



What we have learned about health related quality of life (HRQoL) in children...

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This title and not the one in the program represents the content of our work. Children's HRQoL was studied in joined cooperation by the Leiden University Medical Centre and TNO Prevention and Health. [20sec]

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My name is Nicolet Theunissen and I had the pleasure to work with the people mentioned here (in alphabetical order)/. And there were even many more physicians and psychologists involved. I will make an attempt to summarise four years of research in 20 minutes, so wish me good luck [20sec]

Next slide please [6sec]

Main Themes

- HRQoL versus Health Status (HS)
- Measuring HRQoL in children
- Informant of the child's HRQoL
- Is HRQoL health related or not?

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Although the studies we performed served various purposes, four themes arose repeatedly: The difference between HRQoL and HS; Measuring HRQoL in children; the choice of an informant on the child's HRQoL, and the ideas about how much HRQoL is related to health. [25 sec]

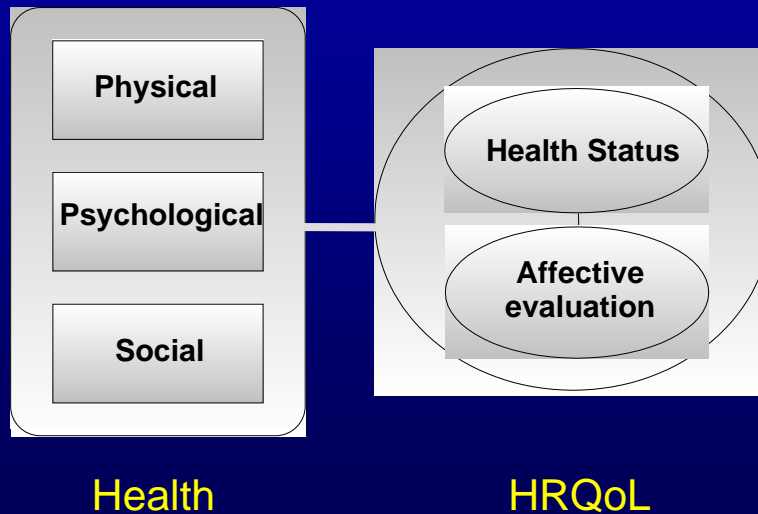
Main Themes

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First, I will discuss the difference between HRQoL and HS. But let me start by introducing the model of HRQoL we used. [10 sec]

A Model on HRQoL



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This slide presents the model we used on children's HRQoL. At the left you see the domains of interest in HRQoL. They are physical-, psychological- and social wellbeing. They all form part of the Child's health, according to the definition of the World Health Organisation.

At the right HRQoL is symbolised. A HRQoL report is composed out of two parts: Firstly, Health Status, which is the assessment by a person of his or her own health functioning. And secondly, affective evaluation of possible problems in Health Status.

This definition of HRQoL incorporates individual and culturally determined differences in coping with HS problems. Moreover, it reflects internal standards about HRQoL. Although these are factors which are emphasised by many authors, in HRQoL research it is not yet common to make this affective addition. [72 sec]

The TACQOL questionnaire

Have you had difficulty running?

never sometimes often
[4]

During this I felt:

(very)good not so well rather bad bad
[3] [2] [1] [0]

- a child form and a parent form
- seven scales of eight items each: 56 items
- physical complaints, motor functioning, autonomy, cognitive functioning, social functioning, positive emotions, negative emotions.

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This is an example of a TACQOL Child Form item from the motor functioning scale. the question is 'Have you had difficulty running?'

'The answers 'never, sometimes or often' measure Health Status. If a HS problem is reported, it leads to a question about the emotional responses: "During this I felt: Very good, not so well, rather bad and bad"

'The TACQOL-Parent Form equivalent of the question is "Has your child had difficulty running?" and "During this my child felt:". No affective responses about positive and negative emotional functioning were assessed, since this would have led to illogical items.

After extensive HOMALS analyses, HS was weighted by the affective evaluation to obtain HS scores. Since the TACQOL provided HRQoL as well as HS scores, we could compare these two constructs.[80]

HRQoL versus HS

- HRQoL results differ from HS results
- relation between HRQoL and physical health \neq relation between HS and physical health
- concerning agreement between informants: HRQoL \neq HS

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Firstly, it was found that sometimes the HRQoL score was better than the HS score, and sometimes poorer. For instance, it was shown in samples from the general population, that only half of the reported HS problems were combined with negative affective responses to those problems.

In a group of children with idiopathic short stature the standard deviations of the mean HS scores were in general somewhat lower than that of the HRQoL scores. This means that the differences between children became larger when the affective evaluations were taken into account.

Secondly, HRQoL and HS differ in their relationship with physical health. For instance, correlations between perinatal factors and QoL motor functioning were lower and less often significant than between perinatal factors and HS motor functioning.

Finally, agreement between informants was sometimes different for HRQoL and HS. For instance, the global judgement of a neonatologist correlated with HS, but not with HRQoL. It was concluded that the neonatologists did not include the affective evaluation of HS problems in her judgement, which the parents did.
[83sec]

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The next theme to be discussed is Measuring HRQoL in children.

Measuring HRQoL in children requirements

- multi-factorial (statistical confirmed and useful)
- subjective
- content relevant for children of a certain age
- self-administered (feasibility may be limited by the child's cognitive skills)
- parallel child and parent form

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Instruments that measure HRQoL in children should be multi-factorial: physical, psychological and social well-being should be covered;

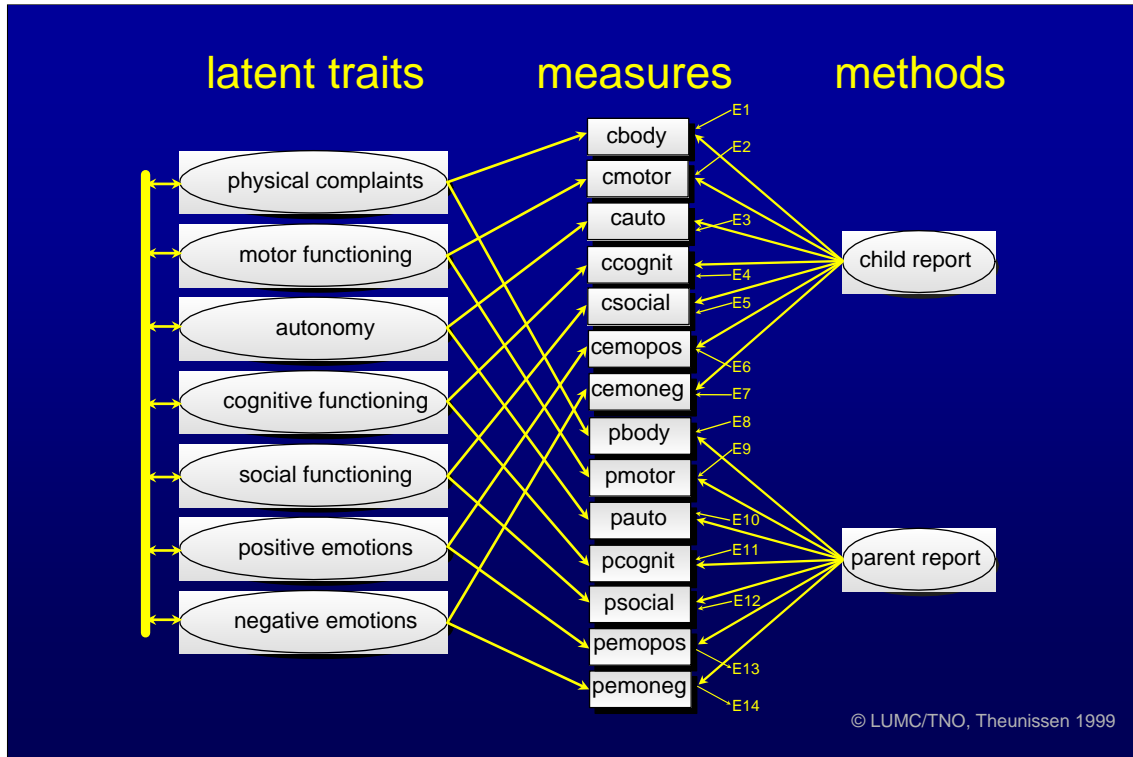
They should assess the subjective opinion of the child's HRQoL;

The content must be relevant for children of a certain age;

The instruments preferably are self-administered.

The feasibility may be limited by the child's cognitive skills, which is one of the reasons why we developed a parallel child and parent form.

I restrict the illustration to the multi-factorial character, which was confirmed by statistical analyses. [60 sec]



Validity was extensively tested by Multi-Train Multi-Method modelling on the TACQOL scores of 8 to 11 year olds and their parents.

The Hays-Hayashi (MTMM) Quality Index was .94, which represents a rather good discriminant validity.

The model presented on this slide was fitted on the data. Seven latent variables for the seven scales were constructed with no restrictions on their correlations and two additional sources of variations: child versus parent reports and measurement error. [35 sec]

Percentage explained variance in MTMM model

Measure	Method	Latent trait	Method	Error
Physical complaints	child	68	8	24
	parent	65	14	21
Motor functioning	child	59	10	30
	parent	67	24	9
Autonomy	child	41	22	37
	parent	38	5	57
Cognitive functioning	child	42	17	40
	parent	54	6	40
Social functioning	child	38	30	32
	parent	39	21	40
Positive emotions	child	55	6	39
	parent	65	2	32
Negative emotions	child	50	5	45
	parent	73	0	26
Total (M%)	child	51	14	35
	parent	57	10	32

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This slide presents the percentage explained variance in the fitted Multitrait- Multimethod model. The model did have a good fit.

Children and parent scores were determined primarily by the latent trait and much less by method and error. Therefore, the validity of both methods seemed to be high.

The multi-factorial structure was not only confirmed by statistical analyses, it appeared useful as well. Children reported different HRQoL at different scales.

For instance, children born very preterm had a low HRQoL in different scales to children with other problems in the neonatal period. Furthermore, children with idiopathic short stature had the same HRQoL as children from a reference group, except for social functioning in which the HRQoL was lower. [55 sec]

Main Themes

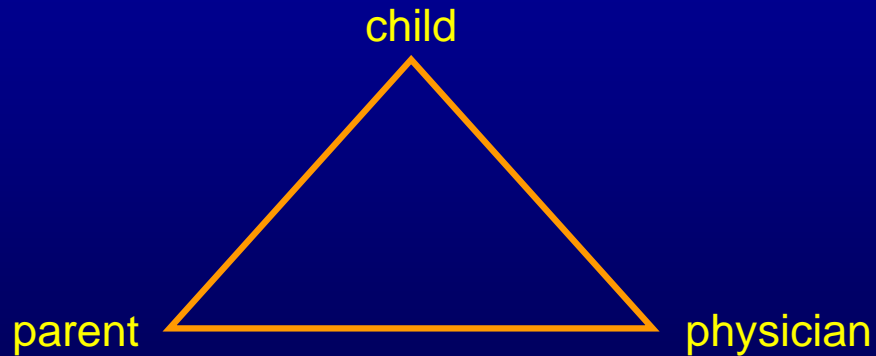
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Although HRQoL aims for the individual's perception, children cannot always serve as informant. They may lack the cognitive skills or could be too ill to fill in questionnaires.

Informant of the child's HRQoL

Key figures

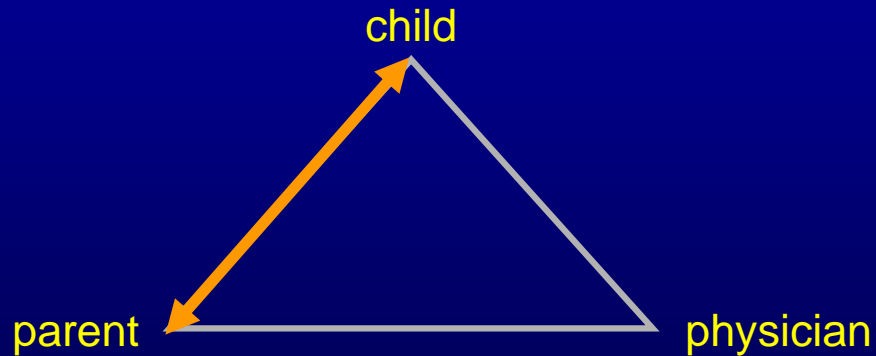


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In that case someone has to act as a proxy, for instance the parent or the physician. Proxies may not have the same knowledge and internal standard as the child him or herself, which can influence their reports. [30sec]

Informant of the child's HRQoL

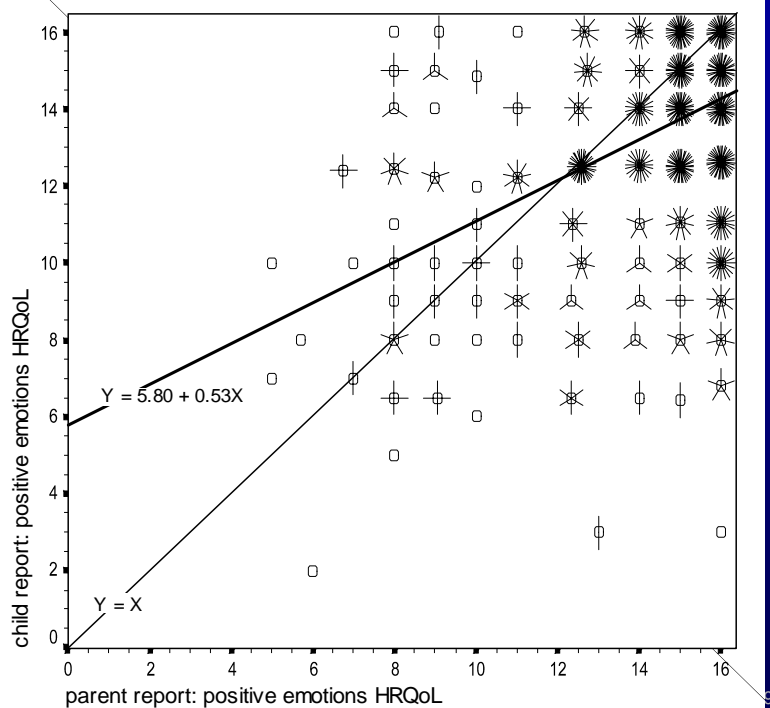
Key figures



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Parent reports were only moderately correlated to child reports. And the mean differences between parent and child at group level depend heavily on the sample studied. Therefore it is not possible to give a formula for 'translating' parent scores into child scores. [10 sec]

Regression
of the child's
pos.emotions
on the
parent's
pos.emotions

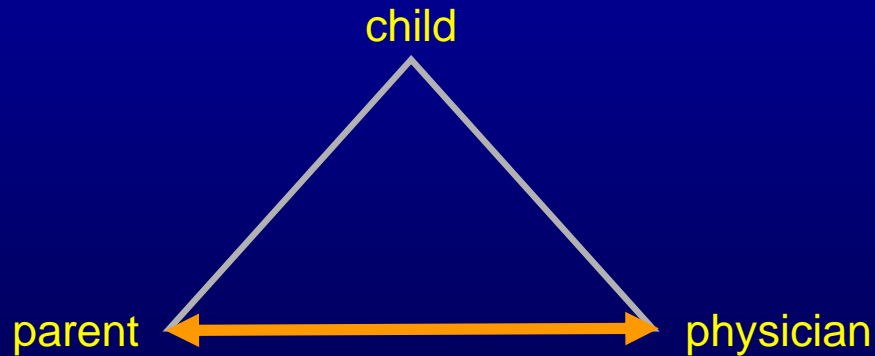


This slide is an example of the relation we found between parent-child agreement and the height of the HRQoL score. The regression on positive emotions is presented here. The x-axis represents the parents scores, the y-axis represents child'scores. The greater the distance is between the regression line and the $Y=X$ line, the lower the agreement is between parents and children.

If child and parent both reported low QoL, a child reported higher than its parent. If child and parent both reported a high QoL, then the child socres were lower that the parent socres. All scales followed the same pattern. Overall, child scores were less extreme than the parent scores. [60sec]

Informant of the child's HRQoL

Key figures

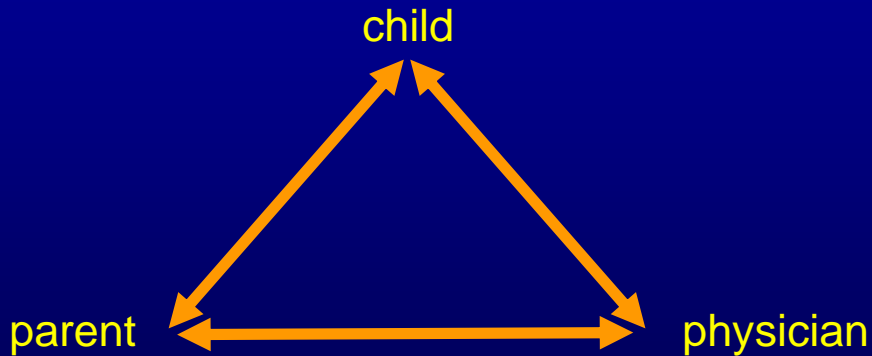


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If a proxy is needed, the parent is the preferable proxy. In a clinical situation, however, the treatment program is established in concordance between physician and parent. We studied differences between parents and neonatologists, concerning the QoL of pre-school children born preterm. We concluded, for instance, that a neonatologist will be surprised that a parent does not want to have full treatment for the child's motor functioning problems. The parent simply does not consider the motor problems to be as serious as the neonatologist does. In reverse, parents evaluated problems with lungs, stomach and sleeping as being more of an emotional burden than the neonatologist does. The parent therefore does not understand why these problem in their child receive less attention from the neonatologist. [55 sec]

Informant of the child's HRQoL

Key figures



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Child, parent and physician's perception on the child's HRQoL was studied in a group of children with idiopathic short stature. Half of the group was treated with growth hormone, the other half acted as a control group. The physician reported an improvement of HRQoL in the children treated with growth hormone, the parents reported no change, whereas according to the children themselves the treatment group had the same or sometimes even poorer HRQoL than the control group. [30 sec]

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We now arrive at the last main theme which is perhaps not restricted to HRQoL in children: is HRQoL health related? [18 sec]

Is HRQoL health related or not?

health related quality of life
implies a relationship with health

- **cross-sectional hypothesis:**
children with a health problem have poorer HRQoL than healthy children
- **longitudinal hypothesis:**
changes in the child's health can result in changes in HRQoL

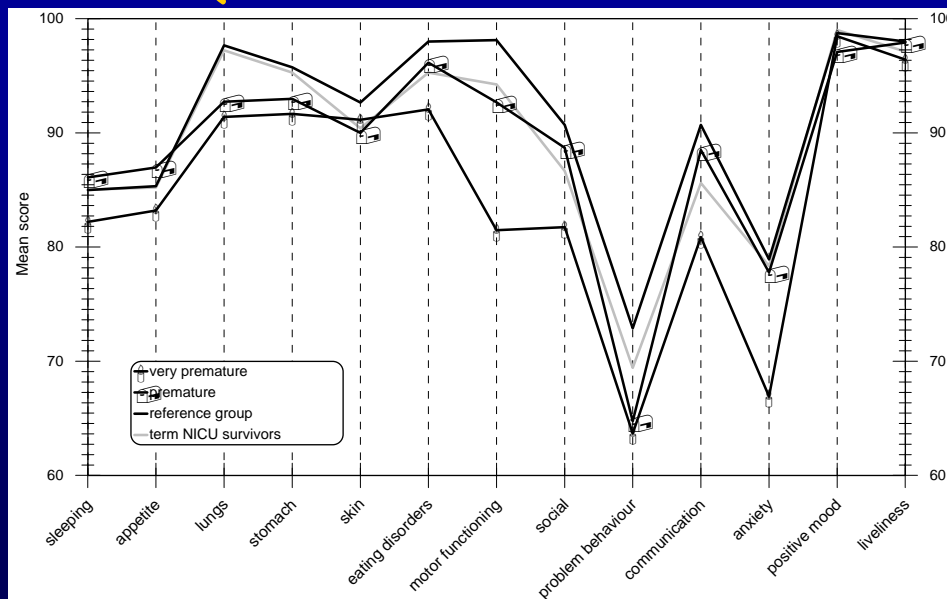
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The term health related quality of life implies a relationship with health. This relation can be studied in a cross-sectional design by testing the hypothesis that children with a health problem had poorer HRQoL than healthy children.

In a longitudinal design the hypothesis could be formulated as: changes in the child's health can result in changes in HRQoL, or improved health will give improved HRQoL. Health is defined as 'physical, mental or social wellbeing. This implies that children with a physical, mental or social health problem could be subject of study.

In our research we limited ourselves to study in children with a physical health problem. [60 sec.]

Is HRQoL health related or not?



We found support for the hypothesis that children with a physical health problem had poorer HRQoL than physically healthy children.

For instance, children born very preterm had lower HRQoL than a health reference group.

The x-axis represents the scales of the TNO-AZL-Preschool QOL questionnaire, of the TAPQOL.

The y-axis represents the scale scores which ranged from 0 to 100, with higher scores representing better HRQoL.

The order of the groups is often as expected: lowest HRQoL in the very preterm, medium in the preterm, and highest in the reference group. Furthermore, the HRQoL of the term survivors of neonatal intensive care (the grey line) is worse than that of the reference group (the top line)

[70 sec.]

Is HRQoL health related or not?

longitudinal research in children with a physical health problem

- **predictability**
maintenance of relative position on particular characteristics over time
- **plasticity approach**
the ability of children to change over time as a result of changes in health

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The goal of a longitudinal study reveals implicit ideas of the investigators about the changeability of QoL. Two main approaches to change can be distinguished in advance: Predictability and plasticity.

Predictability is the maintenance of relative position on particular characteristics over time. This approach is expressed in studies that are conducted, for instance, to identify determinants of QoL, or to evaluate the test-retest reliability of new QoL instruments. This type of research depends on a certain amount of invariability in time, without which QoL cannot be predicted.

Plasticity is the ability of an individual to change in characteristics over time. This approach is expressed in studies that are conducted, for instance, to evaluate the effect of an intervention or treatment on QoL, or to evaluate the responsiveness to change of a new QoL instrument. These research aims emphasise the possibility of QoL to change over time [75 sec]

Is HRQoL health related or not?

predictability approach in children with a physical health problem

- **stability (no change):**
 - personality traits
 - adaptation
- **continuity (consistency in relative rank over time):**
 - cognitive development
 - developmental stages or tasks

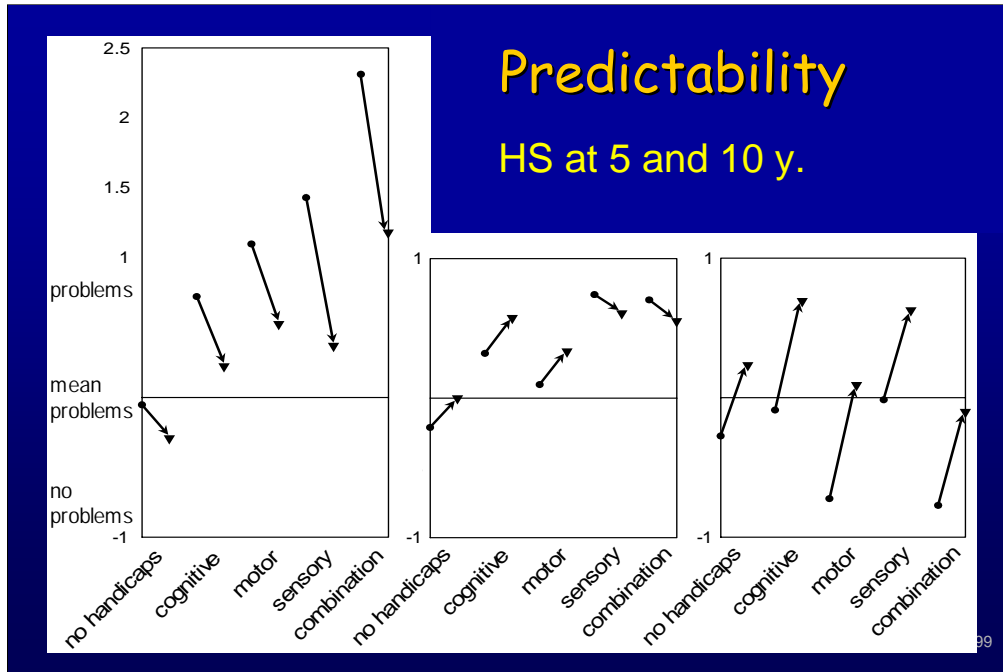
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Evidence obtained from studies on adults suggests that QoL is quite stable anyway, and often does not reflect changes in life. Stability in QoL is influenced by personality traits and the human capacity to adapt to changes in life circumstances.

The effects of the child's age and level of development probably interfere with stability but can still imply continuity, the consistency in relative rank over time.

The level of cognitive development influences the ability of the child to read and understand the QoL questions, to recall the relevant information and to formulate the answer. More important it influences the child's concepts of health and illness.

The specific impact of a medical situation also varies with age. For example, hair loss associated with chemotherapy of childhood cancer, may be especially disturbing during adolescence and less during childhood. [85 sec]



This slide represents an example of continuity effect. It involved a longitudinal study in children born preterm between 5 and 10 years of age.

The most remarkable was the decrease in basic functioning problems in children with more than one handicap, as can be seen on this slide. We concluded that the changes were mainly age-related instead of health-related.

The normal development of children at a certain age was reflected in the type of problems that the parents reported. Basic functioning is important from early childhood onwards. At 'kindergarten age', concentration problems become more important. When children grow older they start to communicate more about their moods. As a result, problems in basic functioning decreased while negative moods increased, and concentration problems increased slightly. In this study changing parental standards appeared to be the motor of change, and not physical health. [95 sec]

Is HRQoL health related or not?

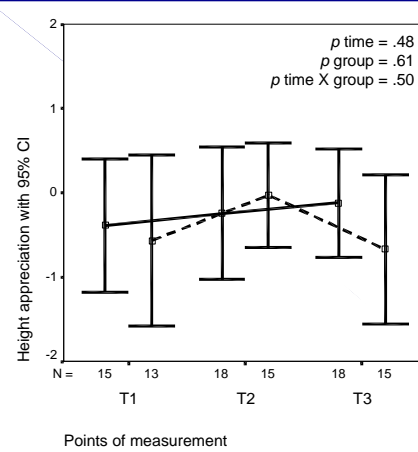
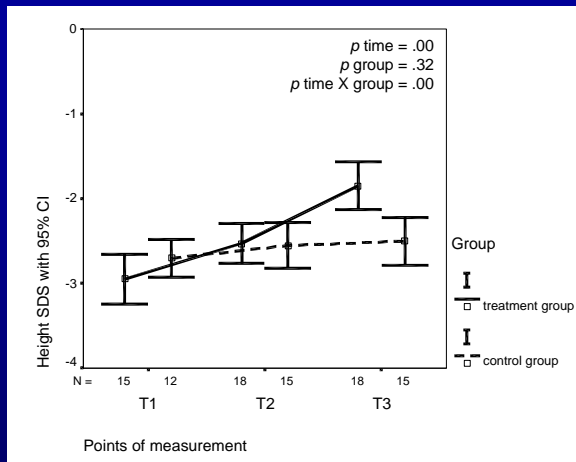
plasticity approach in children with a physical health problem

- **biomedical model:**
changes in the child's **physical health** can result in changes in HRQoL
- **biopsychosocial model:**
changes in the child's **physical, psychological or social health** can result in changes in HRQoL

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The plasticity of changeability of QoL can be approached with two models: A biomedical model, which states that changes in the child's physical health can result in changes in HRQoL. And a biopsychosocial model, that states that changes in the child's physical, psychological or social health can result in changes in HRQoL. [30 sec]

Plasticity height SDS versus height appreciation



This slide gives an example of plasticity that need a biopsychosocial model. We studied the HRQoL of children with idiopathic short stature (ISS), which is short stature without an underlying disease. By treating these children with growth hormone, it is not their physical health which is changed but rather their physical appearance. It was hoped that increasing height would give more age appropriate reactions to the children, which in turn would improve their HRQoL. In other words, not physical health but psychological or social health was object of study.

It appeared that, although height improved in the treatment group only, the appreciation of height did not differ between groups. This is and indication of a psychological phenomenon called coping, the ability to adjust to difficult situations.

Changes in HRQoL of these children did hardly relate to growth. Instead, changes in some HRQoL scales were related tot the height appreciation by the child her/him self. [80 sec]

What we've learned about HRQoL...

conclusions

- children as well as parents distinguish between HS and HRQoL
- scientific tools are available to measure HRQoL in children
- parents and physicians have limited insight into the way a child perceives his/her HRQoL
- HRQoL is related to physical, psychological and social health

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Four main themes in HRQoL were discussed and four main conclusions can be drawn out of it.

- children as well as parents distinguish between HS and HRQoL

-scientific tools are available to measure HRQoL in children

-parents and physicians have limited insight into the way a child perceives his/her HRQoL

-HRQoL is related to physical, psychological and social health

[30 sec]

What we've learned about HRQoL... recommendations

- incorporate an emotional evaluation of HS problems in HRQoL
- use a valid and reliable generic HRQoL instrument: they are available!
- obtain child's, parent's and physician's HRQoL evaluations whenever possible
- do not limit HRQoL studies to changes in physical health

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And these four main conclusions coincides with four main recommendations:

-incorporate an emotional evaluation of HS problems in HRQoL

-use a valid and reliable generic HRQoL instrument: they are available!

-obtain child's, parent's and physician's HRQoL evaluations whenever possible

-do not limit HRQoL studies to changes in physical health.

I think we have learned a lot about HrQoL in children and hopefully we are right in our conclusions. If children are right about their perceptions of their QoL is maybe less important, because ...[50 sec]

People may not be right about themselves,
but their self-evaluations are the ones
that most powerfully affect their future behavior.

Byrnes, J.F. (1984) *The Psychology of Religion*, New York: The Free Press.

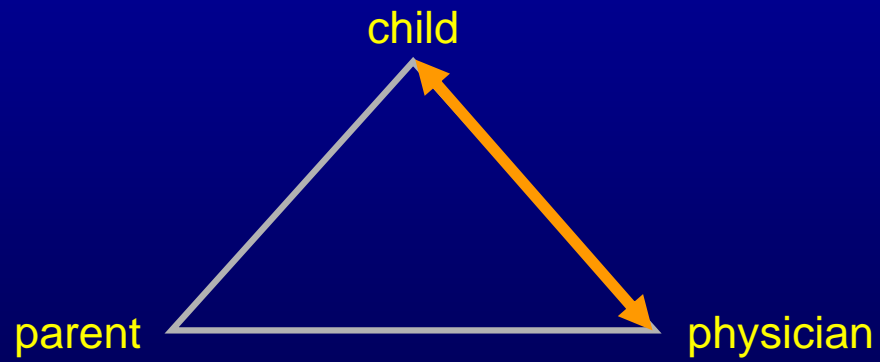
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People may not be right about themselves,
but their self-evaluations are the ones
that most powerfully affect their future behavior.

Thank you for your attention [10 sec]

Informant of the child's HRQoL

Key figures



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