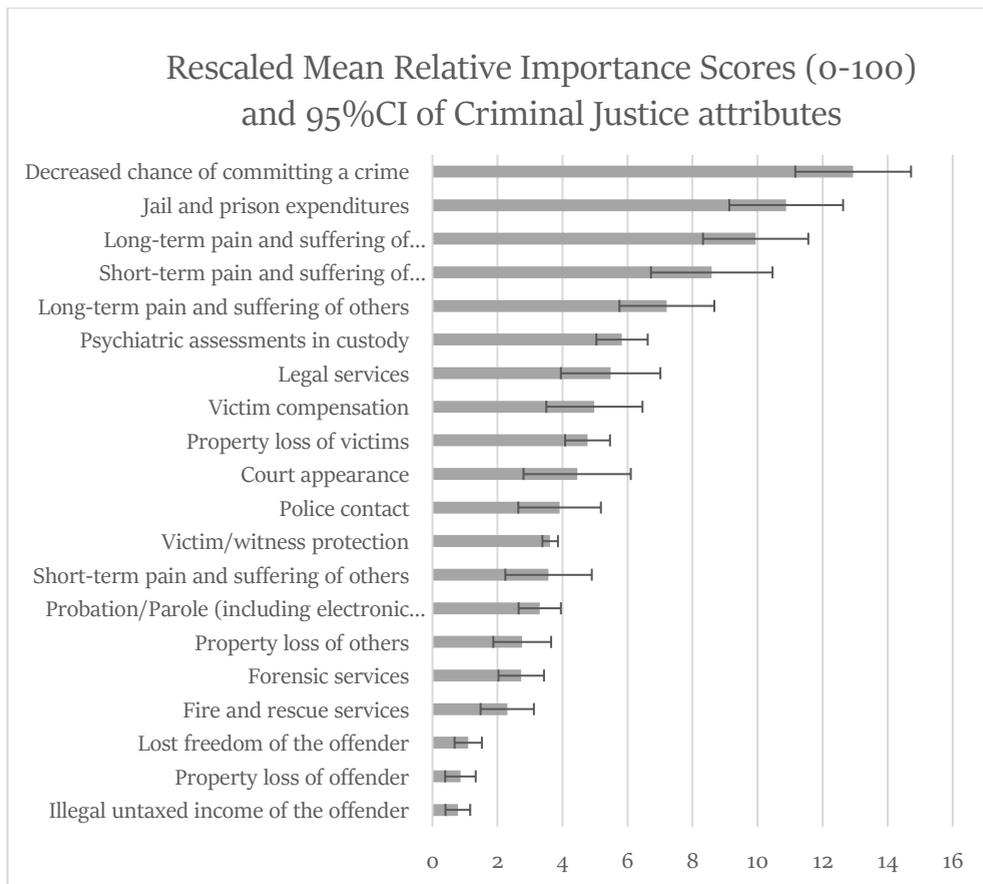


Figure 3. Relative importance scores of the criminal justice attributes (n=38)

BWS analysis – subgroup analysis

The RIS per attribute of two expert subgroups, one with years of experience above 7.1 (the mean number of years of experience in health economics among the respondents in this study) and the other below or equal to 7.1 were compared. In addition, the differences in the RISs per attribute were explored in relation to the country of employment of the experts (the Netherlands versus elsewhere). Subgroup analysis did not reveal notable differences in the ranking (Supplementary File 6).

Missing attributes

Three experts reported three additional criminal justice ICBs that they thought were missing from the attribute list: “long-term effects for the offenders after release”, “income earned while in prison”, and “additional benefits/entitlements to family members/third parties (of/to victims)”. Two experts reported additional education ICBs that could be added to the attribute list: “dropout”, “costs of training teachers”, “costs of adapting facilities”, “out-of-pocket payments for education beyond standard home schooling”.

Discussion

The economic impact of MBDs is substantial and affects many societal sectors, including the education and the criminal justice sectors.⁵⁻⁷ Nevertheless, economic evaluations in the area of MBDs rarely capture this impact, which can be attributed to the lack of knowledge about the relevance of ICBs and which ICBs are the most important to include in economic evaluations. This study, using data obtained from an online survey, provides insight into health economists' and HTA experts' opinions towards and experiences with the inclusion of education and criminal justice ICBs in economic evaluations. Furthermore, BWS was employed to assess the relative importance of these ICBs, to help researchers in selecting the most important ICBs for inclusion in economic evaluations of MBDs. Thirty-nine experts participated in the survey and assessed the relative importance of the education and criminal justice ICBs in a BWS experiment.

While the majority of the respondents agreed that education and criminal justice ICBs could be relevant for economic evaluations, far fewer of them reported having previously included these ICBs in economic evaluations. This could be attributed to the experts' focus on a disease area, for which education and criminal justice ICBs are less important (e.g. palliative care). Furthermore, national pharmaco-economic guidelines play an important role in framing economic evaluations in a given country. Of the participating experts' countries of employment, only the Dutch guidelines specifically recommend the inclusion of education and criminal justice ICBs if these are relevant to the context of the study,¹³ whereas other national guidelines recommend either a healthcare system perspective or a societal perspective without specifying education or criminal justice ICBs.²⁸ Therefore, to facilitate the adoption of a broader societal perspective in economic evaluations, a re-examination of existing national pharmaco-economic guidelines is needed, in addition to further methodological developments. Furthermore, the adoption of a societal perspective does not preclude conducting additional analyses from narrower health care or payer perspectives as these costs would already be available for the analysis.

While various arguments have been put forward as to why a societal perspective could be considered superior to narrower healthcare and payer perspectives,¹² choosing a broader perspective has implications for the funding of new healthcare interventions. Health interventions are generally subsidized by the health care sector alone and policy silos currently hinder effective intersectoral coordination and efficient use of resources to enhance societal welfare and improve population health.²⁹ If an intervention delivers benefits in sectors outside the health care, it is imperative to consider the possibility of multi-sectoral funding. This highlights the growing importance of broader integrated approaches to policy-making, in particular in relation to health, such as Health in All Policies, which emphasize intersectoral accountability and joint funding.^{30,31} Furthermore, research that demonstrates the intersectoral benefits of health interventions could act as a catalyst for setting up such initiatives.

The most common education ICBs in resource-use measurement instruments, as identified by Mayer et al.¹⁸ were "absenteeism from school", "tutoring", "classroom assistance", "special school/boarding school", and "social and school functioning", which includes some of the most

highly ranked education ICBs. However, while the items “reduced school attainment” and “reduced school performance” were considered important by the experts, these were not often included in the existing RUM instruments. The most common criminal justice ICBs were “lawyer/legal assistance”, “police custody/prison detainment”, “court appearance”, “injury”, and “police contact”, which is less comparable to the ranking in this study. Judicial services were ranked much lower in comparison with less tangible consequences of crime (e.g. “long-term pain and suffering of victims/victimization”), with the exception of “jail and prison expenditures”. Furthermore, the attributes related to the consequences of crime for the victims were ranked more important in comparison with the consequences for the offenders. Although from a societal perspective it makes no difference where costs and benefits associated with an intervention occur, bias towards favoring victims of crime rather than offenders might lead to prioritizing the costs and benefits that fall on victims rather than those that fall on offenders. This could lead to biased results, inequitable resource allocation, and loss of societal welfare (i.e. financial and productivity losses).³² The differences between these two lists could be due to the specific focus on MBDs, while Mayer et al. reviewed all existing RUM instruments. In addition, the most important ICBs in this study were not only resource-use items that could be measured by a self-reported questionnaire (e.g. “jail and police expenditures”), but also outcomes, which could be measured by either reviewing administrative data (e.g. “reduced school attainment”)³³ or by employing quality of life measurement instruments (e.g. “long-term pain and suffering of victims”).

Several experts provided additional education and criminal justice ICBs that could complement the master attribute list. Some of the suggested ICBs have been incorporated in the attributes that were already included in the attribute list. For example, the attribute “reduced school attainment” included school dropout/premature leave, refusal of admission, exemption from compulsory education, and change in the educational level. Hence, including “dropout” would compromise the mutual exclusivity of the attributes. Prior to including additional ICBs in the attribute list, an assessment of each attribute in relation to other attributes must be completed to ensure the mutual exclusivity of the attributes. Employing a group consensus method (e.g. Delphi method)³⁴ could allow to develop a comprehensive and mutually exclusive attribute list.

To our knowledge, this is the first study to date that aimed to evaluate the relative importance of education and criminal justice ICBs in the context of MBDs among health economists and HTA experts. Nevertheless, this study is subject to several limitations. First, despite numerous efforts to reach potential respondents, the sample size of this study was relatively small (n=39). While sample size calculations are not required for studies using BWS methods, a sample size of fifty respondents is recommended as a rule of thumb.³⁵ The small sample size and the overrepresentation of Dutch respondents in the sample could limit the generalizability of the results. Second, experts were able to access the questionnaire an unlimited number of times, which means they could have started it multiple times before eventually completing it. This might have led to an overestimated non-response rate; however, due to privacy regulations, it was not possible to account for this factor when analyzing the responses. In addition, we were not able to distinguish the experts with and without experience in the MBD domain, while this could have provided additional insights into the relationship between

experience in this domain and the attitudes regarding the relevance of the inclusion of ICBs in economic evaluations. Third, completing the questionnaire was rated relatively difficult (7 out of 10). This might explain that fewer respondents completed the object case survey for education (n=35) in comparison with the object case for criminal justice (n=39). Because the object case survey for criminal justice was completed prior to the one for education, several respondents might have quit without finishing the education survey due to survey fatigue.

The findings of this study could facilitate the inclusion of ICBs in economic evaluations in several ways. First, it could help researchers with selecting the most relevant ICBs for inclusion in economic evaluations in the area of MBDs, while limiting the burden associated with survey fatigue. It is important to note that education and criminal justice ICBs can be incorporated in economic evaluations on both the cost and the effect side. Researchers need to be aware of potential double counting.³⁶ Second, this could help advance methodological research into the development of measurement and valuation instruments. Furthermore, it is important to note that a general prioritization of education and criminal justice ICBs does not provide sufficient information regarding the importance of these items in a specific context, because the importance of cost items will always be dependent on the patient population, disease and/or intervention of interest, among other factors.³⁷ Using other methods, such as reviewing literature or consulting experts, might help determine the most important ICBs in a specific context. Further research could also focus on conducting a similar prioritization exercise with other stakeholder groups (e.g. mental health professionals, policy makers, patients) to help facilitate the inclusion of relevant ICBs in economic evaluations and to contribute to preference-sensitive decision-making.

Conclusions

The ranking of the most important education and criminal justice ICBs identified in this study can help select relevant ICBs for developing measurement and valuation tools and for the inclusion in economic evaluations in the domain of MBDs. Furthermore, while the majority of the respondents found education and criminal justice ICBs relevant, only a few reported previously including them in economic evaluations.

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Supplementary files

Supplementary File 1. Master attribute list of education and criminal justice costs and benefits

Education costs and benefits	Criminal justice costs and benefits
Absenteeism from school	Court appearance
Additional education services provided at a regular school during operating hours	Decreased chance of committing a crime as a consequence/effect of mental health programs/interventions
Additional education services provided at a regular school outside operating hours	Fire and rescue services
Attendance officer	Forensic services
Counselling services offered in school	Illegal untaxed income of the offender
Grade retention	Jail and prison expenditures
Home education/Homeschooling	Legal services
Negative school experiences	Long-term pain and suffering of others
Presenteeism at school	Long-term pain and suffering of victims/victimization
Problems with school entry	Lost freedom of the offender
Reduced school adaption	Police contact
Reduced school attainment	Probation/Parole (including electronic monitoring)
Reduced school competence	Property loss of offender
Reduced school engagement	Property loss of others
Reduced school performance	Property loss of victims
Reduced school readiness	Psychiatric assessments in custody
School completion programs for adults	Short-term pain and suffering of others
School-based health promotion intervention	Short-term pain and suffering of victims/victimization
Special education school attendance	Victim compensation
Student transport to school	Victim/witness protection

Supplementary File 2. Best-worst scaling survey

Included below is the complete questionnaire. Please note that questionnaire was administered via Qualtrics; hence, there are some deviations from the format and the skip logic and validation methods, which were applied in the online version.

Background questions

Please state whether you have experience in the field of health economics. If you selected "yes", specify your year(s) of experience.

- Yes (please specify) _____
- No

Please state your country of employment:

In which field(s) of study have you conducted health economics research? Please tick all relevant fields of study.

- Demand and utilization of health services
- Economic evaluation of health and care interventions
- Evaluation of policy, programs and health system performance
- Health care financing and expenditures
- New developments in methodology
- Organization of health care markets
- Production of health, health behaviors and policy interventions
- Specific population, i.e. health in developing countries, health and ageing, child health, mental health, disability, infectious diseases, non-communicable diseases
- Supply of health services
- Other

Have you conducted economic evaluations?

- Yes
- No

Which field(s) of study have you conducted economic evaluations in? Please select all that apply.

- Economic evaluations within clinical trials
- Economic evaluation using decision analytic modelling and discrete event simulation
- Policy design and evaluation
- Health insurance and health care financing
- Prevention
- Diagnostics
- Medical devices
- Long-term care
- Forensics
- Other

Have you ever included costs and benefits from the education sector (e.g. reduced school attainment, special education school attendance, reduced school engagement) in previously conducted economic evaluations?

- Yes
- No

Do you think that costs and benefits from the education sector (e.g. reduced school attainment, special education school attendance, reduced school engagement) could be relevant for economic evaluations conducted from a societal perspective?

- Yes
- No

Please motivate your previous response.

Have you ever included costs and benefits from the criminal justice sector (e.g. police contact, legal services, psychiatric assessments in custody) in previously conducted economic evaluations?

- Yes
- No

Do you think that costs and benefits from the criminal justice sector (e.g. police contact, legal services, psychiatric assessments in custody) could be relevant for economic evaluations conducted from a societal perspective?

- Yes
- No

Please motivate your previous response.

Best-worst scaling experiment: criminal justice services

Now, we ask you to evaluate costs and benefits in the criminal justice sector in a series of trade-off scenarios. In each scenario, we will show you five potential costs and benefits in the criminal justice sector to include in economic evaluations conducted in the disease area of mental and behavior disorders.

We will ask which cost and benefit in the criminal justice sector (among this set of five) do you believe is the **MOST** important to include, and which is the **LEAST** important. We need to ask you repeated trade-off scenarios (involving different costs and benefits each time) so that we can learn which costs and benefits you truly find important. You can find the complete list of costs and benefits in the criminal justice sector below. For the definitions, please refer to Appendix I.

Complete list of criminal justice services

- Court appearance
- Decreased chance of committing a crime as a consequence/effect of mental health programs/interventions
- Fire and rescue services
- Forensic services
- Illegal untaxed income of the offender
- Jail and prison expenditures
- Legal services
- Long-term pain and suffering of others
- Long-term pain and suffering of victims/victimization
- Lost freedom of the offender
- Police contact
- Probation/Parole (including electronic monitoring)
- Property loss of victims
- Property loss of offender
- Property loss of others
- Psychiatric assessments in custody

- [Short-term pain and suffering of others
- [Short-term pain and suffering of victims/victimization
- [Victim compensation
- [Victim/witness protection

You are asked to complete 12 trade-off scenarios, where you will identify what you believe is the MOST and LEAST important cost and benefit in the criminal justice sector to include in economic evaluations conducted in the disease area of mental and behavioral disorders. Please tick ONLY ONE item in the most and least important column.

Example: here 'Court appearance' as the most important cost and benefit and 'Lost freedom of the offender' is regarded as the least important cost and benefit to include in an economic evaluation conducted in the disease area of mental and behavioral disorders.

Most important		Least important
x	Court appearance	
	Probation/Parole (including electronic monitoring)	
	Long-term pain and suffering of victims/victimization	
	Psychiatric assessments in custody	x
	Lost freedom of the offender	

Trade-off scenario 1

Please identify which cost and benefit in the criminal justice sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important		Least important
	Property loss of offender	
	Long-term pain and suffering of others	
	Probation/Parole (including electronic monitoring)	
	Court appearance	
	Property loss of victims	

Trade-off scenario 2

Please identify which cost and benefit in the criminal justice sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important	Least important
<p>Legal services</p> <p>Police contact</p> <p>Victim compensation</p> <p>Short-term pain and suffering of others</p> <p>Decreased chance of committing a crime as a consequence/effect of mental health programs/interventions</p>	

Trade-off scenario 3

Please identify which cost and benefit in the criminal justice sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important	Least important
<p>Long-term pain and suffering of others</p> <p>Victim compensation</p> <p>Lost freedom of the offender</p> <p>Victim/witness protection</p> <p>Jail and prison expenditures</p>	

Trade-off scenario 4

Please identify which cost and benefit in the criminal justice sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important	Least important
<p>Court appearance</p> <p>Property loss of offender</p> <p>Legal services</p> <p>Property loss of others</p> <p>Short-term pain and suffering of victims/victimization</p>	

Trade-off scenario 5

Please identify which cost and benefit in the criminal justice sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important	Least important
Lost freedom of the offender Jail and prison expenditures Property loss of others Fire and rescue services Long-term pain and suffering of victims/victimization	

Trade-off scenario 6

Please identify which cost and benefit in the criminal justice sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important	Least important
Victim/witness protection Short-term pain and suffering of victims/victimization Psychiatric assessments in custody Forensic services Illegal untaxed income of the offender	

Trade-off scenario 7

Please identify which cost and benefit in the criminal justice sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important	Least important
Forensic services Long-term pain and suffering of victims/victimization Property loss of victims Decreased chance of committing a crime as a consequence/effect of mental health programs/interventions Psychiatric assessments in custody	

Trade-off scenario 8

Please identify which cost and benefit in the criminal justice sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important	Least important
Short-term pain and suffering of others Illegal untaxed income of the offender Jail and prison expenditures Property loss of victims Legal services	

Trade-off scenario 9

Please identify which cost and benefit in the criminal justice sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important	Least important
Police contact Probation/parole (including electronic monitoring) Illegal untaxed income of the offender Fire and rescue services Short-term pain and suffering of others	

Trade-off scenario 10

Please identify which cost and benefit in the criminal justice sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important	Least important
Property loss of others Victim/witness protection Police contact Long-term pain and suffering of others Forensic services	

Trade-off scenario 11

Please identify which cost and benefit in the criminal justice sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important	Least important
Probation/Parole (including electronic monitoring) Decreased chance of committing a crime as a consequence/effect of mental health programs/interventions Long-term pain and suffering of victims/victimization Victim compensation Short-term pain and suffering of victims/victimization	

Trade scenario 12

Please identify which cost and benefit in the criminal justice sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important	Least important
	Fire and rescue services
	Lost freedom of the offender
	Court appearance
	Psychiatric assessments in custody
	Property loss of offender

Are there any additional costs and benefits in the criminal justice sector, which you consider important for economic evaluations conducted in the disease area of mental and behavioral disorders that have not been listed? If yes, please specify.

- Yes (please specify) _____
- No

Best-worst scaling experiment: education services

Now, we ask you to evaluate costs and benefits in the education sector in a series of trade-off scenarios. In each scenario, we will show you five potential education services to include in economic evaluations conducted in the disease area of mental and behavior disorders.

We will ask which cost and benefit in the education sector (among this set of five) do you believe is the **MOST** important to include, and which is the **LEAST** important. We need to ask you repeated trade-off scenarios (involving different costs and benefits in the education sectors each time) so that we can learn what is truly important to you when deciding important items. You can find the complete list of costs and benefits in the education sector below. For the definitions, please refer to Appendix I.

Complete list of education services

- Absenteeism from school
- Additional education services provided at a regular school during operating hours
- Additional education services provided at a regular school outside operating hours
- Attendance officer
- Counselling services offered in school
- Grade retention
- Home education/Homeschooling
- Negative school experiences

- [Presenteeism at school
- [Problems with school entry
- [Reduced school adaption
- [Reduced school attainment
- [Reduced school competence
- [Reduced school engagement
- [Reduced school readiness
- [Reduced school performance
- [School-based health promotion intervention
- [School completion programs for adults
- [Special education school attendance
- [Student transport to school

You are asked to complete 12 trade-off scenarios, where you will identify what you believe is the MOST and LEAST important cost and benefit in the education sector to include in economic evaluations conducted in the disease area of mental and behavioral disorders. Please tick ONLY ONE item in the most and least important column.

Example: here 'Absenteeism from school' is regarded as the most important and 'School-based health promotion interventions' as the least important education cost and Benefit to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important	Least important
	Reduced school performance
	School-based health promotion interventions x
	Student transport to school
	Additional education services outside of regular school operating hours
x	Absenteeism from school

Trade-off scenario 1

Please identify which cost and benefit in the education sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important	Least important
	Reduced school competence
	Negative school experiences
	Reduced school attainment
	Absenteeism from school
	Reduced school performance

Trade-off scenario 2

Please identify which cost and benefit in the education sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important		Least important
	Home education/ Homeschooling	
	Reduced school adaptation	
	Special education school attendance	
	School completion programs for adults	
	Additional education services provided at a regular school during operating hours	

Trade-off scenario 3

Please identify which cost and benefit in the education sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important		Least important
	Negative school experiences	
	Special education school attendance	
	Problems with school entry	
	Student transport to school	
	Grade retention	

Trade-off scenario 4

Please identify which cost and benefit in the education sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important		Least important
	Absenteeism from school	
	Reduced school competence	
	Home education / Homeschooling	
	Reduced school engagement	
	School-based health promotion intervention	

Trade-off scenario 5

Please identify which cost and benefit in the education sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important		Least important
	Problems with school entry	
	Grade retention	
	Reduced school engagement	
	Additional education services provided at a regular school outside operating hours	
	Presenteeism at school	

Trade-off scenario 6

Please identify which cost and benefit in the education sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important		Least important
	Student transport to school	
	School-based health promotion intervention	
	Reduced school readiness	
	Attendance officer	
	Counselling services offered in school	

Trade-off scenario 7

Please identify which cost and benefit in the education sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important		Least important
	Attendance officer	
	Presenteeism at school	
	Reduced school performance	
	Additional education services provided at a regular school during operating hours	
	Reduced school readiness	

Trade-off scenario 8

Please identify which cost and benefit in the education sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important		Least important
	School completion programs for adults	
	Counselling services offered in school	
	Grade retention	
	Reduced school performance	
	Home education/ homeschooling	

Trade-off scenario 9

Please identify which cost and benefit in the education sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important		Least important
	Reduced school adaptation	
	Reduced school attainment	
	Counselling services offered in school	
	Additional education services provided at a regular school outside operating hours	
	School completion programs for adults	

Trade-off scenario 10

Please identify which cost and benefit in the education sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important		Least important
	Reduced school engagement	
	Student transport to school	
	Reduced school adaptation	
	Negative school experiences	
	Attendance officer	

Trade-off scenario 11

Please identify which cost and benefit in the education sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important	Reduced school attainment Additional education services provided at a regular school during operating hours Presenteeism at school Special education school attendance School-based health promotion intervention	Least important
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Trade-off scenario 12

Please identify which cost and benefit in the education sector you believe is the **MOST** important and which is the **LEAST** important to include in economic evaluations conducted in the disease area of mental and behavioral disorders.

Most important	Additional education services provided at a regular school outside operating hours Problems with school entry Absenteeism from school Reduced school readiness Reduced school competence	Least important
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Are there any additional costs and benefits in the education sector, which you consider important for economic evaluations conducted in the disease area of mental and behavioral disorders that have not been listed? If yes, please specify.

Yes (please specify) _____

No

Closing questions

Please indicate on a scale from 1 to 10, how easy or difficult this questionnaire has been for you.

Easy									Difficult
1	2	3	4	5	6	7	8	9	10

Do you wish to be informed of the study results? If so, please provide your name and email address.

[Yes _____

[No

If you know a health economist that might be interested in completing this questionnaire, please provide their contact details.

We thank you for your time spent taking this survey. Your response has been recorded.

Supplementary File 3. The number of experts who found education and criminal justice costs and benefits relevant to include in economic evaluations vs the number of experts who included these costs and benefits in economic evaluations

	Relevant	Included	N	%
Education costs and benefits	n=31	Yes	10	32.3
		No	21	67.7
Criminal justice costs and benefits	n=29	Yes	10	34.5
		No	19	65.5

Supplementary file 4. The results of the two-sided Fisher's exact test comparing the respondents based on the country of employment and the years of experience in health economics in relation to whether they found education and criminal justice costs and benefits relevant and whether they had previously included them in economic evaluations; $p < 0.025$ was considered statistically significant

	Subgroups	
	Country of employment (Netherlands vs elsewhere)	Years of experience (below 7.1 vs above 7.1)
Relevance of education costs and benefits	p=1.000	p=0.492
Relevance of criminal justice costs and benefits	p=1.000	p=0.113
Previously included education costs and benefits in economic evaluations	p=0.070	p=0.712
Previously included criminal justice costs and benefits in economic evaluations	p=0.458	p=0.453

Supplementary File 5. Best-worst scaling: mean relative importance scores and corresponding 95% confidence intervals for education and criminal justice attributes

Attribute	Average	95% Lower	95% Upper
Education			
Special education school attendance	11.21	9.71	12.71
Absenteeism from school	10.08	8.28	11.89
Reduced school attainment	9.43	7.83	11.04
Additional education services provided at a regular school outside operating hours	7.49	5.88	9.09
Reduced school performance	7.44	5.69	9.2
School-based health promotion intervention	6.2	4.76	7.64
Counselling services offered in school	6.01	4.4	7.62
Reduced school competence	5.89	4.51	7.28
Additional education services provided at a regular school during operating hours	4.86	3.43	6.29
Presenteeism at school	4.42	2.83	6.02
School completion programs for adults	4.39	3.29	5.5
Reduced school readiness	4.04	2.62	5.45
Grade retention	3.69	2.18	5.2
Negative school experiences	3.17	1.98	4.36
Problems with school entry	3	1.89	4.11
Home education/Homeschooling	2.8	1.63	3.96
Attendance officer	2.54	1.52	3.55
Reduced school adaption	1.6	0.98	2.23
Reduced school engagement	0.91	0.4	1.42
Criminal Justice			
Decreased chance of committing a crime	12.94	11.16	14.72
Jail and prison expenditures	10.88	9.14	12.63
Long-term pain and suffering of victims/victimization	9.94	8.31	11.56
Short-term pain and suffering of victims/victimization	8.59	6.72	10.46
Long-term pain and suffering of others	7.21	5.75	8.67

Attribute	Average	95% Lower	95% Upper
Psychiatric assessments in custody	5.83	5.03	6.62
Legal services	5.48	3.95	7.01
Victim compensation	4.98	3.51	6.46
Property loss of victims	4.77	4.09	5.46
Court appearance	4.45	2.8	6.1
Police contact	3.91	2.63	5.18
Victim/witness protection	3.62	3.38	3.86
Short-term pain and suffering of others	3.57	2.23	4.9
Probation/Parole (including electronic monitoring)	3.3	2.65	3.95
Property loss of others	2.76	1.88	3.65
Forensic services	2.73	2.04	3.43
Fire and rescue services	2.3	1.48	3.12
Lost freedom of the offender	1.1	0.67	1.52
Property loss of offender	0.86	0.39	1.33
Illegal untaxed income of the offender	0.78	0.39	1.16

Supplementary File 6. Best-worst scaling: comparison of mean relative importance scores between the subgroups of experts based on the number of years of experience in health economics and the country of employment

Attribute	Over all ranking	Years of experience		Country of employment	
		Ranking subgroup <7.1	Ranking subgroup >7.1	Ranking subgroup employed in the Netherlands	Ranking subgroup employed elsewhere
Education costs and benefits					
Special education school attendance	1	1	2	1	3
Absenteeism from school	2	2	3	2	1
Reduced school attainment	3	4	1	3	2
Additional education services provided at a regular school outside operating hours	4	3	5	4	6
Reduced school performance	5	7	4	6	4
School-based health promotion intervention	6	5	8	5	9
Counselling services offered in school	7	6	7	8	5
Reduced school competence	8	8	6	7	8
Additional education services provided at a regular school during operating hours	9	9	11	9	11
Presenteeism at school	10	10	13	10	13
School completion programs for adults	11	11	12	11	12
Reduced school readiness	12	13	10	13	10
Grade retention	13	16	9	17	7
Negative school experiences	14	12	16	12	16
Problems with school entry	15	16	14	15	14
Home education/Homeschooling	16	15	15	14	17
Attendance officer	17	14	17	16	15
Reduced school adaption	18	18	18	18	18
Reduced school engagement	19	19	20	19	20
Student transport to school	20	20	19	20	19

Attribute	Over all ranking	Years of experience		Country of employment	
		Ranking subgroup <7.1	Ranking subgroup >7.1	Ranking subgroup employed in the Netherlands	Ranking subgroup employed elsewhere
Criminal justice costs and benefits					
Decreased chance of committing a crime as a consequence/effect of mental health programs/interventions	1	1	1	1	1
Jail and prison expenditures	2	3	2	3	2
Long-term pain and suffering of victims/victimization	3	2	3	2	3
Short-term pain and suffering of victims/victimization	4	4	4	4	4
Long-term pain and suffering of others	5	5	5	5	5
Psychiatric assessments in custody	6	6	7	6	8
Legal services	7	7	6	7	6
Victim compensation	8	8	8	9	7
Property loss of victims	9	9	9	8	11
Court appearance	10	10	10	11	9
Police contact	11	13	11	14	10
Victim/witness protection	12	12	13	12	12
Short-term pain and suffering of others	13	11	15	10	17
Probation/Parole (including electronic monitoring)	14	14	12	13	13
Property loss of others	15	15	16	15	14
Forensic services	16	16	14	16	15
Fire and rescue services	17	17	17	17	16
Lost freedom of the offender	18	19	18	18	19
Property loss of offender	19	18	19	20	18
Illegal untaxed income of the offender	20	20	20	19	20

Chapter 5. Development and Harmonization of a Resource-Use Measurement Instrument for the Assessment of Health-Related Multi-Sectoral Resource Use in Europe: The PECUNIA Project

Chapter 5 draws upon:

Pokhilenko, I., Janssen, L.M.M., Paulus, A.T.G., Drost, R.M.W.A., Hollingworth, W., Thorn, J.C., Noble, S., Simon, J., Fischer, C., Mayer, S., Salvador-Carulla, L., Konnopka, A., Hakkaart van Roijen, L., Brodzsky, V., Park, A., Evers, S.M.A.A. Development of an instrument for the assessment of health-related multi-sectoral resource-use in Europe: the PECUNIA RUM.

Submitted for publication.

Abstract

Background. Measuring objective resource-use quantities is important for generating valid cost estimates in economic evaluations. In the absence of acknowledged guidelines, measurement methods are often chosen based on practicality rather than methodological evidence. Furthermore, few resource-use measurement (RUM) instruments focus on the measurement of broader resource use and their development process is rarely described. Thorn and colleagues proposed a stepwise approach to the development of RUM instruments, which has been used for developing cost questionnaires for specific trials. However, it remains unclear how this approach can be translated into practice and whether it is applicable to the development of generic self-reported RUM instruments and instruments measuring resource use in multiple sectors. This study provides a detailed description of the practical application of this stepwise approach to the development of a multi-sectoral RUM instrument developed within the ProgrammE in Costing, resource use measurement and outcome valuation for Use in multi-sectoral National and International health economic evaluAtions (PECUNIA) project.

Methods. For the development of the PECUNIA RUM instrument, the methodological approach was based on best practice guidelines. The process included six steps, including the definition of the instrument attributes, identification of cost driving elements in each sector, review of methodological literature and development of a harmonized cross-sectorial approach, development of questionnaire modules and their subsequent harmonization.

Results. The selected development approach was, overall, applicable to the development of the PECUNIA RUM instrument. However, due to the complexity of the development of a multi-sectoral RUM instrument, additional steps such as establishing a uniform methodological basis, harmonization of questionnaire modules and involvement of a broader range of stakeholders (healthcare professionals, sector-specific experts, health economists) were needed.

Discussion. This is the first study that transparently describes the development process of a generic multi-sectoral RUM instrument in health economics and provides insights into the methodological aspects and overall validity of the instrument. This study could also serve as a guide for researchers who undertake RUM instrument development.

Background

Resource-use measurement (RUM) is known to be challenging and time-consuming, but an essential step in economic evaluations of health care interventions.^{1,2} Measuring valid and reliable quantities of resources utilized is of major importance for generating valid cost estimates.^{1,3} A wide variety of measurement methods exists, including the use of self-reported data by patients collected via questionnaires or cost diaries, the use of administrative data, and the use of expert panels.⁴ However, methodological guidance on developing an appropriate measurement method is limited. There remains a striking difference between the small amount of research dedicated to methods for the appropriate measurement of resource use compared with the large amount of evidence on the measurement of outcomes within economic evaluations.^{1,5,6} Due to the absence of a gold standard and acknowledged guidelines among other factors, the measurement method is often selected based on practicality rather than methodological evidence.¹ Although administrative data sources can provide accurate resource-use estimates, they often fail to capture potentially relevant intersectoral costs from a societal perspective (e.g. out-of-pocket expenses or costs associated with lost productivity).¹ Furthermore, linking multiple databases containing patient-level data can be challenging due to privacy regulations. Therefore, it can be argued that the use of self-reported data, i.e. data collected by administering questionnaires to patients and/or their caregivers, is currently the most feasible method of generating valid resource-use estimates for the measurement of broader resource use.¹

A number of self-reported RUM instruments are available, many of them accessible via the Database of Instruments for Resource-use Measurement (DIRUM).⁷ DIRUM is a repository of RUM instruments based on patient recall for use in trial-based economic evaluations. As of August 2021, it contained 97 RUM instruments developed by researchers worldwide, available in the form of diaries, logs or questionnaires. Out of these instruments, the majority are designed for measuring disease-specific resource use, while only six instruments are meant for generic use.⁸⁻¹³ In addition, most instruments focus on resource use in a limited number of sectors (e.g. health care and social care);¹⁴⁻¹⁷ few also include questions about broader resource use in other sectors (e.g. informal care, education, and (criminal) justice).^{8,11,18,19} Research shows that for some disease areas resource use in sectors outside the health care sector can constitute a large proportion of the total costs and have a significant impact on the study results.²⁰⁻²² The shift towards adopting a societal perspective in economic evaluations calls for appropriate measurement tools that enable measurement of broader resource use.²³ There is little existing research on the feasibility, validity and reliability of RUM instruments,^{3,10,18,19,24} and even fewer studies report the process of instrument development.²⁵⁻²⁷ This is particularly relevant because the decisions taken during the development process of an instrument, such as the framing and order of the questions, can ultimately affect the instrument and responses to its questions.²⁸ Furthermore, in several health-related disciplines (e.g. psychology, health promotion) it is common to report how new instruments were developed,^{29,30} while this is not yet customary in health economics, in particular in relation to RUM instruments.^{27,31,32}

Thorn and colleagues (2013)² proposed a stepwise approach for the development and testing of RUM instruments in trial-based economic evaluations, based on discussions among a large

group of health economists with expertise in RUM. The authors outlined an “ideal way of approaching instrument development”, which entails several stages including planning, development, piloting and deployment of the instrument. Several studies reported using this approach for developing a cost questionnaire for specific trials³³⁻³⁵ without, however, describing the process of applying this approach in detail. Therefore, it remains unclear how this approach can be translated into practice and to what extent it is applicable to the development of a) generic self-reported RUM instruments and b) instruments measuring resource use in multiple sectors.

The current article aims to provide a transparent detailed description of the practical application of the approach proposed by Thorn et al. (2013)² to the development process of the RUM instrument in the ProgrammE in Costing, resource-use measurement and outcome valuation for Use in multi-sectoral National and International health economic evaluations (PECUNIA) project. The PECUNIA RUM instrument aims to address the above-mentioned research gaps by capturing generic resource use in several societal sectors, including 1) health care, 2) social care, 3) education, 4) (criminal) justice, 5) employment and productivity, and costs borne by 6) patients or 7) families including informal care. This instrument will support the measurement of broader resource use and the adoption of a societal perspective in future economic evaluations. The added value of this article is two-fold. First, to the best of our knowledge, this is the first article that describes the development process of a generic multi-sectoral RUM instrument for the use in health economics research. This can be helpful for the researcher who would consider using the PECUNIA RUM in the future by providing insights into the feasibility, reliability and validity of the instrument. Second, by exploring the applicability of the approach described by Thorn et al. (2013) this article aims at providing guidance to researchers who set out to develop a new RUM instrument.

Context: PECUNIA project

The PECUNIA project is a network of ten partners in six countries (Austria, Germany, Hungary, Spain, the Netherlands, and the United Kingdom (UK)) that aims to tackle the dramatic increase in health care costs by establishing standardized costing and outcome assessment measures.³⁶ To reduce unnecessary variations in the resource-use input data of economic evaluations, the PECUNIA project aims to develop an internationally standardised, harmonised and validated, self-reported, multi-sectoral RUM instrument consistent with a harmonised unit costing approach intended for the measurement of generic resource use. Furthermore, to address the variations in the input cost data and to facilitate cross-country transferability of economic evaluations, the PECUNIA RUM instrument is intended to be applicable in multiple European countries. More information about the PECUNIA project structure can be found on the website <https://www.pecunia-project.eu/>.

Methods

The development process of the PECUNIA RUM instrument was based on the approach proposed by Thorn et al. (2013).² The proposed approach covers all stages of instrument development and testing starting from the identification of relevant cost drivers to the piloting and deployment of the instrument. It also suggests optimal time points for considering

important instrument attributes (e.g. recall period, length of questionnaire) as well as consultations with various stakeholders (healthcare professionals, patients, healthy volunteers) at multiple development and testing stages. The PECUNIA group translated this theoretical method into a structured work plan in scientific discussions during multiple joint teleconferences over six months. The development and testing process of the PECUNIA RUM was structured into nine steps with two milestones outlined in Figure 1. The current paper focuses on the application of this approach in practice to the development of the instrument, i.e. the first six steps of the process up until the instrument was ready to be tested.

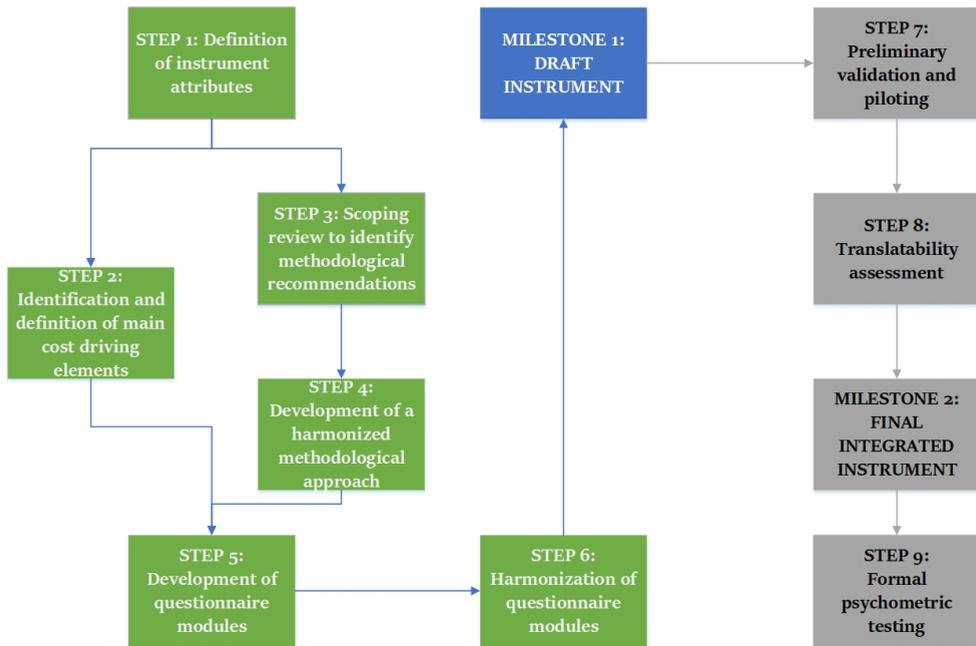


Figure 1. Methodological steps of PECUNIA resource-use measurement instrument development; the green boxes illustrate methodological steps undertaken to develop the instrument, the blue box indicates the milestone of the instrument development, the grey boxes indicate further steps of instrument validation that are not covered in this article (own illustration)

Step 1: Definition of instrument attributes

In step one, the main attributes of the PECUNIA RUM instrument were conceptualized by the PECUNIA consortium in line with the general objectives of the PECUNIA project. General attributes of RUM instruments that ought to be considered were outlined by Thorn et al. (2013) and included perspective, setting of care, disease, population, and nature of intervention. A plan was set up to develop a generic multi-sectoral instrument, which would allow for collecting broader resource-use data (i.e. from a societal perspective) and would be applicable to any setting of care, disease, population and intervention. The instrument would initially be developed in the form of a pen-and-paper version for the use in the adult population and in the English language. While the PECUNIA RUM instrument was intended to be a generic

instrument, mental health was used as an illustrative disease area for the development and preliminary testing (reported elsewhere) of the instrument.

Step 2: Identification, definition and classification of main cost driving elements

In step two, cost-driving elements were identified and defined. First, an independent search of peer-reviewed and grey literature was conducted to identify main cost-driving elements in health care, social care, education, (criminal) justice, employment and productivity, patient, family, and informal care sectors. Second, the list of cost drivers was reviewed by sector-specific experts selected by purposive sampling via a survey (e.g. health care experts reviewed the list of main cost drivers in the health care sectors). The details of this process for the health and social care, (criminal) justice, and education sectors have been described elsewhere.³⁷⁻³⁹ The draft list of cost drivers needed to be defined and organised in conceptually harmonised clusters of units of analysis, which was done according to the PECUNIA Care Atom, a conceptual framework for semantic standardization.⁴⁰ Furthermore, the identified services were classified according to the Description and evaluation of services and directories in Europe - long term care (DESDE-LTC) as modified for the PECUNIA project. DESDE-LTC is a standard taxonomy for description, mapping and comparison of services that was developed over the past twenty years with the aim of improving the comparability of services across settings based on the content of the service rather than semantic equivalence.⁴¹ Additional characteristics of services such as related interventions and professionals were classified according to the International Classification of Health Interventions (ICHI) and the multilingual classification of European Skills, Competences, Qualifications and Occupations (ESCO) as part of the overall PECUNIA Coding system.⁴²

Step 3: Scoping review to identify methodological recommendations

In step 3, a scoping review was conducted to review methodological literature pertaining to RUM and to identify methodological recommendations.⁴³ The results of the scoping review provided an overview of methodological issues in RUM that need to be considered when selecting a measurement method. These included such issues as what type of resource use to measure, how to frame questions and how to deal with missing resource-use data. While the literature was not able to provide clear-cut recommendations regarding RUM, the overview of challenges guided the methodological decisions made in step 4.

Step 4: Development of a harmonised methodological approach

In step 4, a harmonised approach towards the development of a RUM instrument from its separate modules was developed. For each module, cost drivers identified and classified in step 2 were to be matched to questions from existing RUM instruments. If no suitable questions were available, new questions were to be developed. Furthermore, consistent use of units of analysis depending on the setting of care was to be implemented throughout the instrument. Outpatient services corresponded to the measurement unit “per day”, day care services corresponded to the measurement unit “per day” and residential care services corresponded to the measurement unit “per night”. Decisions on the methodological aspects of the instrument were based on the findings of the scoping review (step 3). The questions were to

be developed to be clear and easily understandable by lay audience avoiding the use of double negatives, asking about more than one type of resource-use per question and ensuring mutual exclusivity of the questions. Consistent recall period was to be implemented throughout the instrument. The methodological approach was presented to health economists and health technology assessment experts at the PECUNIA satellite workshop at the annual conference of the International Health Economics Association (iHEA) in Basel (Switzerland) in 2019.

Step 5: Development of questionnaire modules

In step 5, the harmonised approach (step 4) was employed to develop RUM instrument modules. The process described in step 4 was applied to the development of all PECUNIA RUM instrument modules with the exception of the module on employment and productivity which was based on an existing instrument, the Institute of Medical Technology Assessment (iMTA) Productivity Cost Questionnaire (iPCQ). iPCQ has been shown to be an optimal instrument for measuring productivity losses in economic evaluations conducted from a societal perspective⁴⁴ and it has been extensively used to measure productivity losses (i.e. absenteeism and presenteeism) of paid and unpaid work. The development process and the validation of the iPCQ have been described in previous studies.^{27,45}

Also in step 5, the DESDE-LTC codes developed in step 2 were used to establish the connection between the services included in the PECUNIA RUM instrument. The PECUNIA costing concept based on the PECUNIA Care Atom linked the PECUNIA RUM instrument and the other PECUNIA costing tools including the PECUNIA Reference Unit Cost (RUC) Templates and the PECUNIA RUC Compendium⁴⁶ to ensure that sufficient level of detail of resource-use data is collected for costing purposes. Some questions were expanded to accommodate the collection of detailed resource-use data, which is necessary for accurate costing. For example, the unit costs of additional educational support might differ depending on the setting of service provision (public vs private). Therefore, this differentiation was introduced in the module on education.

Step 6: Harmonization of questionnaire modules

Because the PECUNIA RUM instrument modules were developed by different working groups, the format, phrasing, order of the questions and answer options needed to be harmonised. This was carried out iteratively in multiple teleconferences and rounds of revision. The setup of each questionnaire module was standardized. The text of the draft PECUNIA RUM instrument underwent a formal wording review in February 2020 followed by a professional English language editing in June 2020. Based on these assessments, the instrument was further revised regarding the phrasing.

Results

PECUNIA RUM instrument

The PECUNIA RUM instrument was designed to contain eight modules corresponding to various types of resource use (Table 1). Each module begins with a short description of its content including the types of resource use measured and the applicable recall period. The suggested recall period was overall set at three months. However, since the evidence on the

optimal recall period, in particular regarding employment and productivity,^{47,48} remains inconclusive, this methodological choice will have to be further scrutinized in the testing phase. Consistent format, phrasing, wording, order of the questions and the answer options were implemented.

Table 1. Content of the PECUNIA RUM modules

Module	Types of resource-use measured
Place of living and overnight stays	Usual living situation, residential care, and institutional stay selected from a range of residential, health, social, educational and correctional facilities
Non-residential health and social care	Use of health and social services including outpatient, daycare, helplines, and vocational services
Medication	Use of medications
Unpaid help (informal care)	Informal care provided by the respondent's friends, relatives, neighbors or volunteers
Education	Highest level of education, current educational status, absenteeism and presenteeism during education, the use of education services (e.g. tutoring)
Employment and productivity	Current employment status, absenteeism and presenteeism at paid and unpaid work
Safety and justice system	Contacts with police, fire-and-rescue and legal services, material damage caused by the respondent (e.g. theft, vandalism), incarceration
Out-of-pocket and other expenses	Personal expenses including expenses for household help, childcare, purchase of goods (e.g. wheelchair)

It is important to note that the number of questionnaire modules (eight) does not correspond to the number of sectors (seven) included in the PECUNIA RUM instrument. This was done to facilitate the adaptability of the instrument to various contexts. For example, the module on medication use is separate from the module on resource use in health and social care sectors as it might not be relevant to include it in every study. This allows omitting it entirely without compromising the coherence of the instrument.

Several techniques were employed to facilitate the feasibility and understandability of the questionnaire for the respondents. First, the questions were accompanied by instructions such as:

“Please tick all answers that apply and indicate the number of contacts you had with a given service”.

Second, examples were provided to aid recall such as:

“Specialist medical care (e.g. orthopaedist, psychiatrist, gynaecologist)”

Third, an additional explanation was provided to make sure that the respondents understand what type of resource use they need to recall, for example:

“Have you used legal services (e.g. contact with a lawyer) in the past 3 months? This could be a face-to-face meeting, an online consultation or a phone call with someone working in legal services.”

Also, skip logic was introduced to direct the respondents through the questionnaire (please refer to Figure 2 for an example). For most types of resource use, respondents were first asked whether they have used a resource; if yes, follow-up questions were asked; if not, the respondent could jump to the following module. This allows the respondents to focus on only relevant questions given their resource use, minimizing burden on respondents.

E6	Have you used education support services in the past 3 months?	
	<i>This includes any additional educational support you received, such as tutoring, additional lessons, etc.</i>	
<input type="checkbox"/>	Yes	<i>Please go to question E6.1</i>
<input type="checkbox"/>	No	<i>Please go to question E7</i>
<input type="checkbox"/>	I don't remember/I would rather not say <i>Please go to question E7</i>	
E6.1	How much did you use other education support services in the past 3 months?	
		Average number of hours per week
1	Education support at your place of study	_____ hours
2	Education support in a private setting (e.g. private tutoring)	_____ hours
3	Other, please specify: _____	_____ hours

Figure 2. PECUNIA RUM (draft version): example questions from the section on education resource-use

Expert feedback

At the PECUNIA satellite workshop in July 2019, the experts indicated that the development of a multi-sectoral RUM instrument that would be applicable to a variety of settings was a difficult process requiring a lot of consensus building. Nevertheless, it was agreed that such an instrument is needed to facilitate the comparability of the results of health economic evaluations.

When the draft instrument was presented to health economists in November 2019, the experts agreed that the availability of a standardized measure was important. They also argued that the full version of the instrument would often not be applicable to a specific setting, as the relevance of cost categories is dependent on the nature of the intervention in question. The experts suggested that the instrument could be used as a basis that can be further adapted to specific research requirements. Developing a manual with suggestions regarding the selection of relevant cost categories and hence relevant modules of the PECUNIA RUM instrument (e.g. consulting the patients and/or the literature) was recommended. The experts also raised concerns regarding the applicability of the PECUNIA RUM instrument to all countries in Europe due to the differences in, e.g., health care system structure. The planned validation activities and the link to the compatible method for the unit cost calculation were perceived as major strengths of the PECUNIA RUM instrument.

Application of the process

The method by Thorn et al. (2013) was, overall, applicable to the development of the PECUNIA RUM instrument. As proposed in the method, cost drivers and instrument attributes were defined at the start. Stakeholders (sector-specific experts and health economists) were involved at an early stage of the development process and potential respondents (patients and caregivers) will be involved at later stages when the instrument will be tested. Furthermore, the PECUNIA RUM instrument was developed in conjunction with a standardized costing tool which allows for harmonized collection of resource use and cost data. However, the development process of the PECUNIA RUM instrument deviated from the method of Thorn et al. (2013) in several ways. First, the method recommends that healthcare professionals are involved in the development process. However, due to the multi-sectoral nature of the PECUNIA RUM instrument, a broader range of experts were involved in the development and testing, including experts from other sectors (e.g. education, (criminal) justice, productivity) and health economists. Second, while the method of Thorn et al. (2013) prescribes reliance on existing RUM instruments when developing a new one, the development of the PECUNIA RUM instrument is guided by existing RUM instruments only to a limited extent. This can be attributed to the lack of multi-sectoral RUM instruments comprehensive RUM instruments that incorporate cost categories beyond the health care. Existing instruments were used as illustrative examples, but no parts of questionnaires or questions were used verbatim, with the exception of the module on employment and productivity, which was fully based on an existing RUM instrument (iPCQ). Furthermore, many existing RUM instruments were developed for use in specific contexts,⁴⁹ while the PECUNIA RUM instrument was meant to be a generic internationally applicable instrument. Third, two additional steps in the planning phase of the Thorn et al. (2013) method were needed to account for the complexity of developing a multi-sectoral RUM instrument, i.e. scoping review (step 3) and establishing a uniform methodological basis (step 4), and one additional step in the development phase, i.e. harmonization of instrument modules (step 6).

Discussion

The aim of this article was to provide a transparent detailed description of the practical application of the method described by Thorn et al. (2013) for the development of the PECUNIA

RUM instrument. To our knowledge, this is the first study that transparently describes the development of a generic multi-sectoral RUM instrument in detail. The development process was based on best practices² and consisted of six steps starting from the identification of cost drivers up to harmonization of the instrument modules. The methods described by Thorn et al. (2013) were, overall, applicable to the development of this generic multi-sectoral RUM instrument. However, because the instrument comprises eight modules developed by four distinct research teams, additional steps such as establishing a sound methodological basis (step 4) and harmonization of the instrument modules (step 6) were necessary. On the other hand, a scoping review (step 3) is not likely to be a necessary step for developing new RUM instruments, as the current scoping review's findings have broader applicability beyond this study. Furthermore, in addition to healthcare professionals, other sector-specific and health economics experts needed to be involved in the development process. This is due to the inclusion of more cost categories beyond health care and the overall complexity of the instrument, for which the involvement of healthcare professionals would not be sufficient to get insights into the feasibility of the instrument.

Compared with other studies that reported using the same method for the development of a RUM instrument,³³⁻³⁵ the current study provides more detail and insights into the development process. This could be helpful by providing guidance to researchers undertaking the development of RUM instruments, but also by providing insight into the context in which methodological choices behind the current instrument were made. Several previous studies reported the development process of either a disease-specific RUM instrument²⁵ or an instrument intended for measuring resource use related to productivity losses,²⁷⁻⁵⁰ which limits the generalizability of the process to the development of generic multi-sectoral RUM instruments. In contrast, the current study provides a structured stepwise approach to instrument development based on available best practices² and presents the development process of an instrument that relies significantly less on existing RUM instruments. Furthermore, this study demonstrates that the methodological approach to the development of a RUM instrument is determined by the type of a RUM instrument being developed. For example, in case of the PECUNIA RUM, the involvement of a broader range of experts in the instrument development process was needed due to the multi-sectoral nature of the instrument.

The PECUNIA RUM instrument contains questions on resource use in several societal sectors that could be affected by health interventions including health and social care, education, (criminal) justice, employment and productivity, patient, family, and informal care. The instrument is intended for measuring self-reported generic resource use in trial-based economic evaluations in Europe. Compared with other generic RUM instruments available in DIRUM,⁷ the PECUNIA RUM instrument allows for measuring resource use in more sectors (e.g. education, (criminal) justice) and for collecting standardized evidence for informing optimal decision-making. Connection to the DESDE-LTC coding, which facilitates the comparability of services, and to the compatible PECUNIA costing tools allows for producing comparable resource-use and cost estimates across studies. The RUM instrument was developed by a multinational consortium and will be tested in several European countries in

addition to formal translatability assessment to ensure its applicability to various geographical, linguistic, cultural and system settings. In addition, the flexible structure of the instrument makes it possible to adapt the instrument to a specific setting by selecting only relevant modules. This allows researchers to make a trade-off between, on the one hand, having an incomplete instrument that is easier to complete and, on the other hand, a comprehensive instrument that is too lengthy and difficult for the respondents to fill in.

The PECUNIA RUM instrument can be compared with other commonly used comprehensive RUM questionnaires covering broader resource use such as the Client Service Receipt Inventory (CSRI),¹² the Treatment Inventory of Costs in Patients with psychiatric disorders (TiC-P),¹⁰ or the Health Economics Questionnaire (HEQ).⁸ The aim of developing the PECUNIA RUM instrument is not to replace other existing instruments, but to offer a broader alternative, which has certain benefits such as a valid, structured and documented development process, inclusion of more resource-use items and cost categories in- and outside of the healthcare sector, and a modular structure, which makes the instrument more easily adaptable to various contexts. Furthermore, the PECUNIA RUM is intended to be a multicountry instrument with broad applicability in Europe and its link to a standardized costing tool allows for producing harmonized cost data.

Reflections on the development process

Several aspects of the development process need to be mentioned. First, using a different methodological approach to the development of the PECUNIA RUM instrument could have resulted in a different instrument both in terms of the content as well as the structure. The PECUNIA group selected the method developed by Thorn and colleagues (2013) as, to the best of our knowledge, it is the only available systematic method for the development of a RUM instrument in health economics. This approach was generally applicable given the aim of the PECUNIA project. However, due to the complexity of the instrument, additional steps were needed. Second, as argued by the health economics experts, the applicability of the instrument to various settings (e.g. patient groups, disease areas) is difficult to ensure during the development phase. This would require additional testing of the instrument in different patient groups with respect to disease and country-specific context. Although the PECUNIA RUM instrument will initially be tested among former mental health service users and carers, the cost categories included in the instrument are also relevant for other conditions (e.g. chronic diseases).⁴⁹ Third, instruments used for data collection in research are developed by researchers with high educational level and specialized knowledge, while the instruments are usually meant to be completed by patients and caregivers with varying levels of education and (health) literacy. To ensure understandability of the text, a formal wording review and revision of the text by a professional English editor were conducted. Fourth, RUM instruments are commonly used to collect resource-use data in randomized controlled trials alongside many other measures. This means that participants are often asked to fill in multiple questionnaires at each measurement point. Therefore, it would be important to determine the appropriate length of the questionnaire by finding a balance between sufficient detail to obtain necessary data and patient burden. Testing the instrument with the potential end-users will provide more insights into whether the current length is optimal.

Implications for further steps

To optimize the draft PECUNIA RUM instrument, a piloting exercise will be conducted in the form of think-aloud interviews. Mental health service users and informal caregivers of patients with mental illness will be consulted in relation to the completeness, difficulty and feasibility of filling in the instrument. Furthermore, the instrument will undergo linguistic translatability assessment to prepare the instrument for formal translation to multiple languages. The results of the preliminary validation and piloting will be used to develop the final, integrated, multi-sectoral PECUNIA RUM instrument.⁵¹ In the future, the PECUNIA RUM instrument is planned to undergo formal psychometric testing and several further versions of the instrument will be developed (e.g. child and proxy versions). More extensive, i.e. midi and maxi versions of the instrument will also be developed that would allow for a more detailed data collection for specific health care systems and costing exercises. In addition, as suggested by the health economics experts, a user guide will be developed with the aim of providing guidance on how to use the PECUNIA RUM instrument and how to apply it to a specific setting for the researchers who plan to use the instrument in the future.

Conclusion

This study describes the practical application of the method outlined by Thorn et al. (2013) to develop the PECUNIA RUM instrument, a multi-sectoral generic RUM instrument. The described process can be used as a guide for researchers who want to undertake the development of a RUM instrument. The process was divided into six steps starting from the identification of general attributes of the instrument (e.g. perspective, disease) up to the harmonization of the instrument modules in relation to the format, phrasing and order of the questions to form a coherent instrument. While the method by Thorn et al. (2013) was, overall, suitable for the development of the PECUNIA RUM instrument, additional steps were needed to account for the complexity of developing an instrument to measure broader resource use. Further steps are needed to test the validity of the instrument in different settings and population groups.

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Chapter 6. International Comparability of Reference Unit Costs of Education Services: When Harmonizing Methodology Is Not Enough (PECUNIA Project)

Chapter 6 draws upon:

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Abstract

Background. Health problems can lead to costs in the education sector. However, these costs are rarely incorporated in health economic evaluations due to the lack of reference unit costs (RUCs) of education services and of validated methods to obtain them. In this study, a standardized unit cost calculation tool developed in PECUNIA project, the PECUNIA RUC Template for services, was applied to calculate the RUCs of selected education services in five European countries.

Methods. The RUCs of special education services and of educational therapy were calculated using the information collected via an exploratory grey literature search and contact with service providers.

Results. The RUCs of special education services ranged from €57 to €189 per day. The RUCs of educational therapy ranged from €6 to €27 per contact and from €5 to €77 per day. Variation was observed in the type of input data, service unit, representativeness, and the components included in the RUCs.

Discussion. Despite the use of a standardized tool, the RUCs still varied. The tool helped to gain insights into the aspects beyond methodology that could contribute to the variability. Further research is required to reduce the variability of the RUCs attributed to these aspects.

Introduction

Health and education are interrelated. On the one hand, education is one of the social determinants of health, which is associated with better health outcomes.¹⁻³ This association can be explained by the healthier lifestyle and higher (health) literacy of people with a higher education, along with other mechanisms.¹⁻³ On the other hand, a person's ability to participate in education can be affected by their health status.⁴ Poor health has been associated with lower academic achievement⁵ and lower educational attainment.⁶ Accordingly, interventions with the primary intent of improving health also have the potential to improve educational outcomes. For example, Belot and James⁷ find that healthy school meals were associated with higher test scores and lower rates of absenteeism. Similarly, the findings of a study by Levine and Schanzenbach⁸ suggest that health insurance coverage was associated with improved test scores in reading.

Health problems also often have economic implications for the education sector, particularly in the population of children and adolescents. For example, the study by Le and colleagues⁹ has shown that costs associated with providing additional education for children and adolescents with attention deficit hyperactivity disorder could account for up to 60% of the total costs associated with the condition. At the same time, health interventions can lead to economic benefits in the education sector, as demonstrated in the study by Wellander and colleagues,¹⁰ using the example of an intervention to reduce mental health problems among students. Accordingly, taking education costs into account in health economics research could be crucial for informing optimal decision-making at the societal level, particularly in the context of interventions targeting children and adolescents with health problems.

Yet the number of health economics studies that take into account costs in the education sector is limited.^{11,12} This can be attributed to the predominance of the narrow healthcare perspective in the majority of economic evaluations performed,^{11,12} but also to the limited methodological guidance on how to properly value education costs in health economics studies. As demonstrated in the recent review, studies in which education costs and/or consequences were taken into account vary widely, in particular in terms of the valuation methods.¹³ For example, in two recent studies in Sweden, the costs of additional educational support were valued differently: Lenhard et al.¹⁴ based their estimate on national-level data and Jolstedt et al.¹⁵ used the average market price of this service. Moreover, many studies did not report how the education costs and/or consequences were valued, which further hampers the comparability between the estimates and hence the study results.¹³ To improve the comparability of health economics cost data, several countries developed national unit costs for common components of resource-use (e.g. health and social care services, medication prices).^{16,17} However, the unit costs of education services are rarely available. Furthermore, while national unit costs facilitate the comparability of national evidence, the cross-country comparability of unit costs remains an issue.

The standardization of valuation methods and the development of multi-national unit costs in health economics research has the potential to improve the comparability, applicability, and transferability of health economic evidence within and across countries. Therefore, the ProgrammE in Costing, resource use measurement, and outcome valuation for Use in multi-

sectoral National and International health economic evaluations (PECUNIA) project set out to develop standardized methods and tools for resource-use measurement and unit cost calculation, with the aim of producing comparable cost data for health economic evaluations across Europe.¹⁸ PECUNIA also broadened the scope beyond the healthcare sector and incorporated other societal sectors that can be relevant for health economic evaluations: health and social care, education, criminal justice, labor and productivity, and patient, family and informal care. This particular study focuses on the education sector. The aim of this study is to provide the reference unit costs (RUCs) for selected educational services by applying the standardized unit cost calculation tool, the PECUNIA RUC template for services, in five European countries. These RUCs are available for use in future national and multi-national economic evaluations in Europe in an online database, the PECUNIA RUC Compendium.¹⁹

Methods

PECUNIA RUC template for services

The study aimed to calculate and compare the RUCs of selected services in the education sector by applying the PECUNIA RUC template, a methodologically validated Excel template for a guided stepwise calculation of RUCs.²⁰ The template for calculating the RUCs of services allows for developing unit costs using a top-down micro-costing or a top-down gross costing approach, using either primary or secondary input data. A detailed description of the template and its development is available elsewhere.²¹ In this study, the template for the RUC calculation of services using a top-down gross costing was used. This template was validated by the PECUNIA group by calculating RUCs of various services, including one service provided in the education sector in five of the six countries that participated in the PECUNIA project (Austria, Germany, Hungary, the Netherlands, and the United Kingdom (UK)) in November 2019.

Selection of services

For this study, the template for calculating the RUCs of services was applied to three selected education services in Austria, Germany, Hungary, the Netherlands, and the UK. The services were selected from a comprehensive overview of costs and consequences in the education sector, developed at an earlier stage of the PECUNIA project. The list was developed by conducting a systematic review of peer-reviewed and grey literature and a survey completed by an international group of experts in the education sector. For a detailed description of the development process of the list of services, we refer the reader to Pokhilenko et al.²² From this list, three education services were selected: “education services provided in a special education school (either primary or secondary)”, “educational therapy provided in primary schools”, and “educational therapy provided in secondary schools”. Based on previous research, it was expected that these three services would exist in all participating PECUNIA countries and be frequently used by children and adolescents with mental problems.¹³ Furthermore, each of the services was accompanied by a more detailed description developed by the authors of the study in consultation with the expert in the education sector (Table 1).

Table 1. Education services and service descriptions selected for reference unit cost calculation

Service name	Service description
Education services provided in a special education school (either primary or secondary)	Education services provided in the special education school, as a day care facility, for students with mental or physical disabilities
Educational therapy provided in primary schools	Additional educational support provided as an outpatient service at a regular primary school to students with learning difficulties (often referred to as remedial teaching)
Educational therapy provided in secondary schools	Additional educational support provided as an outpatient service at a regular secondary school to students with learning difficulties (often referred to as remedial teaching)

Data collection

For this study, the top-down gross costing approach was selected by the PECUNIA group as it was the only method feasible given the timeframe of the project and the ongoing COVID19 pandemic. Top-down gross costing relies on aggregated data gathered, for example, in the reports of national ministries or national statistics organizations.²³ To collect relevant input data, an exploratory search of grey literature (e.g. ministry reports, and reports of national statistics organizations) was conducted in all five countries that participated in this study, namely Austria, Germany, Hungary, the Netherlands, and the UK. The minimally required data for calculating the RUCs using this approach included total costs associated with the provision of each service for a given period of time (i.e. per year) and the total number of client contacts for the same period of time. For insights into the comparability of the estimates, additional information regarding the components of the RUCs (e.g. staff costs, overhead costs) is required: in case the source did not include information in the required level of detail, the authors contacted the data provider directly. Any additional assumptions necessary for calculating the RUCs (e.g. regarding the average number of school days per year or the average number of students per classroom) are transparently documented and motivated by empirical data where possible.

In case no secondary data was available, the alternative strategy was to collect primary data. Relevant service providers, i.e. the providers of the service for which the RUC was developed, were contacted and asked to fill out a separate data collection sheet. In the data collection sheet, the service providers were asked to provide information regarding the costs incurred due to providing the given service, such as annual direct and overhead costs and the number of client contacts for a given period of time (e.g. in a given year). These data were then fed into the PECUNIA RUC template.

The RUCs of the three selected education services were compared based on several aspects, including the service unit, costing method and the type of input data used for the calculation, components included in the unit cost, and the representativeness of the estimates. To facilitate

the comparison, all values were converted to 2019 Euros using the national Consumer Price Index.

Results

Education services provided in a special education school

The RUCs of the education services provided in a special education school ranged from €57 per day in the Netherlands to €190 per day in Austria (Table 2). All RUCs corresponded to the service unit of “school day per student” and were calculated based on secondary data. The Dutch government report used for the RUC calculation allowed for the calculation of separate estimates for the education services provided in primary and secondary special education schools.²⁴ The RUC of the services provided in secondary special education schools was approximately twice as high as the RUC of the services provided in primary special education schools. Four out of five RUCs were national-level estimates; the RUC in the UK was a local authority level estimate. The components of the RUCs were known for four out of five estimates (with the exception of the UK).

Table 2. Reference unit costs of education services provided in a special education school (2019 EURO)

Country	Reference unit cost (EUR)	Service unit	Input data	Representativeness	Cost components included
Austria	188.85	School day per student	Secondary data	National	Staff costs, investments, overhead costs
Germany	99.54	School day per student	Secondary data	National	Staff costs, overhead costs, costs for allocation of grants and exemptions
Hungary	64.02	School day per student	Secondary data	National	Staff costs, other direct costs, overhead costs
The Netherlands	57.09 ¹	School day per student	Secondary data	National	Average expenditure per student excluding the cost of housing
	128.17 ²	School day per student	Secondary data	National	Average expenditure per student excluding the cost of housing

Country	Reference unit cost (EUR)	Service unit	Input data	Representativeness	Cost components included
The United Kingdom	73.12	School day per student	Secondary data	Local authority	No breakdown of the costs reported

¹education services provided in a primary special education school

²education services provided in a secondary special education school

Educational therapy provided in primary schools

The RUCs for the educational therapy provided in primary schools ranged from €7.73 to €27 per contact and from €4.51 to €34.56 per school day (Table 3). In Austria no data on educational therapy was available. Therefore, the RUC was based on the proxy service – education services provided in a regular primary school. Since approximately 61% of students with special needs attend regular primary and secondary schools,²⁵ this service was considered to be the closest alternative given the absence of appropriate data. The RUC was based on the average staff costs per hour of teaching per student in a regular primary school. Similarly, in Germany the majority of the students with special needs attend regular primary schools. The RUC of educational therapy was based on the average cost per hour of teaching per student in a regular primary school. In Hungary the RUC of educational therapy was calculated as the difference in daily costs between schools with and without programmes for students with special education. Furthermore, since primary and secondary education is typically provided in the same institution, a single estimate was calculated for educational therapy provided in primary and secondary schools. In the Netherlands, the RUC was calculated for a proxy service - remedial teaching provided in a private setting after school instead of in a daycare facility (as in the original definition). This estimate was based on primary data collected from the service provider and it was the same regardless of the education level, i.e. the cost was the same for primary and secondary school students.

Table 3. Reference unit costs of educational therapy provided in a primary school (2019 EURO)

Country	Reference unit cost (EUR)	Service unit	Input data	Representativeness	Cost components included
Austria	7.73	Contact per student	Secondary data	National	Staff costs
Germany	34.56	School day per student	Secondary data	National	Staff costs, overhead costs, costs for allocation of grants and exemptions

Country	Reference unit cost (EUR)	Service unit	Input data	Representativeness	Cost components included
Hungary	4.51 ¹	School day per student	Secondary data	National	Expenditure on teachers' salaries, expenditure on other salaries, material costs, investment, renovation, other expenditure
The Netherlands	26.17 ¹	Contact per student	Primary data	Regional	Staff costs, costs for purchasing equipment
The United Kingdom	23.99	School day per student	Secondary data	Local authority	No breakdown of the costs reported

¹The RUCs for educational therapy provided in primary and secondary schools are the same.

Educational therapy provided in secondary schools

The RUCs of educational therapy provided in secondary schools ranged from €5.82 to €26.17 per contact and from €4.51 to €13.36 per school day (Table 4). The calculation of the RUCs for Austria and Germany followed the same principle as described in paragraph 3.2 using the data on regular secondary schools. The Hungarian and Dutch RUCs for this service were the same regardless of the educational level.

Table 4. Reference unit costs of educational therapy provided in a secondary school (2019 EURO)

Country	Reference unit cost (EUR)	Service unit	Input data	Representativeness	Cost components included
Austria	5.82	Contact per student	Secondary data	National	Staff costs
Germany	14.64	Contact per student	Secondary data	National	Staff costs, overhead costs, costs for allocation of grants and exemptions

Country	Reference unit cost (EUR)	Service unit	Input data	Representativeness	Cost components included
Hungary	4.51 ¹	School day per student	Secondary data	National	Expenditure on teachers' salaries, expenditure on other salaries, material costs, investment, renovation, other expenditure
The Netherlands	26.17 ¹	Contact per student	Primary data	Regional	Staff costs, costs for purchasing equipment
The United Kingdom	13.36	School day per student	Secondary data	Local authority	No breakdown of the costs reported

¹The RUCs for educational therapy provided in primary and secondary schools are the same.

Differences and similarities of RUCs

The RUCs corresponded to either of the two harmonised units of measurement: “per (school) day” (n=10, 67%) or “per contact” (n=4, 33%). Ten RUCs were nationally representative, one RUC was representative of a specific region, and three RUCs were representative of the local authority level. The majority of the RUCs (n=13, 87%) were calculated based on publicly available secondary data; one RUC was calculated based on primary data obtained from the service provider. The RUCs varied in terms of the cost components included. The most frequently included cost components included staff costs (n=11) and overhead costs (n=6). For the three RUCs from the UK, the components included in the unit costs were unknown. Furthermore, there were differences in the definition of the services for which the RUCs were developed. For example, the Dutch RUC of educational therapy was calculated based on the proxy service of remedial teaching provided in the private setting. Further details of the RUCs can be obtained from the PECUNIA RUC Compendium¹⁹ or from the corresponding author of this study.

Discussion

This study applied the novel standardized methods and tool for unit cost calculation developed in the PECUNIA project, the PECUNIA RUC Template for services, to three selected education services in Austria, Germany, Hungary, the Netherlands, and the UK. The RUCs calculated in this study could be used by researchers in future economic evaluations, although the

remaining limitations of the estimates need to be taken into account. The results also suggest that despite an attempt to standardize the methodological approach, many other potential sources of variability affect the comparability of the RUCs - for example differences in the definition of the services and in the type of data available for the calculation. The PECUNIA RUC template has the potential to facilitate the comparability of the RUCs by offering a standardized and transparent methodological approach to RUC calculation. However, researchers should be aware that other aspects beyond methodology, such as those related to data availability and composition, can hinder the comparability of the RUCs and require further international standardization and harmonization of data collection methods. For example, if the components of RUCs are different (e.g. overhead costs are (not) included), the estimates cannot be directly compared.

The absence of a universally accepted gold standard for the valuation of services in health economics might lead to variability in the RUCs. For example, Mayer and colleagues calculated a unit cost of general practitioner consultation in Austria using six costing methods, and found an overall variation of 173%.²⁶ Despite the use of a standardized methodological approach, the results of this study still demonstrate substantial variation. For example, the RUCs of education services provided in a special education school ranged from €57 to €190. Variability of RUCs can lead to lack of comparability of the results of economic evaluations and can affect the decisions made based on the results. Variability can be attributed to various reasons. Mogyorosy and Smith²³ outline eight potential sources of variability of RUCs of health services, including differences in costing methodologies (e.g. differences in resource use measurement and valuation), differences in cost accounting systems and practices, differences in inclusion and exclusion of particular costs, differences in controlling regional and seasonal variations during costing, structural/organisational differences between providers, differences in patient case-mix and socio-demographic characteristics, and differences in financial and non-financial incentive systems, as well as geographical differences in input prices.

In this study, several potential explanations for the variability could be observed. First, the description of services differed across the five countries. This can be attributed to many factors, including differences in national educational systems, differences in the interpretation of the services and their descriptions, and the lack of (suitable) input data to calculate the RUC for the originally selected service. Second, differences in the type of data used for the calculation could contribute to the variation. When using aggregate secondary data for top-down gross costing or relying on existing estimates, it is not always clear what components are included in the calculation. This was the case for all the RUCs provided in the UK. On the other hand, calculating the RUCs on the basis of primary data might produce an estimate that is only relevant for a specific context and is not directly transferable. Differences in the type of input data available was also one of the major observed sources of variability related to the differences in cost accounting practices. This was reflected in the differences in the service unit, the representativeness of the data and the components included in the data. When available data were limited, the researchers had to make assumptions relying on the published literature but also on their own judgement, which further limited the comparability of the estimates. Geographical differences in input prices could also potentially explain some of the

variability. However, the RUCs calculated in this study were not adjusted for purchasing power parity, which could explain the variability attributed to the differences in price level across countries.

Methodological reflection

This study presents the first attempt to apply standardized and harmonized method and tool, the PECUNIA RUC template for services, to the calculation of the RUCs of education services in health economics. The tool was tested in five European countries and provides insights into different sources of variability of the unit costs. It is important to note that while the PECUNIA RUC Template provided a structured methodological approach to the calculation of RUCs, the input data for the calculation needed to be obtained from various primary or secondary sources. Therefore, the quality of the RUCs remains dependent on the quality of the input data used for the calculation. The RUCs calculated based on reliable nationally representative secondary data are expected to be of higher quality in comparison with the RUCs calculated based on primary data collected from a single service provider (e.g. the RUC of educational therapy in the Netherlands).

Setting research agenda

The current study and the overall PECUNIA Project was prompted by the lack of harmonization, standardization and overall guidance on how to approach unit cost calculation in health economics research. This is important because a standardized approach can help produce comparable health economics data for informing policy decision-making. Although the PECUNIA RUC template for services provides an important contribution to standardizing unit cost calculation in health economics, there are other research gaps that could guide future research in this domain. First, focusing on the aspects that can affect variability of the RUCs proposed by Mogyorosy and Smith,²³ further insight is needed into the aspects that could not be detected in the current study. Also, shedding light on the aspects that affect the variability the most can help concentrate efforts on optimizing the approach to unit cost calculation more effectively. Second, variation was observed in the description of services across countries. The use of a standardized terminology and classification system can help facilitate national and international comparison of services. One example of such a system that has been used in health economics is the standard taxonomy for description, mapping and comparison of services for long-term care, DESDE-LTC.²⁷ DESDE-LTC was developed to classify services in different societal sectors, including services in the education sector. This system provides common terminology and codes to enable comparison of services across settings. The use of DESDE-LTC for the classification of services in the education sector in health economics research has been explored in the PECUNIA project; however, further work to develop and test the classification of services in the education sector is needed to ensure that the system is fully functional.

Third, the results of the current study clearly demonstrate that both the availability and the type of data available for unit cost calculation are major contributors to unit cost variability. This highlights the need for promoting uniform accounting and data reporting standards across Europe. Fourth, as available RUCs for education services are limited, establishing a RUC

database that includes education costs among costs in other sectors could facilitate the inclusion of intersectoral costs and consequences in health economics studies. This was tackled in the PECUNIA project by developing the PECUNIA RUC Compendium. The PECUNIA RUC Compendium is a multi-sectoral, multi-country database of RUCs comparable across countries and sectors developed using standardized and scientifically validated methods.¹⁹ It is designed to be a living database planned to be updated and expanded both in terms of the services covered and countries. Finally, as unit cost calculation in health economics often involves a trade-off between accuracy and practicality, transparent reporting of the methodological approach is crucial for correct interpretation. This can be facilitated by the use of the PECUNIA costing tools – the PECUNIA RUC Templates and the PECUNIA RUC Compendium.

Conclusion

This study presents the first attempt at using a standardized unit cost calculation tool, the PECUNIA RUC Template for services, to calculate RUCs of education services for use in health economic evaluations across Europe. While the use of a standardized costing tool helped harmonize the methodological approach towards unit cost calculation, the RUCs still varied; differences in the RUCs can be attributed to aspects beyond methodology (e.g. the type of input data used for the calculation). While the PECUNIA RUC Template for services can help reduce variability attributed to the methodological approach, further research and harmonization is required to tackle other potential sources of variability.

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Chapter 7. General Discussion

Mental problems have a wide impact on various sectors of society, including the education sector.¹ In comparison with their healthy peers, children and adolescents with mental problems are more likely to have trouble with schooling, which can be reflected in difficulties concentrating, impaired school productivity, school absenteeism, lower educational outcomes, and lower educational attainment.^{2,3} Often, mental problems are also associated with higher costs for the education sector, due to the need for providing additional support to these students in the form of tutoring, classroom assistance, and special education services.^{4,5} Interventions to prevent or treat mental problems can lead not only to better health outcomes for these students, but also improve their educational outcomes and ultimately provide economic benefits (i.e. savings) to the education sector. Taking these costs and benefits into account in economic evaluations of mental health interventions targeting children and adolescents is essential for informing policy decisions regarding optimal allocation of resources. Nevertheless, costs and benefits in the education sector are rarely included in health economic evaluations.^{6,7} This can be attributed, among other factors, to lack of awareness regarding the relevance of these costs and benefits to economic evaluations, as well as the lack of evidence-based standardized methods for including them. Furthermore, with the increase in multi-national health economics studies, the demand for internationally applicable costing methods and tools is urgent.

Given the situation noted above, the aim of this dissertation is two-fold. The first aim is to investigate the available methods for including education costs and benefits in health economics research, and to explore the attitude among health economists towards their inclusion (Chapters 2, 4). The second aim of the dissertation is to propose new standardized internationally applicable identification, definition, measurement, and valuation methods that can enable the inclusion of education costs and benefits in health economics studies in a transparent way and enhance the comparability of study results (Chapters 3-6). This final chapter summarises the main findings of this dissertation, placing them in the context of broader research, discusses methodological considerations, and provides recommendations for policy and further research.

Main findings

To address the first aim of this dissertation, a systematic literature review of health economics studies was conducted to explore the available methods for including education costs and benefits in health economics research (Chapter 2). The results show substantial variation in the methodological approaches to the identification, definition, measurement, and valuation of education costs and benefits in the reviewed studies. Self-reported data was most commonly used to measure education costs and benefits, with a variety of resource-use measurement (RUM) instruments (e.g. Treatment Inventory of Costs in Patients with psychiatric disorders {TiC-P}, Child and Adolescent Service Use Schedule {CASUS}) having been employed. Eight methods were identified for developing the unit costs of education costs and benefits. These include the calculation of unit costs based on secondary national data or literature, the use of national costing guidelines, and own unit cost calculation, among others. The majority of identified studies lacked transparency in reporting the methods used. Seventy percent of the studies did not report the methods used for identifying education costs and benefits. Thirty-

three percent of the studies that collected self-reported data for measuring education costs and benefits did not report which instrument was used to collect the data. In terms of valuation, no unit costs were reported for 33% of the extracted items, and no sources of the unit costs were reported for 8% of the items. To further address the first aim of this dissertation, a survey was conducted among health economists and health technology assessment experts to investigate their attitudes towards including education costs and benefits in health economics studies (Chapter 4). Although the respondents were generally aware of the relevance of including education costs and benefits in their research, only one-third of the respondents reported having included these costs and benefits in previous studies. Several reasons for not including education costs and benefits were mentioned (e.g. the irrelevance of these costs and benefits to the budget holder and the difficulty of obtaining information).

The second aim of this dissertation was addressed by proposing new standardized internationally applicable methods and tools to support the identification, definition, measurement and valuation of education costs and benefits. A list of twenty-four education costs and benefits that could be relevant for inclusion in health economics studies in the mental health domain was developed based on the systematic review of peer-reviewed and grey literature in six European countries and on a survey with experts in the education sector (Chapter 3). Furthermore, education costs and benefits on this list were categorised into 'inputs', i.e. resources introduced in the education system such as services and staff (e.g. special education services); 'throughputs', i.e. the use of resources by consumers and activities resulting from the delivery of services (e.g. home education); and 'outputs', i.e. the consequences of using education system resources such as changes in functioning (e.g. reduced school attainment). This categorization is aimed to facilitate the comparability of education costs and benefits and support the subsequent development of costing tools. The list of education costs and benefits was used in the consecutive study to determine the most important items for inclusion in health economics studies in the mental health domain (Chapter 4). This was done by conducting a best-worst scaling survey with an international group of health economists. 'Special education school attendance', 'absenteeism from school', and 'reduced school attainment' were identified as most important for inclusion in economic evaluations in the mental health domain. The list of education costs and benefits presented in Chapter 3 was also used as a basis for the development of two costing tools (Chapters 5-6). A multi-sectoral multi-national RUM instrument was developed in the course of the Programme in Costing, resource use measurement and outcome valuation for Use in multi-sectoral National and International health economic evaluations (PECUNIA) project, to address variation in obtaining resource use data for multi-sectoral economic evaluations. The PECUNIA RUM is a questionnaire for collecting self-reported data; it contains modules on resource use in multiple sectors, including the education sector. Furthermore, this study can provide guidance for researchers regarding the development of RUM instruments. Chapter 6 describes the application of a standardized unit cost calculation tool, the PECUNIA Reference Unit Cost Template for costing services, to develop unit costs of selected education services across five European countries. The study shows that despite a harmonized methodological approach, other factors (e.g. the availability of data or national data collection practices) can contribute to variation in the unit costs.

Contribution to health economics research

Inclusion of the broader costs and consequences of health interventions is in line with the theory of welfarism, which posits that the goal of health interventions is to improve social welfare; this underpins health economic evaluation.⁸ According to this theory, the aim of health economic evaluations is to help decision-makers achieve the most optimal societal resource allocation that would maximize social welfare. Compared to a more narrow (e.g. health system) perspective, a societal perspective is preferred for supporting optimal societal decision-making in relation to health and resource allocation, to provide care in a more cost-conscious manner and achieve better health outcomes for the population.⁹ The findings of this dissertation, as well as the methods and tools presented in this dissertation, have the potential to facilitate the adoption of a societal perspective in health economics studies. By demonstrating that education costs and/or benefits can constitute a significant proportion (up to 67% in the reviewed studies) of total costs and/or benefits associated with a mental health condition or a mental health intervention, the results of the systematic literature review (Chapter 2) provide a rationale for the importance of considering education costs and/or benefits for inclusion in health economics studies. Health economists who participated in the best-worst scaling survey (Chapter 4) also indicated the importance of considering these costs and benefits.

The systematic literature review (Chapter 2) highlighted the existing research gap in the availability and use of methods and tools to support the inclusion of education costs and benefits in health economics studies. The results showed substantial variation in the application of identification, definition, measurement, and valuation methods. This hampers the comparability and transferability of generated evidence, leading to inefficient use of resources in research. This dissertation contributes to the methodological body of research in health economics by proposing methods and tools to support the inclusion of education costs and benefits in health economics studies in a standardized and transparent way.

Identification

Several classifications of cost categories in health economics have been suggested over the years. The prominent handbook for conducting health economic evaluations by Drummond and colleagues (2015) outlined four categories of costs to be considered in economic evaluations, namely costs in the health sector, costs in other sectors, patient and family costs, and productivity losses.¹⁰ However, this classification does not provide any further specification regarding what other sectors and specific types of costs in other sectors should be considered. The US Panel on Cost-Effectiveness published an impact inventory listing health and non-health impacts of health interventions that should be considered in economic evaluations conducted from a societal perspective.¹¹ The impact inventory specifies seven non-health care sectors, including the education sector, but it does not list the types of costs to be considered. The classification scheme by Drost and colleagues (2013) is focused on intersectoral costs and benefits associated with mental health interventions.¹² The list contains fourteen items related to costs and benefits in the education sector, identified in the literature search and in consultation with experts in the field of health promotion and disease prevention.

This dissertation, particularly the studies presented in Chapters 3 and 4, build on and complement previous research and provide an important contribution to the development of methods for identifying education costs and benefits in health economics. Compared to previous classifications,¹⁰⁻¹² the one presented in Chapter 3 incorporates the international perspective from international peer-reviewed and grey literature and international experts in the education sector. The list of education costs and benefits presented in Chapters 3 and 4 can be used to select relevant cost items for economic evaluations for which education costs and benefits could be relevant. Furthermore, the prioritization exercise presented in Chapter 4 identified the most important costs and benefits in the education sector for inclusion in the economic evaluation of mental health interventions. This study could help researchers focus on the “big ticket” items when research resources (e.g. time, funding) are limited.

Definition

Establishing common descriptions and definitions of cost items is crucial for ensuring the comparability of health economics studies. Nevertheless, the lack of terminological comparability has been highlighted in the literature of health economics and health services research.¹³ The same issue has been identified in several studies of school-based mental health promotion interventions set in the education sector.^{14,15} The findings of the systematic literature review (Chapter 2) are in line with previous research and indicate the lack of clarity regarding what the included cost items entailed. The categorization of intersectoral costs and benefits in the education sector conducted in Chapter 3 presents the first step in the disambiguation of item descriptions and definitions. Categorizing costs and benefits into ‘inputs’ (e.g. services, professionals), ‘throughputs’ (e.g. interventions), and ‘outputs’ (e.g. consequences) provides a formal framework for disentangling the heterogeneity of the identified items. This was an important step for the subsequent development of costing tools in the PECUNIA project, as for each category of the items an appropriate measurement and valuation method needed to be identified. For example, a separate costing template was developed for the valuation of services (Chapter 6).

Measurement

There is currently no gold standard for the measurement methods of costs and benefits in health economics. Although a large number of self-reported RUM instruments are available, little research has been done on the proper development of RUM instruments as well as on their measurement properties including feasibility, validity, and reliability.^{16,17} Based on consultation with health economists, Thorn and colleagues (2013)¹⁶ proposed best practices for developing RUM instruments in health economics. However, the application of this approach has never been described in detail. The study in Chapter 5 describes the development process of the PECUNIA RUM, based on the best practice approach by Thorn and colleagues (2013).¹⁶ The PECUNIA RUM is intended for use in multi-sectoral trial-based economic evaluations, as it facilitates the collection of data on resource use in multiple societal sectors. In comparison with existing RUM instruments, the PECUNIA RUM includes a wider range of resource-use categories. The module that covers resource use in the education sector is of particular importance, as generic, internationally applicable RUM instruments that cover use of resources in the education sector are scarce.¹⁸ An important contribution of the study

presented in Chapter 5 is in the transparent reporting of the development process of the PECUNIA RUM, thereby offering guidance to researchers who set out to develop a RUM instrument. Furthermore, transparent reporting of how the PECUNIA RUM was developed provides insight into the methodological choices made during the development process. This could be important for assessing the validity and reliability of the instrument.

Valuation

Similar to resource-use measurement, little guidance is available on the proper valuation of education costs and benefits in health economics.¹⁹ This implies that researchers rely on their judgment in selecting appropriate valuation methods, which can lead to substantial variation and therefore lack of comparability across studies, as demonstrated by the systematic literature review in Chapter 2. Earlier research also shows that the choice of costing methodology can have significant impact on the unit costs.²⁰ Several countries (e.g. the Netherlands and the UK) established reference unit costs for harmonizing health economic analyses in the national context. However, they vary in terms of costing methodologies, which makes it difficult to conduct international comparisons between studies. Furthermore, the majority of the reference unit cost programs focus on costs and benefits in the health and social care sectors, while there is a scarcity of unit costs of intersectoral costs and benefits, including those in the education sector. The availability of valid and reliable unit costs can help facilitate the inclusion of costs and benefits in the education sector in economic evaluations. The study by Drost et al. (2017)²¹ proposed four valuation methods for developing unit costs of intersectoral costs and benefits, including those in the education sector. Based on the methods proposed in this study, the unit costs of selected education costs and benefits were developed and included in the Dutch costing manual for intersectoral costs and benefits.²² However, the unit costs included in this manual are based on the Dutch data and are applicable only in the Netherlands.

The development of internationally applicable unit costs of education costs and benefits was explored in Chapter 6. Using a standardized unit cost calculation tool developed in the PECUNIA Project,²³ the unit costs of three selected education services were calculated in five European countries. In comparison with previous research, the unit costs presented in this study are more easily comparable for two reasons. First, the unit costs were developed using the same methodological approach, and second, the unit cost calculation was transparently documented. These unit costs can be used in future economic evaluations that consider resource use in the education sector; they are available in the PECUNIA Reference Unit Cost Compendium, free of charge for non-commercial use.²⁴

Methodological reflections

Considering the methodological approach adopted in the studies included in this dissertation, several strengths can be mentioned. First, as research presented in this dissertation was conducted alongside a larger EU-funded project, steps for the development of the methods and tools were conceptualized and peer-reviewed before their application. The methodological approach was developed by the PECUNIA Consortium, a multi-national multidisciplinary group of experts with expertise in the field of health economics, health technology assessment,

mental health, and health services research, with particular focus on methodological expertise. Furthermore, a structured standardized approach was applied to the development of costing methods and tools in all sectors explored in the PECUNIA project, including health and social care, education and criminal justice, employment and productivity, and patient, family, and informal care. This was particularly important for the development of the PECUNIA RUM because of the instrument's multi-sectoral structure.

Second, a wide range of stakeholders was involved in developing the methods presented in this dissertation. Experts in the education sector were consulted during the development of the list of relevant education costs and benefits in Chapter 3. Health economists were involved in the prioritization of education costs and benefits in Chapter 4 and in the development of the PECUNIA RUM instrument in Chapter 5. Furthermore, external feedback for the work presented in this dissertation was sought from academic peers at international health economics conferences. The input of external experts was instrumental in ensuring that rigorous standards were followed in the development and application of the methods and tools presented in this dissertation, particularly given the novelty of this research.

Third, an important strength of this dissertation is in the standardization and international orientation of the methods and tools presented here. The majority of the tools in health economics are applicable to a particular (national) context, which hampers the transferability of the evidence produced. The PECUNIA tools are intended to be generic and applicable in a broader European context. This was ensured by involving international experts, consulting international peer-reviewed and grey literature, and testing methods in multiple European countries - although it is important to note that the applicability of the tools to countries not involved in the PECUNIA project and to health conditions outside the mental health domain would need to be explored in future research. A major strength of the PECUNIA methods is their complementarity. The PECUNIA RUM and the PECUNIA costing templates are intended for joint use, to produce more comparable health economics evidence.

This dissertation has several limitations. First, while the methods and tools presented in this dissertation are intended to be generic and applicable to all disease areas, mental health conditions were used as an illustrative example. Mental health was chosen because of its wide societal impact and associated costs in multiple societal sectors, including the education sector. Other health domains could be explored to complement the development of the methods presented in this dissertation.

Second, research in this dissertation was conducted through the prism of health economics, focusing on the impact of health conditions and interventions in the healthcare sector on other sectors. This might have resulted in a somewhat limited perspective as interventions in other sectors can also have an impact on the healthcare sector. For example, interventions in the education sector, such as free school meals, can lead to costs and/or benefits for the healthcare sector.²⁵ Given the context of this dissertation, reviewing methods and tools for identifying, measuring and valuing costs used in education research could have further complemented the development of the methods and tools presented in this dissertation.

Third, the aim of this dissertation was to broaden the societal perspective in health economics by facilitating the inclusion of education costs and benefits on the cost side of economic evaluations. However, the impact of health conditions and health interventions on the education sector can also be included on the outcome side of an economic evaluation. For example, educational attainment can be used as one of the outcome measures in a cost-effectiveness analysis. Further research is needed to explore the appropriate methods to account for educational outcomes in health economics research. In addition, this might have implications for double counting in economic evaluations, and researchers need to be aware of this potential bias.

Finally, the research presented in this dissertation pertains to a relatively new research area in health economics. Intersectoral costs and benefits and the methods for their inclusion in health economics studies have not yet been extensively explored. On the one hand, this highlights the relevance of this dissertation to the field of health economics. On the other hand, the methodology of the studies included in this dissertation was somewhat exploratory due to the absence of established methods to guide the development of the tools. Nevertheless, as mentioned above, the applied methodology was developed by an international group of health economists with a wide range of methodological expertise based on the best available albeit scarce evidence and was subjected to peer review at every stage.

Implications and recommendations for further research

This dissertation addresses the research gap in the availability of methods and tools for including relevant costs and benefits from the education sector in health economics studies. The list of education costs and benefits presented in Chapters 3 and 4 can be used to select relevant items for future economic evaluations. Furthermore, the PECUNIA tools, the PECUNIA RUM and the PECUNIA Costing Templates presented in this dissertation can help facilitate the adoption of a societal perspective in health economics by supporting the inclusion of the broader costs and consequences of health interventions, including those in the education sector, in health economics evaluations. The PECUNIA tools are intended to be used in conjunction to produce comparable and transferable cost data for multi-national economic evaluations, which are currently lacking standards.²⁶

This dissertation also has important implications for developing standards for conducting economic evaluations in national and international contexts. The methods for conducting economic evaluations are regulated through national costing guidelines, such as the costing manual developed by the National Health Care Institute in the Netherlands²⁷ and the guide to the methods of technology appraisal developed by the National Institute of Care and Excellence in the United Kingdom.²⁸ National costing guidelines aim to standardize economic evaluations in a given country by outlining the methodological requirements (e.g. study perspective, discount rate for costs and outcomes) to be adhered to. On the one hand, these guidelines are useful for providing an evidence-informed uniform standard in terms of methods and reporting, thereby improving the quality of the studies. On the other hand, national guidelines have certain limitations. First, these guidelines are generic and they do not take into account any disease-specific factors that can shape economic evaluations in various health domains. Several attempts have been made to develop additional standardized guidance for different

areas, including osteoarthritis²⁹ and immunization programmes.³⁰ This guidance is more specific than generic costing guidelines and can facilitate the comparability of studies in specific health domains. Further research could explore the feasibility of developing best practices for conducting economic evaluations in the mental health domain. The results of this dissertation can support the development of these standards, particularly in the context of interventions for which the inclusion of costs and benefits in the education sector would be relevant (e.g. mental health interventions targeting children and adolescents). Furthermore, it is not yet clear to what extent the standard methods and guidelines for conducting economic evaluations are applicable to specific populations, including the paediatric population.³¹ Given the increase in the number of economic evaluations conducted in the population of children and adolescents,³¹ the results of this dissertation could be used to develop guidance on conducting economic evaluation in this target group.

Furthermore, national costing guidelines vary in terms of their recommendations, even in countries that are geographically close to each other.³² For example, guidelines in the Netherlands recommend adopting a societal perspective, while in Germany a health sector perspective is primary. Differences in recommendations hamper the international transferability of health economic evidence. The lack of standardization to support economic evaluations across countries motivated the PECUNIA project.^{33,34} In addition to proposing standardized methods, the results of the project laid the foundation for developing overarching guidance (i.e. on the European level) to promote uniform standards for conducting economic analyses.

Finally, the current guidelines that do recommend adopting a societal perspective rarely specify including costs and benefits from the education sector when those can be relevant for inclusion, and provide limited methodological guidance on how to include these costs and benefits in health economic evaluations. By proposing standardized internationally applicable methods and tools for the identification, definition, measurement, and valuation of education costs and benefits, this dissertation can contribute to expanding the current guidelines to develop a more defined methodological guidance for their inclusion in economic evaluations.

Variation in the use of methods and lack of transparency in reporting on the methods point to the need for incentivising good research practices in health economics research. Transparent reporting has the potential to facilitate decision-makers' confidence in health economics methods, thereby promoting the uptake of health economic evidence in decision-making.³⁵ Furthermore, transparent reporting allows for assessing the appropriateness of the selected methodology, reducing bias, facilitating comparability between studies, and generally improving the quality of economic evaluations.^{10,36} In recent years, several initiatives have been put forward to tackle the lack of transparency in various domains of health economics. One particularly relevant example of such an incentive is the introduction of the Consolidated Health Economic Evaluation Reporting Standards (CHEERS) to stimulate standardized reporting of economic evaluations.³⁷ It lists twenty-four items and provides recommendations on how to approach reporting on each item. Fourteen out of the twenty-four items refer to the methods for conducting economic evaluations. However, only two items concern the cost side of an economic evaluation, namely 'study perspective' and 'estimating resource use and costs';

guidance is limited on what information has to be reported, leaving this choice up to the researchers' discretion. Defining reporting standards in relation to the choice of costing methods in health economics research could help facilitate transparency. In addition to reporting checklists, the use of Health Economic Analysis Plans has recently been advocated as a useful tool to facilitate transparency and minimize bias in trial-based economic evaluations.³⁸

The results of this dissertation also provide directions for future methodological research with regard to the inclusion of costs and benefits from the education sector in health economic evaluations. In terms of identification methods, further research could focus on identifying and prioritizing the most important cost items in the education sector in relation to specific disorders. This could help researchers focus on including the most relevant cost items given the specific context of their study. Furthermore, this could help facilitate the development of disease-specific guidance for conducting health economic analyses.

The systematic literature review presented in Chapter 2 identified the lack of clarity and comparability regarding the descriptions and definitions of cost items included in the reviewed studies. This issue is not unique to the education sector and has been emphasised in other research domains. The current situation with terminological variability has led to increased interest in and awareness of the need for standardized classifications. One example of such a classification that has the potential to tackle this issue is a standard taxonomy for description, mapping and comparison of services for Long-Term Care (DESDE-LTC). DESDE-LTC is an international classification system of care services based on the European Mapping Service Schedule (ESMS) for coding adult mental health care, which was extended to incorporate other care sectors such as child and adolescent mental health, drugs and alcohol, ageing, disabilities, long-term care, and social care. It includes an international, standardised tool for describing care teams within health services, and compiles information on the inputs and processes of care at the meso-level (e.g. health/social catchment areas) and micro-level (e.g. individual services).¹³ DESDE-LTC has been validated and extensively used in the field of health economics for comparing resource utilization across different settings – for efficiency analysis and the calculation of unit costs, among other research purposes.³⁹ The use of DESDE-LTC has been explored in the PECUNIA project for developing standard descriptions and codes for health and social care services. Further research could explore the harmonization and standardization of descriptions and definitions of education costs and benefits by exploring whether this classification could be extended to the education sector. This could help facilitate the comparability and transparency of health economics studies including broader costs and benefits.

Further research is needed to test the applicability of the PECUNIA tools in and outside of Europe as well as in other disease areas outside of mental health. The PECUNIA RUM still needs to undergo formal validity and reliability testing. It is important to note that although the module on the resource-use in the education sector would be particularly relevant for the interventions targeting children, adolescents, and young adults, the original version of the PECUNIA RUM was developed for use in the adult population only. To be able to use the PECUNIA RUM in the paediatric population, the development and validation of a version of

the instrument suitable for children is necessary. Chapter 6 presents the first application of the PECUNIA costing tool to the development of unit costs of education services. Further research could use this tool to expand the current compendium and develop unit costs of other services in the education sector in more European countries beyond those that are currently included. Furthermore, methods used in other domains of health economics for which costs in the education sector could be relevant (e.g. immunization, chronic care) as well as methods used in other disciplines (e.g. the economics of education) could provide valuable input for the development of valid methods to support broader cross-sectoral economic analyses.

Another interesting area pertaining to the valuation of education costs and benefits is the valuation of children's time. Similar to adults, children's productive and leisure time can be affected by health conditions. For example, anxiety disorders in children were found to be associated with absenteeism.⁴⁰ However, most of the existing literature focuses on the valuation of time in the adult population, while little is known about how to value children's productive and leisure time. Andronis and colleagues (2019) began to explore this gap in the literature by reviewing available theories on valuing past time and concluded that existing approaches were not suitable for the valuation of children's time.⁴¹ Further research is needed to explore and propose appropriate methods to value children's productive and leisure time.

Implications and recommendations for policy

While the results of this dissertation are primarily of interest for researchers, several implications and recommendations for policy need to be mentioned. Health and health interventions have a broad societal impact that goes beyond the healthcare sector and affects other sectors. This has been clearly demonstrated by the COVID-19 pandemic and its impact on society, including the education sector.⁴² The implications of (mental) health in the education sector are indisputable and this calls for broader approaches to policy and economic analyses to inform policy around (mental) health and education. Despite accounting for a significant proportion of health expenditures,⁴³ mental health is severely underfunded.⁴⁴ By demonstrating the broader societal impact of mental health on sectors beyond the healthcare sector, the results of this dissertation provide a rationale for facilitating intersectoral approaches to policymaking and joint funding for mental health interventions. One example of such initiative is Health in All Policies (HiAP). HiAP is a public policy approach that promotes consideration of health implications across all policy domains and stresses the importance of cross-sectoral collaboration and coordinated funding.^{45,46}

To support broader approaches to policymaking, a broader approach to conducting economic analyses for informing policy is also needed. The majority of national costing guidelines still recommend adopting a narrow (e.g. health system) perspective. A narrow perspective does not account for important costs and consequences of health interventions, thereby leading to suboptimal policy recommendations.⁹ Also, as demonstrated in Chapter 3, a narrow perspective was indicated as one of the barriers to the inclusion of broader costs and benefits in economic evaluations. While researchers may be aware of the importance of including intersectoral costs and benefits, they are restricted by the recommendations of the national costing guidelines. This implies that there is a shared responsibility between policymakers and researchers in promoting good research practices to support optimal societal decision-making.

The findings of this dissertation can be used to support advocacy for broadening the perspective of economic analyses in national costing guidelines.

Another important policy implication was highlighted in Chapter 6 of this dissertation. During the development of unit costs of education services across five European countries, the differences in data collection practices have been noted to be a major obstacle to the comparability of the unit costs. Policy efforts to incentivise the development and adoption of harmonized data collection standards in Europe could help facilitate the comparability of study results. This would be especially relevant for multi-national studies and could help contribute to improving overall research efficiency.

Conclusion

The main aim of this dissertation was to facilitate the inclusion of education costs and benefits in health economics research in the mental health domain by providing an overview of available methods and proposing new methods for their inclusion. The results suggest that education costs and benefits can make up a significant proportion of total costs associated with mental problems. However, there is a lack of structured methodological guidance regarding the inclusion of these costs and benefits. Previous studies vary in how education costs and benefits are identified, defined, measured and valued, which leads to the lack of comparability of the economic evidence. The methods and tools proposed in this dissertation can contribute to reducing this variation by offering a standardized transparent approach to support the inclusion of education costs and benefits in health economics studies. The knowledge developed in this dissertation also provides the basis for further harmonization and standardization of the methods for the inclusion of education costs and benefits and for broadening the societal perspective in health economics research.

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Summary

Mental problems often begin to develop during childhood and/or adolescence and can be associated with high costs for multiple societal sectors, including the education sector. Children with mental problems are more likely to require additional support at school to help them achieve the same educational outcomes in comparison with their peers without mental problems. This support is often associated with substantial costs for the education sector. On the other hand, interventions to prevent or treat mental problems can lead to benefits to the education sector in the form of cost savings. Taking costs and benefits in the education sector into account in economic evaluations is important for informing policy decisions in relation to optimal resource allocation. Nevertheless, costs and benefits in the education sector are rarely included in economic evaluations of health interventions. This can be attributed to limited knowledge on the importance of including these costs and benefits, as well as the scarcity of methods and tools to support their inclusion in economic evaluations. Therefore, the aim of this dissertation is two-fold. The first aim is to investigate the available methods for including education costs and benefits in health economics research, and to explore the attitude towards their inclusion among health economists. This aim is addressed by the studies presented in Chapters 2 and 4. The second aim is to propose new standardized internationally applicable identification, definition, measurement, and valuation methods that can enable the inclusion of education costs and benefits in health economics studies in a transparent way and enhance the comparability and transferability of the results within and across countries. The second aim was addressed by the studies presented in Chapters 3-6, each of which corresponded to one or more of the four steps of costing in health economics, namely identification, definition, measurement, and valuation.

Chapter 1 introduces the scope and main concepts related to this dissertation. It discusses the rationale for conducting the research presented in this dissertation by outlining the economic impact of mental problems on society and within societal sectors. This chapter explores in more detail how mental problems affect the education sector. Furthermore, the chapter introduces the discipline of health economics with particular focus on the choice of perspective in health economics studies. The choice of perspective determines what costs and consequences of health interventions are included in the analysis. When a health sector perspective is adopted, only costs and consequences relevant for the health sector are included. When a societal perspective is adopted, all costs and consequences are considered regardless of the sector where they occur. A societal perspective is preferred for informing optimal resource allocation decisions. Chapter 1 also outlines key methodological research on costs and benefits in the education sector, highlighting gaps in the literature. Chapter 1 concludes with the context, aims, and outline of this dissertation.

Chapter 2 presents the results of a systematic literature review of economic evaluations and cost of illness studies conducted in the mental health domain over the population of children and adolescents. This study aimed to gain insights into the methods used for including education costs and benefits in health economics studies and into the proportion of education costs and benefits in relation to the total costs associated with a condition or an intervention. Forty-nine studies were included in the analysis. The results show considerable variation in the choice of identification, definition, measurement, and valuation methods for including

education costs and benefits in health economics studies, as well as the general lack of transparency in reporting on the methods. This limits meaningful comparison between the studies and the transferability of the generated evidence. Furthermore, the results show that education costs and benefits can comprise up to 67% (range 0-67%) of the total costs associated with a condition or an intervention. This provides a strong rationale for the importance of considering education costs and benefits for inclusion in health economics studies.

Chapter 3 explores the methods for identifying and defining costs and benefits in the education sector. The aims of this study were to identify costs and benefits in the education sector that could be relevant to health economics studies set in the mental health domain, to validate the identified costs and benefits in the European context, and to categorize them into conceptually harmonized clusters of terms. To identify costs and benefits in the education sector, a systematic review of peer-reviewed and international grey literature was conducted. The list of costs and benefits in the education sector, comprised of twenty-four items, was reviewed by an international group of fourteen experts in the education sector for completeness, clarity, and relevance. The list of costs and benefits was subsequently categorized into inputs (e.g. services, professionals), throughputs (e.g. interventions) and outputs (e.g. consequences). The main contribution of this study is in developing a comprehensive, validated list of costs and benefits in the education sector that could be relevant to health economics studies. Furthermore, the categorization of costs and benefits presents the first step towards the development of costing tools for the proper inclusion of these costs and benefits in health economics studies.

Chapter 4 addresses both aims of this dissertation. In line with the first aim, this study explores the attitudes of health economists towards including education costs and benefits in economic evaluations of interventions targeting mental health and behavioural disorders. This chapter further explores the methods for identifying education costs and benefits by quantifying their relative importance in the context of mental health and behavioural disorders. To collect the data for this study, an online survey with open-ended questions and a best-worst scaling object case study was developed. Thirty-nine experts in the field of health economics and health technology assessment completed the survey. The most important education costs and benefits were “special education school attendance”, “absenteeism from school”, and “reduced school attainment”. The majority of the respondents (68%) reported that education costs and benefits were relevant, but only a few (32%) included them in economic evaluations. The experts also indicated that irrelevance of these costs and benefits to the budget holder, the difficulty of obtaining information, the difficulty of quantifying these costs and benefits, and the difficulty of defining the boundaries of a broader (societal) perspective were the main barriers to the inclusion of education costs and benefits in economic evaluations. The results of this study can be used to select items for the development of measurement and valuation tools and for including education costs and benefits in economic evaluations in the mental health domain. Furthermore, by highlighting the barriers to adopting a societal perspective in health economics, this study proposed directions for further research to facilitate the inclusion of broader costs and benefits in economic evaluations.

Chapter 5 focuses on the methods for broader resource-use measurement (RUM) in health economics. This chapter describes the development process of a multi-sectoral multi-national RUM instrument developed in the Programme in Costing, resource use measurement and outcome valuation for Use in multi-sectoral National and International health economic evaluAtions (PECUNIA) project, the PECUNIA RUM. The methodological approach was based on best practice guidelines for the development of RUM instruments in health economics. The process consisted of six steps, including the definition of the instrument attributes, identification of cost-driving elements in each sector, a review of methodological literature, development of a harmonized cross-sectoral approach, development of questionnaire modules and their subsequent harmonization. The selected methodological approach was, overall, applicable to the development of the PECUNIA RUM instrument. However, due to the complexity of developing multi-sectoral RUM instruments, additional steps were needed. Establishing a uniform methodological basis and harmonization of questionnaire modules were necessary steps to ensure cohesion of the instrument, as it is made up of eight modules. Furthermore, the involvement of a broader range of stakeholders (healthcare professionals, sector-specific experts, health economists) was deemed important given the inclusion of resource-use in multiple sectors. The draft version of the PECUNIA RUM was presented to the focus group of sixteen health economists. The participants indicated that having a standardized RUM instrument would be important, although the full version of the instrument would not be likely to be applicable to every setting. This is the first study that transparently describes the development process of a generic multi-sectoral RUM instrument in health economics and provides insights into the methodological aspects and overall validity of the instrument. This study could also serve as a guide for researchers who undertake RUM instrument development.

Chapter 6 explores the methods for the valuation of education costs and benefits. This study describes the application of a standardised costing tool developed in the PECUNIA project, the PECUNIA Reference Unit Cost Template for services, for calculating the unit costs of services in health economics research. The tool was used to calculate the unit costs of three selected services in the education sector, namely special education services, educational therapy provided in primary schools, and educational therapy provided in secondary schools, in five European countries (Austria, Germany, Hungary, the Netherlands, and The United Kingdom). The unit costs of special education services ranged from €57 to €189 per day. The unit costs of educational therapy provided in primary schools ranged from €6 to €26 per contact and from €5 to €35 per day. The unit costs of educational therapy provided in secondary schools ranged from €8 to €27 per contact and from €5 to €13 per day. Variation was observed in the type of data used for the calculation, service unit, representativeness of the unit costs, and the components included in the unit costs. The majority of the unit costs were nationally representative and were calculated based on national secondary data. The most frequently included cost components included staff costs and overhead costs. This study fills the research gap by providing the unit costs of selected education services that can be used in future national and multi-national economic evaluations in the European context. The results of this study also indicate that despite the use of a standardized tool, the unit costs and their composition still varied. Other aspects, such as the choice of the input data for calculating unit

costs, and structural differences in educational systems in different countries could contribute to this variability.

Finally, in Chapter 7 the main findings of this dissertation are discussed in relation to the broader research context. The results of this dissertation demonstrate substantial variation in the methodological approaches to the identification, definition, measurement and valuation of education costs and benefits in the reviewed studies. This variation can lead to the lack of comparability and transferability of health economic evidence, inefficient research practices, and suboptimal resource allocation decisions within and across countries. The main contribution of this dissertation in bridging this research gap is in proposing identification, definition, measurement and valuation methods for including education costs and benefits in health economics studies in a standardized way. This chapter also provides reflections on the methodology used for conducting the studies included in the dissertation, directions for further research, and implications of this dissertation for policy.

Impact

Mental problems have an economic impact on all aspects of the affected individuals' lives, generating costs and benefits in multiple social sectors.^{1,2} Costs and benefits associated with mental problems that occur in the education sector can be particularly relevant in the context of interventions targeting children and adolescents. However, these costs and benefits are not often considered in health economic evaluations.^{3,4} This can be attributed to limited knowledge on the importance of including these costs and benefits in health economics studies, but also to the scarcity of methods and tools for including these costs and benefits. The aim of this dissertation was to investigate available methods for including education costs and benefits in health economics research, and explore the attitude towards their inclusion among health economists. Furthermore, the studies included in this dissertation propose new standardized identification, definition, measurement, and valuation methods that can enable the inclusion of education costs and benefits in health economics studies in a transparent way and enhance the comparability and transferability of the costing evidence generated. This section reflects on the scientific and social impact of this dissertation.

Scientific impact

In practice, most health economic studies tend to adopt a narrow perspective, focusing on a limited range of costs and consequences,^{3,4} which can potentially lead to inefficient resource allocation decisions.⁵ The results of this dissertation can facilitate the adoption of a broader societal perspective in health economics and contribute in several ways to more optimal decision-making on the allocation of resources. First, the evidence presented in the systematic literature review of economic evaluations and cost-of-illness studies (Chapter 2) suggests that education costs and benefits can make up a large proportion of the total costs associated with a condition or an intervention in the mental health domain. These results provide the rationale for the importance of considering the inclusion of these costs and benefits in health economic studies when their inclusion can be expected to be relevant (e.g. in economic evaluations of interventions that target children and adolescents).

Second, health economists and health technology assessment experts who participated in the survey (Chapter 4) indicated that they found it important to consider including education costs and benefits in health economic evaluations. However, they also mentioned several barriers to their inclusion, such as the irrelevance of broader costs and consequences to the budget holder, the difficulty of obtaining the necessary data and of quantifying the intersectoral impact of health interventions, and the difficulty of defining the boundaries of a societal perspective. The results of this study can help direct future research towards the development of methodological guidance to overcome these barriers.

Third, the costing tools presented in this dissertation (Chapters 3-6) can contribute to bridging the current gap in the availability of methods for including education costs and benefits in economic evaluations. The results of the studies presented in Chapters 3 and 4 can help researchers select relevant resource-use items in the mental health domain for inclusion in health economics studies. In comparison with previous research, the list presented in this dissertation incorporated evidence from the international grey literature and feedback from experts in the education sector for its validation. The resource-use measurement (RUM) instrument, the PECUNIA RUM (Chapter 5), can be used for collecting resource-use data for

multi-sectoral health economic evaluations, including data on resource use in the education sector, in a standardized way. The PECUNIA RUM can therefore not only support the inclusion of broader costs and benefits in economic evaluations, but also contribute to the overall structural enhancement of national and multi-national economic evaluations. By outlining the development process of the PECUNIA RUM in detail, this study can also be of interest to researchers who set out to develop a RUM instrument. The unit cost calculation tool, the PECUNIA Costing Template for services, presented in Chapter 6, can support the calculation of unit costs of services, and the presented unit costs can be incorporated in economic evaluations that include costs in the education sector. Furthermore, by providing a standardized transparent method for calculating unit costs, this costing tool can help identify and explain variability in the unit costs that stems from aspects other than the methodological approach. Finally, the joint use of the tools presented in this dissertation can contribute to producing comparable health economic evidence within and across countries in Europe.

The relevance of this dissertation is not limited to the discipline of health economics. The methods and tools presented here can support economic evaluations of public health and educational interventions in the school setting. Experts in the education sector were one of the target groups for research presented in this dissertation; their input was sought for the validation of costs and benefits as described in Chapter 3. Researchers in the area of the economics of education can also utilize the methods and tools presented in this dissertation for conducting economic analyses of interventions in the education sector. Furthermore, by demonstrating the impact of the costs of mental problems on the education sector, the studies included in this dissertation can facilitate cross-sectoral collaboration between health and education economists.

Social impact

This dissertation also provides important insights into social challenges and can be of interest to policymakers, particularly those in the health and education sectors. By highlighting the cross-sectoral economic impact of mental problems, this dissertation is aligned with broader approaches to policymaking,⁶ such as Health in All Policies⁷ and systems thinking,⁸ that stress the importance of collaboration across societal sectors to improve population health. Broader acceptance and the adoption of a societal perspective in health economics can facilitate the inclusion of relevant costs and consequences of health interventions in economic analysis and thereby contribute to reducing bias in study results and ensuing policy recommendations.⁹ Adopting a broader perspective can also incentivise intersectoral coordination and collaboration and promote the development of joint funding arrangements across the affected societal sectors. The latter can be particularly important for improving mental health care in Europe, given its severe underfunding.¹⁰

Furthermore, as indicated in the expert survey (Chapter 4), the irrelevance of broader costs and consequences of health interventions to the budget holder was one of the barriers to adopting a societal perspective in health economics studies. Currently, the majority of national costing guidelines recommend the adoption of a narrow perspective for conducting health economic analyses.¹¹ This highlights the responsibility of policymakers and regulatory bodies in promoting good research practices to support optimal decision-making. The results of this

dissertation, in conjunction with other methodological research, can be used for revising the existing guidelines to incorporate guidance for adopting a broader perspective in health economics.

Dissemination

To reach the scientific community, the results of this dissertation were presented at major health economics conferences in the form of abstracts, oral presentations, and parts of organized sessions. Some of the conferences targeted a broader multi-disciplinary audience including policymakers, and representatives of patient organizations, industry, and regulatory bodies in addition to academics. Studies presented in this dissertation have been published in or submitted to peer-reviewed journals. Please refer to Dissemination Activities for the full overview.

The results of this dissertation were also disseminated as part of the communication and dissemination strategy of the PECUNIA project. Two satellite workshops were organized to present the results of the project to the broader multi-disciplinary audience. The first satellite workshop took place in July 2019 at the International Health Economics Association congress in Basel. Its aim was to present intermediate project results, including the steps taken to identify relevant cost items in the education sector (Chapters 3 and 4) and to develop the PECUNIA RUM (Chapter 5). The second satellite workshop was organized online due to the COVID-19 pandemic, and advertised to a broad group of stakeholders. The workshop was dedicated to presenting the results of the PECUNIA project, including the final version of the PECUNIA RUM (Chapter 5) and the unit costs of education services (Chapter 6). The recording of the workshop is available on YouTube.¹² The tools developed in the PECUNIA project, the PECUNIA RUM and the PECUNIA Reference Unit Cost Templates are available for non-commercial use free of charge. They can be accessed via the website of the PECUNIA project.¹³ The PECUNIA RUM is currently available in English as a pen-and-paper questionnaire for the adult population. Further plans include the development of a digital version, proxy and child versions, and formal translation into other European languages. The PECUNIA RUM will also be included in the Database of Instruments for Resource Use Measurement (DIRUM), which acts as an open-access repository of RUM instruments for use in economic evaluations.¹⁴

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Dissemination Activities

Scientific articles in peer-reviewed journals

Pokhilenko, I., van Esch, T. E., Brabers, A. E., & de Jong, J. D. 2021. Relationship between trust and patient involvement in medical decision-making: A cross-sectional study. *PlosONE*, 16(8), e0256698.

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Submitted articles

Pokhilenko, I., Kast, T., Janssen, L.M.M., Evers, S.M.A.A., Paulus, A.T.G., Simon, J., Mayer, S., Berger, M., Konnopka, A., Muntendorf, L., Brodsky, V., Serrano-Aguilar, P., Park, A., Salvador-Carulla, L., Drost, R.M.W.A. International comparability of reference unit costs of education services: when harmonizing methodology is not enough (PECUNIA Project).

Pokhilenko, I., Janssen, L.M.M., Paulus, A.T.G., Drost, R.M.W.A., Hollingworth, W., Thorn, J.C., Noble, S., Simon, J., Fischer, C., Mayer, S., Salvador-Carulla, L., Konnopka, A., Hakkaart van Roijen, L., Brodzsky, V., Park, A., Evers, S.M.A.A. Development of an instrument for the assessment of health-related multi-sectoral resource-use in Europe: the PECUNIA RUM.

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Invited talks

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As the tradition goes, I would like to begin with my supervisory team – Silvia, Aggie, and Ruben. First of all, I want to thank you for giving me the chance to join the PECUNIA team. This experience has in many ways shaped me as an academic, and I couldn’t imagine any project that would suit me better. It has been a pleasure working with you over the years. Silvia, I truly admire your ability to stay on top of everything despite your busy schedule. Thank you for your support and trust in me, and also, very importantly, for teaching me about the value of professional networking and using Twitter! Aggie, you were always there ready to help with a kind supportive word and a wise advice – thank you for this. Ruben, you have always had an eye for detail and gave very helpful thorough feedback; I truly learned a lot from working with you.

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И теперь по-русски. Последними по счету но не по значимости, я хочу поблагодарить свою семью, в особенности маму, папу и бабушку. Я благодарна вам за вашу поддержку и веру в меня и за те возможности, которые вы мне дали, которые позволили мне оказаться там, где я сейчас. Без вас это было бы невозможно. Люблю вас очень сильно! И наконец, отдельные слова любви и поддержки хочу посветить моей семье в Украине. Вы всегда в моих мыслях.

About the author



Irina Pokhilenko was born on February 21st in 1994 in Petrozavodsk, Russia. After graduating from high school in 2012, she moved to Jyväskylä, Finland, to pursue her undergraduate degree in Health Care at the Jyväskylä University of Applied Sciences. During her studies, she spent one semester doing her internship at the Mater Dei Hospital in Malta. In 2017-2018, Irina completed her postgraduate studies in Healthcare Policy, Innovation and Management at Maastricht University.

After graduation, Irina joined the department of Health Services Research as a junior researcher. She was involved in the ProgrammE in Costing, resource use measurement and outcome valuation for Use in multi-sectoral National and International health economic evaluations (PECUNIA) project, which focused on the development of methods for conducting multi-sectoral multi-national economic evaluations in Europe. Irina also contributed to the teaching activities at the department of Health Services Research, mainly in the MSc program Healthcare Policy, Innovation and Management as a tutor, trainer, and placement supervisor for the students working on their dissertations. Irina was also a member and a president of the Maastricht University ISPOR student chapter, as part of which she organized health economics and outcomes research-related events. In 2019-2021, Irina was involved in supporting the European guideline education and Cost-Conscious Healthcare (CoCoCare) project which aimed to develop a guideline training for medical residents in Europe. Since 2019, Irina has been a convener for the mental health economics special interest group of the International Health Economics Association. In 2021, Irina spent two months at the Loyola University Andalucía as a visiting research fellow to collaborate in the project on developing standardized methods for classification of services in health economics and health services research. Irina's research has led to numerous scientific publications and presentations at national and international health economics conferences.

Irina is currently working as a research fellow at the Health Economics Unit at the University of Birmingham. She is involved in research to tackle the wider determinants of obesity focusing on the impact of school food policies on children's health and quality of life outcomes.