Anorectal malformations and Hirschsprung disease
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Chapter 4
Quality of life and anxiety in parents of children with an Anorectal Malformation or Hirschsprung disease: The first year after diagnosis

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Abstract

Introduction
In 2012, we started the KLANKbord-study. A QoL study that follows patients with an anorectal malformation or Hirschsprung disease and their parents from diagnosis till the age of 18 years. We hypothesized that the diagnosis of an anorectal malformation or Hirschsprung disease initially has a negative influence on QoL and anxiety levels of parents, but that this influence will diminish over time. The aim of this study is to see whether QoL and anxiety levels of parents change within the first year after the diagnosis.

Methods
Parents of all children born with an anorectal malformation or Hirschsprung disease, were eligible for this study. Within 3 months after the diagnosis, parents received a set of validated QoL questionnaires (measurement 1). Measurement 2 was 12 months after the first questionnaire.

Main Results
During measurement 1 mothers (n=20) scored significantly higher on the social (p value: 0.01; 95% CI 0.3946 – 3.1528) and environmental domain (p value: 0.01; 95% CI 0.4449 – 2.2851) of the World Health Organisation Quality of Life-BREF (WHOQOL-BREF) compared with the known reference values. Fathers (n=19) scored significantly higher on the physical (p value: 0.01; 95% CI 0.2964 – 1.8072), psychological (p value: 0.001; 95% CI 0.7697 – 2.4757), and environmental domain (p value: 0.003; 95% CI 0.5586 – 2.4214) than the reference values. Comparison of all domains of the WHOQOL-BREF for mothers and fathers between measurements did not show a significant difference. Anxiety levels of mothers were lower during measurement 2 compared with measurement 1. Anxiety levels of fathers were higher during measurement 2 compared with measurement 1. These differences are not significant. Anxiety levels of mothers were significantly higher than anxiety levels of fathers during measurement 1 (p value: 0.002; 95% CI 0.808 – 2.956). During measurement 2 this difference in anxiety of mothers versus fathers did not exist (p value: 0.373 ; 95% CI-1.157 – 2.922).

Conclusion
A negative influence on the QoL of parents having a child with an anorectal malformation or Hirschsprung disease, compared with the reference population, was not seen in this population. QoL did not change significantly during the first year for both fathers and mothers. Anxiety levels of mothers did decline during this first year. The number of parents included in this study is still small, which might influence our results. Therefore, we will continue the KLANKbord-study indefinitely.
Quality of life and anxiety in parents: the first year after diagnosis

Introduction

In the Netherlands, approximately 80 children are born each year with either an anorectal malformation or Hirschsprung disease. Both diseases are relatively common congenital anomalies in pediatric surgery. These anomalies are mostly treated within the first months after the birth of the child. The medical treatment often consists surgery. Parents sometimes have to perform interventions before and/or after their child’s surgery, for example, rectal washouts multiple times a day or anal dilation. The diagnosis, the surgery, as well as the interventions that have to be performed in a daily routine can be of influence on the parents. The mental health status of parents can be influenced when they are confronted with a medical condition of their child.

We postulate that both the anxiety and the QoL of parents of a child diagnosed with an anorectal malformation or Hirschsprung disease can be impaired.

In recent literature QoL of parents, of a child diagnosed with an anorectal malformation or Hirschsprung disease has never been investigated. One study amongst parents of children with anorectal malformations showed that parents do have difficulties in coping with the implications of the disorder and express a need for support. Anxiety of parents has been investigated by Funakosi et al. They describe that the anxiety of the mothers corresponds with the age of the child. Anxiety is more experienced when the child is younger.

QoL is a multidimensional concept. In literature many different definitions can be found. The World Health Organization Quality of Life Group has defined QoL as “an individual’s perception of his/her position in life in the context of the culture and value systems in which he/she lives and in relation to his/her goals, expectations, standards, and concerns”. In other words, QoL is one’s evaluation of his/her functioning in a wide range of areas. Thus, QoL is subjective and refers to satisfaction. Since QoL refers to (dis)satisfaction with functioning it can vary over time depending on life events or due to coping strategies. While parenting a child with a chronic disease, such as an anorectal malformation or Hirschsprung disease, coping strategies may lead to changes in QoL. Worrying about diagnosis, surgery and other aspects of the treatment of your child will change over time.

This is why we started a prospective longitudinal multicentre study in the Netherlands: the KLANKbord-study (a Dutch acronym for QoL analysis of children diagnosed with an anorectal malformation or Hirschsprung disease and their parents). This study investigates QoL of children born with an anorectal malformation or Hirschsprung disease and their parents.
This study is the first analysis of the anxiety and QoL of parents of patients with an anorectal malformation or Hirschsprung disease. Our hypothesis is that both anxiety and QoL change during the first year after the diagnosis.

**Material and Methods**

**Patients**

Parents of all children, born in one of the three participating pediatric surgical centres, with an anorectal malformation or Hirschsprung disease, were eligible for this study. One centre started inclusion in January 2012, one started in July 2012 and one started in November 2012. Within 3 months, after the diagnosis anorectal malformation or Hirschsprung disease was confirmed, parents were asked to participate in the study and received a set of validated QoL questionnaires. After one year, measurement 2 took place. In this study we intended to investigate whether the diagnosis had an effect on the anxiety and QoL reported by parents briefly after the diagnosis and after one year.

Exclusion criteria were the inability to speak the Dutch or English language, and not (yet) completing a follow-up questionnaire one year after the first questionnaire. The study was approved by the institutional ethics review board of all the hospitals.

**Questionnaires**

Parents completed 2 questionnaires, the WHOQOL-BREF (26 items) and the Spielberger State-Trait Anxiety Inventory (STAI-trait 10 items, STAI-state 6 items). These questionnaires are validated for the normal population and for parents of children with a chronic disease. However, they are not specifically validated yet for parents of children with an anorectal malformation or Hirschsprung disease.7,8

The WHOQOL-BREF is a QoL assessment developed by the WHOQOL group. This questionnaire has 26 items in four different domains and a general QoL facet. The domains are physical health, psychological health, social relationships, and environment. Every domain focuses on different facets of life. For instance, the physical health domain focuses on activities in daily life, pain, energy, sleep, mobility, medication, and work. The response scales are 5-point Likert scales. In general, a higher score represents a good QoL.9

The Spielberger State-Trait Anxiety Inventory (STAI) is an instrument for measuring anxiety in adults. The STAI was used to assess state anxiety and the personality characteristic trait anxiety. State anxiety is a momentary emotional condition characterized
by subjective feelings of apprehension and tension, and heightened autonomic nervous system activity. Trait anxiety concerns differences in individuals in the disposition to respond to stressful situations, such as having a child with an anorectal malformation or Hirschsprung disease, with varying amounts of stress. The trait anxiety is only measured once per person. The original and short versions of the STAI-trait and STAI-state were used. The psychometric properties of both versions are well established and considered good.10-13

**Statistical analysis**

All continuous variables are presented as means ± standard deviation or median values with interquartile ranges, and analyzed using either the independent $t$-test with 95% confidence intervals or paired sampled $t$-test. We used the paired sampled $t$-test to compare the different domains of the WHOQOL-BREF of the parents with each other and over the course of time, and to compare the state and trait anxiety. The one sample $t$-test was used to compare the domains of the WHOQOL-BREF with reference values. Statistical significance was declared at the 0.05 level. All data management and statistical analyses were performed using IBM SPSS Statistics, version 19 (SPSS Inc., Chicago, Illinois, United States).

**Results**

From January 2012 to December 2014, 68 patients were eligible for the study. Parents of 7 children were excluded because of the inability to speak Dutch or English. Twenty parents decided not to participate in the study after receiving the questionnaires. Thus, in total 41 parents were included in our study. Out of which 21 had not (yet) completed the questionnaire at one year follow-up. This means that 20 parents were included in this first analysis.

First we compared the four different domains of the WHOQOL-BREF (physical, psychological, social and environmental) with the reference values of the general population. Mothers scored significantly higher on the social ($p$ value: .014; 95% CI 0.3946 – 3.1528) and environmental domain ($p$ value: .006; 95% CI 0.4449 – 2.2851) during measurement 1.
During measurement 2 the mothers scored higher on the psychological ($p$ value: .023; 95% CI 0.1969 – 2.3152) and the environmental domain ($p$ value .024; 95% CI 0.1386 – 1.7914) compared with the reference values (Table 1).

Fathers scored significantly higher on the physical ($p$ value: .009; 95% CI 0.2964 – 1.8072), psychological ($p$ value: .001; 95% CI 0.7697 – 2.4757) and environmental domain ($p$ value: .003; 95% CI 0.5586 – 2.4214) than reference values during measurement 1. During measurement 2 the physical domain was not significant anymore (Table 2).

When we compare measurement 1 with measurement 2 for mothers, they score lower on the social and environmental domain and higher on the physical, psychological and overall domain after one year (measurement 2). Fathers scored higher on the psychological and overall domain during measurement 2. These differences between the measurements were not significant for both fathers and mothers.

We also performed an analysis to measure QoL between fathers and mothers during the first measurement and after 1 year. Results can be found in table 3. Mothers scored lower than fathers on the physical domain, and higher on the social domain.
Trait anxiety of mothers was significantly higher than trait anxiety of fathers (p value: .012 ; 95% CI 0.606 – 4.283). State anxiety of mothers was lower during measurement 2 compared with measurement 1 (10.60 vs 10.95). Anxiety levels of fathers were higher during measurement 2 compared to measurement 1 (9.76 vs 9.06). Anxiety of mothers was significantly higher than anxiety levels of fathers during measurement 1 (p value: 0.002 ; 95% CI 0.808 – 2.956). During measurement 2 this difference in anxiety of mothers versus fathers did not exist (p value: 0.373 ; 95% CI-1,157 – 2.922) (Table 4).

### Table 3: WHOQOL-BREF mothers versus fathers

<table>
<thead>
<tr>
<th>Domain WHOQOL-BREF</th>
<th>Mothers (n=20)</th>
<th>Fathers (n=20)</th>
<th>p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physical (mm 1)</td>
<td>15.60</td>
<td>16.91</td>
<td>0.021</td>
</tr>
<tr>
<td>Psychological (mm1)</td>
<td>15.37</td>
<td>16.00</td>
<td>0.187*</td>
</tr>
<tr>
<td>Social (mm1)</td>
<td>16.80</td>
<td>15.67</td>
<td>0.009</td>
</tr>
<tr>
<td>Environment (mm1)</td>
<td>17.13</td>
<td>17.25</td>
<td>0.733*</td>
</tr>
<tr>
<td>Overall (mm1)</td>
<td>8.70</td>
<td>8.65</td>
<td>0.834*</td>
</tr>
<tr>
<td>Physical (mm2)</td>
<td>17.20</td>
<td>16.33</td>
<td>0.501*</td>
</tr>
<tr>
<td>Psychological (mm2)</td>
<td>15.54</td>
<td>16.28</td>
<td>0.237*</td>
</tr>
<tr>
<td>Social (mm2)</td>
<td>15.86</td>
<td>15.16</td>
<td>0.205*</td>
</tr>
<tr>
<td>Environment (mm2)</td>
<td>16.71</td>
<td>17.05</td>
<td>0.385*</td>
</tr>
<tr>
<td>Overall (mm2)</td>
<td>8.84</td>
<td>10.58</td>
<td>0.418*</td>
</tr>
</tbody>
</table>

**Abbreviations:** mm, measurement; ref, reference; WHOQOL-BREF, World Health Organisation Quality of Life-BREF

\*The values are non-significant

### Table 4: Anxiety measured with STAI-trait and STAI-state

<table>
<thead>
<tr>
<th></th>
<th>Mothers</th>
<th>Fathers</th>
<th>p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>STAI-Trait</td>
<td>16.39</td>
<td>13.94</td>
<td>0.012</td>
</tr>
<tr>
<td>STAI-State (mm1)</td>
<td>10.95</td>
<td>9.06</td>
<td>0.002</td>
</tr>
<tr>
<td>STAI-State (mm2)</td>
<td>10.60</td>
<td>9.76</td>
<td>0.373*</td>
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</table>

**Measurement 1**

<table>
<thead>
<tr>
<th></th>
<th>Measurement 2</th>
<th>p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mothers</td>
<td>10.95</td>
<td>10.60</td>
</tr>
<tr>
<td>Fathers</td>
<td>9.06</td>
<td>9.76</td>
</tr>
</tbody>
</table>

**Abbreviations:** mm, measurement; ref, reference; WHOQOL-BREF, World Health Organisation Quality of Life-BREF

\*The values are non-significant
Discussion

This study shows that the QoL of parents of children with an anorectal malformation or Hirschsprung disease is not negatively influenced by this diagnosis during the first year of the child’s life. We do see that mothers are more anxious than fathers in the first months after the diagnosis was made, but this difference cannot be measured anymore after a year.

The social and the environmental domain of mothers are even better than those of our reference population during measurement 1. This could be the result of the attention that parents, and especially mothers, receive from their friends and family after birth. Friends and family are a big support when receiving bad news, or when being diagnosed with a disease. This opinion is supported by the fact that the social domain is no longer significantly better compared to the reference population during measurement 2. Fathers scored significantly higher on all domains except the social domain shortly after the birth of their child, compared to the reference population. The reference values available for the WHOQOL-BREF are from a mixed population of men and women, which might be of influence on our results.

When comparing parents with each other we see that mothers are significantly less satisfied about their physical functioning than fathers are during measurement 1. This may be a result of giving birth and the lack of sleep during these first months. Mothers are more positive about their social life than fathers are. After a year fathers and mothers do not score different anymore on any of the domains of our QoL questionnaire. Some trends are seen in our results, but the study population is too small to draw firm conclusions.

The trait anxiety of mothers is higher than fathers, meaning that mothers have a more anxious character than their partners. State anxiety, that is the momentary emotional condition, also differs between fathers and mothers during measurement 1. We do not see this difference anymore after 1 year. If the state anxiety was influenced by the trait anxiety, we would not expect the state anxiety levels for fathers and mothers to be the same after a year. Instead the difference would sustain over time. This means that the diagnosis anorectal malformation or Hirschsprung disease does have an influence on state anxiety of parents.

In an earlier cohort study of our study group, parents of newborns compared to parents of older children (10-13 years), we have noticed that mothers of newborns diagnosed with an anorectal malformation or Hirschsprung disease scored significantly worse on state anxiety than fathers did. This difference in anxiety was no longer present in the group of parents of older children.\textsuperscript{14} This study shows that 1 year after birth the state
anxiety is not different anymore between fathers and mothers. This means that parents were able to cope with their anxiety within a year after the diagnosis anorectal malformation or Hirschsprung disease was made. We are anxious to know whether this is the result of coping, or that the children function better after several months.

Studies show that parents of premature children or children with a disease have high levels of anxiety at time of diagnosis and birth. A study analysing parents of children with cancer demonstrated that mothers were the most anxious family member immediately after diagnosis. A year later a decrease in state anxiety was reported. Programs focussing on psychological interventions and counselling of parents showed a positive influence on anxiety. Subjects that underwent these psychological interventions and counselling were characterised by a better use of problem-focused coping strategies.

A limitation of our study is that numbers of parents included are still small. Therefore we will continue our study and hope to have more solid data in the future. Then it may be possible to conclude more about the mechanisms of QoL and anxiety in the parents of patients with an anorectal malformation or Hirschsprung disease.

In conclusion, a significant negative influence on the QoL of parents having a child with an anorectal malformation or Hirschsprung disease, compared with the reference population was not seen in this population. In addition QoL did not change significantly during the first year for both fathers and mothers. Anxiety levels of mothers however, were high immediately after diagnosis, but diminished within the first year.

It may be worthwhile to develop psychological interventions for those parents whose anxiety levels do not return to reference levels.
Chapter 4

Reference List