Anorectal malformations and Hirschsprung disease

Functional and psychosocial challenges

Witvliet, M.J.

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Chapter 3
Anxiety and quality of life of parents with children diagnosed with an Anorectal Malformation or Hirschsprung Disease

Marieke J Witvliet
Christine Sleeboom
Justin R de Jong
Anton H van Dijk
Sander Zwaveling
Alida FW van der Steeg

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Chapter 3

Abstract

Aim
The aim of this study was to investigate the difference in anxiety and QoL between mothers and fathers of children with an anorectal malformation or Hirschsprung disease. In addition, anxiety of parents of newborns was compared with parents of children in the age-group of 10 to 13 years.

Methods
Parents of 44 patients with an anorectal malformation or Hirschsprung disease completed the World Health Organisation Quality of Life (WHOQOL)-BREF (26 items) and the Spielberger State-Trait Anxiety Inventory (STAI-trait 10 items, STAI-state 6 items). They were divided into two groups according to their children’s age (0-4 years and 10-13 years).

Results
Overall mothers scored significantly worse than fathers on state anxiety (p = 0.005). Trait anxiety was not significantly different between mothers and fathers. The psychological domain of the WHOQOL-BREF was significantly different between parents (p = 0.016), with mothers scoring worse. Dividing the group in newborns and school going children, mothers of newborns showed significantly more state anxiety compared with fathers of newborns (p = 0.016). In the group of older children, both anxiety and QoL were not significantly different between fathers and mothers. Comparing mothers of newborns with mothers of older children, the first group scored higher on state anxiety, but this difference was not significant (p = 0.138; 95% CI – 0.535 – 3.717).

Conclusions
Mothers of newborns have a higher level of anxiety than fathers of newborns with an anorectal malformation or Hirschsprung disease. When children with an anorectal malformation or Hirschsprung disease become older this difference in anxiety between parents is not significant anymore. This may be explained by coping strategies.
Introduction

Anorectal malformations and Hirschsprung disease are relatively common congenital anomalies in pediatric surgery. The incidence of anorectal malformations reported worldwide is between 1 in 1000 and 1 in 5000 live births.\(^1\) The incidence of Hirschsprung disease is approximately 1 in 5000 to 1 in 10000 live births.\(^2\) This leads to a total of approximately 80 children born in the Netherlands each year with either an anorectal malformation or Hirschsprung disease.

In 2005, an international classification system for anorectal malformations was reached by consensus. This classification system does not focus on anatomical or embryological features. It divides anorectal malformations in “major clinical groups” and “rare / regional variants”. This system is based on frequency of occurrence and allows management outcomes to be measured.\(^3\)

In Hirschsprung disease a colonic segment lacks intrinsic nerves (ganglion cells). The affected segments lead to mechanical obstruction and severe constipation. The surgical treatment of anorectal malformations and Hirschsprung disease is well defined and described. After surgery, often seen problems are constipation, soiling and fecal incontinence. These problems can influence QoL of children and their families. Anorectal malformations and Hirschsprung disease can thus be of influence on the entire social system of the child.

Hartman et al. have described that patients with an anorectal malformation or Hirschsprung disease believe that they are psychosocially impaired compared with their healthy peers, and they have less positive emotions. During the second measurement after 3 years the “good” patients showed an improvement in health-related functioning, while the “bad” patients scored even worse.\(^3,6\)

Having your newborn child diagnosed with an anorectal malformation or Hirschsprung disease can cause fears, stress and worrying for parents. Anxiety of parents with a child diagnosed with an anorectal malformation or Hirschsprung disease has only been investigated by Funakosi et al. He investigated the psychological status of Japanese children and their mothers, using the Spielberger State-Trait Anxiety Inventory (STAI) and the Zung Self-rating Depression Scale (SDS). The mothers of preschool children showed higher levels of anxiety and depression than mothers of school-aged children.\(^7\)

We believe that higher levels on anxiety of parents can be of influence on the child. This has not been investigated for children suffering from an anorectal malformation or Hirschsprung disease, but has been studied in children with functional constipation.
Especially the influence of parent-child interaction on the clinical problems has been studied in children with functional constipation. A study performed in the Netherlands by van Dijk et al. showed that children of parents with high self-pity scores and high scores on overprotection have more fecal incontinence than parents with low self-pity scores. They conclude that parenting issues should be incorporated in the treatment of children with constipation. Amendola et al. feel that early psychological intervention in parent-child interaction is of great importance in the treatment of functional constipation. After 3 months of combined treatment (psychological intervention and medical treatment) 88% of their patients had regular bowel movements. It is important to investigate the parent-child interaction in children with an anorectal malformation or Hirschsprung disease. But first we need to know whether the parents of these children in The Netherlands suffer from anxiety and an impaired QoL.

We started a prospective longitudinal multicentre study in the Netherlands called the KLANKbord-study. This study investigates QoL of both children born with an anorectal malformation or Hirschsprung disease and their parents. QoL is a multidimensional concept. In literature, many different definitions can be found. The World Health Organization Quality of Life (WHOQOL) 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 because of 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 in time. Because of the possibilities of coping strategies we also included parents of older children in our study. These are parents of children in the age-group of 10 to 13 years.

The hypothesis of this study is that parents of children with an anorectal malformation or Hirschsprung disease will experience stress and fears (anxiety). Also, we assume that anxiety levels will be different between mothers and fathers because of a difference in worrying about their child. This stress and these fears can lead to a decreased QoL of parents. According to the definition of QoL, it refers to (dis)satisfaction and can vary over time. We assume that fear of parents will decline in time, which may have an influence on their QoL.
The aim of this study was to investigate the difference in anxiety and QoL between mothers and fathers of children with an anorectal malformation or Hirschsprung disease. In addition, we wanted to investigate the difference in anxiety between parents of newborns compared with parents of children in the age group 10 to 13 years old.

**Patients and Methods**

**Patient population**
Since January 2012, we started including all patients and their parents born with an anorectal malformation or Hirschsprung disease in two of the participating hospitals in Netherlands. Since September 2012 we also started including patients and their parents in two other participating hospitals. The only exclusion criterion is the inability to speak Dutch or English.

All parents of patients born between January 1st 1999 and December 31st 2002 with an anorectal malformation or Hirschsprung disease in two of the participating hospitals were sent questionnaires after receiving an invitation for this study. The study was approved by the medical ethical committee of all participating hospitals.

**Questionnaires**
Parents had to complete two questionnaires, the WHOQOL-BREF (26 items) and the Spielberger State-Trait Anxiety Inventory (STAI-trait 10 items, STAI-state 6 items).

The WHOQOL-BREF is a QoL assessment developed by the WHOQOL-group. This questionnaire has 26 items in 4 different domains and a general QoL facet. The domains are physical health, psychological health, social relationships and environment. Every domain comprises different facets. For instance, the physical health domain focuses on activities of daily living, dependence on medical substances and medical aids, energy and fatigue, mobility, pain and discomfort, sleep, rest and work capacity. The questions include the following: “To what extent do you feel that physical pain prevents you from doing what you need to do?” or “How would you rate your quality of life?” The response scales are 5-point Likert scales. In general, a high score represents a good QoL.

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 dis-
ease, with varying amounts of stress. 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.\textsuperscript{11-14} Examples of questions in the STAI-State and STAI-Trait are: “I feel calm”, “I feel safe” and “I worry too much about not important things”. Although both questionnaires are validated for parents of children suffering from a disease\textsuperscript{15,16}, they have never been validated specifically for parents of children with an anorectal malformation or Hirschsprung disease.

**Statistical analysis**

All continuous variables are presented as means ± standard deviation or median values with interquartile ranges and analysed using either the independent $t$-test with 95% confidence intervals or paired sampled $t$-test. We used the paired sampled $t$-test to compare mothers with fathers. The independent $t$-test was used to compare the parents of newborns with the parents of older children. 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, IL, United States).

**Results**

In total 24 patients with an anorectal malformation or Hirschsprung disease were included in four participating hospitals. One patient was included on own request after reading the outline of our study in the patient magazine for Hirschsprung disease. Two patients and their parents were excluded because of the inability to speak Dutch or English. Three parents decided not to participate with the study after receiving the questionnaires. Between January 1\textsuperscript{st} 1999 and December 31\textsuperscript{st} 2002, 91 patients born with anorectal malformations or Hirschsprung disease were treated in 2 of the participating centres. A total of 39 patients were lost in follow-up, 11 patients decided not to participate after receiving the announcement of our study. Twenty-four parents returned the questionnaires. Seventeen parents did not return the questionnaires.

In total the parents of 44 children were included in the study, 20 parents of newborns (20 mothers, 20 fathers) and 24 parents of children in the age-group of 10 to 13 years (24 mothers, 22 fathers).

When we compared mothers and fathers of the overall group (newborns and older children) mothers scored significantly worse on state-anxiety ($p = 0.005$). The trait-anxiety was not significantly different between mothers and fathers ($p = 0.113$).
Table 1: Comparison between mothers and fathers

<table>
<thead>
<tr>
<th></th>
<th>Value</th>
<th>Mothers (mean)</th>
<th>Fathers (mean)</th>
<th>p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Overall group</td>
<td>Trait anxiety</td>
<td>17.40</td>
<td>16.10</td>
<td>0.113</td>
</tr>
<tr>
<td></td>
<td>State anxiety</td>
<td>10.74</td>
<td>9.36</td>
<td>0.005</td>
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<tr>
<td></td>
<td>WHOQOL-Bref (psychological domain)</td>
<td>14.86</td>
<td>15.74</td>
<td>0.016</td>
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<tr>
<td>Newborns</td>
<td>Trait anxiety</td>
<td>17.63</td>
<td>15.68</td>
<td>0.059</td>
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<tr>
<td></td>
<td>State anxiety</td>
<td>11.44</td>
<td>9.89</td>
<td>0.016</td>
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<tr>
<td></td>
<td>WHOQOL-Bref (psychological domain)</td>
<td>14.60</td>
<td>15.63</td>
<td>0.067</td>
</tr>
<tr>
<td>Older children</td>
<td>Trait anxiety</td>
<td>17.19</td>
<td>16.48</td>
<td>0.578</td>
</tr>
<tr>
<td></td>
<td>State anxiety</td>
<td>10.14</td>
<td>8.90</td>
<td>0.104</td>
</tr>
<tr>
<td></td>
<td>WHOQOL-Bref (psychological domain)</td>
<td>15.09</td>
<td>15.84</td>
<td>0.130</td>
</tr>
</tbody>
</table>

On the psychological domain of the WHOQOL-BREF mothers scored worse than the fathers (p = 0.016).

After dividing the group in newborns and children between 10 to 13 years, we performed the same statistical analysis. In the newborn group the state anxiety was significantly different between mothers and fathers (p = 0.016). The trait anxiety showed a trend towards more anxiety in mothers (p = 0.059). As did the psychological domain of the WHOQOL-BREF (p = 0.067).

In the group of older children both anxiety and QoL were not significantly different between fathers and mothers (Table 1).

We also compared mothers of the newborns with mothers of the older children. And we did the same with the fathers. All domains of the WHOQOL-BREF, the trait anxiety and state anxiety were not significantly different between these groups. However, we did see that mothers and fathers of newborns scored worse on state-anxiety than mothers and fathers of older children (mothers: 11.9 vs 10.3 (p = .138 ; 95% CI-.536 – 3.717) ; fathers: 10.3 vs 8.8 (p = .132 ; 95% CI-.470 – 3.451)).

Discussion

Our results show that mothers experience more anxiety than fathers. This difference in anxiety level is most profound in parents of newborns. The trait anxiety in this group is not significantly different. This means that the mothers do not have a more anxious char-
acter than the fathers do. But the mothers do suffer from more anxiety during the time their children are diagnosed with an anorectal malformation or Hirschsprung disease. This anxiety is thus not a result of their character, but solely a result of having a child with an anorectal malformation or Hirschsprung disease. When the children are in the age-group 10 to 13 years old, we do not have a difference in anxiety between mothers and fathers anymore.

Comparing mothers of newborns to mothers of older children we see the trend that mothers of newborns report more state anxiety, than mothers of older children do.

Our results show that over time anxiety has become less in mothers with children with an anorectal malformation or Hirschsprung disease. This can be a result of coping strategies. We know from literature that coping strategies will reduce anxiety in patients and in their care-givers over time.17,18 A recent article about anxiety and depression in parents of sick neonates revealed that mothers suffer from more anxiety and depression than fathers do.19

We hope to determine with our longitudinal study the moment that mothers do not suffer from more anxiety than fathers do anymore. When we define this moment, psychosocial intervention programs for mothers can be of help in managing the situation with a child with an anorectal malformation or Hirschsprung disease.

A Cochrane review by Barlow et al. has revealed that parenting programs can make a significant contribution to the short-term psychosocial health of mothers. We have limited data on long-term follow-up, and the data that are available suggest that these results may not maintain over time.20

Looking at our data, it is suggested that anxiety and QoL will improve over time. This may be because of coping strategies. This also suggests that psychosocial health of mothers can be improved in mothers of newborns with an anorectal malformation or Hirschsprung disease with parenting programs, and that over time coping will help these mothers with reducing anxiety and improving QoL.

Limited data are available for fathers, but these also show significant short-term improvement of paternal stress.21

A shortcoming of this study is that the patient groups are still small. Because of these small groups comparing the data with each other or to the norm values of the normal population is difficult. We hope that in the future we can perform these analysis again when we have included more patients and their parents. Also, it has not been possible yet to follow the parents of the newborns in time to see if their anxiety levels will decline. The data in this study are derived from a postal questionnaire. A disadvantage of postal questionnaires is that more people are lost to follow-up in comparison to face-to-face
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It is also known that postal questionnaires are more prone to confusion and misunderstandings. On contrary, in face-to-face interviews, an interviewer may influence the answers. This might particularly be the case when satisfaction, a very subjective feeling, is measured. In order to prevent this, questionnaires used for measuring QoL must be self-administered.

The aim of this study was to investigate the difference in anxiety and QoL between mothers and fathers of children diagnosed with an anorectal malformation or Hirschsprung disease, together with the difference between parents of newborns and older children.

**Conclusion**

In conclusion mothers do suffer from more anxiety and a lower psychological QoL than fathers. Data also suggest that mothers and fathers of newborns suffer from more anxiety than parents of older children do.

Anxiety and a decreased psychosocial QoL are an important factor during the first period after birth and during the first treatment of anorectal malformations and Hirschsprung disease. Because of the results of this study we expect that coping strategies will help in reducing this anxiety, but further research is important. In our longitudinal, prospective, multicentre centre study (KLANKbord-study) we will follow mothers, fathers and children from birth till 18 years of age. These data will provide us hopefully with answers about anxiety and QoL in patients and parents over time.
Reference List
