Summary
Background

The use of patient reported outcomes (PROs) in routine clinical practice is becoming increasingly common. PROs are answers to questionnaires based on direct reporting by patients without the intervention of an observer. Health-related quality of life (HRQOL) assessment is a form of PRO which often includes physical, emotional, social and cognitive functioning of the patient. By using PROs in clinical practice, the HRQOL of patients can be systematically monitored. PRO studies are usually conducted in adult patients and the results are promising. Thus far however, there has been little attention for PROs in paediatric practice. This is remarkable since it is known that children with a (chronic) disease are at risk for suffering from HRQOL problems and that physicians are not always aware of these problems.

Children with cancer can also experience HRQOL problems. PRO tools could bridge the gap between child and paediatric oncologist and serve as an aid in identifying and discussing HRQOL problems. Additionally, PRO tools may contribute to effective patient–physician communication, which is of crucial importance to psycho-social outcomes in children with cancer.

This thesis covers the Quality of Life In Childhood ONcology (QLIC-ON) study. The main aim of the QLIC-ON study was to investigate the effectiveness of an intervention that provides a PRO about HRQOL (the QLIC-ON PROfile, p. 173) to the paediatric oncologist.

More information about the background of the QLIC-ON study is given in the general introduction (chapter 1) of this thesis. It elaborates on childhood cancer, HRQOL and psychosocial functioning of children with cancer. A summary of research findings regarding the effectiveness of PROs in clinical practice is also provided. The study aims, study design and intervention are described in this chapter as well.

Methods

A sequential cohort study design was applied in the QLIC-ON study. Paediatric oncologists participated in the control (March 2006 to January 2008) as well as the intervention period (January 2008 to November 2009); children with cancer (0-18 years, shortly after completion of treatment) and their parents participated in either the control period or the intervention period. First, a control period was introduced, in which 99 children or parents (if the child was younger than 8 years of age) completed a HRQOL questionnaire on a standalone computer while waiting in the out-patient clinic prior to three consecutive consultations with the paediatric oncologist. Answers on the HRQOL questionnaire were summarized in the QLIC-ON PROfile. In the control period the QLIC-ON PROfile was not provided to the paediatric oncologist, the child or the parent. The paediatric oncologist started the intervention period – with a training - only as soon as he had seen all patients
participating in the control period (to prevent contamination bias). The intervention period consisted of a group of 94 children or parents filling out the HRQOL questionnaire. This time however, the QLIC-ON PROfile was provided to the paediatric oncologist, the child and the parent, to be discussed during the three consultations. All participants completed (evaluating) questionnaires after each consultation; there was also a baseline and follow-up assessment. In addition, the consultations were audio taped. The effectiveness of the QLIC-ON PROfile was assessed with the outcome measures communication about HRQOL domains, identification of HRQOL problems, the child’s HRQOL, the amount of psychosocial referrals and satisfaction with communication.

Results

Before presenting the main results of the QLIC-ON study, this thesis first reports on four related studies.

To begin with, the reference data of the Dutch version of the PedsQL are described in chapter 2. The PedsQL is a widely used generic paediatric HRQOL measure (which was applied as a PRO in the QLIC-ON study). It was therefore important to investigate the PedsQL within the general Dutch population. For 496 children (5 to 18 years) PedsQL reference data were collected. The psychometric properties of the PedsQL proved to be adequate and it can therefore be used as a HRQOL instrument in paediatric research in the Netherlands.

Secondly, chapter 3 demonstrates that the HRQOL of children with cancer (0 to 18 years, N=191) shortly after completion of treatment can be impaired compared with the norm. This is also the case for parents of children with cancer aged 0 to 7 years. HRQOL was assessed by means of the ITQOL (generic, proxy-report, 0 to 4 years), the CHQ PF 50 (generic, proxy-report, 5 to 7 years), the Kidscreen (generic, self-report, 8 to 18 years) and the Disabkids (chronic generic, self-report, 8 to 18 years). The findings in this chapter underline the importance of systematically monitoring HRQOL in paediatric oncology, not only during but also after end of treatment and not only in children but also in parents.

Thirdly, 24 paediatric oncologists were interviewed for the purpose of the QLIC-ON study in chapter 4. Data were qualitatively analyzed by means of a framework analysis. In general, paediatric oncologists believed that it was their task to discuss psychosocial functioning, to identify psychosocial problems and to provide emotional support to children with cancer. However, they also indicated that this task is limited due to e.g. their inherent lack of expertise, lack of time or by the physician’s personality. It is concluded that paediatric oncologists feel responsible for providing psychosocial care to the best of their ability.

Fourthly, an extensive description of the development and implementation of the QLIC-ON intervention as well as the rationale behind the study design and the content of the training is provided in chapter 5. This chapter can be used as an initial guide for
researchers and other professionals, who are interested in setting up PRO interventions in clinical practice.

The main results of the QLIC-ON study are reported in chapter 6. They demonstrate that the QLIC-ON PROfile increased discussion of emotional and psychosocial functioning without lengthening the consultation duration. Additionally, more emotional problems were identified in the intervention group compared to the control group e.g. problems related to anger, fear, and sadness. Furthermore, the QLIC-ON PROfile improved HRQOL of patients 5 to 7 years of age with respect to self-esteem, family activities, and psychosocial functioning. However, the QLIC-ON PROfile had no effect on the amount of psychosocial referrals. Neither did the QLIC-ON PROfile improve parent and physician satisfaction regarding communication, possibly because this was already high. Overall, it is concluded that a PRO is a useful tool for systematic monitoring HRQOL of children with cancer.

Finally, the findings of a qualitative analysis of one third (N=155) of the audio recorded consultations are presented in chapter 7. It appeared that the QLIC-ON PROfile did not change the psychosocial content of communication. The nature of the psychosocial topics in the intervention group was equal to the nature of the psychosocial topics in the control group. However, the discussion of psychosocial topics increased in the intervention group compared to the control group. Thus, pediatric oncologists already address psychosocial issues, regardless of the use of a PRO. However, with a PRO available they address these issues more systematically and more often.

Discussion

The findings presented in this thesis are discussed in the general discussion (chapter 8). The findings lead to three key messages:

- PROs in clinical practice increase identification and discussion of psychosocial problems
- Successful use of the QLIC-ON PROfile in clinical practice: making the right choices
- Pediatric oncologists feel responsible for the identification of psychosocial problems

Chapter 8 reflects on these three key messages. Limitations in study design, logistics, and study population are described as well. Additionally, the general discussion elaborates on the clinical implication to implement PROs in routine clinical practice, incorporating the use of internet, during all treatment stages of childhood cancer. It is also advised to implement PROs within other pediatric populations. The chapter ends with future perspectives that focus on PRO implementation research (instead of investigating effectiveness), a PRO for parents of children with a (chronic) disease, and developing a PRO screening tool (in addition to the current monitoring tool).
Conclusion

The QLIC-ON study is the first study to investigate the effectiveness of PROs about HRQOL in paediatric oncology practice. It concerned a multicentre project with a large sample size. The PRO intervention was carefully developed, using innovative methods with respect to the design and the use of the PRO. Positive effects were found for communication and identification of psychosocial problems. PROs are recommended to be implemented in paediatric oncology practice.