Broad Consultation as Part of the Standardization of Economic Evaluation Research in the Youth Sector

ZonMw

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We would like to thank the stakeholders for their valuable input and feedback, both during the written consultation, as well as during the stakeholder consultation meeting (for an overview of the names and affiliations see Table 3). Additionally, several colleagues gave much-appreciated input during two general meetings (Kosteneffectiviteit in de Jeugdzorg, ZonMw 21 april 2015 and Congres Jeugd in Onderzoek 14 maart 2016) where standardization of the economic evaluation in the field of youth was discussed. A special word of thanks to the Netherlands Organisation for Health Research and Development (ZonMw) program “Effectief werken in de jeugdsector” for commissioning this consultation and to the members of the six consortia which have been established under this program.

Summary

Introduction

Stakeholders are increasingly interested in the societal impact of psychosocial interventions in the youth sector, in terms of costs and quality of life, as well as in outcomes research. As a result, increasing attention is being focused on economic evaluations studies in the youth sector. However, methods and instruments which are used in economic evaluations have been developed for somatic (health) care and moreover for an adult population, making it challenging to perform economic evaluations in the youth sector.

Objective

The aim of this broad consultation is to reach consensus regarding the steps which have to be undertaken to set a research agenda which will lead to further methodological development and the standardization of economic evaluations in the youth sector.

Method

The broad consultation consisted of an 8-step procedure, including the conceptualization of a consultation document consisting of a scoping review of (mainly) international opinion/ methodological literature and an inventory of existing Dutch guidelines and manuals for economic evaluation, a written consultation procedure among a broad range of stakeholders, and a consultation meeting with these stakeholders. In the consultation document, the methodological issues and challenges emerging from the scoping review, as well as potential solutions for these issues and challenges offered by existing guidelines and manuals, have been categorized by framing aspects. In the written consultation procedure and in the consultation meeting, stakeholders have been asked to rank a maximum of the ten most important methodological issues and challenges for economic evaluations in the youth sector in order of importance, and to provide possible solutions or directions for research.

Results

In total 21 documents (18 articles, 1 white paper, 2 conference proceedings) were included in the scoping review. A total of 24 stakeholders participated in the written consultation procedure and 14 stakeholders participated during the consultation meeting.

The methodological issues and challenges which were ranked in the top 5 by the stakeholders are 1) outcome measurement, 2) outcome identification, 3) cost valuation, 4) outcome valuation, and 5) time horizon / analytical approach. The existing guidelines and manuals provided guidance for some,

but not all, issues and challenges. For the outcome side of the economic evaluation, normative questions have been posed such as: what goal of psychosocial care for youth should the outcome(s) comply with, and whose values count when obtaining preference weights for the outcome? Furthermore, respondents urged that, in order to perform economic evaluations in the youth sector, they needed instruments specifically developed for youth, such as instruments to measure costs, preference-based instruments to measure quality of life , and cost prices (for education, social care, and justice).

For other methodological challenges, overall consensus has been revealed which is in line with existing guidelines. For instance, regarding the perspective, most stakeholders agreed that economic evaluations should, in principle, be performed from the broad societal perspective, and, regarding the type of economic evaluation, that the cost-utility analysis is preferred. For the time horizon the stakeholders agreed that a long term time horizon is needed, but that in order to achieve this, more research is needed looking at the relationship between intermediate short term outcomes and long term final outcomes.

Discussion

This broad consultation has led to a research agenda which will in the long run lead to the standardization and methodological improvement of economic evaluations in the youth sector.

1 Introduction

The Netherlands Organisation for Health Research and Development (ZonMw) program "Effectief werken in de jeugdsector", call for "Knowledge advancement on the effectiveness of psycho-social interventions in the field of youth"[1], has led to the establishment of six consortia aiming at the condensation of interventions regarding youth on six themes, namely: 1) social skills/insecurity/ resilience; 2) anxiety, depression, dysthymic problems and other internalising behavioural problems; 3) boisterous behaviour and ADHD; 4) externalising behavioural problems/-disorders; 5) parenting uncertainty - prevention and mild problems; 6) severe problems with parenting/multi-problem families[2].

In the first months of 2015 these six consortia have performed an evidence synthesis regarding the current evidence of interventions looking at effectiveness research relating to these six themes. Based on these results, the consortia were asked to develop a research proposal for Phase 2 of the program. Phase 2 is aimed at condensing the number of interventions in each theme. The focus of these research proposals has been defined by the preliminary studies performed during Phase 1. Currently the consortia are busy with developing research proposals for Phase 2.

In addition to research aimed at reducing the numbers of interventions, other themes have been identified which have to be studied by all consortia jointly during Phase 2. One of these themes is economic evaluation research. In a coordinated action it is suggested that one project in the field of youth services should be organized to provide the sector with guidance for the standardization of economic evaluation research. As it is currently unclear in which form this standardization will take place, ZonMw finds it important to first develop a broadly supported vision of what is necessary and which steps need to be undertaken in order to develop guidance for the standardization of economic evaluation research within the youth sector. Moreover, it was considered important that this standardization be attuned to other methodological developments and to the existing guidelines on economic evaluation research, such as the Dutch guidelines for economic evaluation research, which have recently been revised (1, 2).

[1] ZonMw-oproep 'Kennisbevordering over de effectiviteit van psychosociale interventies die zijn opgenomen in de DEI, fase 1 (consortium en voorstudie)'

[2] Sociale vaardigheden/onzekerheid/weerbaarheid; Angst, depressie, stemmingsproblemen en andere internaliserende gedragsproblemen; Druk gedrag en ADHD; Externaliserende gedragsproblemen/-stoornissen; Opvoedonzekerheid – preventief en lichte problematiek; Zware opvoedproblemen/multiprobleemgezinnen

ZonMw has therefore commissioned a broad consultation, which will include the six consortia, as well as economic evaluation experts and other stakeholders, with the aim of reaching consensus regarding the steps which have to be undertaken in order to come to a standardization of economic evaluation research for the youth sector.

2 Methods

In order to reach this aim a number of activities have been undertaken which are listed in this chapter (see Figure 1).

Figure 1: Steps undertaken in the broad consultation

2.1 Step 1: Methodological considerations based on existing literature

First, an overview has been made of the methodological issues and (practical) challenges for economic evaluation research in the youth sector. The definition of the youth sector in this consultation has been (in accordance with the focus of the ZonMw program "Effectief werken in de jeugdsector") limited to psycho-social care for children and youngsters. This implies that issues and considerations specifically related to economic evaluations of *somatic* care in youth, such as hospital care, are not the focus of this broad consultation. The mission statement of the ZonMw program

"Effectief werken in de jeugdsector" is to increase, consolidate and disseminate knowledge in order to improve psycho-social care for children and adolescents, which is usable for the following (current) sectors: mental health care in youth, local preventive youth care policy, and/or clients on the cutting edge between indicated youth care/ mental health in youth/youths with a mild intellectual disability. The term youth is used for both for children and adolescents.

In order to get an overview we performed a scoping review, which included, in addition to published literature, also conference proceedings, abstracts, and relevant presentations. The scoping review focused mainly on opinion/methodological papers, and did not include empirical studies (i.e. economic evaluations performed in the field of youth research). Search terms for retrieving potentially relevant papers were: challenges, issues, methods/methodological, considerations, problems AND youth, children, infants, youngsters, paediatric AND costs, cost-effectiveness, economic evaluation, quality of life, QALY. Furthermore, the references of the papers were checked for additional papers. The issues that were retrieved from the literature search were complemented with suggestions provided by members of the consortia during the earlier meeting (April, 2015). In this overview the methodological issues and (practical) challenges have been categorized by the framing aspects of an economic evaluation (perspective, time horizon, analytical approach, outcomes, costs, type of economic evaluation, and target population). The 'framing aspects' outcomes and costs have been further categorised in the following classifications: identification, measurement, and valuation. The issues and considerations that were retrieved from the literature were not judged for their relevance; we merely collected, classified and tabulated them.

2.2 Step 2: Inventory of guidelines, manuals, and instruments for economic evaluation research

Second, an inventory and substantive study has been performed looking at existing Dutch guidelines, manuals, and instruments for economic evaluation research. The aim of this inventory was to reveal - if any and if so - which methodological issues and challenges, identified in Step 1, have already been addressed in the existing Dutch guidelines, manuals, and instruments for economic evaluation research. We intentionally focused on Dutch documents only, because these are specifically aimed at providing guidance for conducting economic evaluations in the Netherlands, and for supporting healthcare decisions in the Dutch context.

2.3 Step 3: Identification of relevant stakeholders

During Step 1 and Step 2, and in cooperation with ZonMw, relevant stakeholders were identified. In this identification step, two groups of stakeholders were distinguished, i.e. the “performing” stakeholders and the “using” stakeholders. The latter group are those who can stipulate the conditions and the methods of economic evaluation research in the field of youth research. The “performing” stakeholders are the researchers who carry out economic evaluation research in the youth sector; the “using” stakeholders are those who will use the results of economic evaluation research for (research) policy and practice, e.g. at a national, municipal or institutional level.

Regarding the first group of stakeholders -the “performing” stakeholders - we included the following organisations:

- Academic researchers performing economic evaluation research in the field of youth research;
- The members of the six consortia;
- Knowledge institutes, such as the Trimbos Institute: the National Institute of Mental Health and Addiction (Ti); The Netherlands Youth Institute (NJI); the Dutch Centre for Youth Healthcare (NCJ); the National Institute for Public Health and the Environment (RIVM), the Netherlands Organisation for Applied Scientific Research (TNO), the National Health Care Institute (ZiNL);
- Organisations for professionals in the field of health technology assessment/economic evaluation research, such as the Dutch/ Flemish Organisation of Health Economics (VGE) and the Dutch Society for Health Technology Assessment (NVTAG).

Regarding the second group of stakeholders -the “using” stakeholders - we identified the following groups:

- Umbrella organisations for practice, such as the umbrella organisations for the Centres for Youth and Practice (CJG), Public Health Services (GGD) and the Dutch Association of Mental Health and Addiction Care (GGZ Nederland), which is the sector umbrella organisation of specialist mental health and addiction care providers in the Netherlands (including the Organisation for Outpatient Mental Health Care (RIAGG), the Organisation for Youth Mental Health Care, Mental Health Clinics, etc.);
- Umbrella organisations for schools and education;

- (Umbrella organisations) for the municipalities and provinces; the Ministry of Health, Welfare and Sport; the Ministry of Education, Culture and Science; the Ministry of Social Affairs and Employment; the Ministry of Security and Justice.

2.4 Step 4: Conceptualization of the document for consultation

Based on Step 1 and Step 2, a consultation document has been conceptualized, which gives a systematic overview of the methodological issues identified and (practical) challenges for economic evaluation research in the youth sector, and the way these have already been addressed in existing Dutch guidelines, manuals, and instruments for economic evaluation research.

2.5 Step 5: Written consultation

The consultation document has been sent to the "performing" stakeholders: i.e. the academic researchers performing economic evaluation research in the field of youth research, the members of the six consortia, and the knowledge institutes.

In this written consultation these stakeholders have been asked to:

- give an overall impression of the consultation document;
- suggest additional methodological issues and (practical) challenges for economic evaluation research in the youth sector in addition to those noted in Table 2 (for Step 1);
- suggest additional existing Dutch guidelines and manuals for economic evaluation research, which should be consulted in addition to those noted in Table 3 (for Step 2);
- suggest additional literature, which is relevant for the scoping review (for Step 1);
- prioritise methodological issues and (practical) challenges for economic evaluation research in the youth sector in order of importance; this means that the stakeholders have been asked to rank a maximum of the ten most important methodological issues and challenges listed in Table 2 (including any additional ones suggested by the stakeholder themselves)
- suggest possible (procedural) solutions for the methodological issues and challenges which are included in the top 10 - these suggestions might include adapting the existing guidelines

(including adding module(s) to the existing guidelines), adapting existing sets of instruments, suggestions for additional research, etc.

In this written consultation the stakeholders were asked to state their exact title and name and organisation (see Table 2).

2.6 Step 6: Ranking of the most relevant methodological issues and challenges for economic evaluation research in the youth sector

All stakeholders were asked to rank their top 10 most important issues. Issues that were ranked most important were assigned 10 points; least important issues were assigned 1 point. If for example, only 3 issues were ranked, a maximum of 3 points was assigned to the most important issue. The issues were subsequently clustered into the framing aspects. Furthermore, based on feedback from the respondents, the final consultation document was adapted.

2.7 Step 7: Consultation meeting with the stakeholders

The written consultation document was discussed in a consultation meeting with all experts on February 18, 2015 at the office of ZonMw. The “performing” stakeholders and “using” stakeholders were invited to this consultation meeting and had a discussion based on the consultation document (see Step 6). The purpose of this meeting was to further prioritize the steps which have to be taken in order to come to a standardization of economic evaluation research for the youth sector. The aim and results of the broad consultation were furthermore presented and discussed during a masterclass which was held at the conference Youth in Research on March 14th 2016[3].

[3] Congres Jeugd in Onderzoek 2016, maandag 14 maart 2016, Congrescentrum Brabanthallen 's Hertogenbosch

2.8 Step 8: Final report and consultation

Finally, a report has been composed, describing:

- the results coming from each step of this consultation;
- the main points discussed during the written consultation and the consultation meeting;
- a common vision of the steps (research agenda) which have to be taken in order to come to a standardization of economic evaluation research in the youth sector.

3 Results

3.1 Methodological issues and (practical) challenges

3.1.1 Results of scoping review

For the scoping review we included 18 papers, 1 white paper and proceedings from 2 Dutch conferences. Table 1 (in conjunction with the references) lists the methodological issues and (practical) challenges for economic evaluation research in the youth sector, categorized by the framing aspects. Some of the issues are presented on a detailed level (and may show some overlap), whereas others are more general (and overarching). Furthermore, although some issues may not be unique for the youth sector, they are nevertheless included because they have been addressed in the context of youth.

Table 1: Methodological issues and challenges for economic evaluation research in the youth sector categorized by the framing aspects

Framing aspect	Problem / issue / challenge
1. Perspective	1. The societal perspective does not consider the distribution of required resources or benefits among stakeholders/agencies and sectors (3). 2. Although the societal perspective is preferred, often the funding organization is interested only in those aspects which affect their particular agency (4) 3. There is a danger of identifying consequences both as costs and effects, i.e. double-counting (3); there is even potential for double-counting of economic benefits (e.g., those related to criminal activity and juvenile justice services) (5)
2. Time horizon/ analytic approach	1. Although a lifetime horizon is considered necessary, follow-up periods are relatively short or differ among studies, and there may be an absence of longitudinal data from long-term follow-up studies, e.g. due to limited resources, high non-response and stakeholder demand for quick answers (3, 5-7). 2. With a long-time horizon, it is difficult to establish whether positive outcomes are still the result of the original intervention (7). 3. Improved techniques are needed for valuation and extrapolation of costs and outcomes over time, in order to study the long-term consequences of an intervention (8). 4. Although it is tempting to forecast benefits beyond the follow-up period, forecasts are often unreliable in terms of actual data (5).
3. Outcomes: identification	1. In addition to outcomes targeted by interventions, there may be other non-targeted (positive or negative) outcomes that may have short-term and/or long-term financial impacts (9); accordingly, the scope of relevant outcomes is difficult to determine (7). 2. QALYs of the child do not capture benefits in other domains (7, 10, 11).

		3. Generic HRQL may be insufficiently sensitive to the kinds of change observed for people with mental health problems, and particularly for children and adolescents (12). 4. QoL / QALYs of the child do not capture benefits to others (unitary approach), e.g. family members ((7, 10, 11, 13). 5. Unit of analysis is unclear; should this be the individual child/youth or the total system (parents, sibling, peers) (3, 6)?
4.	**Outcomes: measurement**	
I	***General***	1. There is a lack of instruments which include full range of (economic) effects; it may be necessary to augment clinical instruments with other measures (5, 6). 2. There is a great variety of assessment tools and scales as each stakeholder (society, healthcare policy, patient) has a different desired outcome and value system, with corresponding implications for choice of assessment tool or method (14). 3. There is a lack of generalised outcome measures / QoL instruments that can be applied generally (15, 16); no generic instrument is available for children younger than 5 years (10). ---
II	***Concept/dimensions of Health/QoL in children***	4. There is considerable diversity in the conceptualisation of and operational approach to QoL in children (14). 5. There is a lack of consensus regarding the fundamental construct / definition of QoL in children/adolescents and what domains to encompass (16, 17); Pal, 1996, (14). 6. It's necessary to specify which items comprise the key outcome domains of the child's health, functioning and HRQL, the causal relationships between items and outcome domains, and also specify the factors (family, developmental level) that influence key outcome domains (18). 7. There may be additional attributes or domains related to a child's social and physical functioning that are not present in adult instruments (10, 11). 8. Age-specific dimensions *versus* a core set of dimensions relevant to all ages for all children and adults? (19) 9. Items in a HRQL instrument should correspond to experiences, activities and contexts that are directly relevant to the age of the sample, i.e. be relevant to the child (14, 20); most instruments do not enquire about context, e.g. the family / family functioning and social environment (21). 10. Many of the dimensions on the MAU scales do not characterize the experiences of very young children (19). 11. It is very hard to develop hypothetical health state descriptions [for valuation studies] *for very young children* because it's impossible to know the experience of a particular health state (13). ---
III	***Children's limited abilities***	12. It is a challenge to create (preference-based) instruments that are child-centred with respect to measurement and valuation; these instruments are lacking (11, 21). 13. The wording of (indirect preference-based) instruments may pose (language) comprehension challenges to young children (11, 20). 14. Linguistically adapted versions of adult instruments may have questionable applicability to paediatric populations (10). 15. Adapting adult measures for use in research on children is complicated due to rapid developmental changes that take place in childhood and adolescence (10).

	16. Children at various ages have different cognitive abilities and linguistic skills for reporting and evaluating [see also under valuation] their health status, and the type of (abstract) health concepts that they can comprehend varies as they age; bias is possible due to their perception of time, the way they are questioned, the length of the instrument (burden) and the influence of the setting (10, 14, 19-21). 17. There is no consensus regarding at what age a child can report his/her QoL (due to developmental issues, language development, reading ability, recall ability). Children below age of 8 have restricted abilities with respect to introspection, meta-communication, abstract thinking, and reflection; older children struggle with the problem of social desirability, the influence of adults, and the wish to conceal feelings (17). 18. Research is required to establish the psychometric integrity of the measurement approach when applied to children, i.e. practicality, internal consistency, reliability, validity and responsiveness (14, 18, 19, 21). 19. There is a lack of adequate and validated (health state classification and/or preference-based) instruments specifically for children/youth (6, 10, 13, 15, 21).
IV Proxy-measurement	--- 20. There is a methodological issue / no consensus about how to identify the appropriate respondent for the description and valuation process in preference-based measures / information on children's QoL (17, 19-21). 21. Strong relations between the welfare of the family and the child complicate judgement regarding whose opinion should be sought (21). 22. A proxy report is somewhat inconsistent with the concept of HRQL, which is defined according to the patient's subjective view (16, 20). 23. Proxy reports may be problematic due to weak agreement between the child and parent proxies, especially for the subjective, social, emotional domains of QoL (11, 14, 15, 17, 19-21). 24. There is a question whether low parent-child concordance is due to limitations in abstract reasoning [the child's limited abilities] or to true difference in perspective or opinion (21). 25. A parent's assessment of the impact of an illness on a child may be biased by how they and others in the family, are affected (17, 20). 26. The parents' views are themselves affected by their own health status, knowledge, experiences and expectations (19). 27. Collecting data from both child and a parent [multiple sources] may provide the most complete picture (18)), but will be more costly and raises several methodological questions, e.g. whose reports are more accurate (20).
V Outcomes in others	--- 28. The measurement of QoL in significant others should be performed independently, without considering the QoL trade-off among family members (11).
5. Outcomes: valuation ***I General***	1. There is a lack of a valuation sets for youth. Valuation sets originally derived from adult preferences may not be appropriate for reflecting the experience of health states by children (6, 7, 11). 2. The methods used to calculate QALYs for children and adolescents vary extensively (10).

II Proxy-valuation	3. Direct health state valuation by children/adolescents raises difficulties, due to their lack of cognitive skills for understanding TTO/SG, difficulty in identifying indifference points, [age-dependent] differences in their attitudes towards risk, difficulty in grasping the concept of time [violation constant proportional trade-off] or possibility of death (11, 13). --- 4. The methodological issue is to identify the appropriate respondent for the description and *valuation* process in preference-based measures (19). 5. Proxy valuations (e.g. by parents) may be influenced by competing priorities (other children in family, guilt, other beliefs) and changes with regard to their own Health Related Quality of Life (HRQoL)/ interdependence of HRQL (10, 11, 13). 6. Adult descriptions of the child's health status are commonly reinforced by adult valuations, resulting in constructs that overlook the child's subjective perceptions and preferences (19). 7. Perspective of the proxy may influence valuation (imagine being a child, or value from own adult perspective) (11, 13). 8. There is a problem of how to combine sets of utility values (e.g. parent and child); simple aggregation is not valid solution because of utility interdependence (10). 9. There is weak agreement between child and parent proxies for direct and indirect utility elicitation approaches (11).
6. Costs: identification	1. There is no clear typology of youth services; terms relating to services are used differently (4). 2. Relevant issues should be identified through a literature review and a pilot study (22), but this may be hampered by time and money constraints (4). 3. There is no gold standard for identifying the broad range of services that might be used (4)→ use classification (23). 4. The unit of analysis is unclear; should this be the individual child/youth or the total system (parents, sibling, peers) (4)?
7. Costs: measurement ***I General***	1. There is no clear approach or valid/reliable instrument to measure resource use (4, 6, 8, 24). 2. Existing resource use instruments are primarily applied to adult programs (e.g. DATCAP) (5). 3. There is a lack of time and resources for developing sound methods for measuring resource use (4). ---
II Source for measurement	4. Self-reported instruments may suffer from recall bias (4). 5. Obtaining permission to use databases for measuring costs is difficult (4). 6. Proxy reports or data should be issued from multiple sources (parent, child) (4).
8. Costs: valuation	1. There is a lack of (uniform valuation/national applicable) unit costs for social services, school services, and criminal justice services(4-6, 25) (focus on education). 2. Should unit costs be figured nationally or locally (5)? 3. Some unit cost estimates may be applicable only to adults, e.g. criminal activity (5). 4. There is no standardized method for calculating unit cost (building blocks) (4). 5. Unit prices may not be transferable across settings and countries (8).
9. Type of economic evaluation	1. Selection of appropriate type of economic evaluation (i.e. CEA, CUA, CBA, cost consequences) may not be obvious (7).

	2. Focusing on one target outcome in CEA may be too narrow (5, 7, 12). 3. Determining cost-consequences poses a challenge if ratios point to different interventions being more cost-effective (12). 4. Selection and conversion of outcomes to money (e.g. school absenteeism, school expulsion) may be difficult in cost-benefit analysis (CBA) (5, 7, 12). 5. WTP (in CBA) suffers from income effects and other biases, and all other difficulties related to preference assessment in children apply (11).
10. Target population	1. The population is heterogeneous, leading to uncertainty regarding the cost-effectiveness of the intervention (6). 2. The population may not be accessible, due to a youngster's recognition of the problem/condition, (un)willingness to participate in the intervention and study, and it may be difficult to find an appropriate control intervention/situation. The intervention may spread among the target group (6).

3.1.2 Summary of the scoping review

Perspective

Although most seem to agree on the perspective which, in principle, should be the broad societal perspective, the distribution of costs and effects over different stakeholders should receive more explicit attention. Furthermore, taking a societal perspective raises the challenge of identifying and measuring the broad range of resources used and outcomes, as well as the potential danger of double-counting costs and consequences.

Time horizon

Most economic evaluations performed thus far lack a long-term follow-up which is essential for evaluations in the youth sector. Although it is recognised that modelling techniques may be necessary to estimate the long-term cost-effectiveness, improved techniques and long-term data are considered necessary to do this in valid way.

Costs: identification, measurement and valuation

Regarding the measurement of *resource use and the valuation of costs,* it is mentioned that there is no standard available for identifying the broad range of services and types of resources that might be relevant for the analysis. Instruments for measuring resource use in youth are either lacking or based on instruments developed for adults and have not been properly validated for use in youth. Furthermore, it can be difficult and time-consuming to gain access to existing databases or registries, and self-reported measures may suffer from recall bias and raise the question whether data from

youngsters themselves, proxy reports or multiple sources/informants should be used for analysis. Finally, it is noted that no (Dutch) standardized unit costs are available, nor is there a standardized method for calculating costs which fall in sectors other than health care, such as social services, education/school services and criminal justice.

Outcomes: identification, measurement, and valuation

With respect to *outcome(s)* in economic evaluation in the youth sector, it is put forward that the scope of relevant (targeted and non-targeted) outcomes may be difficult to determine, and that each stakeholder may focus on different desired outcomes. Consequently, it is unclear which type of economic evaluation should be the standard. Moreover, in a cost-utility analysis (CUA), Quality Adjusted Life Years (QALY) of the youngsters (based on preference-based measures like the EuroQol-5 dimensions EQ-5D) do not capture outcomes beyond health, nor do QALYs include benefits gained by other persons, e.g. parents, family, or other stakeholders. This raises the question of what the appropriate unit of analysis is - should this be the youngster, their family, or broader? In a cost-effectiveness analysis (CEA), the focus is on one target (natural) outcome and this may be too narrow to capture all relevant (targeted and non-targeted) outcomes. In a cost-consequence analysis (CCA), the incremental cost-effectiveness ratios based on different outcomes may point to different interventions being cost-effective. In a cost-benefit analysis (CBA), both the selection and conversion of outcomes to money may be problematic.

Much of the literature (see Table 1) has focused on the problems and challenges in the *measurement* of health and (health-related) Quality of Life (QoL) in youth. In general, there is a lack of validated (preference-based) QoL instruments specifically developed for youth, and if available, more research should be dedicated to establishing the feasibility and measurement properties of these instruments.

Adapting adult measurements for use in youth research may be questionable, as the concept and relevant dimensions of health/QoL, especially in children, are likely to differ from those in adults, and may even be age-dependent. Furthermore, children at various ages have difficulties understanding and reporting their health/QoL, due to limited cognitive abilities and linguistic skills, in comparison with adults.

Although it is generally agreed that QoL is subjective, and that a QoL instrument should reflect the perspective of the child, proxy reports of health/QoL may be necessary to replace or complement the self-reports of children. However, proxy reports of a child's health or QoL may be confounded by the proxy's own value system and how the proxy (or others in the family) is affected by the child's condition. Furthermore, proxy reports may be problematic due to weak agreement between child

and proxy (e.g. parent and child), or between different proxy respondents (father or mother), the latter raising the question who the appropriate proxy is.

If, in addition to the QoL measurement of children, the QoL is also measured for people who are close to the child, e.g. the parents, then generally this done without consideration that there is a QoL trade-off (i.e. utility interdependence) between family members.

With respect to *valuation* of outcomes, there is a lack of valuation sets specifically developed for youth in order to construct utility scores. Existing valuation sets based on adults' preferences may not be appropriate for reflecting the experiences of children and adolescents. Health state valuations performed by children themselves raise similar problems as in health/QoL measurement, due to children's limited cognitive and linguistic abilities. In addition, children may have a different attitude towards risk, or may have difficulty comprehending the concept of time, or the possibility of death. Alternatively, proxy valuations can be used. However, proxy valuations raise the same issue as mentioned earlier in regard to the health/QoL measurement, with respect to who the appropriate proxy is, weak agreement between the child's and proxy valuations, and confounding due to the proxy's own value system and utility interdependence, the latter also being influenced by the perspective of the proxy.

Time horizon / analytical approach

Although it is recognized that the time horizon should be long enough to capture all downstream costs and benefits over time, most of the economic evaluations in the youth sector have applied a short time horizon. The reasons for the short time horizon are that stakeholders may request a swift answer, resources may be limited, there may be a limited time horizon in the call for proposals, and limited possibilities for a valid long-term follow-up due to a high nonresponse. As a consequence, there is a lack of data available which can be used as input for long-term modelling studies, and there is the danger of forecasts being unreliable. Furthermore, due to the many transitions that a youngster goes through over time, it may also be difficult to establish whether there is still a causal relationship between the original intervention and (positive) effects over time.

Target population

The youth population is heterogeneous with respect to age, ethnicity and cause of the problem (behaviour)/condition, which may impact the results of economic evaluations. Also, there may be problems in gaining access to youth.

3.2 Inventory of existing guidelines, manuals and instruments for economic evaluation

3.2.1 Included existing Dutch guidance documents

For this consultation the following guidelines, manuals and instruments for economic evaluation have been studied:

- Bouwmans CAM, Schawo SJ, Jansen DEMC, Vermeulen KM, Reijneveld SA, Hakkaart-van Roijen L. Handleiding Vragenlijst Intensieve Jeugdzorg: Zorggebruik en productieverlies. Erasmus Universiteit Rotterdam, 2012 (26);
- Bouwmans C, Schawo S., Hakkaart-van Roijen L. Handleiding Vragenlijst TiC-P voor kinderen. Rotterdam: iMTA, Erasmus Universiteit Rotterdam, 2012 (27);
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3.2.2 Results of review of existing Dutch guidance documents

Table 2a lists how the methodological issues and (practical) challenges have been addressed in existing guidelines and manuals for economic evaluation. Table 2b gives an overview of how Dutch

instruments for costs and the EQ-5D-Y address the issues that are specifically mentioned under the framing aspects, i.e. identification, measurement and valuation (of costs and outcomes). In Tables 2a and 2b, the issue number in the first column refers to Table 1, where the first number refers to the "framing aspect", and the second number (behind the point) refers to the specific "problem/issue/challenge". For instance 1.1 refers to the framing aspects "Perspective" and to the problem/issue/challenge "Although societal perspective is preferred, often the funding organization is interested only in those aspects which affect their particular agency".

Issue no. Table 2	Topic	Guideline EE in healthcare, 2015	Guideline societal CBA	Guideline societal CBA, Health	Guidance outcomes research	Guideline cost research, 2015	Guideline ICB, 2014

Table 2a: Methodological issues addressed in Dutch guidelines and manuals

Perspective							
1.1	**Distribution of costs and effects**	Page 16: States that all costs and benefits should be considered, irrespective of who carries costs or gains benefits	Page 30, etc.: Throughout the report it is reflected that all stakeholders and actors should be included in the societal CBA. Page 33 and 34: Explicitly statess that the distribution of costs and benefits among stakeholders and agencies should be made explicit Page 34: States that the wealth of individuals should be aggregated to the wealth of the society	Page 44, etc.: States that the societal costs are preferred. In addition it is explicitly mentioned that the distribution of costs and benefits among stakeholders and agencies should be made transparent.	Page 34: States that all costs should be considered taking all actors into account, irrespective of who carries the costs	Page 14: Makes a reference to guideline EE in healthcare 2015. States that all costs should be considered irrespective of who carries them.	Page 22: States that the societal costs are preferred Page 48: States that all costs and benefits should be considered, irrespective of who carries costs or gains benefits
1.2	**Stakeholder prefers narrow perspective**	Page 16: In addition to the societal perspective, results can be presented from other perspectives, if justified. Page 39: States that the budget impact analysis (BIA, Chapter 5) is performed from the perspective of the budget holder (payer's perspective)		Page 44: In addition to the societal perspective, results can be presented from other perspectives; however, the societal perspective is preferred.	Page 25: Justification is needed for using perspective other than the societal perspective.	Page 14: In addition to the societal perspective, results can be presented from other perspectives, if relevance is justified.	Page 22: In addition to the societal perspective, results can be presented from other perspectives; however, the societal perspective is preferred.

Issue no. Table 2	Topic	Guideline EE in healthcare, 2015	Guideline societal CBA	Guideline societal CBA, Health	Guidance outcomes research	Guideline cost research, 2015	Guideline ICB, 2014
1.3	**Double-counting**	Page 29,31: States that double-counting of costs should be avoided (but not how)	Page 41: States that double-counting is less common in a societal CBA in comparison with MCDA Page 63,64,91,123: States that double-counting of costs and benefits should be avoided and offers some suggestions to do so	Not explicitly issued	Not issued	Not issued	Page 52-54: States that double-counting of costs and benefits should be avoided
Time horizon/analytical approach							
2.1	**Short follow-up periods**	Page 11,18: States that EEs should preferably have a lifetime horizon. The time horizon should be long enough to make a valid and reliable estimation of the difference in costs and effects between interventions. Page 18: States that deviation from lifetime horizon should be justified	Page 87: Defining a time horizon is an explicit step in societal CBA; state that the time horizon is defined by the period for which the problems are relevant, in which bottlenecks will occur and the period for which opportunities will occur.	Page 50: The societal perspective indicates that the time horizon should be long enough to include all costs and benefits, even when they are indirect, and occur after a long time period.	Page 17: Note that extrapolation over time, or from intermediate to final outcomes, is necessary if the patient follow-up period is too short	Page 20,21: Recognizes the problem of short follow-up periods in clinical trials and observational studies	Not explicitly stated
2.2	**Blurred relation - intervention with long-term outcomes**	Not issued	Not issued	Page 47 and page 66: Explicitly mentions the difficulty of causality in the analysis, i.e. the link between intervention and effects needs to be quantified	Not issued	Not issued	Page 70-71: Explicitly mentions the difficulty of causality between intervention, health and other societal costs

Issue no. Table 2	Topic	Guideline EE in healthcare, 2015	Guideline societal CBA	Guideline societal CBA, Health	Guidance outcomes research	Guideline cost research, 2015	Guideline ICB, 2014
2.3	**Improved techniques for economic modelling necessary**	Page 21-25 (Chapter 2): Distinguish between several (advanced) modelling techniques and analyses and make a reference to ISPOR/SMDM guidelines for modelling and modelling handbooks	Not issued	Not issued	Not issued	Not issued	Not issued
2.4	**Forecasts unreliable due to lack of long-term data**	Page 24: Note that extrapolation techniques should be used in case of missing long-term data. Uncertainty around these parameters should be addressed in probabilistic sensitivity analysis and scenario analyses. Make a reference to NICE DSU technical support document.	Page 155: Extrapolation methods are an explicit part of the risk analysis of the societal CBA.	See 2.2	Page 20-22 Address uncertainty in decision analytical modelling	Page 14: Makes a reference to simple extrapolation or econometric / statistical models and the importance of sensitivity analysis	Page 70-71: Explicitly mentions the difficulty of causality between intervention health and other societal costs
Outcomes: identification (see also type of economic evaluation)							
3.1	**Scope of relevant outcomes difficult to determine/ External effect**	Page 18: Note that choice for a particular outcome (in effectiveness research) depends on the patient population, disease/condition and aim of treatment. Page 44: Suggest using an outcome measure (next to QALY in reference case) in forensic interventions which complies with the primary objective of	Page 56: Note that external effects are an important aspect to consider; provide an example of a classification of external effects.	Page 30: Mention external effects. Page 83: Provide an example of an external effect, i.e. tax revenues.	Page 38: Refer to additional clinical outcomes (in medication research) such as efficacy, effectiveness, side-effects, patient experience, feasibility, ease of use (not specified for youth).	Not applicable	Not applicable

Issue no. Table 2	Topic	Guideline EE in healthcare, 2015	Guideline societal CBA	Guideline societal CBA, Health	Guidance outcomes research	Guideline cost research, 2015	Guideline ICB, 2014
		intervention, e.g. Criminal Activity-free Years or Drug Abuse-free Years (not specified for youth).					
3.2	**QALYs do not capture outcomes in other domains**	Page 30: Note that other instruments for QoL can be used in addition to the EQ-5D-5L, but this should be justified (not specified for youth). Page 43, 44: Note that wellbeing / broader QoL can be used if an intervention is not aimed at improving health. The ICECAP is recommended for this (but has not been developed for youth). Page 44: Suggest using an outcome measure (next to QALY in reference case) in forensic interventions which complies with the primary objective of intervention, e.g. Criminal Activity-free Years or Drug Abuse-free Years (not specified for youth).	Not issued	Page 10: Notes that the current QALY concept is not suitable for quantifying the health gain of long-term care and that new concepts are needed. Page 67: Notes that the current 5 domains of EQ-5D do not cover all relevant health domains.	Not issued	Not applicable	Not applicable
3.3	**Generic HRQoL may be insufficiently sensitive in some populations**	Page 33: Note that other instruments for QoL can be used if EQ-5D-5L is not sensitive, but the EQ-5D-5L should also be included (not specified for youth).	Not applicable	Not applicable	Not issued	Not applicable	Not applicable

Issue no. Table 2	Topic	Guideline EE in healthcare, 2015	Guideline societal CBA	Guideline societal CBA, Health	Guidance outcomes research	Guideline cost research, 2015	Guideline ICB, 2014
3.4	**QALYs do not capture outcomes in others**	Not issued	Not applicable	Not applicable	Not issued	Not applicable	Not applicable
3.5	**Unit of analysis?**	Not issued	Not issued	Not issued	Not issued		
Outcomes: measurement							
4.1	**Lack of comprehensive instruments in youth**	Not issued	Not applicable	Not applicable	Not issued	Not applicable	Not applicable
4.2	**Variety in measurement tools**	Page 32: States that EQ-5D-5L should always be used to reduce variety (but has not been developed / validated for youth). Page 32,33 Note that other instruments for QoL can be used in addition to EQ-5D-5L.	Not applicable	Not applicable	Page 47: Note that QALY can be based on EQ-5D, HUI and SF-6D (not specified for youth). Page 51,52: Note that several descriptive and disease or domain-specific QoL measures are available, although less relevant for EE (not specified for youth).	Not applicable	Not applicable
4.3	**Lack of generalised outcome measure**	Page 11: Table 1 states that that at least EQ-5D-5L should be used (but has not been developed for / validated in youth) in reference case. Page 13, 18,32: States that QALY is the standard outcome measure in EEs within health care (and if relevant also life years gained).	Not applicable	Not applicable	Page 48: Note that HUI2 and HUI3 are instruments developed for youth.	Not applicable	Not applicable

Issue no. Table 2	Topic	Guideline EE in healthcare, 2015	Guideline societal CBA	Guideline societal CBA, Health	Guidance outcomes research	Guideline cost research, 2015	Guideline ICB, 2014
		Page 36: States that (costs per) life years gained should also be reported.					
4.4 – 4.11	**Concept and dimensions of health/QoL in youth**	Not issued	Not applicable	Not applicable	Not issued	Not applicable	Not applicable
4.12 – 4.19	**Children's limited cognitive and linguistic abilities**	Not issued	Not applicable	Not applicable	Not issued	Not applicable	Not applicable
4.20 – 4.27	**Proxy measurement**	Page 32: States that QoL instruments should be filled out by patients (but no reference to children).	Not applicable	Page 68: States that QALY can be based on a valuation in patient or in the general population, which can lead to difference (but no reference to children).	Page 46: States that patient fills out the QoL questionnaire (PROM; no reference to children).	Not applicable	Not applicable
4.28	QoL in significant others (utility interdependence)	Not issued	Not applicable	Not applicable	Not issued	Not applicable	Not applicable
Outcomes: valuation							
5.1	**Lack of valuation set**	Not issued Page 11,13,32: Dutch valuation set for EQ-5D-5L should be used in reference case (which has not been developed specifically for youth)	Not applicable	Not applicable	Page 48: Note that there is no Dutch valuation set for HUI2/3 (so also not for youth).	Not applicable	Not applicable
5.2	**Variation in methods for calculating QALYs**	Page 11,13,32: Dutch valuation set for EQ-5D-5L should be used in reference case (which has not been developed	Not applicable	Not applicable	Page 47: Note that QALY can be based on EQ-5D, HUI and SF-6D (not specified for youth).	Not applicable	Not applicable

Issue no. Table 2	Topic	Guideline EE in healthcare, 2015	Guideline societal CBA	Guideline societal CBA, Health	Guidance outcomes research	Guideline cost research, 2015	Guideline ICB, 2014
		specifically for youth)					
5.3	**Children's limited cognitive and linguistic abilities**	Not issued Page 32: States that health state valuations should be based on preferences of Dutch (adult) general population	Not applicable	Not applicable	Not issued	Not applicable	Not applicable
5.4-5.9	**Proxy valuation**	Not issued Page 32: States that health state valuations should be based on preferences of Dutch general population.	Not applicable	Not applicable	Page 46: States that society values health state descriptions, e.g. for EQ-5D and Health Utility Index (HUI) Page 50: If no valuation is available, valuation can be performed by clinical experts. Note that such valuations are subjective and less trustworthy.	Not applicable	Not applicable
Costs: identification							
6.1	**No uniform typology/terminology of youth services**	Not issued	Not issued	Not issued	Not issued	Not issued	Page 24: Provide a standard set of items, e.g. education and justice, with an explicit mention of the factors relevant for children.
6.2	**Proper identification limited by time/money constraints**	Not issued	Not issued	Not issued	Not issued	Not issued	Not issued
6.3	**Lack of gold standard for identification**	Page 29: Make a reference to guideline for cost research	Not issued	Not issued	Page 25: Make reference to guideline	Page 15: Distinguish between healthcare costs,	Page 24: Provide a standard set of items, e.g. education

Issue no. Table 2	Topic	Guideline EE in healthcare, 2015	Guideline societal CBA	Guideline societal CBA, Health	Guidance outcomes research	Guideline cost research, 2015	Guideline ICB, 2014
		2015 for identification. Page 30-32: Distinguish between health care costs, patient and family costs and costs in other sectors, the latter e.g. special education, and justice. Page 31: Make a reference to guideline ICB 2014 for costs in other sectors.			pharmacoeconomic research and guideline cost research for identification, measurement and valuation. Page 34: Present overview cost categories with some examples, e.g. special education and juridical costs.	patient and family costs and costs in other sectors, the latter e.g. special / special education, police & justice, damage caused by a patient. Page 18: States that the importance of separate identification of an item depends on its relative contribution to total and incremental costs. Page 20: Makes a reference to clinical practice guidelines, treatment protocols/standards, existing literature, expert opinion and diagnosis treatment combination (DBC) information for identification of relevant items in health care. Page 71: Make a reference to the classification scheme of the guideline ICB 2014 for costs in other sectors	and justice.
6.4	**Unit of analysis?**	Not issued	Not issued	Not issued	Not issued	Not issued	Not issued
Costs: measurement							

Issue no. Table 2	Topic	Guideline EE in healthcare, 2015	Guideline societal CBA	Guideline societal CBA, Health	Guidance outcomes research	Guideline cost research, 2015	Guideline ICB, 2014
7.1	**No valid/reliable instrument for youth available**	Not issued	Not issued	Not issued	Page 29: Notes that there are only a few standardized instruments available, and research into convergent validity is limited (not specified for youth).	Page 21: Refers to Medical Consumption Question-naire (iMCQ) and Treatment Inventory of Costs in Psychiatric Patients (TiC-P) for costs of healthcare consump-tion (not specific for youth)	Not issued
7.2	**Existing instruments developed for adults/adult programs**	Not issued	Not issued	Not issued	Not issued	Not issued	Not issued
7.3	**Instrument development hindered by time/money constraints**	Not issued	Not issued	Not issued	Not issued	Not issued	Not issued
7.4 – 7.6	**Source for cost measurement**	Page 29: Make a reference to guideline cost research 2015, which presents methods for measuring resource use.	Not issued	Not issued	Page 25: Make reference to guideline for pharmacoeconomic research 2006 and guideline for cost research 2010 for identification, measurement and valuation Page 28-32: Refer to several sources, such as patients (self-reporting; not specified for youth), healthcare	Page 20: States that choice should be guided by representativeness and generalisability of the data, impact on total and incremental costs, availability of the data. Page 20-33: Refer to several sources such as clinical studies, local/national registries, expert opinion, DBC system, literature or self-reporting by patients (not specified for youth) for use of	Not issued

Issue no. Table 2	Topic	Guideline EE in healthcare, 2015	Guideline societal CBA	Guideline societal CBA, Health	Guidance outcomes research	Guideline cost research, 2015	Guideline ICB, 2014
					professionals, registries. Page 29: States that cost diaries are more reliable than retrospective questionnaires or interviews (not specified for youth).	healthcare resources. Page 43-59: (Chapter 4): Present sources for measuring cost items within (mental) health care (not specified for youth). Page 62,63: Offers some guidance on methods for measuring patient and family costs (not specified for youth) Page 71: Make a reference to guideline ICB 2014 for costs in other sectors	
Costs: valuation							
8.1	**Lack of national unit costs**	Page 30: Makes a reference to the guideline for cost research 2015 for valuation / guideline prices	Not issued	Not issued	Page 25: Makes a reference to the guideline for pharmaco-economic research 2006 and the guideline for cost research 2010 for identification, measurement and valuation	Page 43-59: (Chapter 4): Presents sources for valuation / guideline prices for cost items within (mental) health care (not specified for youth). Page 59-61: Presents a table with guideline prices for (mental) health care, reference year 2014	Page 35-50 (Chapter 3 and 4) Presents sources for valuation / guideline prices for cost items for the justice and education sector.

Issue no. Table 2	Topic	Guideline EE in healthcare, 2015	Guideline societal CBA	Guideline societal CBA, Health	Guidance outcomes research	Guideline cost research, 2015	Guideline ICB, 2014
						(not specified for youth). Page 63-65: Offers some guidance on methods of value-tion and a table with standard calculation units / reference prices for some patient and family costs, e.g. travel costs and time costs (not specified for youth). Page 71: Offers guidance on valuation of productivity loss and some standard calculation units (not specified for youth). Page 71: Makes a reference to ICB 2014 for costs in other sectors Page 81-84 (Appendix 2): Offers a list of standard calculation units / guideline prices for (mental) health care (not specified for youth).	
8.2	**National or local unit costs?**	Page 30 Makes a reference to the guideline for cost research	Not issued	Not issued	Not issued	Page 24: States it is important to find a balance between	Not issued

Issue no. Table 2	Topic	Guideline EE in healthcare, 2015	Guideline societal CBA	Guideline societal CBA, Health	Guidance outcomes research	Guideline cost research, 2015	Guideline ICB, 2014
		2015 for obtaining national guideline prices or performing own unit price calculation.				standardisation and comparability between studies on the one hand, and the specific context of the EE on the other. Note that guideline prices are preferred to support decision-making at the national level. Page 23-28: Refers to several sources for unit prices for use of healthcare resources, like financial registries within institutions, tariffs (not DBC tariffs), market prices, literature, guideline prices, own unit price calculation.	
8.3	**Existing unit costs applicable for adults**	Not issued	Not issued	Not issued	Not issued	Not issued	Page 35-40: Chapter3 Unit costs for education
8.4	**No standardized methodology for calculation**	Page 30: Makes a reference to the guideline for cost research 2015 for methods for performing own unit price calculation.	Not issued	Not issued	Not issued	Page 29-42 (Chapter 3) Provides methodology/ building blocks for calculating integral unit prices in health care, e.g. if guideline price is not available or not specific enough.	Page 27: Provides a crude system for determining costs
8.5	**Transferability of unit costs**	Page 32: Note that if Dutch cost data are not available, foreign data need to be validated for Dutch situation.	Not issued	Not issued	Not issued	Page 27: If unit prices are obtained from literature, it's important to check	Not issued

Issue no. Table 2	Topic	Guideline EE in healthcare, 2015	Guideline societal CBA	Guideline societal CBA, Health	Guidance outcomes research	Guideline cost research, 2015	Guideline ICB, 2014
						applicability for own research.	
Type of economic evaluation (see also outcome identification)							
9.1 – 9.4	**Choice type of EE not obvious**	Page 11,13,19,32: State that a CUA should always be performed with QALY as an outcome measure (reference case). Page 13, 36: States that incremental costs per life years gained (if relevant) should also be reported (=CEA). Page 36: States that incremental cost-effectiveness ratios (ICERs) should be presented for additional relevant health outcomes. Page 44: Suggests that in forensic interventions (not specifically for youth) CEA should be performed (as well as the CUA reference case), based on an outcome related to the primary objective of the intervention, e.g. Criminal Activity-free Years (CAFY) or Drug Abuse-free Years (DAFY).	Not applicable, refers to (societal) CBA	Not applicable, refers to (societal) CBA	Page 33: States that CEA can be performed in addition to CUA, or CEA can be performed alone if there is no expected effect on QoL.	Not issued	Not issued
Target population							

Issue no. Table 2	Topic	Guideline EE in healthcare, 2015	Guideline societal CBA	Guideline societal CBA, Health	Guidance outcomes research	Guideline cost research, 2015	Guideline ICB, 2014
10.1	**Heterogeneity**	Page 17,22,23: Notes that differences in e.g. age, gender and condition can have great impact on results, and that subgroup analyses (preceded by a priori hypotheses) can be performed.	Not issued	Not issued	Page 20: Refers to subgroup analysis to address heterogeneity in modelling studies	Not issued	Not issued
10.2	**Accessibility**	Not issued	Not issued	Not issued	Not issued	Not issued	Not issued

Table 2b: Methodological issues addressed in existing Dutch instruments

Issue no Table 2	Topic	EQ-5D-Y, 2014 (development paper: 2010)	Questionnaire Intensive Youth Care, 2012	Questionnaire TIC-P for children, 2012
Outcomes: identification				
3.1	Scope of relevant outcomes / external effects	EQ-5D-Y reflects generic HRQoL	Not applicable	Not applicable
3.2	QALYs do not capture outcomes in other domains	EQ-5D-Y reflects generic HRQoL	Not applicable	Not applicable
3.3	Generic HRQoL may be insufficiently sensitive in some populations	EQ-5D-Y reflects generic HRQoL	Not applicable	Not applicable
3.4	QALYs do not capture outcomes in other persons	EQ-5D-Y reflects generic HRQoL in child	Not applicable	Not applicable
3.5	Unit of analysis	Child	Not applicable	Not applicable
Outcomes: measurement				
4.1	Lack of comprehensive instruments in youth	Not applicable	Not applicable	Not applicable
4.2	Variety in measurement tools	Not applicable	Not applicable	Not applicable
4.3	Lack of generalised outcome measure	EQ-5D-Y is generalized outcome measure	Not applicable	Not applicable
4.4-4.11	Concept and dimensions of health/QoL in youth	Adaptation from the adult EQ-5D	Not applicable	Not applicable
4.12-4.19	Children's limited cognitive and linguistic abilities	Page 877 (paper): Developed for self-reporting of children from 8 years onwards under the assumption that it can be correctly understood as from this age.	Not applicable	Not applicable
4.20-4.27	Proxy measurement	Not applicable Developed for self-reporting of children	Not applicable	Not applicable
4.28	QoL in significant others (utility interdependence)	Not applicable	Not applicable	Not applicable

Issue no Table 2	Topic	EQ-5D-Y, 2014 (development paper: 2010)	Questionnaire Intensive Youth Care, 2012	Questionnaire TIC-P for children, 2012
Outcomes: valuation				
5.1	Lack of valuation set	Not available for EQ-5D-Y Page 884 (paper): Interesting question of how social preferences should be elicited and who they should be elicited from	Not applicable	Not applicable
5.2	Variation in methods for calculating QALYs	Valuation set not available for EQ-5D-Y	Not applicable	Not applicable
5.3	Children's limited cognitive and linguistic abilities	Not issued	Not applicable	Not applicable
5.4-5.9	Proxy valuation	Not issued	Not applicable	Not applicable
Costs: identification				
6.1	No uniform typology/ terminology of youth services	Not applicable	Page 11-12: Provides terminology for several youth services in the Netherlands	Not issued
6.2	Proper identification limited by time/money constraints	Not applicable	Not applicable	Not applicable
6.3	Lack of gold standard for identification	Not applicable	Page 11/12: Provides a standard set of items, but notes that items can be omitted or added	Provides a standard set of items (not specified for youth)
6.4	Unit of analysis	Not applicable	Page 6: Unit of measurement is child and their parent(s) / caretaker(s)	Unit of measurement is child
Costs: measurement				
7.1	No valid/reliable instrument for youth available	Not applicable	Not issued	Not issued
7.2	Existing instruments developed for adults/adult programs	Not applicable	Page 29-42: Manual includes a questionnaire which is specifically developed for intensive youth care (children/adolescents aged 4-18)	Questionnaire is specifically developed for children.
7.3	Instrument development hindered by time/money constraints	Not applicable	Not applicable	Not applicable

Issue no Table 2	Topic	EQ-5D-Y, 2014 (development paper: 2010)	Questionnaire Intensive Youth Care, 2012	Questionnaire TIC-P for children, 2012
7.4 – 7.6	Source for cost measurement	Not applicable	Page 6-8: Self-report questionnaire to be filled out by parent/caretaker who spends most time with the child/adolescent, with recall period of 3 months for service use and 1 month for productivity losses. Recall period can be adapted based on characteristics of population / intervention.	Page 3: Self-report questionnaire to be filled out by parent/caretaker who spends most time with the child/adolescent, with recall period of 3 months.
Costs: valuation				
8.1	Lack of national unit costs	Not applicable	Page 13-16: For healthcare services, a reference to the 2010 guideline for cost research is made. For medication, a reference to CVZ Medicijnkosten.nl is made. Tables are provided which contain several guideline prices within (Table 1) and outside (Table 2) health care. Page 16-17: Note that some unit costs have not yet been calculated or are based on a shadow process.	Not issued
8.2	National or local unit costs	Not applicable	Page 13: Advises performing unit price calculation if there is an indication that unit costs deviate from the guideline price.	Not issued
8.3	Existing unit costs applicable for adults	Not applicable	Page 13-16: Unit costs for child services, if available, logically applicable to child/adolescent population.	Not issued
8.4	No standard methodology for calculation	Not applicable	Not issued	Not issued
8.5	Transferability of unit costs	Not applicable	Not issued	Not issued

3.3 Summary of how issues and challenges are addressed in existing guidelines, manuals and instruments

Overall, very few methodological problems/issues/challenges are solved in the existing guidelines and manuals, especially if we focus on the target population of this consultation, i.e. youngsters. What is remarkable is that some of the guidance documents touch upon some similar issues as in the scoping review, albeit mostly without providing any concrete solutions or alternatives.

For measuring resource use in children, two instruments are available. These instruments have specifically been developed for children, but are both to be filled out by a parent/caretaker. Where the TiC-P primarily focuses on resource use related to (mental) health care, school absence and leisure activities, the questionnaire Intensive Youth Care also includes resource use items outside the healthcare sector, such as youth care, social care, residential care and contact with judicial authorities. The IYC questionnaire also includes resource use by the parents due to the antisocial behaviour of the youngster; this includes health care resource use (by the parents), absence from work and reduced efficiency at work.

With respect to measuring outcomes, the Dutch version of the EQ-5D-Y is available. This instrument is an adaption of the EQ-5D adult version. It is a generic instrument which is restricted to measuring health-related quality of life. No valuation set is yet available for the EQ-5D-Y.

3.4 Written consultation and stakeholders meeting

3.4.1 Response / attendance

The consultation document was sent out to 34 experts, including the two organisations for professionals in the field of health technology assessment/economic evaluation research. In addition, ZonMw sent out the documents to the 6 consortia. Consortia leaders were asked to forward the consultation document to the consortia members and “using” stakeholders. Nineteen feedback instruments were received from 24 stakeholders. Respondents consisted of 13 HTA/HE researchers, 5 knowledge institutes, and the Dutch/Flemish Health Economics Association.

Following the written consultation procedure, a stakeholders meeting was organized on February 18, 2015 to discuss the results. At this meeting, 14 stakeholders were present. Although the consortia

were asked to approach the "using" stakeholders for the broad consultation, none of the "using" stakeholders participated in the written consultation or in the stakeholders meeting.

None of the consulted stakeholders preferred to stay anonymous; for an overview of the stakeholders during the (written) consultation and the consultation meeting, see Table 3.

Table 3: Overview of stakeholders included in the (written) communication and the consultation meeting (in alphabetical order).

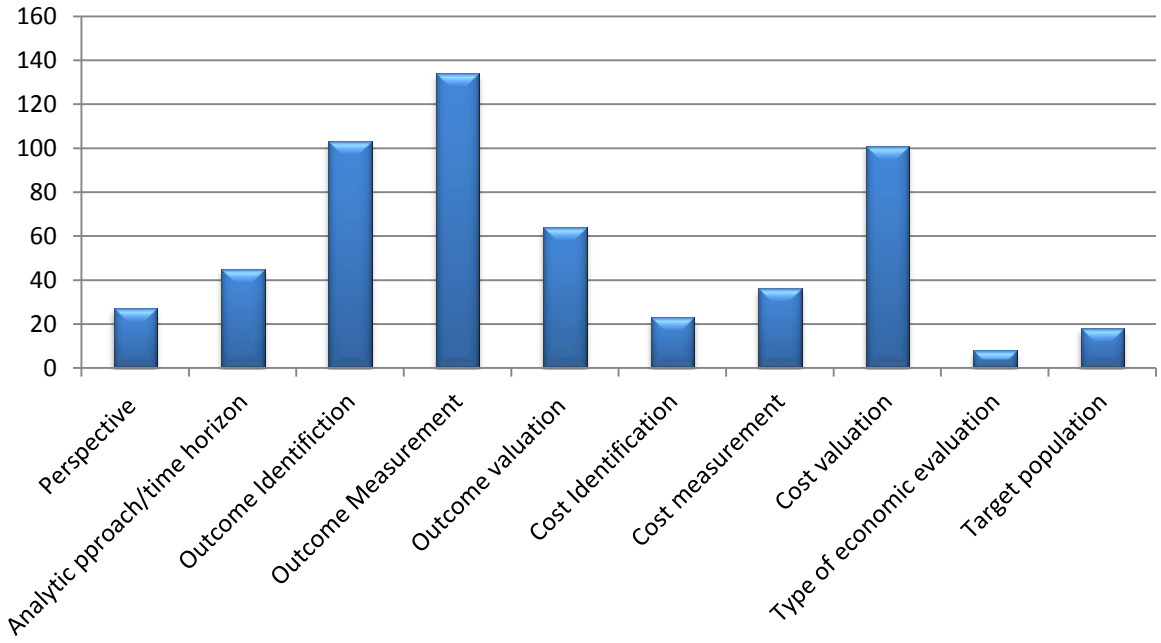
Name	Organization (English)	Written consultation	Stakeholders meeting
Dr. E.M.M. Adang	Radboud University Medical Center, Department Health Evidence	X	
Dr. M.E. van den Akker	Leiden University Medical Center (LUMC) Dutch Flemish Association of Health Economists (VGE)	X	X
Dr. T.A.D.I. van Asselt	University Medical Center Groningen (UMCG), Department of Epidemiology, unit HTA. University of Groningen (RUG), Groningen Research Institute of Pharmacy, unit PharmacoEpidemiology & PharmacoEconomics.	X	
Dr. G. van den Berg	Program Leader (Cost) Effectiveness and Integrated Youth at the Netherlands Youth Institute (NJi) Program Leader Research Youth System in Amsterdam.	X	X
Dr. J.E. Bosmans	Vrije Universiteit Amsterdam (VU) Department of Health Sciences and EMGO Institute for Health and Care Research	X	X
Prof. Dr. E. Buskens	University Medical Center Groningen (UMCG)	X	X
P. Dijkshoorn	Accare, Child and Youth Psychiatry Dutch Knowledge Centre for Child and Adolescent Psychiatry (KJP)	X	X
H. van Eeren	De Viersprong: Institute for Studies on Personality Disorders (VISPD), Halsteren, The Netherlands. Erasmus Medical Center (Erasmus MC), Department of Psychiatry, section Medical Psychology and Psychotherapy	X	X
Dr. K.E. Evenboer	University Medical Center Groningen (UMCG)		X
Dr. M. Fekkes	Netherlands Organisation for Applied Scientific Research (TNO), Child Health		X
M. Goorden	Erasmus University Rotterdam (EUR)		X
Dr. L. Hakkaart	Erasmus University Rotterdam (EUR) institute for Medical Technology Assessment (iMTA) Institute Health Policy and Management (iBMG)	X	

Name	Organization (English)	Written consultation	Stakeholders meeting
Prof. Dr. P.J. Hoekstra	University Medical Center Groningen (UMCG) Accare, Child and Youth Psychiatry	X	X
Dr. B.J. van den Hoofdakker	Accare, Child and Youth Psychiatry University of Groningen (RUG)	X	
R. Hornstra	Accare, Child and Youth Psychiatry		X
Prof. dr. C. Hosman	Maastricht University, Department of Health Promotion. Radboud University, Department of Clinical Psychology. CIKEO consortium (parenting interventions), Erasmus University and AMPHI Academic Workplace, (Public Health), Radboud University.	X	
Dr. D.E.M.C. Jansen	University Medical Center Groningen (UMCG), Department of Health Sciences	X	
Dr. H. Jonkman	Verwey Jonkers Institute		X
Dr. M.A. Joore	Maastricht University Medical Center Department of Clinical Epidemiology and MTA	X	
D. Kann	Netherlands Youth Institute (NJI)	X	X
Dr. J. Lokkerbol	Trimbos Instituut (Ti) University Medical Center Groningen (UMCG) University Center for Psychiatry (UCP)	X	
Dr. J.O. Mierau	University of Groningen (RUG)	X	
T.M.D. Ngo	Dutch Association Of Mental Health And Addiction Care (GGZ Nederland)	X	
Dr. F.I.M. Pijpers	National Institute for Health and the Environment (RIVM), Centre of Health and Society, Department of Care and Prevention	X	
Dr. K.C.P.J. van der Ploeg	Netherlands Organisation for Applied Scientific Research (TNO), Child Health	X	X
Prof. Dr. J.J. Polder	National Institute for Public Health (RIVM) Tilburg University	X	
Dr. E.A. Stolk	EuroQol Research Foundation	X	
Dr. K.M. Vermeulen	University of Groningen (RUG), Department of Epidemiology	X	
Dr. A. de Wit	National Institute for Public Health (RIVM) Utrecht University	X	

In the written consultation procedure, all stakeholders were asked to rank their top 10 most important issues. Results are presented in Figure 2. In order of importance, the topics were: outcome measurement, outcome identification, cost valuation, outcome valuation, time horizon / analytical

approach, cost measurement, perspective, cost identification, target group, type of economic evaluation. Remarkably, outcome identification, outcome measurement and outcome valuation all ranked in the top five. This confirms the findings of the scoping review, in which many issues and considerations related to this topic have been put forward. Cost valuation and time horizon/analytical approach were also among the five most important topics.

Figure 2: Prioritization of topics



Stakeholders also provided suggestions for possible (procedural) solutions for the methodological issues and challenges. These are listed in Table 4 below.

Table 4: Potential solutions

Top 10 issues/considerations	Potential (procedural) solutions
1. Outcome measurement	• More fundamental research necessary for definition of QoL in youth (including measurement method) • Development of a QoL instrument for youth • Suggestion: Rotterdam Wellbeing of Youth Scale (R-WYS; not yet available), TAPQOL (not preference-based), HUI • Adapt outcome measurement according to age • Add dimensions to generic instruments • 'Best' proxy may vary per child and even per measurement (example CBCL) • Guidance/guideline with respect to proxy measurement • Focus only on outcomes in child, as no accepted methods are available for including outcomes in others (in QoL research) • Guidance/guideline with respect to unit of analysis • Additional analysis with outcomes in family or others as unit of analysis • Need for an overview of existing instruments and their psychometric properties and drawbacks
2. Outcome identification	• Answer normative question first: what is the objective of psycho-social interventions for youth • Broader range of outcomes in early intervention and prevention studies (and the relation between outcomes) • Subjective wellbeing as broader outcome • Need for qualitative studies to explore (relation between) outcomes
3. Cost valuation	• Need for standard for uniform cost prices and a method for calculating these in the youth sector (among others criminal behaviour, school absence in terms of further education and career perspective, (quality) time spent by parents) • Identify where existing manuals can be extended
4. Outcome valuation	*Health-related quality of life* • First answer normative question whose valuation counts (perspective): societal preferences or children's preferences

	• More fundamental research into valuation method (SG, TTO, VAS) • Develop Dutch valuation set for available instruments • More research into valuation of R-WYS *Other outcomes* • Monetize finished education (relation to work/career over the longer term) • Monetize improved scores on self-confidence / anxiety • Monetize death at young age
5. Time horizon / analytical approach	• Lifetime in accordance with guidelines • Time horizon depending on objective intervention (e.g. family functioning) • Establish relationship between intermediate (which) and final outcomes • Establish relationship between early factors/conditions and (mental) health in adolescence/adulthood • More focus on evidence synthesis / meta-analysis to strengthen the validity of results of economic evaluations • More economic modelling studies • Project on appropriate discount rate
6. Cost measurement	• Development of a uniform validated questionnaire for youth which can be adapted for specific target group • Validation of TiC-P for children
7. Perspective	• Distinguish economic evaluation from budget impact analysis; EE is not focused on distribution of costs and benefits • Perspective dependent on budget holder / commissioning agency
8. Cost identification	• Add taxonomy of cost items as appendix to cost manual
9. Target group	• Awareness regarding cost-effectiveness based on study population versus real-world cost-effectiveness • Awareness for representativeness of study population in relation to informed consent procedure up to age 18. • Awareness that cost-effectiveness may be dependent on moderators (context)
10. Type EE	• CUA is default, do not reconsider default for youth sector

3.4.3 Additional literature, guidelines and documents

Literature that was additionally suggested by the stakeholders consisted mainly of economic evaluations performed in the youth sector, which we intentionally excluded from the review. A few suggestions for other methodological papers were followed, and in following a suggestion we additionally included the TiC-P instrument for children (included in Table 2b). We also received some references to unpublished literature / documents / instruments.

3.4.4 Additional issues

Some additional issues besides those presented in Table 1 were also put forward. One was directed at the possibility of transferring economic evaluations from other jurisdictions to the Netherlands. It was also noted that attention should be paid to design issues, as a randomized design is not always attainable in the youth sector (25). In case of alternative designs, like observational studies, one should be aware of potential biases. In relation to this, the potential use and validity of 'routine outcome measurements' (ROM), databases and registries for economic evaluations was put forward. Some issues were not directly related to economic evaluation, but to the choice, working mechanisms and moderators of interventions and programs under evaluation.

3.4.5 Further prioritization during the stakeholders consultation meeting

At the stakeholders meeting, first the results of the written consultation procedure were presented. Following this, stakeholders were asked to further prioritize the steps to take, by means of group work.

Most of the issues brought forward during the stakeholders meeting confirm the results of the written consultation: most were about outcomes assessment (the need to develop a generic, preference-based QoL instrument focussing on well-being, including psychosocial aspects and broader outcomes, and to develop a module for youth in guidelines for economic evaluation), followed by costs (build on existing manuals and instruments, adapt instruments for measuring resource use in the youth sector), perspective (include other perspectives as well as the societal perspective), time horizon (follow the life course, using existing databases / data-linking) and unit of analysis (child and family). Again the 'routine outcome measurement' (ROM) was emphasized; on the one hand it was suggested that it be simplified by using shorter questionnaires, on the other hand it was suggested that ROM be aligned with instruments for use in economic evaluations. In this respect, it was recommended that a 'core set' of indicators for effectiveness be developed, to be used in addition to cost-utility analysis. With respect to perspective / time horizon, it was suggested

that economic evaluations be performed taking a municipality (budget holder) perspective up to age 18, and separately an economic evaluation with a lifetime societal perspective.

One important new issue was brought up as well, being that in the first place, attention should be paid to the context in which economic evaluations are currently being performed in the youth sector. As from 2015, the central government has transferred the budget and organisation of psychosocial care for youth to the municipalities. Municipalities may have different wishes and views towards the relevance and content of economic evaluations in the youth sector. It is therefore key to explore their views, but at the same time to inform them regarding good quality economic evaluations, even if performed in a 'quick and dirty' manner (develop a minimal standard).

4 Conclusion, discussion, and recommendations

4.1 Conclusion

The objective of the broad consultation procedure was to reach consensus regarding the steps which have to be undertaken towards further methodological development and the standardization of economic evaluations in the youth sector.

In order to reach this objective a systematic approach was chosen, which included a scoping review of the international opinion/methodological literature and an inventory of existing Dutch guidelines/manuals for economic evaluation. On two occasions, stakeholders had the possibility to provide their input: i.e. in the written consultation 24 stakeholders gave their input and 14 stakeholders participated in the consultation meeting.

This broad consultation resulted in a clear ranking of the methodological issues which were regarded as being most important for the further development of economic evaluation in the youth sector. The issues ranked in the “top 5” by the stakeholders are: 1) outcome measurement, 2) outcome identification, 3) cost valuation, 4) outcome valuation, and 5) time horizon / analytical approach. Existing Dutch guidelines and manuals provide guidance for some, but not all, issues and challenges. For the outcome side of the economic evaluation, normative questions have been posed such as: what is the goal of psychosocial care for youth which the outcome(s) in economic evaluations should comply with, and whose values count when obtaining preference weights for the outcome? Furthermore, respondents urged that they are in need of instruments specifically developed for youth to perform economic evaluations, such as instruments for measuring costs, preference-based instruments for measuring quality of life (utilities), and cost prices (e.g. for interventions, education, social care, and police/justice).

With respect to other methodological challenges, stakeholders generally agreed that the overall guidelines should be applied to the youth sector. For instance, regarding the perspective, most stakeholders agreed that economic evaluations should be, in principle, performed from the broad societal perspective, and, regarding the type of economic evaluation, that cost-utility analysis is preferred. With respect to the time horizon, the stakeholders agreed that a long-term time horizon is needed, but that in order to achieve this, more research is needed looking at the relationship between intermediate short-term outcomes and long-term final outcomes.

4.2 Discussion

To our current knowledge this is (inter)nationally the first broad consultation issuing methodological challenges and providing the groundwork for the standardization of economic evaluations in the youth sector. This broad consultation has several strengths. First, we included a large group of (academic) experts from different backgrounds. Second, this consultation was based on a systematic approach, in which the authors were transparent about each step undertaken. Third, during the scoping review and the consultation, we deliberately took a *non-normative* approach, meaning that all issues were included during the scoping review and during the consultation, without judging or selecting the issues according to their relevance.

Although overall the stakeholders considered the consultation document to be complete, transparent, detailed, consistent, useful and interesting, some limitations of this work were also put forward and need to be considered. First, during the inventory of existing guidelines/manuals for economic evaluation, we included materials only from the Netherlands, as this consultation focusses on the Dutch situation. As was mentioned during the written consultation, a systematic analysis of the international guidelines for economic evaluation might reveal additional ideas and solutions which are not reflected in the Dutch documents. In relation to this, consulting the broader international literature, outside the scope of economic evaluations, was recommended, for potential guidance and 'best practices' with respect to some methodological issues, such as proxy measurement. Second, although we included a large diversity of experts, not all relevant stakeholders were present during the broad consultation. For example, we did not include children and their parents as stakeholders in the broad consultation. In addition, during this consultation we received input mainly from the first group of stakeholders - the "performing" stakeholders (academic researchers, members of the six consortia, and knowledge institutes), while the "using" stakeholders (umbrella organisations from practice, school, and the government) did not attend the broad consultation. Third, although the aim of the broad consultation was to reach consensus regarding steps to take towards the standardization of economic evaluations in the youth sector, there was no time to complete a full consensus procedure, which should be done e.g. by means of several Delphi rounds. In this broad consultation, we completed only one round (exploration of issues and ranking in the written consultation procedure and discussion in the stakeholders meeting). Nevertheless, we obtained a clear prioritization of issues which serves as guidance for further actions.

4.3 Setting the (research) agenda: recommendations

Based on this broad consultation, the following recommendation can be made relating to context and the standardization of economic evaluation in the youth sector.

Context

For the Dutch context it is important to realize that as from 2015, the organization and financing of mental health and social care for youth has been placed in the hands of the municipalities. This transition from central to local government was accompanied by serious budget cuts, making the need for economic evaluations crucial. The leading organization of methodological standardization of economic evaluations in (mental) health care for the youth sector has traditionally been the National Health Care Institute (ZiNL), an organization which informs the Ministry of Health regarding the content of the insurance package. Due to this transition, the question should be raised as to who is the lead organization to standardize economic evaluations in the domain of youth? It would be logical that that the municipalities, or the association of municipalities, would have a leading position in this. The question is whether the municipalities are willing to comply with guidelines for economic evaluation in health care which recommend cost-utility analysis, or if they prefer to follow the guidelines for societal cost-benefit analysis coming from other organizations and sectors. These two approaches to economic evaluation have notable differences which should not be disregarded, as they will impact upon the results. Therefore, we strongly recommend that uniform methodology be applied for economic evaluations in the youth sector. Furthermore, policymakers in the municipalities have often not been trained in the field of economic evaluations. As the “using stakeholders” were underrepresented in the consultation procedure, an important first step towards standardization is to perform a needs assessment to explore their views and wishes. Although this exploration is a necessary first step towards standardization, it should be accompanied by schooling / education of the relevant stakeholders in the municipalities and the ‘translation’ of guidelines (EE / SCBA) to their context and understanding. In order to succeed, this should be a joint effort between researchers, practice organizations and policymakers.

Standardization of economic evaluation in the youth sector

Not reinvent the wheel

It is strongly recommended that existing guidelines be complied with, especially regarding perspective and time horizon. Researchers should preferably take a societal perspective, and can additionally address (a) narrow perspective(s). A budget impact analysis can additionally be performed to address the financial streams for specific budget holders. In case of a SCBA, it should be clarified how the costs and benefits fall on different sectors / stakeholders. The time horizon should preferably be lifetime, or at least long enough to capture all downstream costs and outcomes, or differences in costs and outcomes between interventions. A secondary analysis from a municipality perspective with a time horizon up to 18 years could be performed if preferred by municipalities. Additional research is required into the selection of and relation between intermediate outcomes and final outcomes, and prognostic models should be developed clarifying the course of risk factors and conditions over time. In the Netherlands, a rich pool of researchers with knowledge and experience in (model-based) health economic evaluations is available and ready to provide guidance and support.

Normative discussion on the objective of psychosocial care for youth

During the consultation several stakeholders stressed that in order to standardize the economic evaluation in the youth sector, a normative discussion is needed at the outset, to reveal what is the ultimate objective of psychosocial care for youth. This broad consultation has revealed that the focus on health and health-related quality of life is too narrow; this is obvious from the literature review and the stakeholders' comments regarding the inclusion of outcomes beyond health for the youngster, the inclusion of outcomes/benefits to others/other sectors than the youngster, as well as unit of analysis. This normative discussion touches upon many aspects of economic evaluation, such as the target population, type of analysis and the identification/measurement/valuation of both costs and outcomes.

Type of economic evaluation / analysis

Although in this consultation it was suggested that cost-utility analysis should be the default, no clear consensus emerged regarding the choice for either cost-utility analysis (CUA) or social cost-benefit analysis (SCBA). In the (Dutch) health economic community (34) a new discussion has blazed whether the cost-utility analysis is still the preferred type of analysis, as (social) cost benefit analysis (SCBA) is increasingly mentioned as a good alternative. Accordingly, in order to reach standardization a clear

recommendation/standardization on the type of analysis is needed; this should align with the objective of psychosocial care for youth (normative discussion) and be supported by the municipalities.

Both CUA and SCBA bring some important methodological challenges. A CUA will not automatically do justice to the broad range of outcomes of psychosocial interventions for youth, that is, outcomes beyond health in the child, and outcomes in others or other sectors, also known as spill-over effects. In the current guidelines for economic evaluations, it is recommended that the EQ-5D be used for calculating quality-adjusted life years (QALYs). The EQ-5D-Y has recently been developed for use in economic evaluations in the youth sector. Although the EQ-5D focuses on health-related quality of life, a valuation set is not yet available for the EQ-5D-Y; one should be developed. In order to capture outcomes beyond health, broader QoL/wellbeing instruments for youth need to be developed and validated. This should be accompanied by an overview of existing preference-based QoL measures and their psychometric qualities. Furthermore, insight into the concept of QoL/wellbeing in youth, which might be age-dependent, should inform instrument development. A new, preference-based instrument (R-WYS) has been suggested for capturing broader outcomes, but this instrument is currently being validated and is not yet broadly available. QoL in others or from a family perspective is currently best addressed separately in additional (sensitivity) analyses, as no generally accepted methods are yet available to deal with utility interdependence in a QALY framework. In general, including outcomes in others or in other sectors in a utility framework will require methodological work on how to conjointly value these multiple outcomes.

In SCBA, in theory the full range of outcomes (both outcomes beyond health and spill-over effects) should be monetized, as is recommended in the guidelines for social cost-benefit analysis. If adopting SCBA, one should be aware that some of the challenges raised with respect to CUA also apply when performing a SCBA, as the guidelines for SCBA (31) recommend including the QALY (monetized) as the measure of outcome for health, without a critical appraisal of its background and usefulness in particular contexts. Furthermore, for a SCBA, guidance should be provided as to how the full range of broader outcomes (both short- and long-term) should be identified, measured (core set of outcomes) and monetised, to avoid doublecounting.

Cost analysis

Irrespective of whether a CUA or SCBA is performed, several stakeholders stressed, on the costing side, in line with societal perspective, to build on existing instruments in order to cover intersectoral cost and benefits. This implies developing a taxonomy of resource use items for identification purposes and developing a resource use questionnaire for broad use in psychosocial care in the

youth sector, in which these relevant items can be selected. As a starting point, the available framework of Drost (23), TiC-P for youth (27), and the questionnaire Intensive Youth Care (26) can be used. Furthermore, it is recommended that unit prices for interventions and for child (services) resource use be calculated. Finally, existing cost manuals (2, 29) should be extended to capture these specific cost prices.

Evidence-based care for youth

At several instances throughout the consultation, it was stressed that sound evidence should be the basis for care in the youth sector, i.e. there was a strong plea for evidence-based youth care. In order to achieve evidence-based youth care, additional steps have been suggested. Several relate to design issues, as a randomized controlled trial (RCT) is not always attainable in the youth sector. This urges the use/development of alternative designs, instead of the RCT, which should be free of bias as much as possible. A related design issue is that cost-effectiveness based on a selected study population does not always reflect real world cost-effectiveness, urging the necessity to further explore real-world economic evaluation designs in the youth sector. In relation to real-world cost-effectiveness results, the potential use and validity of 'routine outcome measurements' (ROM), databases and registries for economic evaluations was also put forward. Finally, in order to reach evidence-based youth care, more research effort should be put on evidence synthesis and meta-analysis in the youth sector to strengthen the validity of the results of economic evaluations in this sector.

HTA methodology call in the sector youth

This broad consultation revealed that in order to reach standardization of economic evaluation in the youth sector, several issues (see above) have to be solved. In order to stimulate the development and application of uniform methodology, a call for HTA methodology in the youth sector could be considered, similar to the he HTA-methodology program in ZonMw Health Care efficiency research, ZonMw prevention, and ZonMw GGG.

In summary, based on this broad consultation the following point-by-point recommendations can be made.

Context:

1. As the organization and financing of mental health and social care for youth in the Netherlands has been placed in the hands of the **municipalities**, they should be actively **included in further agenda setting**, so that their needs, views, and wishes are included in the standardization of economic evaluation of the youth sector.
2. As the organization and financing of mental health and social care for youth in the Netherlands has been placed in the hands of the municipalities, further standardization should be accompanied by the **schooling / education** of relevant stakeholders at the municipality level and the **translation of these guidelines** to their context and understanding. The association of Dutch municipalities (VNG) can be an important partner in this.

Standardization of economic evaluations in the youth sector:

3. For the standardization of economic evaluation in the youth sector, it is strongly recommended that **existing guidelines be complied with**, especially in regard to perspective and time horizon.
4. For the standardization of economic evaluations in the youth sector, a **normative discussion** among researchers, practice organizations and policymakers (i.e. municipalities) is needed, to reveal what is the ultimate **objective of psychosocial care for youth.** This discussion will inform the type of economic evaluation, as well as the identification, measurement and valuation of outcomes and costs. Based on this discussion, additional methods might have to be developed.
5. Following 4, for the standardization of economic evaluation in the youth sector, it is important to reach **consensus about the preferred type of analysis** - the (social) cost benefit analysis (SCBA) or cost-utility analysis (CUA).
    a. Regarding the CUA, it is specifically recommended:
        - to develop a **valuation set for the EQ-5D-Y** instrument
        - to obtain an **overview of existing preference-based QoL instruments** for youth and their psychometric properties
        - to define the **concept of QoL/wellbeing** in youth, which may be **age-dependent**
        - to develop and validate a self-reported, **generic, preference-based QoL instrument** for youth which captures outcomes beyond health for broad use in economic evaluations in the psychosocial care for youth

- to develop a **core set of outcome measures** to be used in addition to cost-utility analysis (e.g. for use in additional cost-effectiveness analyses)
- to provide **guidance for proxy measurement** for young children
- to develop a **module for youth** which is integrated in the existing guidelines for economic evaluation (covering the above topics, among others)
- if **outcomes in others** than the child, such as family, or even in other sectors are to be included in a cost-utility framework, then **appropriate methodology** for this should be developed/applied.

b. Regarding the SCBA it is specifically recommended:

- to develop a **core set of outcome measures** for use in SCBA (also suitable for use in other types of analyses)
- to provide guidance as to how **(long-term) outcomes should be monetized.**

6. For the standardization of the **cost analysis** in economic evaluations in the youth sector, it is recommended that a **taxonomy of resource use items** for identification purposes be developed, and that **a resource use questionnaire** be adapted /developed, in which these relevant items can be selected, for broad use in psychosocial care in youth. Finally, existing **cost manuals should be extended** to capture these specific cost prices.
7. In order to stimulate **evidence-based youth care**, it is recommended to
    - to use/develop **alternative designs**, instead of the RCT, which should be free of bias as much as possible
    - to explore **real-world economic evaluation designs** in the youth sector
    - to explore the potential use and validity of '**routine outcome measurements' (ROM), databases and registries** for (real-world) economic evaluations
    - to put more research effort on **evidence synthesis and meta-analysis** of the results of economic evaluation in the youth sector.
8. In order to stimulate the development and application of uniform methodology regarding economic evaluation in youth a **call for HTA methodology** in the youth sector could be considered.

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Appendix 1 Mail to the stakeholder (in Dutch)

December 2015

Geachte heer, mevrouw

In vervolg op onze vorige mail, aangaande de 'Brede consultatie in het kader van standaardisatie van economisch evaluatieonderzoek in de jeugdsector', sturen wij u hierbij het consultatiedocument toe.

Het doel van deze consultatie is om te verkennen in hoeverre de huidige (geactualiseerde) richtlijnen, standaarden en handleidingen voldoende toereikend zijn om economische evaluaties in de jeugdsector op een kwalitatief goede manier uit te kunnen voeren.

Wilt u voor uw feedback gebruik maken van het bijgevoegde document “feedback consultatie”.

Dit document “feedback consultatie” bestaat uit 6 onderdelen:

1) Een deel waarin u uw algemene indruk over het consultatiedocument kunt geven;
2) Een deel waarin u aanvullende methodologische uitdagingen/problemen kunt aangeven;
3) Een deel waarin u aanvullende Nederlandse richtlijnen/handleidingen/instrumenten voor economisch evaluatieonderzoek kunt aangeven;
4) Een deel waarin u aanvullende literatuur kunt suggereren, die relevant is voor de scoping review;
5) Een deel waarin wij u vragen om een prioritering aan te brengen in de methodologische uitdagingen/problemen; we vragen u om hier (maximaal) een top tien samen te stellen van de methodologische uitdagingen/problemen;
6) Een deel waarin wij u vragen om mogelijke (procedurele) oplossingsrichtingen voor deze top tien aan te geven

Wij vragen u om hier uiterlijk 25 januari 2016 schriftelijk op te reageren. Via email naar: effectiefjeugd@zonmw.nl

Ter herinnering, op 18 februari 2016 (van 13.00 -16.00 uur) vindt een consultatiebijeenkomst plaats bij ZonMw. Tijdens deze bijeenkomst wordt het consultatiedocument en de reacties daarop besproken. Het doel van deze meeting is tevens om consensus te bereiken over de vraag welke stappen nodig zijn om tot standaardisatie economisch evaluatieonderzoek in de jeugdsector te komen.

Extra vraag voor de consortia: Graag willen we voor de consultatie ook gebruikers van economisch evaluatieonderzoek binnen jeugd benaderen. Graag willen we de al bij de consortia betrokken gebruikers hiervoor benaderen. We denken hierbij aan (koepelorganisaties) praktijkinstellingen (Centrum voor Jeugd en Gezin, GGD, RIAGGs, APZ, Jeugdinstellingen) GGZ Nederland, (koepelorganisaties van) gemeenten, provincies, ministeries van VWS, Justitie, Sociale Zaken en Onderwijs en (koepelorganisaties) onderwijsinstellingen. Daarom vragen we de consortia om in hun netwerk te kijken wie gebruikers zijn die willen meedenken over de standaardisatie van economisch evaluatieonderzoek in de jeugdsector en deze mensen te benaderen om deel te nemen aan de

consultatie. Graag horen we voor 22 januari welke gebruikers we op 18 februari bij de consultatiebijeenkomst zullen zijn.

Graag horen we zo spoedig mogelijk, doch uiterlijk 22 januari 2016 of u en/of uw collega bij deze consultatiemeeting op 18 februari 2016 aanwezig kunt zijn. Via email: effectiefjeugd@zonmw.nl

Met vriendelijke groet,
prettige kerstdagen en een gelukkig nieuwjaar,
mede namens prof. Silvia Evers en prof. Carmen Dirksen,

Valesca Kuling
Programmasecretaris Jeugd

Appendix 2 Feedback consultation document (in Dutch)

Onderdeel 1:

In het consultatiedocument willen we graag uw **titel, voorletters, naam en affiliatie(s)** correct vermelden in tabel 1. Graag in onderstaande tabel uw titel, voorletters, naam en affiliatie(s) noteren, zoals die uiteindelijk dient te worden opgenomen in het consultatiedocument.

Verder kunt u hieronder ook aangeven indien u anoniem wilt blijven.

Titel	
Voorletters	
Naam	
Affiliatie (1)	
Affiliatie (2)	
Affiliatie (3)	
Affiliatie (4)	

0 Graag aankruisen indien u anoniem wilt worden opgenomen in het document

Algemene indruk/opmerkingen	

Onderdeel 2:
Hieronder kunt u uw **algemene indruk en opmerkingen** geven over het consultatiedocument

Onderdeel 3:

Bekijk tabel 2 op pagina 21 en verder

Hieronder kunt u aanvullende methodologische uitdagingen/problemen suggereren

Framing aspect	Additional Problem / issue / challenge

Onderdeel 4:

Bekijk paragraaf 3.4 op pagina 27 en 28

Hieronder kunt u aanvullende Nederlandse richtlijnen/handleidingen/instrumenten voor economisch evaluatieonderzoek suggereren

Aanvullende Nederlandse richtlijnen/handleidingen/instrumenten

Onderdeel 5:

Bekijk referentielijst op pagina 46

Hieronder kunt u aanvullende literatuur suggereren, die relevant is voor de scoping review

Aanvullende literatuur

Onderdeel 6:
In onderstaande tabel vragen we u om (maximaal) een top tien samen te stellen van de belangrijkste uitdagingen/problemen en om daarnaast mogelijke (procedurele) oplossingsrichtingen voor deze top tien aan te geven. We vragen u om hier (maximaal) een top tien samen te stellen van de methodologische uitdagingen/problemen, gebruik hiervoor de nummer zoals gepresenteerd in tabel 2, bijvoorbeeld 1.1 verwijst naar “Perspective” en het probleem van de verdeling van kosten en baten over de verschillende stakeholders/sectoren. In de laatste kolom kunt u een mogelijke oplossingsrichting aangeven. Hierbij kunt u denken aan: aanpassing van de richtlijn/handleidingen (en zo ja welke), aanpassing van bestaand instrumentarium, verdiepingsmodule, suggesties voor aanvullend onderzoek, etc.

Prioritering (1 meest belangrijk en 10 minst belangrijk)	Problem / issue / challenge geef hier het nummer weer, zie tabel 2, pagina 21 en verder	Mogelijke (procedurele) oplossingsrichtingen
1		
2		
3		
4		
5		
6		
7		
8		
9		
10		

Maastricht University

Maastricht UMC+

Trimbos instituut

ZonMw