CHAPTER 7

EXPLORING THE RESPONSE SHIFT PHENOMENON IN CHILDHOOD CANCER PATIENTS AND ITS EFFECT ON HEALTH-RELATED QUALITY OF LIFE

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ABSTRACT

Purpose/Objectives: To explore the response shift phenomenon in childhood cancer patients and to determine its effects on the ratings of health-related quality of life (HRQOL).

Design: Retrospective pretest-posttest design.

Setting: Pediatric oncology department in the northern part of the Netherlands.

Sample: 37 children newly diagnosed with cancer and 80 parents.

Methods: The then-test method was used to determine response shift. HRQOL was assessed within two weeks after diagnosis (pretest) and three months later (post-test) using both child- and parent report of PedsQL and Cantril’s ladder. The post-test and then-test were administered concurrently.

Main research variables: Overall and multidimensional HRQOL.

Findings: Scores on Cantril’s then-test were lower than the pretest in both child and parent reports, indicating response shift in the assessment of overall HRQOL. Children experienced a greater response shift than parents. No differences were found between the PedsQL then- and pretests.

Conclusions: Both child- and parent-report ratings of overall HRQOL were affected by response shift, resulting in an underestimation of the improvement in overall HRQOL between diagnosis and three months post-diagnosis. No response shift was demonstrated in the more specific domains of HRQOL (PedsQL).

Implications for nursing: Knowledge of the response shift phenomenon helps nurses to better interpret the outcomes of HRQOL. The use of the PedsQL instruments is recommended in future studies that aim to demonstrate changes in HRQOL.
INTRODUCTION

The Health Related Quality of Life (HRQOL) is important for understanding the impact of cancer on a child’s life. HRQOL is defined as “a multi-dimensional construct that includes physical, social and emotional functioning of the child, measured from the perspective of both the child and his/her family, and sensitive to the changes that occur throughout development.” To determine deterioration or improvement in HRQOL during treatment, the reference point for good or poor HRQOL should be the same at all measurement points. However, because the measurement of HRQOL relies on self-report, the rating depends on an individual’s perception of HRQOL at the time of measurement. This perception might change over time. Being confronted with cancer, for instance, can change an individual’s perception about good and poor HRQOL due to adaptation to the imperfect health status. Consequently, individuals may report good levels of HRQOL despite deterioration in their health status, and in contrast to what nurses expect. Several studies have demonstrated that cancer patients reported levels of HRQOL similar to healthy persons. In childhood cancer patients, psycho-social HRQOL was often found to be even higher than in healthy children. This phenomenon of adaptation to a change in health status is called response shift. The response shift phenomenon has been found in adult cancer patients. However, research on the influence of response shift on HRQOL ratings in childhood cancer patients is lacking. Response shift is defined as “a change in the meaning of one’s self-evaluation of a target construct as a result of changes in internal standards (scale recalibration), values (reprioritization), or a redefinition of the target construct (reconceptualization).”

Although response shift is a natural way to cope with and adapt to changes in health status, it generates a bias in consecutive measurements of HRQOL. As a result, treatment effects on the child’s HRQOL can be reduced or inflated. Moreover, when studying a nursing intervention aiming to improve HRQOL the outcome might be underestimated when the endpoint parameter is subject to response shift.

Because the HRQOL of children is preferably measured using both child and parent report, both ratings might be influenced by response shift. Proxy-measurement is considered to be less sensitive to response shift, because proxy-raters do not experience the imperfect health status themselves. However, whether this is true for parents of critically ill children is unknown because childhood cancer has a major impact on the parents as well.
In this study we will assess the impact of response shift on the assessment of HRQOL in childhood cancer at the level of overall impression of HRQOL, because this level is known to be sensitive to response shift in adults, and second, at the more specific level of the domains of HRQOL. This level, consisting of the physical, social and emotional domains of HRQOL, is most commonly used in HRQOL assessment. This study aims: (1) to determine the change in health status and HRQOL during the first three months after diagnosis; (2) to examine whether the HRQOL ratings are affected by response shift; and (3) to determine similarities and differences with regard to the presence, direction, and magnitude of response shift between child- and parent-report ratings.

**METHODS**

**Participants**
Data were collected as part of a prospective cohort study of newly diagnosed cancer patients at the University Medical Center Groningen (UMCG) in the Netherlands. Ethical approval was gained from the Medical Ethics Committee of the UMCG. Between September 2007 and December 2009, all patients aged 2-18 years consecutively admitted to the Pediatric Oncology Department were asked to participate. Exclusion criteria were: having insufficient command of the Dutch language or being in a palliative phase of treatment. The response shift study was restricted to children aged 8 years and older and their parents for child- and parent-report. Parents of children between 2-7 years old were included for parent report only. A total of 121 parents and 61 children were eligible and were invited to participate. Fifty-one children and 100 parents gave informed consent. Response rate was 83.6% for child report and 83% for parent report. Reasons for declining child report were: too burdensome (n=6) and lack of motivation (n=4), and for parent report: too burdensome (n=17) and lack of motivation (n=4). After inclusion, 7 children dropped out (too burdensome (n=2), lack of motivation (n=3), or being too ill (n=2)), and the data of another 7 children were incomplete. Fifteen parents dropped out (too burdensome (n=5), lack of motivation (n=8), child is too ill (n=2)), and the data of 5 parents were incomplete. Finally, 37 child-parent pairs completed all measurements as well as 43 parents of younger children. Only complete data sets including all measurements were used for analysis.
**Procedure**

The most widely used method to examine response shift is the then-test method, also known as the retrospective pretest-posttest design method.\(^{14,18,19}\) In this method, individuals are asked to evaluate their HRQOL at the pretest and the post-test. Immediately following the post-test, the then-test is administered; whereby individuals are instructed to reassess their pretest HRQOL. They are not asked to remember their pretest rating, but to retrospectively give a renewed judgment about their HRQOL at the pretest. Because the then-test is completed at the same time as the post-test, respondents would likely use the same internal standards. A difference between the then-test and the pretest provides evidence of recalibration of HRQOL between pre- and post-test measurement. In this study, HRQOL was assessed within two weeks after diagnosis (pretest) and three months later (post-test). For administering the then-test, parents received written instructions to take the first week after diagnosis in mind when filling out the then-test. The researcher asked children to recall the period shortly after diagnosis and to name special events from that period. When children indicated they could vividly remember this period, the then-test was administered by interview.

**Measures**

Health status was assessed by the Lansky Play Performance scale (PPS),\(^{20}\) a 10-point parent-rated Likert-scale recording the daily play activity of the child ranging from fully active (100), to completely disabled, not even passive play (10). The PPS has adequate reliability (mother versus father ratings, \(r = 0.71\)) and content validity (parent versus nurses ratings, \(r = 0.75\)).\(^{20}\) The Memorial Symptom Assessment Scale (MSAS)\(^{21,22}\) was used for reporting the number of symptoms children experienced. The MSAS is a child- and parent-rated instrument consisting of 30 of the most common symptoms experienced during cancer treatment and has demonstrated reliability (\(r = 0.83-0.87\)) and validity (high correlations with other symptom instruments and higher symptom number among patients who had recent chemotherapy) in childhood cancer populations.\(^{21}\) Crohnbach alpha in the present sample was \(r = 0.80\) and 0.75 for child-reported pre- and post-test and \(r = 0.85\) and 0.83 for parent-reported pre- and post-test, respectively.

HRQOL was assessed by means of Cantril’s ladder and PedsQL instruments. Cantril’s ladder\(^{23}\) is a single item Visual Analog Scale rating the overall impression of Quality of Life on a scale of 0-10, where 10 represents the best possible quality of life and 0 the worst possible quality of life. Cantril’s
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ladder was used as pre-, post-, and then-test. Although Cantril’s ladder has frequently been used in many studies, no data for reliability and validity have been reported.

The PedsQL 4.0 Generic Core Scale\(^{24,25}\) is a 23-item multidimensional scale designed to measure HRQOL in children and adolescents aged 2-18 years old. The PedsQL Cancer Module\(^{26}\) is a 27-item scale developed to measure pediatric cancer specific HRQOL. The PedsQL instruments are comprised of parallel child self-report formats (ages 5 and older) and parent proxy-report formats (ages 2 and older) and have high levels of internal consistency ranging from \(r = 0.72-0.88\) for child report and \(r = 0.86-0.90\) for parent report. Validity was demonstrated using the known-groups method; the PedsQL instruments distinguished between healthy children and children with cancer.\(^{24-27}\) For the then-test a selection of 14 items from both PedsQL instruments (noted as adjusted PedsQL) was used to diminish the burden of filling out the entire HRQOL measures twice at the time of the post-test. Items were selected according to the criteria mentioned by Schwartz and Sprangers.\(^{14}\) The authors selected items that might be remembered well, like pain or fear of injections. Next, items expected to be prevalent shortly after diagnosis were selected, covering the domains of HRQOL. The sum scores of the adjusted PedsQL (aPedsQL) were used for the comparison of pre-, post-, and then-test ratings. Because some domains of HRQOL are more sensitive to recalibration response shift than others,\(^{17}\) the domains of the aPedsQL were analyzed separately as well. The authors divided the aPedsQL into domains according to the categorization of the original instruments (see Figure 1). For ease of interpretability, the items of the 5-point Likert scale were reversed and converted to a 0-100 scale according to standard procedures so that higher scores indicated better HRQOL. The internal consistency of the aPedsQL was satisfactory (Cronbach alpha \(r = 0.74, 0.77, \) and \(0.78\) for child report pre-, post-, and then-test; and \(r = 0.78, 0.87, \) and \(0.76\) for parent report pre-, post-, and then-test, respectively). The aPedsQL was representative for the PedsQL instruments, with \(R^2 = 0.86\) for child report and \(R^2 = 0.88\) for parent report.

**Analyses**

Changes in HRQOL were examined by comparing the pre- and post-test scores (reported change) and then- and post-test scores (adapted change). To determine whether response shift had occurred, then-tests scores were compared with pretest scores. As the assumptions for normalcy were not met, all comparisons were analyzed using Wilcoxon Signed-Rank Test with an
alpha level of 0.05. To test the relationship between child- and parent-report Spearman’s correlation coefficient was used.

**RESULTS**

*Change in health status and HRQOL over time*

Thirty-seven children (8-17 years), their parents, and 43 parents of children 2-7 years old (80 parents in total) participated in the study (see Table 1). The median parent-reported Play Performance Scale (PPS) values increased ($Z = -3.54, p < .001$), indicating that the children’s health status improved over the period shortly after diagnosis till three months later. Furthermore, both children and parents reported less symptoms on the Memorial Symptom Assessment Scale (MSAS) at the post-test than at the pretest (child report $Z = 3.23, p < .001$; parent report $Z = 4.46, p < .001$, see Table 2).

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**Physical Functioning**
1. I had low energy.
2. I hurt or ached.
3. Food did not taste very good to me.
4. I became sick to my stomach when I thought about medical treatments.

**Emotional Functioning**
5. I felt afraid or scared.
6. I worried about what would happen to me.
7. I felt angry.
8. I felt sad and blue.
9. I got scared about having needle sticks.

**Cognitive Functioning**
10. It was hard for me to pay attention to things.
11. I forgot things.

**Social Functioning**
12. I had problems getting along with other kids.

**Communication**
13. It was hard for me to ask the doctors and nurses questions.
14. It was hard for me to explain my illness to other people.

Note. Items 3, 4, 9, 13, and 14 are from the PedsQL cancer module. All other items are from PedsQL 4.0 Generic Core Scale.
Table 1. Patient characteristics.

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Child-report ((n=37))</th>
<th>Parent-report ((n=80^a))</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Median</td>
<td>Range</td>
</tr>
<tr>
<td>Age of children (years)</td>
<td>14</td>
<td>(8-17)</td>
</tr>
<tr>
<td>Gender of children</td>
<td>n (%)</td>
<td>n (%)</td>
</tr>
<tr>
<td>Female</td>
<td>20 (54.1)</td>
<td>44 (55)</td>
</tr>
<tr>
<td>Diagnosis</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hematological</td>
<td>12 (32.4)</td>
<td>36 (45)</td>
</tr>
<tr>
<td>Solid tumors</td>
<td>18 (48.6)</td>
<td>26 (32.5)</td>
</tr>
<tr>
<td>Brain tumors</td>
<td>7 (18.9)</td>
<td>18 (22.5)</td>
</tr>
</tbody>
</table>

\(^a\)Both parents of 37 children \(\geq 8\) years and of 43 children 2-7 years old.

Table 2. Change in scores between pre-tests and post-tests (reported change) and between post-tests and then-tests (adapted change).

<table>
<thead>
<tr>
<th>Variable</th>
<th>Pretest</th>
<th>Post-test</th>
<th>Then-test</th>
<th>Reported change</th>
<th>Adapted change</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mdn</td>
<td>IQR</td>
<td>Mdn</td>
<td>IQR</td>
<td>Mdn</td>
</tr>
<tr>
<td>HRQL</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Child-report</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Cantril</td>
<td>7</td>
<td>2</td>
<td>8</td>
<td>2</td>
<td>5***3</td>
</tr>
<tr>
<td>aPedsQL</td>
<td>71.43</td>
<td>19.64</td>
<td>76.79</td>
<td>24.11</td>
<td>73.21</td>
</tr>
<tr>
<td>Parent-report</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Cantril</td>
<td>6</td>
<td>2</td>
<td>7</td>
<td>2</td>
<td>5*3</td>
</tr>
<tr>
<td>aPedsQL</td>
<td>60.71</td>
<td>25.30</td>
<td>73.21</td>
<td>23.96</td>
<td>60.71</td>
</tr>
<tr>
<td>Health status</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Child-report</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>MSAS</td>
<td>11</td>
<td>9</td>
<td>7</td>
<td>10</td>
<td>.001</td>
</tr>
<tr>
<td>Parent-report</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>MSAS</td>
<td>13</td>
<td>9</td>
<td>9</td>
<td>9</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>PPS</td>
<td>60</td>
<td>30</td>
<td>80</td>
<td>30</td>
<td>&lt;.001</td>
</tr>
</tbody>
</table>

Note. Cantril scores range from 0-10; higher scores indicate higher HRQL. aPedsQL (= adjusted PedsQL) scores range from 0-100; higher scores indicate higher HRQL. MSAS (= Memorial Symptom Assessment Scale) scores represent the frequency of symptoms and range from 0-30. PPS (= Lansky Play Performance Scale) scores range from 0-100; higher scores indicate a higher level of play or daily activities. Mdn = median; IQR = inter quartile range. * Wilcoxon Signed-Rank Test \(^p<.05\), *** \(^p<.001\) Wilcoxon Signed-Rank Test then-test versus pre-test.
The response shift phenomenon in HRQOL

The median child report ratings of aPedsQL improved (Z = -2.81, \( p < .001 \)). However, the increase of the ratings on Cantril’s ladder were not significant (Z = -1.23, \( p = .219 \)). The parent-reported scores demonstrated improved HRQOL for both aPedsQL (Z = -4.33, \( p = .028 \)) and Cantril’s ladder (Z = -3.10, \( p < .001 \)). In keeping with the improved health status, HRQOL ratings improved as well.

**Response shift**

**Overall HRQOL**

To determine the influence of response shift on the child- and parent-report ratings, the scores of the then-tests and pretests were compared. Wilcoxon Signed-Rank Tests indicated that both children and parents rated the overall HRQOL (Cantril’s ladder) retrospectively lower than at the pretest (see Table 2, Figure 2). The median value of the child report pretest was 7 and of the then-test 5 (Z = -4.40, \( p < .001 \)). The median value of the parent report pretest was 6 and of the then-test 5 (Z = -2.52, \( p = .012 \)). In the response shift literature this difference is interpreted as an overestimation of overall HRQOL at the pretest. The improvement in overall HRQOL according to the then-test versus post-test design (adapted change) was significantly greater than based on the pretest versus post-test design both for child- and parent report (Z = -3.90, \( p < .001 \); Z = -4.17, \( p < .001 \)) (see Table 2, Figure 2).

**Domains of HRQOL**

The aPedsQL then-test of both child report and parent report did not differ from the pretest (Z = -0.57, \( p = .572 \) for child report, Z = -0.08, \( p = .935 \) for parent report, see Table 2); thus indicating no response shift for the aPedsQL. Separate analyses of the domains of the aPedsQL demonstrated the largest difference between pre- and then-test in the domain emotional functioning, namely 75.00 at pretest and 69.32 at then-test. However, this difference was not significant (Z = -1.74, \( p = .081 \)). Therefore, no response shift could be confirmed for emotional functioning. Then-test ratings and pretest ratings for the other child report domains and for all parent report domains showed no differences.

These results demonstrate that only the ratings of Cantril’s ladder were affected by response shift, resulting in an underestimation of the extent of improvement in overall HRQOL between diagnosis and three months post-diagnosis. The aPedsQL ratings were not affected.
Table 3. Comparison child- and parent-report ratings (n=37).

<table>
<thead>
<tr>
<th>Variable</th>
<th>Child report</th>
<th>Parent report</th>
<th>Differencea</th>
<th>Correlationb</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mdn</td>
<td>IQR</td>
<td>Mdn</td>
<td>IQR</td>
</tr>
<tr>
<td>Cantril’s ladder</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Pretest</td>
<td>7</td>
<td>2</td>
<td>6</td>
<td>3</td>
</tr>
<tr>
<td>Post-test</td>
<td>8</td>
<td>2</td>
<td>7</td>
<td>2</td>
</tr>
<tr>
<td>Then-test</td>
<td>5</td>
<td>3</td>
<td>5</td>
<td>3</td>
</tr>
<tr>
<td>Response shift</td>
<td>-2</td>
<td>3</td>
<td>-1</td>
<td>4</td>
</tr>
<tr>
<td>PedsQL</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Pretest</td>
<td>71.43</td>
<td>19.64</td>
<td>59.62</td>
<td>26.79</td>
</tr>
<tr>
<td>Post-test</td>
<td>76.79</td>
<td>24.11</td>
<td>69.64</td>
<td>25.00</td>
</tr>
</tbody>
</table>

Note. Mdn = median; IQR = inter quartile range
a Wilcoxon Signed-Rank Test
b Spearman’s rho
** p < .01

Figure 2. Response shift of overall HRQL of a) child report ratings (n=37) and b) parent report ratings (n=80). Then-test ratings are lower than pretest ratings both for child report (p<.001) and parent report (p<.05). The improvement in overall HRQL according to then- and post-test design is larger than the improvement according to pre- and post-test design (p<.001 both for child- and parent-report). Child report response shift a) is larger than parent report response shift b) (p = .001).
Note. Median values are presented.
The response shift phenomenon in HRQOL

**Similarities and differences between child- and parent-ratings**

For this section, the data of 37 child- and parent-reports were compared pairwise. The pre- and post-test ratings of child- and parent-report of both Cantril’s ladder and aPedsQL were positively related: Spearman’s rho varied between 0.48 and 0.70 (see Table 3). Parent-reported ratings of HRQOL were lower than child-reported ratings both for Cantril’s ladder and for aPedsQL at pre- and post-test.

Because no response shift was found for the PedsQL, further analyses were performed for Cantril’s ladder only. The child- and parent-report then-test and response shift of Cantril’s ladder were positively related (see Table 3). The Cantril’s then-tests did not differ between child- and parent-report. Although at the pretest children and parents rated the overall HRQOL differently, in retrospect their perception of overall HRQOL shortly after diagnosis was nevertheless the same. Comparison of the magnitude of the Cantril’s response shift revealed a scale recalibration of -2 points in child report and -1 point in parent report (see Figure 2). The effect-size for child report was -0.74, for parent report -0.30. Children over-estimated the pretest more than parents did ($Z = -3.27, p = .001$).

**DISCUSSION**

This is the first study to explore response shift in the assessment of HRQOL in childhood cancer patients. Our findings are threefold. First, in keeping with an improved health status, HRQOL improved within three months after diagnosis. Second, both child- and parent-report ratings of overall HRQOL were affected by response shift, while the more specific domains of HRQOL were insensitive to response shift. Third, children experienced a greater response shift than parents did. These three results will be discussed in detail below.

1. Because the health status improved within three months after diagnosis, an improved HRQOL was to be expected. However, one measure did not show up significantly. The change in overall HRQOL (Cantril’s ladder) was underestimated due to response shift. After taking response shift into account, the improvement of overall HRQOL was profound for both child- and parent-report. That the children’s health status and HRQOL improved so quickly in
the three months after diagnosis is a positive finding, despite the fact that they were still in treatment. Only a few studies have assessed HRQOL in this early phase of treatment, and reported either an improvement,\textsuperscript{28-30} or no changes in HRQOL.\textsuperscript{31} Unfortunately, these studies used different time intervals which make a comparison with our results difficult.

2. The finding of response shift for overall HRQOL is consistent with previous research in adult patients.\textsuperscript{9,15,32,33} It corresponds with the then-test hypothesis in the literature stating that adjustment to an improved health status may lead to lower then-test than pretest ratings.\textsuperscript{7,19} The pretest overestimation of HRQOL might be explained by children’s and parents’ coping style to be positive despite the severe illness.\textsuperscript{34,35} The fact that the aPedsQL seems to be unresponsive to response shift can be explained by its concreteness. Concrete items are known to be less sensitive to response shift compared to broad domains like overall HRQOL,\textsuperscript{15} because a concrete item offers less room for personal interpretation. Other studies using HRQOL instruments at overall and domain level found comparable results.\textsuperscript{7,31} The only exception in our findings was the aPedsQL domain of emotional functioning which showed the largest difference between pre- and then-test. However, due to the small sample size statistical significance could not be confirmed. Further research is needed to demonstrate whether child report of emotional functioning is sensitive to response shift. Because the aPedsQL was very representative for the PedsQL instruments, we believe comparable results would have been found when using the total PedsQL. Therefore, for future studies that aim to demonstrate changes in HRQOL the use of the PedsQL instruments is recommended; whereas the use of a global measure like Cantril’s ladder is not advisable.

The finding of response shift in parent report is congruent with a study examining response shift in children with middle ear infection. While the health of these children had improved six weeks after surgery, parents rated their child’s HRQOL at the then-test more negative than at the pretest.\textsuperscript{36} An improved health status resulted in a shift in internal standards when parents realised that the initial HRQOL was worse than perceived at the moment itself. Although recall-bias might explain the differences between pre- and then-test ratings, some facts argue against recall-bias. A study among stroke patients that researched the influence of memory, for instance, found that those with good memory reported the largest response shift.\textsuperscript{15} In another study the memory ratings of the pretest turned out to be very similar to the pretest itself, while the then-test ratings differed significantly.\textsuperscript{18} Furthermore, it is unlikely that recall-bias would influence the ratings of only the overall HRQOL and not
of both measures of HRQOL. The fact that the then- and pretest ratings of the PedsQL were the same indicates that child and parent were perfectly able to remember the child’s condition shortly after diagnosis. Research has demonstrated that children aged 8 years and older could handle a 4-week recall period accurately. In measures assessing life events a 3 months period resulted in valid and reliable outcomes. Moreover, because being diagnosed with cancer is such an overwhelming and daunting experience, we believe children and parents have a vivid memory of this period even after 3 months, thus diminishing recall-bias. Nevertheless, further research on the reliability of recalling a period of 3 month ago is desirable. Another risk factor that should be taken into consideration when using self-report measures is reporting bias. Children and parents might have rated the then-test lower than the pretest, because they feel the situation of the child should have improved as a result of the intensive treatment.

3. We investigated similarities and differences in child and parent reports. Consistent with other studies in childhood cancer patients, parents rated the HRQOL of their children lower than the children themselves did, whereas child- and parent-ratings were moderately to strongly and positively related. The effect-size of the child- or parent-reported response shift differed considerably. According to Cohen’s criteria, the effect-size of parent report was small and comparable with mean effect-sizes of overall HRQOL demonstrated in adult patients, while the child report effect-size was large. Hence, the child report of overall HRQOL was more severely biased by response shift than the parent report ratings. The difference in effect-size might be explained by the fact that parents more often have substantial information about the disease and treatment than their child, and are more aware of possible complications and risks. This information and the uncertainty about the prognosis result in lower parent-reported than child-reported ratings of the child’s HRQOL at the pretest. Furthermore, children seem to have different response styles than their parents. Children provide more extreme scores and base their judgment on one single example; whereas parents try to give a more balanced rating.

In contrast to the pretest ratings, the then-test ratings of overall HRQOL of children and parents were the same. They had a similar perception of the past HRQOL, probably because they shared the same experience and because the then-test ratings were not affected by adaptation. Comparable results were found in a study among chronically ill patients in which, contrary to the pretest ratings, the patients’ then-test ratings corresponded to the proxy-ratings.
Limitations

Some limitations of this study should be noted. First, the final sample consisted of motivated respondents who felt able to participate in the study. Although this phenomenon is not uncommon in survey research, this means that the final sample was not entirely representative of the total population of childhood cancer patients. Another point to acknowledge is that patients had different diagnoses and hence underwent different treatment regimens. However, we believe that neither the heterogeneity of the sample nor the non-response affected the magnitude or direction of response shift, because response shift concerns differences within subjects and not differences between subjects. It would be interesting to test this hypothesis in future research. Second, given the large variation in HRQOL scores, the number of included children was too small to demonstrate changes in overall HRQOL or in the separate domains of HRQOL. Nevertheless, the number was adequate to determine response shift. Third, although the adjusted PedsQL was found to be representative of the PedsQL instruments, it has only been tested in this study. Replication in larger samples is warranted for further validation.

In this study we found an overestimation of overall HRQOL at the pretest. However, it is not known whether overall HRQOL at the post-test was overestimated as well. As a way of coping with the severe illness, children and parents might tend to present the child’s quality of life as more positive than it actually is. Unfortunately, performing a then-test at three months after diagnosis is difficult because this time point is difficult to mark. Additional research is needed on coping mechanisms such as repressive adaptation, wishful thinking, and social comparison in childhood cancer patients so as to provide more insight in the mechanisms responsible for response shift. Another important discussion point is which measurement represents HRQOL the best: the actual pre- and post-test, or the retrospective then-test. The authors believe an actual test is preferable. However, researchers should be aware that some self-report measures, including Cantril’s ladder, are sensitive to response shift and that measurements of change are biased and may lead to incorrect conclusions. This study demonstrated that the PedsQL instruments, which are frequently used in the assessment of HRQOL in children, were less sensitive to response shift than Cantril’s ladder and were able to determine unbiased changes in HRQOL. Because the sample size was relatively small, additional research is warranted to determine how insensitive the PedsQL measures are in larger samples.

In summary, the improvement in overall HRQOL between diagnosis and three
months post-diagnosis was underestimated by response shift. No response shift was demonstrated in the more specific domains of HRQOL. Therefore, the use of the PedsQL is recommended in studies that aim to demonstrate changes in HRQOL.

**Implications for nursing practice**

Nurses can learn from the current study, that child- and parent-reported ratings of HRQOL can be biased by response shift. As a result, two or more consecutive measurements are not comparable anymore and it becomes difficult to determine, for instance, the impact of treatment or the effect of a nursing intervention. One might wrongly conclude that severity of treatment has no impact on the child’s HRQOL or that a nursing intervention does not contribute to HRQOL. Also other subjective scales measuring pain, fatigue, or nausea could be sensitive to response shift. These measures are frequently used in nursing practice and knowledge of response shift helps nurses to better interpret the outcomes of such measures. Besides, the phenomenon of response shift offers an explanation for the high ratings of HRQOL despite severe illness. To surprise of nurses and other health care professionals, children diagnosed with cancer are very positive and optimistic, despite the child being severely ill and experiencing many side effects. They rate their HRQOL higher than nurses would expect. Apparently, they adapt very well to the severe illness. Knowledge of the phenomenon response shift helps nurses to understand and interpret these outcomes.
Chapter 7

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