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# **Psychosocial Aspects on Children with Imperforate Anus and Their Parents**

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*To Sven, Miriam, Elias, Hannah*

## ABSTRACT

**Aim:** The overall aim of this thesis was to explore psychosocial consequences on children with high and intermediate imperforate anus and their parents.

**Material/Methods:** *Paper I* was an explorative study involving interviews. The purpose was to illuminate the children's and parents' experiences of imperforate anus (IA) and to create a baseline for further research. In *Papers II-V*, study-specific questionnaires were used to collect data from 25 children (8-13 years old) with high and intermediate imperforate anus and from their parents. The questionnaires comprised 45 items for the children and 59 items for the parents, covering psychosocial issues, items on child bowel function, and on experiences of hospital care. The parents also filled in the Child Behavior Checklist (CBCL), and the children's teachers filled in the Teacher's Report Form (TRF). Thirty children with juvenile chronic arthritis and 32 children with no chronic condition, along with their parents, served as Comparison Groups. In *Paper VI*, the psychometric properties of the child self-reported psychosocial items were evaluated with Rasch analysis.

**Results:** Four categories of effects of the malformation were disclosed in the interviews: physical, emotional, social, and family effects. The emotional effects permeated the interviews; the parents' suffering was evident. In the questionnaires the children with imperforate anus reported being generally happy, they liked school, and had good relationships with peers. According to their parents, the children had fecal incontinence and constipation. In the CBCL, the children with imperforate anus were assessed to have more emotional/behavioral problems than the children with juvenile chronic arthritis. On the CBCL competence scales no differences were found between groups. In the TRF, the teachers reported few emotional/behavioral problems for the children with imperforate anus, though they assessed them to be less adapted in school than were the children with no chronic condition. Differences were found in answers on the psychological items, between the children and their mothers, on the pair level. The mothers of children with imperforate anus rated lower on their child's expression of will and on respecting their child's will, than did the other mothers. Fathers scored school items optimistically. Several parents of children with imperforate anus reported positive experiences in relation to their child's condition involving child development, parental development, and strengthening of family unity. The psychometric properties of the psychological and the social dimension in the child self-report questionnaire were reasonably good.

**Conclusions:** The children with imperforate anus might have some psychosocial difficulties, though informants do not agree. According to self-report questionnaires, the children appear to be well-adjusted, despite functional bowel problems. Parents of children with imperforate anus have to go through difficult experiences associated with their child's malformation, though some parents also report positive experiences. Psychosocial issues seem crucial to children with imperforate anus and to their parents, and qualified individualized assistance should be a central part of their continuing care to ameliorate or prevent difficulties. Collaboration with expertise from child and adolescent psychiatry may be required. Child autonomy needs more attention. Psychosocial interventions are requested as well as a follow-up study on these children and their parents. Further development of the measurement tools, the study-specific questionnaires, is required.

**Keywords:** Psychosocial experiences, Psychosocial function, Children with imperforate anus, Parents, Multiple informant assessment.

## LIST OF PUBLICATIONS

- I. Nisell M., Öjmyr-Joelsson M., Frenckner B., Rydelius P-A., Christensson K.  
(2003). How a family is affected when a child is born with anorectal malformation. Interviews with three patients and their parents. *Journal of Pediatric Nursing*. 18(6), 423-432.
- II. Öjmyr-Joelsson M., Nisell M., Frenckner B., Rydelius P-A., Christensson K.  
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- III. Nisell M., Öjmyr-Joelsson M., Frenckner B., Rydelius P-A., Christensson K.  
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- V. Nisell M., Öjmyr-Joelsson M., Frenckner B., Rydelius P-A., Christensson K.  
Psychosocial experiences of parents of a child with imperforate anus.  
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- VI. Nisell M., Brodin, U., Christensson K., Rydelius P-A.  
The Imperforate Anus Psychosocial Questionnaire (IASPQ): Its construction and psychometric properties. (*Submitted*).

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## LIST OF ABBREVIATIONS

CBCL	Child Behavior Checklist
HRQoL	Health-related Quality of Life
IAPSQ	Imperforate Anus Psychosocial Questionnaire
IBD	Inflammatory Bowel Disease
IRT	Item Response Theory
JCA	Juvenile Chronic Arthritis
MACE	Malone's Antegrade Contenance Enema
NCC	No Chronic Condition
PSARP	Posterior Sagittal Anorectoplasty
QoL	Quality of Life
TRF	Teacher's Report Form
VAS	Visual Analogue Scale



# 1 INTRODUCTION

Psychosocial issues play a dominant role in adaptation to a chronic condition for the affected child as well as for the parents. Psychosocial issues may be as fundamental as the physical concerns in a family with a child with a chronic condition. To have a child with a chronic condition/congenital anomaly can bring about crisis situations in a family, and the physical care may add considerably to the family's burden and stress. Eventually the child and the family will live their life as normally as possible, though psychosocial consequences might ensue.

Imperforate anus is a complicated malformation that includes lack of a normal anorectum, often entailing constipation and fecal incontinence, in spite of successful surgical treatment. This chronic condition affects parts of the body viewed as private. The physical consequences can be seen as shameful, and there may be particular psychosocial consequences of the condition.

When children with chronic conditions and their parents are in contact with pediatric health care, the child's physical well-being is primarily and naturally in focus. Getting a more complete view of the family, an integration of different viewpoints, including child and adolescent psychiatry, will allow a better assessment of the child's functioning.

The overall aim of this thesis was to explore the psychosocial consequences on children with high and intermediate imperforate anus and their parents.

## **2 BACKGROUND**

### **2.1 PSYCHOSOCIAL ISSUES**

Childhood is a flexible time when family and social relations have a big impact on personal development. In a well-known theory about child development, by Erikson (1950), psychosocial issues are included. This theory is based on a multifaceted perspective, where development originates in biological inheritance and psychosocial factors. A biopsychosocial model proposes a transactional relationship between the child, the family, the school, and the health care provider.

The psychosocial concept is extensive and can encompass many aspects. It is not always easy to draw clear distinctions between the commonly used terms to describe consequences of chronic illness, such as “emotional disorders”, “psychosocial maladjustment”, “behavioral disorders”, and “social functioning” (Pless & Nolan, 1991). Other authors add other definitions. For example, school activities and self-esteem are defined as important psychosocial factors for children who have chronic conditions (Vitulano, 2003). Psychosocial factors have been defined as: family, school, friends, activities, hobbies, and emotional life. Through key questions to parents and children about the child’s psychosocial functioning, one can get a picture of the child’s social adjustment and possible psychological problems (Cassidy & Jellinek, 1998).

Psychological aspects are identified and included in social functioning (Adams, Streisand, Zawacki, & Joseph, 2002). Psychosocial problems can also be defined as behavioral, emotional, and educational problems that are highly prevalent among children and can interfere severely with everyday functioning (Brugman, Reijneveld, Verhulst, & Verloove-Vanhorick, 2001).

Child psychosocial adjustment is a reflection of the child’s environment, and the family unit is considered the primary environment of the developing child. The parents might be the most crucial psychosocial indicator for the child’s future and development when a child has a chronic condition (Pless & Nolan, 1991). On the other hand parental functioning and well-being is influenced by their child’s health status (Silver, Westbrook, & Stein, 1998). Furthermore, mother-child interaction was one key predictor of change in both child outcome and parent well-being (Hauser-Cram et al., 2001).

Friends play important roles for all children. For children with a chronic disability the support of classmates and friends might be even more central to psychosocial adaptation. In a study on children with limb insufficiencies, lower levels of depressive symptoms and higher self-esteem were associated with higher classmate support (Varni, Setoguchi, Rappaport, & Talbot, 1992). It has been reported that children with chronic illnesses are less accepted by their peers than healthy children are (Alderfer, Wiebe, & Hartmann, 2001). However, this is not always the case. It has been found that, in general, children with chronic conditions have no more problems in their peer relations than do other young people, except for children that have a stigmatizing disability (La Greca, Bearman, & Moore, 2002).

School is a social factor of great importance. In school a register of psychosocial issues are amalgamated and the child's psychosocial function is tested. It is important to measure limitation in school function among children with chronic conditions and to distinguish the various conditions to obtain a more accurate picture of the impact of health on the children's lives (McDougall et al., 2004). Paying attention to school attitude is of significance in order to improve Quality of Life (QoL) of children and adolescents with anorectal malformations and Hirschsprung's disease (Hartman et al., 2007).

Some may argue that psychosocial functioning and QoL are interchangeable terms and if not identical, they seem to be closely related. Good QoL is a key outcome for the individual child and is desired for all children (Dickinson et al., 2007). In research on QoL, psychosocial aspects are usually included (Eiser & Morse, 2001a). When discrepancies and similarities were compared in ten questionnaires about QoL, the authors divided the domains into physical, psychological, and social (Rajmil et al., 2004).

Moreover, QoL is multidimensional and can be defined as a subjective perception of health in physical, emotional, social, and functional domains (Bullinger, Schmidt, & Petersen, 2002). A similar definition is revealed in another study that states that quality of life is physical, psychological, and social well-being (Trajanovska & Catto-Smith, 2005). When measuring health-related QoL issues, physical, psychological (including

emotional and cognitive), and social health dimensions are defined (Varni, Limbers, & Burwinkle, 2007a). An additional concept is Health Related Quality of Life (HRQoL), which has become more prominent in relation to chronic conditions. Health-related quality of life can be considered to be that part of a person's overall quality of life that is determined primarily by their health status, and the health status refers in general to mental and physical health. (Moorthy, Peterson, Harrison, Onel, & Lehman, 2008)

Initiating and talking about psychosocial issues may be even harder than discussing physical aspects. Parents usually have more questions about physical well-being than about psychosocial issues (Yerkey & Wildman, 2004). Even if children and parents do not need contact with staff specialized in psychiatry, psychosocial issues can be raised by health care staff. Few parents voluntarily voice concerns about psychosocial problems, and therefore health care personnel need to ask both the parents and the children about these issues (Wildman, Kinsman, & Smucker, 2000).

## **2.2 CHILDREN WITH CHRONIC CONDITIONS**

Two of the often-used definitions describing chronic pediatric condition concepts are chronic illness, and chronic health conditions. (van der Lee, Mokkink, Grootenhuis, Heymans, & Offringa, 2007). The prevalence rates vary in severity and there is diversity in the definitions used. This may hinder reliable estimates, and a consensus is called for (Turkel & Pao, 2007). Around 10% of children appear to be suffering from a chronic health condition, and in the United States the number was 12.8% in 2006 (Seferian, Lackore, Rahman, Naessens, & Williams, 2006). The percentage rate of children with long-term chronic illness was 12.9% in Sweden (1996), with an increase from 7.4 % since 1984 (Berntsson & Kohler, 2001a). Criteria for a chronic condition in childhood are that the condition has been present for a longer time than three months or that it will last longer than three months (Mokkink, van der Lee, Grootenhuis, Offringa, & Heymans, 2008).

Children with chronic conditions are known to be at increased risk for psychosocial adjustment problems. Evidence from meta-analyses showed that children with chronic condition were at slightly elevated risk of psychosocial distress, although only a minority experience clinical symptomatology (Barlow & Ellard, 2006). Differences on psychosocial variables in relation to healthy children was found when HRQoL was

studied among 2500 pediatric patients across ten chronic disease clusters from child/patient and parent perspectives (Varni et al., 2007a). Analogous results were found in a Dutch study, which however had fewer (318) children/patients participating (Grootenhuis, Koopman, Verrips, Vogels, & Last, 2007). Aspects of HRQoL in children are negatively affected by chronic illness (Epstein, Stinson, & Stevens, 2005). However, self-reported QoL of children 8-12 years old with cerebral palsy had similar QoL to that of the general population (Dickinson et al., 2007). And in a recent study on HRQoL in children with thoracic insufficiency syndromes, the scores in psychosocial domains were similar to those of healthy children (Vitale et al., 2008).

An important factor is how children with chronic illness manage to cope with the disability. Children in general use coping strategies meaningfully associated with adjustment measures, and denial coping emerges as a considerable moderator of the relation between status and adjustment (Sandberg et al., 2001). It is crucial to assess children's views of their psychosocial functioning as well as their experiences to understand how these experiences influence everyday functioning, and it is valuable to have more than one informant. In a study on QoL, support is given to include both parents and children when evaluating children's QoL, though large differences can exist in proxy agreement at the individual child-parent level (Berntsson & Kohler, 2001b).

### **2.3 PARENTS OF CHILDREN WITH CHRONIC CONDITIONS**

When a person is subjected to illness it has been found that the close family around him/her is often affected. When the patient is a child, special concerns can be distinguished due to the emotional dependencies between parents and children. There are more demands on parents in raising a child with a chronic condition than there are with a healthy child (Ray, 2002). The time of diagnosis is believed to be the critical period, and parents of a child with an impairment are in need of information and support (Rahi, Manaras, Tuomainen, & Hundt, 2004).

Families that have a child with a chronic condition are exposed to stress and special difficulties (Burke, Kauffmann, Harrison, & Wiskin, 1999). Although the children bear the weight of their chronic health condition, studies have documented increased risk for difficulties among the parents (Bauman, Drotar, Leventhal, Perrin, & Pless, 1997).

(Hatzmann, Heymans, Ferrer-i-Carbonell, van Praag, & Grootenhuis, 2008; Mednick, Gargollo, Oliva, Grant, & Borer, 2009). Both children with a chronic condition and their mothers are at increased risk for psychosocial adjustment difficulties, as compared to peers (Wallander & Varni, 1998). In families with a child with fecal incontinence, cognitions and perceptions influence the parent-child relationship negatively, which may result in behavioral problems in the child (van Dijk, Benninga, Grootenhuis, Nieuwenhuizen, & Last, 2007). A dominant predictor of the child's psychosocial adjustment is how the family copes with the child's condition; the better the family functions, the better the adjustment of the child with a chronic condition will be (Hamlett, Pellegrini, & Katz, 1992)

Additionally, there may be financial consequences of having a child with a chronic condition. Caregivers of children with cerebral palsy were less likely to be engaged in fulltime work compared with that of the general population of caregivers (Brehaut et al., 2004) and parents of children with chronic conditions changed their career plans to devote more time to caregiving (Case-Smith, 2004). In contrast, a more optimistic finding was displayed in a study on the impact of newly-diagnosed chronic pediatric conditions on parental quality of life; the parents were more satisfied with their family situations than were healthy controls (Goldbeck, 2006).

## **2.4 CHILDREN WITH IMPERFORATE ANUS AND THEIR PARENTS**

Imperforate anus is an uncommon (1:4-5000) malformation that includes lack of a normal anorectum (Peña, 2000). The degree of severity varies from the most severe, the high form, to the intermediate and the low form. Children born with imperforate anus often have other abnormalities and the higher form of imperforate anus is more associated with other malformations (Peña & Levitt, 2006).

In general, children with high and intermediate imperforate anus receive a colostomy immediately after birth, to empty their bowels. A new anus is constructed surgically when the child is around two-three months old. The posterior sagittal anorectoplasty (PSARP) is generally the surgical method of choice (Levitt & Pena, 2005). After reconstruction, the new anus must be dilated to attain normal width. The follow-up treatment requires daily dilations of the newly constructed anus for two to three months (Peña, 1985).

When the child is around six months old, closure of the stoma is performed and after that the child can pass stools in the normal way. Even though the surgical procedures have been successful, many children with imperforate anus have functional difficulties including constipation and fecal incontinence (Levitt & Pena, 2007). Constipation is the most common functional disorder observed in patients who have undergone PSARP (Levitt & Pena, 2005). Regular enemas are often given as prevention and treatment for several years (Peña et al., 1998). However, even with these treatments the children can continue to suffer constipation as well as fecal incontinence. It is important to start the treatment to achieve continence before the child reaches school age to surmount the distressing social penalty of fecal soiling and try to assimilate the child with his/her peers (Rintala & Pakarinen, 2008).

Imperforate anus is a congenital malformation. Birth defects can present parents with a crisis and may represent a state of suspended animation for many parents. As imperforate anus is an unusual malformation, it is mostly unknown to the parents. The information given at the time of the child's birth may be hard to grasp and even infants with good prognoses induce distress and represent potential disability (Mayes, 2003). Furthermore, the parents are probably unprepared for the anomaly as it is unusual for an anomaly to be diagnosed after the birth of a child. Many fetal abnormalities are diagnosed with prenatal screening and diagnostic technology (Howard, 2006).

When a child is born with high and intermediate imperforate anus, the parents have to care for the child in a special way. They are involved in the care of the stoma from the very beginning, and during the child's growing up they have to take responsibility for special treatment, performing the follow-up treatment and invasive procedure for two to three months and being in charge of giving their children the enemas for several years. The family needs support throughout the child's development, because of the huge impact of the malformation (Hassink et al., 1998).

Psychosocial consequences of imperforate anus have so far been explored in a limited number of studies. Psychiatric symptoms, such as anxiety, low self-esteem, and depression may develop in adolescents with high imperforate anus. The intrusive procedures the child has to go through and the parents have to perform might affect the relationships between children with imperforate anus and their parents (Davies,

Creighton, & Wilcox, 2004). The malformation and the treatment involved can be painful and violate the child's integrity, and thus influence the relationship between child and parents (Diseth & Emblem, 1996). Additionally, it was found that children with imperforate anus had difficulties in relationships with peers (Hassink, Rieu, Brugman, & Festen, 1994; Meijer, Sinnema, Bijstra, Mellenbergh, & Wolters, 2000). The physical consequences of imperforate anus could be regarded as a stigma; the malformation is not visible, but the condition can be revealed by the odor. It was shown to cause psychological problems; fecal incontinence is socially unacceptable, and clinical experience has shown a link between physical and emotional difficulties (Ludman, Spitz, & Kiely, 1994).

In a study by Hamid, Holland, and Martin (2007), on long-term outcome of anorectal malformations on 167 patients/children, 80% had behavioral problems. A negative social impact was found in 52% of the children, and 15% expressed suicidal thoughts. However the children remained hopeful for the future and reported higher than average global hopefulness scores. Another recent study revealed that bowel function was the vital factor that influences the level of depression of children with anorectal malformations and the mothers' psychological status was also of significance (Amae et al., 2008). When Social Quality of Life was analyzed among 20-40-year-old adult patients who had been operated on, fecal soiling was the key factor that disturbed their occupational life (Iwai et al., 2007). Fecal incontinence was also directly associated with the children's QoL in one of the first studies on psychosocial aspects of children with imperforate anus (Ditesheim & Templeton, 1987). On the other hand, when QoL of adults with anorectal malformations was examined in a Dutch population, fecal incontinence had no effect on the patients' QoL, although psychosocial functioning was shown to have the most essential effect (Hartman et al., 2004).

Psychosocial problems were reported by children with anorectal malformations and there is a call for perceived psychosocial competencies for enhancing QoL, as well as for giving these children extra care (Hartman et al., 2008). Multidisciplinary treatment has been found optimal for children with anorectal malformations (van Kuyk et al., 2001), and psychosocial support for the children with anorectal malformations and their mothers may be necessary (Funakosi et al., 2005).

### 3 AIMS

The overall aim of this study was to explore psychosocial consequences on children with high and intermediate imperforate anus and their parents.

Specific aims of included papers were:

- I. To illuminate the experiences and psychosocial situation of three patients with high imperforate anus and their parents, and to create a baseline for further research.
- II. To evaluate the psychosocial effects of high and intermediate imperforate anus on school-aged children.
- III. To examine disagreement on psychosocial functioning in reports between children with high and intermediate imperforate anus and their mothers.
- IV. To examine how social issues of children with intermediate or high imperforate anus may be affected, from the perspective of their mothers, fathers, and teachers.
- V. To examine the psychosocial experiences of parents of children with high and intermediate imperforate anus, and to describe their potential positive experiences.
- VI. To evaluate the psychometric properties of a self-reported questionnaire constructed to assess the psychological and social functioning of children with imperforate anus.

## 4 STUDY DESIGN, MATERIAL, AND METHODS

This study is a product of a fully integrated cooperation between a pediatric surgery unit and a child and adolescent psychiatric unit. Such a close cooperation is needed if all aspects of nursing care for patients with chronic conditions and their families are to be incorporated. Cooperation between pediatric health care and child and adolescent psychiatry has not really been fully established and developed in regular pediatric care. It has been stated that there is an unmet need for consultation/liaison work between these two specialties (Glazebrook, Hollis, Heussler, Goodman, & Coates, 2003).

It is crucial to identify all aspects that may influence psychosocial functioning, for providing apposite interventions (Barlow, Shaw, & Wright, 2001). Therefore, and in the course of the collaboration between the pediatric surgery unit and the child and adolescent psychiatric unit at Astrid Lindgren Children's hospital, Karolinska University Hospital, this research study was set up.

In an interview study with health care personnel in pediatric care in Stockholm, Sweden, a lack of good models for integrated consultation/liaison work between pediatric somatic and psychiatric disciplines was found. Despite this, the majority of staff, and the nurses above all, expressed a wish for a closer cooperation with the child and adolescent psychiatric unit (Nisell & Rydelius, 2007).

This collaboration was developed as a common base for nursing care for patients at the Astrid Lindgren Children's hospital. We decided to focus on patients with imperforate anus because this malformation represents features that could be linked with child psychiatry. Private parts of the body are involved, and the consequences of the malformation and the follow-up treatment include sensitive characteristics.

We choose to interview three selected patients with high imperforate anus, who had severe problems with the malformation, and their parents. Our intention was to explore the experiences of living with imperforate anus and other factors to which it is related. This is similar to what is called an exploratory study, one that investigates the nature of the phenomenon, rather than simply observes and describes it (Polit & Beck, 2004).

The extensive findings from these interviews (*Paper I*) were the basis for a self-report questionnaire addressing the children with high and intermediate imperforate anus and one for the parents. These questionnaires were used as study instruments in *Papers II-VI*. Selected parts of the study-specific questionnaires have been used, with the ambition of focusing and deepening the different aspects of the psychosocial consequences of imperforate anus. Additionally, the Child Behavior Checklist (CBCL) and the Teacher's Report Form (TRF) have been used as measurement tools. The data used in the various papers are presented in Table 1.

**Table 1.** The data used in the respective papers

<b>Data collection</b>	<b><i>Paper I</i></b>	<b><i>Paper II</i></b>	<b><i>Paper III</i></b>	<b><i>Paper IV</i></b>	<b><i>Paper V</i></b>	<b><i>Paper VI</i></b>
<i>Interviews</i>	X					
<i>Child self-report*</i>		X	X			X
<i>Mother proxy report*</i>		X	X	X		
<i>Mother self-report*</i>					X	
<i>Father proxy report*</i>				X		
<i>Father self-report*</i>					X	
<i>CBCL problem scales</i>		X				
<i>CBCL competence scales</i>				X		
<i>TRF problem scales</i>		X				
<i>TRF competence scales</i>				X		

\* *Study-specific questionnaire*

#### **4.1 PAPER I**

In *Paper I*, interviews were done (by MN and P-AR) with three patients aged 12, 14 and 22, with high imperforate anus and with their parents on their experiences of living with the malformation. These patients were recruited at Astrid Lindgren Children's Hospital, a part of the Karolinska University Hospital complex in Stockholm. The informants/patients along with their parents were asked to participate because of their willingness to discuss the excretory problems that they had experienced in childhood. They had also undergone the same kind of primary surgery at St. Görans's Children's Hospital in Stockholm (before 1987), followed by a special operation called MACE (Malone's Antegrade Continence Enema).

The interviews were preceded by telephone contact and written information. Out of the patients who had been treated at St. Göran Children's Hospital and Astrid Lindgren Children's Hospital, there were five patients along with their parents who were of interest. One of these patients was not available and another declined to participate in the study. That left us with eight informants – three children and their parents.

Individual interviews were conducted at the hospital. The interview questions were constructed by specialists in the area of pediatric surgery and child and adolescent psychiatry. The questions covered the child's and parent's experiences of the malformation and its consequences, from the birth of the child. The answers to 17 open-ended questions for the parents and additional nine open-ended questions for the children were used as data.

#### **4.1.1 Analyses of Interviews**

The first step was to read through the data several times to get an impression of the entire body of data. This was done independently by two of the authors (MN & MÖ-J.). The next step was to get data coded for significance. The codes consisted of sentences or paragraphs, and were marked by hand in different colors. The purpose of the coding was to recognize categories, and these sentences were grouped into categories identifying phenomena emerging from the interviews. The interview responses could be broken down into four categories, based on the effects of the malformation: physical, emotional, social, and family.

## **4.2 PAPERS II-VI**

### **4.2.1 Participants**

The participating children and parents are presented in Table 2. The children had been treated at St. Göran Children's Hospital and at Astrid Lindgren Children's Hospital. Twenty-five children (9 boys and 16 girls) with high and intermediate imperforate anus and their parents (25 mothers and 20 fathers) participated. Thirty children and their parents fulfilled the inclusion criteria for participating in this study. One of the families had moved abroad and could not be located and out of the 29 remaining families, four families did not complete the study.

The children were born between 1987 and 1992 and had had operations and follow-up treatments. The mean age of the children was 10.5 (range 8-13.9). This specific group of children and parents was included as the surgical method was modified and refined in 1987. Many (76%) of the children had additional malformations. Furthermore, the youngest child had to be eight years of age in order to be able to read and understand instructions.

Two Comparison Groups were included. We wanted one group of children with another chronic condition who had been through unpleasant treatment since early childhood and one group of children without any chronic condition, and their respective parents. The first Comparison Group, children with juvenile chronic arthritis and their parents were recruited from the rheumatic outpatient clinic at the Astrid Lindgren Children's Hospital. All children born between 1987-1992 who had juvenile chronic arthritis debuting before two years of age and who had joint injections earlier than four years of age, were pertinent for the study. Forty-four families were contacted from the outpatient clinic for patients with rheumatism. They were informed that a study was going on and asked if we could send them further information about the study. Of these families, 35 gave consent and answered that they were interested in receiving more information from us. An information letter was sent to them and they agreed to take part in the study. Five families did not complete the study, and thus 30 children (5 boys and 25 girls) with parents (30 mothers and 25 fathers) were included in the study.

Members of Comparison Group II were admitted to the hospital with comparatively harmless disorders. The children were recruited from the outpatient surgery unit until similar groups were obtained. The children were there for minor surgery problems, were considered otherwise healthy, and had no chronic conditions. Those children who were born 1987-1992 along with their parents were asked by staff at the outpatient surgery unit if they were interested in getting more information about this ongoing study. If they agreed, they received additional oral and written information. Out of 50 children, 32 children (14 boys and 18 girls) and 32 mothers and 20 fathers completed the study.

Demographic characteristics (collected from the CBL) of the participating families are presented in Table 3.

**Table 2.** Participants in *Papers II-VI*

<i>Participants</i>	<b>IA Group</b> n=70	<b>JCA Group</b> n =85	<b>NCC Group</b> n=84	<b>Total</b> N=239
<b>Children</b>	25	30	32	87
<i>Boys/Girls</i>	9/16	5/25	14/18	28/59
<i>Mean age:</i> (years)	10.5 (8.0-13.6)	10.6 (7.8-13.6)	10.9 (8.3-13.9)	10.7 (7.8-13.9)
<b>Mothers</b>	25	30	32	87
<b>Fathers</b>	20	25	20	65
<b>Teachers</b>	12	12	12	36

Note. IA= Imperforate anus; JCA= Juvenile Chronic Arthritis; NCC= No Chronic Condition

**Table 3.** Structures of the families participating.

	<b>IA group</b> n=25	<b>JCA Group<sup>1</sup></b> n=30	<b>NCC Group</b> n=32
<i>Family Structure</i>			
Two-Parent Family	18 (72%)	22 (73%)	23 (72%)
One-Parent Family	7 (28%)	3 (10%)	9 (28%)

Note. IA= Imperforate anus; JCA= Juvenile Chronic Arthritis; NCC= No Chronic Condition

<sup>1</sup> Information on five families is missing

### **4.2.2 Questionnaires**

All respondents answered questions and filled in questionnaires. The children's study-specific questionnaire included psychosocial areas, one physical and one experiential (i.e. hospital-related care) domain. The questionnaire consisted of 45 items, scored on a five-point Likert scale. Item responses depicted either five faces with expressions symbolizing very sad, rather sad, neither sad nor happy, rather happy, and very happy – or responses were written, i.e. always, almost always, sometimes, almost never, and never. One response was written yes or no (Appendix 1).

The study-specific questionnaire that was used for all participating parents contained statements that fell into these domains: three psychosocial, one physical, and one experiential (i.e. hospital-related care). The questionnaire comprised 59 items including free comments on a 10-cm visual analogue scale (VAS). VAS anchor phrases were: very negative and very positive. A neither-nor phrase was sometimes positioned in the middle of the scale. Two responses were written, either yes or no. Mothers and fathers were asked to fill in the questionnaires individually (Appendix 2).

In the Index Group (the children with imperforate anus and their parents), 83% of the families contacted about the study answered the questionnaires and in Comparison Group I the response rate was 74%. In Comparison Group II the response rate was 64%. A reminder was sent to each family who had not returned the questionnaire, from the beginning. An additional reminder was sent to those families who did not answer after the first reminder.

### **4.2.3 Analyses of Questionnaires**

In *Paper II*, the Index Group children's answers on psychosocial and physical variables were compared with one Comparison Group at a time. Pearson's *chi*-square test was used to analyze data. The parents responded to questions on children's psychosocial functioning as well as on physical functioning, and their responses were compared with those of one Comparison Group at time. T-test and Wilcoxon rank-sum test were used to analyze data.

In *Paper III*, the objective was to compare child and parent responses and to look for differences. Fifteen congruent psychosocial variables were chosen in children's and parents' questionnaires, to obtain comparable data. The variables were further categorized as psychological and social. The scores were transformed into dichotomous variables, to enable a comparison of answers from mothers' responses (on VAS) and children's reports (on the Likert scale). Mothers' and children's answers were compared on the pair level within each group and Wilcoxon signed ranks test was used to analyze data.

In *Paper IV*, the parental perspective on child social functioning was focused and the three groups were compared. The global tests for the MANOVAs were applied per questionnaire (CBCL, TRF, IASIQ/Mothers and IASIQ/Fathers). Additionally, ANOVAs for each score were performed to calculate between group contrasts with mean differences and nominal 95% confidence intervals.

In *Paper V*, the psychosocial items that addressed the parents themselves were examined. Reports from the Index Group parents and parents in Comparison Group I were analyzed. The items on VAS were analyzed with Wilcoxon rank sum test and the *chi*-square test was performed to test for differences between groups on dichotomous variables. A qualitative approach, Manifest Content Analysis, was used to analyze the parents' comments on their positive experiences.

In *Paper VI*, the psychometric properties of the psychosocial domains in the children's self-report questionnaire were evaluated. An Item Response Theory model, a Rasch approach, was used to estimate the characteristics and the usefulness of the psychosocial items, in relation to its psychometric properties. Item difficulty and person ability were concurrently approximated with the Rasch methodology.

### **4.3 CBCL AND TRF**

The parents were requested to fill in the Child Behavior Checklist (CBCL) together. CBCL measures children's emotional/behavioral problems and the children's competence. On the CBCL, parents rate 120 items pertaining to emotional and behavioral problems shown by their children during the past six months on a three-point scale. The CBCL comprises a total behavior problem score and two subscales

referred to as internalizing (social withdrawn, depression anxiety) and externalizing (antisocial, aggressive, delinquency) symptom scales. This part of CBCL was used in *Paper II*. The competence scales in CBCL could be considered the “healthy reports” and include activity, social, school, and total competence (*Paper IV*). The parents were asked to assess children’s participation in sports, habits, and job performance on quantity and quality (activity scale). They were also asked to assess them on social relationships with others and their activeness in organization or club participation (social scale) and academic performance in school.

The children’s teachers filled in the Teacher’s Report Form (TRF) to examine the experiences of the teachers. To get in touch with the teachers, the parents were asked for permission to allow their children’s teachers to participate in the study. Altogether 45 teachers were asked and 36 teachers fulfilled their participation (12 teachers from each respective group of children). A reminder was sent to each teacher who did not return the questionnaire and if no answer was received a second reminder was sent to the teacher. Three teachers from each group never completed the study.

TRF is an instrument that measures the child’s emotional/ behavioral problems in school. On the TRF the teachers rate 113 items pertaining to emotional and behavioral problems shown by the children during the past two months on a three-point scale (*Paper II*). Academic and adaptive functioning scales are included in the TRF, and they provide items for obtaining ratings for academic performance and four adaptive characteristics. The teacher’s ratings of performance in academic subjects are scored one to five for categories from far below actual grade to far above actual grade. The teacher’s ratings for the four adaptive characteristics are scored one to seven for categories ranging from much less to much more, compared to typical pupils of the same age (*Paper IV*). Data from the CBCL and the TRF were analyzed according to instructions in the respective manuals (Achenbach, 1991; Achenbach, 1991).

#### **4.4 METHODOLOGICAL ASPECTS**

The methodological considerations of the study are discussed in Chapter 6: “Discussion”, on pages 41-47.

#### **4.5 ETHICAL ASPECTS**

The study was approved by the Ethical Committee at the Karolinska Hospital. The participants gave informed consent and could contact the researchers about any concerns they might have. This study was performed with the awareness that the questions asked in the interviews and in questionnaires were about sensitive and difficult matters. One parent in the Comparison Group of children with juvenile chronic arthritis did not want to continue the study, remarking that the questions posed in the questionnaire were too personal.

In the interviews the parents stated that it was the first time they had really talked about this matter. The disadvantage of interviews as a method could be that the informants might be reminded of difficult experiences. This hardness was confirmed by the parents interviewed as one mother openly cried during the interview. The informants can tell more than their intention was from the beginning, which might entail feelings of having said too much. The patients/children and their parents interviewed were offered the possibility of contacting the interviewers if they were concerned. Additionally, the interviewers were experienced in the field of child and adolescent psychiatry and were used to talking about sensitive issues.

When children participate in research extra sensitivity is required; they need special protection when included in research. Children are immature and are dependant on their parents' custody. In this study the children were informed about the project with an information letter appropriately written for the age of the children. The purpose of obtaining child assent is to show respect for the child and his/her developing autonomy (Rossi, Reynolds, & Nelson, 2003).

The children participating in this study were still minors, and therefore the parents had to give consent for the children to participate. When children are participating in research one has to be especially sensitive. Children with imperforate anus participating in this study had been through the difficult follow-up treatment with insult to body integrity, and that was taken into consideration. In the study-specific questionnaire we did not ask the children themselves about fecal incontinence for ethical reasons, though we did ask their parents.

## 5 RESULTS/ FINDINGS

In *Paper I*, three patients with high imperforate anus and five of their parents were interviewed. The parents were very outspoken when telling about their experiences, but the children's answers were scant. Four categories of effects were disclosed; *physical*, *emotional*, *social*, and *family* effects. The physical effects could be seen as a baseline from which the narratives emanate – the families' experiences of "living with this anomaly" from the first day of the child's life. The emotional effect category was dominant and permeated the result. The emotions revealed were mostly negative and the parents articulated emotions such as sorrow, anger, and disappointment. These emotions were closely connected with experiences/consequences of the malformations, such as anal manipulations (dilations and enemas), constipation, and incontinence. The parents felt as if they had forced themselves on their child when performing dilations and giving enemas. Furthermore, the three adolescents interviewed admitted that they had been depressed, according to themselves and their parents. Their answers on questions about the enemas were terse; one of the patients responded to the question with the word "hard" and another said, "it was nothing, I got used to it".

The social effects demonstrated that everyday life was strained for these families, because of their anxiety about the child's defecation habits. The children had difficulties in participating in physical activities and taking showers afterwards in school, according to parents. The children reported that they had at least one close friend, and two of them had been exposed to bullying. On the family effects, the whole family agreed that the relationship between the mothers and their children was close. The parents expressed wishes that they could have had someone to talk to about their experiences.

In *Paper II*, the children with imperforate anus showed a positive attitude and they reported being generally happy. Furthermore, the children with imperforate anus did not seem to think any more about their disabilities than did children in the Comparison Groups, nor did they experience more negative feelings such as anger and sadness. Variables related to school showed that the children were well adjusted in school. The children in the Index Group were positive about school, and they did not report being bullied. They enjoyed being with friends and spent much time with their best friends. Further, the children in the Index Group reported being more constipated than children in the Comparison Groups. Their parents reported the children to have more fecal

leakage, constipation, and odors than parents of the children in both Comparison Groups reported for their children. No correlation was found between the severity of the malformation and psychosocial outcome and age.

In the CBCL the parents in the Index Group assessed their children to have significantly more total emotional/behavioral problems than the parents of children with juvenile chronic arthritis reported for their children. Teachers of children with imperforate anus reported few problems for the children with imperforate anus in the TRF.

*Paper III* showed discrepancies on the pair level between children's and mothers' responses in all groups and above all on the psychological variables. There were few discrepancies between mothers' and children's responses on the social variables. Children and parents in all groups disagreed on the negative emotions; all mothers reported their children to be sadder and angrier than the children themselves reported. All mothers reported their children to be more bullied and teased than indicated in the children's responses. Two discrepancies were revealed in the Index Group but not in the Comparison Groups. These were: 1) the mothers indicated their children to have better self-confidence than their children reported themselves to have, and 2) the mothers reported thinking more of their children's disabilities than the children assessed that the mothers thought of their disabilities.

In *Paper IV* and according to mothers, the children with IA expressed their will to a lesser extent compared with the reports of mothers of children with no chronic condition. School items were scored optimistically; fathers of children with imperforate anus rated more favorably than fathers of children with no chronic condition. On the CBCL Competence scales, the Index Group parents reported their children to be just as active, social, and well adjusted to school as the children in Comparison Groups were assessed by their parents to be. In the TRF, the teachers of children with IA seemed to indicate the children as not as well adapted and functioning as were the children with no chronic condition, rated by their teachers. No statistically significant differences were found among groups on academic performance.

In *Paper V*, only parents of children with juvenile chronic arthritis served as a Comparison Group. The mothers of children with imperforate anus reported that their social relationships had been more influenced by their child's condition and that they

respected their child's will to a lesser extent than did the mothers in the other group. No differences were found among fathers' reports on identical items. Several parents of children with imperforate anus reported positive experiences in conjunction with their child's condition; 48% of the mothers and 35% of the fathers.

Moreover in *Paper V*, parents' positive experiences related to the child's condition were analyzed with manifest content analysis and three themes were identified. The first theme was *Child's development*; the child had developed into a good, independent, and empathetic person. The second theme, *Development of the parent*, was the most frequent theme and showed that the parent had become a meeker person. The parents looked at life in a different way than before and had developed new perspectives, and they had become more humble, due to their child's condition. The third theme, *Strengthening of family unity*, recognized that, despite difficult experiences the child's impairment had become a force. The family had been strengthened as a unit and troublesome moments were shared.

*Paper VI* disclosed the psychometric properties of the psychosocial domains in the child self-report questionnaire; the Imperforate Anus Psychosocial Questionnaire (IAPSQ). The statistical analyses were done with Rasch analysis. The psychological dimension of the IAPSQ showed sufficient item reliability and moderate person reliability. The social dimension of the IAPSQ showed satisfactory item reliability but person reliability was weak. Construct validity was recognized on the psychological dimension.

In summary, the main results in relation to the aims of the specific papers were:

- I. The children with imperforate anus and especially their parents had experienced psychosocial problems.
- II. The children with imperforate anus seemed to have good psychosocial functioning, according to their own self-reports. On the CBCL the children were assessed to have more emotional and behavioral difficulties. The TRF showed no more emotional and behavioral difficulties for the children with imperforate anus.
- III. Disagreement was found particularly in answers on the psychological items, between children with imperforate anus and their mothers, on the pair level.
- IV. The children with imperforate anus were assessed by their parents to be socially competent. The teachers seemed to indicate that the children with imperforate anus were less adapted and functioning.

V. The social relationships and the respect of their child's will were more affected among the mothers of a child with imperforate anus. Several parents (mothers) of children with imperforate anus reported positive experiences related to their child's malformation.

VI. The psychometric properties of the psychological and the social dimension in the child self-report questionnaire were reasonably good.

## 6 DISCUSSION

The overall aim of this study was to explore the psychosocial consequences on children with imperforate anus and their parents. There seem to be certain psychosocial consequences of high and intermediate imperforate anus among the affected children and their parents who participated in this study. A heavy emotional burden on the parents and their children was seen in *Paper I*. However, almost half of the parents participating reported positive experiences in conjunction with the child's condition in *Paper V*. From self-report, the children with imperforate anus appeared to be as psychosocially competent (*Papers II and III*) as the other children participating. Psychosocial consequences on the children, including emotional and behavioral difficulties, appear to be disclosed by parental and teacher report in *Papers II and IV*, respectively. The psychosocial impact of the child's condition on child-parent communication and interaction can possibly be discerned in *Papers I-V*.

### 6.1 REFLECTIONS OF FINDINGS

It may be hard to identify if the psychosocial consequences of a chronic condition, as in the case of imperforate anus, are caused by the condition itself, or if and in that case how other factors may contribute. Similarities among chronic illness are acknowledged and there is little verification that psychosocial problems are associated with specific diseases (Geist, Grdisa, & Otley, 2003). In contrast, significant discrepancies between chronic pediatric conditions were identified and some of them meet the criteria of having special health care needs (Schmidt, Thyen, Petersen, & Bullinger, 2004). Surgically corrected hypospadias was not a risk factor for psychosocial maladjustment among boys in middle childhood (Sandberg et al., 2001).

Nevertheless, it has been confirmed that patients with anorectal malformations experience psychosocial problems because of the malformation (Hartman et al., 2005). In a study on children with slow transit constipation, including similar consequences such as of those imperforate anus, children's physical and emotional functioning were affected (Clarke et al., 2008).

The outcome might depend on the *severity* of the malformation's physical consequences and an underlying assumption that psychosocial consequences depend on *illness*

*characteristics*. Supposedly the children concerned in *Papers II-V* had lesser problems with fecal incontinence than the children in *Paper I*. In *Paper I*, the informants/patients were chosen because they had had severe problems with fecal incontinence. They had their anal reconstruction before 1987 and were operated on with a technique that was different from the one used today. After 1987, children with imperforate anus at Astrid Lindgren Children's hospital were operated on with the PSARP (Peña, 1985), and a fistula-saving technique (Husberg, Lindahl, Rintala, & Frenckner, 1992).

Psychosocial outcome was not found to be related to the severity of the malformation (*Paper II*). Many (76%) of the children involved in this project were born with other congenital malformations in addition to imperforate anus. The degree of severity could be viewed as a risk indicator for psychosocial maladjustment, though there is no clear evidence that this is true (Pless & Nolan, 1991). Several studies have shown that the severity of the chronic condition is not a predictor of adjustment (Arnaud et al., 2008; Madden, Hastings, & V'Ant Hoff, 2002; Mureau, Slijper, Slob, & Verhulst, 1997). On the other hand, children with inflammatory bowel disease and with higher disease activity scores reported lower HQoL (Perrin et al., 2008).

We did not find any differences in psychosocial outcome in relation to age, and this is perhaps also due to the small study sample. The oldest child involved in *Paper II-V* was under 14. However, it is important to consider children's developmental age when interpreting responses from child QoL instruments (Cremeens, Eiser, & Blades, 2006). In addition, age may be an important factor, especially later in childhood. During adolescence and the teen years the young person has to deal with additional issues, such as sexual identity. It is during adolescence that lifelong patterns of self-management of and adjustment to chronic health conditions are established (Coakley, Holmbeck, & Bryant, 2006).

### **6.1.1 Psychosocial Consequences on the Children**

We have information about the children with imperforate anus from different sources – the children themselves, their parents, and their teachers. The psychosocial consequences on the children with imperforate anus participating in this study appeared to vary, depending on the rater/informant and the measurement used. These varying reports do not only result from one observer being more correct in reporting or assessing child

adjustment, but also probably from actual differences in children's function in different settings. Each report contributes a bit of data to the construction of a more complete picture of the child.

Negative emotional consequences were seen in *Paper I* on the children with imperforate anus, but they were not as evident in *Paper II*. One reason is, of course, that different children were participating. The responses of the children with imperforate anus showed them to have as good psychosocial functioning as the other children participating in *Paper II*. However, the parents rated their children as having totally more behavioral and emotional problems in the CBCL. The results in *Paper II* might verify that a child can be psychosocially competent, but at the same time have some emotional, behavioral, or social difficulties that will not interfere severely with the child's everyday life. This finding is confirmed in a study on the relationship between fecal incontinence and emotional adjustment in a group of children with anorectal anomalies. The prevalence of emotional problems was higher than those in normal populations, though incontinent children were not less well-adjusted than those with bowel control (Ludman & Spitz, 1995). There can also be other reasons for the discrepancies found; when dissimilar views are measured it should be noted that parent- and self-reports of a child's difficulty are the outcome of an accretion from the functioning of the child and the family and parental stress (Sourander et al., 2006).

The proxy results from the CBCL and the TRF might seem contradictory as the dissimilarities between reports by different informants may mirror the perceived variations in child functioning (Achenbach, McConaughy, & Howell, 1987). The children with imperforate anus showed more internalizing syndromes than the children with juvenile chronic arthritis showed on the problem scales in the CBCL (*Paper II*), but they were assessed as being as competent as the other children (*Paper IV*) by their parents. Furthermore, the teachers of children with imperforate anus reported no more emotional and behavioral problems on the problem scales (*Paper II*), though these children were reported, to be somewhat less adapted in school in comparison with the children with no chronic condition (*Paper IV*). This result may indicate that the children with imperforate anus do have some psychosocial difficulties, although the mean scores fell within the normal range in a Swedish youth population (Larsson & Frisk, 1999). Comparable results were found among another group of children with fecal soiling, caused by Hirschsprungs disease. These children had condition-specific psychosocial

problems, though no increase in the rate of clinical psychiatric morbidity (Athanasakos, Starling, Ross, Nunn, & Cass, 2006).

While the teachers of children with imperforate anus seemed to be concerned about the children's function and adaptation, it is likely that teachers do give extra support to these children, though we do not know this. It might be that these children need psychosocial support in school, though they managed well and performed well intellectually (*Paper IV*). Teachers seem to be alert and well informed, and they are considered to provide extremely vital input in the evaluation of several psychiatric symptoms (Constantino et al., 2007).

The differences in reports from parents and teachers of children with imperforate anus (*Papers II and IV*) may possibly be one way to understand the difficulties health care staff can have in observing and noting psychosocial difficulties. The context is, of course, different and usually the meetings are short and are focused on the more physical issues, and this may explain why psychosocial issues are not being recognized. This is confirmed in a study on identification of psychosocial problems; 27.5% of the children were identified as having one or more problems (Horwitz, Leaf, & Leventhal, 1998). Additionally, differences were found in perceptions of children's well-being between health care staff and parents of children with chronic conditions (Janse, Sinnema, Uiterwaal, Kimpen, & Gemke, 2005). When parents and physicians perceived the health status of children with juvenile idiopathic arthritis, parents most frequently provided lower ratings (Sztajn bok et al., 2007).

One cause for the positive results in *Paper II* might be that social attitudes in society toward fecal incontinence have improved in recent years. It is believed to be easier to talk about this subject nowadays, and the taboo has more or less disappeared (Kamm, 2003). In contrast, fecal incontinence was the strongest predictor of QoL scores although QoL was not scored differently between children with Hirschsprung's disease and healthy children (Mills et al., 2008).

As for the children with imperforate anus (*Paper II*), they were not rejected by their peers. On the contrary, it seems as if their friends were supportive and constructive, and not as if the children's disability was a hindrance in the reciprocal process of making friends. Making friends and initiating contact means creating relations with other

children, and close friendship involves relationships with a few others. The children in *Paper I* said they had few close friends. In *Paper II*, the children with imperforate anus reported that they have both close friends and peers. Supportive close friendship has been disclosed to be even more important for children with medical conditions, than peer likeability from classmates (La Greca & Bearman, 2000). Additionally, classmate support may prevent psychiatric symptoms. The children with chronic condition who had high classmate support had lower levels of depression than those children with lower levels of depression (von Weiss et al., 2002). Friends may be one source of support though support from an additional source may give even better outcome. Children (with chronic illness) who were reported to have a great deal of support from both peers and parents showed better adjustment than those with support from only one source (Wallander et al., 1989).

However, there might be an indication of the children with imperforate anus having some difficulties in peer relations (*Paper IV*). On a descriptive level, parental report showed less favorable answers for the imperforate anus children than for the other children, even if no statistical differences were found. The physical consequences of imperforate anus can be regarded as a stigma and may hinder participation in peer relations. Constipation and incontinence are not visible disabilities, but may be revealed by the odor, which is not socially acceptable in most peer groups and is therefore connected to ridicule (Helman, 1995). Though the children with imperforate anus may have a lower ability for initiating contact with friends, this does not necessarily indicate that they have serious problems. However, the result may suggest that parents need to be observant on this issue.

A very interesting finding was that none of the children with imperforate anus admitted to being bullied. Bullying often occurs in school and there is association between having a special healthcare need as a child, and being bullied (Van Cleave & Davis, 2006). Maybe the non-existence of bullying is one explanation of the children's and their parents' positive perception of school issues. However, coping mechanisms such as dissociation might be involved for children with problems like fecal incontinence (Clayden & Wright, 2007).) There are speculations that denial coping means to not acknowledge feelings of being harmed or rejected, thus probably generating no unfavorable opinions about their peers (Sandstrom, 2004). Another reason why all children with imperforate anus denied being bullied may be a methodological matter of

how the question was posed. Subsequently this item should be reformulated in a future study, as is discussed in *Paper VI*.

We assume that the children's peers and their teachers knew about the disability, although we did not ask the children or the parents if they had told the classmates and teachers about the children's disabilities. It is not fully understood when to disclose information about a child's medical condition to classmates. It may have a positive or a negative influence on other children's attitudes, depending on the visibility of the condition (La Greca & Bearman, 2000).

### **6.1.2 Psychosocial Consequences on the Parents**

The psychosocial consequences on the parents of children with imperforate anus seen in this study seemed to be linked with their child's congenital anomaly. In *Paper I*, the negative emotional consequences of the malformation were clear and profound. In particular, the parents' suffering was closely connected to the physical effects of the malformation, including the necessity of performing intrusive procedures such as dilations of the anal canal and giving enemas. It was not surprising that the families had revealed difficulties in *Paper I*. However, the intensity of parental suffering was greater than expected. According to the parents, they had never been asked before about their psychosocial experiences associated with the child's malformations, and they seemed to have a need to voice their concerns.

Maybe the parents in *Paper I* were embarrassed and not used to talking about these private body regions connected with excretory and sexual functions. If they had had the opportunity to talk about their burdensome experiences at an earlier stage, their suffering might have been lessened. One reason for the parents not having the opportunity to talk about their experiences could be health professionals' own attitudes, which are crucial in an encounter with parents. Shame is an affect that has been connected to the malformation. A shameful disease may be one factor that contributes to health professionals' avoidance of direct consultations with patients (Gardner & Cook, 2004). If a subject is hard to talk about, it could inhibit the information and communication process between patients/parents and staff. Additionally, imperforate anus could be regarded as an intricate malformation and that could further complicate information exchange between staff and parents (Noll, Spitz, & Pierro, 2001). Some chronic

conditions are visible and easily understood by others while others require constant explanation. Some illnesses are viewed with great sympathy and others are highly stigmatized (Shapiro, 2002).

An additional explanation for the suffering revealed in *Paper I* could be the method used to collect data. In *Paper I*, the parents and their children were interviewed, and in *Papers II-V*, questionnaires were used. The parents told about their experiences for the first time; they were asked and someone listened to their answers. In a study of bereaved parents they said that it helped to talk, that it was healing (Dyregrov, 2004). In an interview study on family health associated with children's chronic illnesses, parents reported that discussions helped them to work through their grief (Hopia, Paavilainen, & Astedt-Kurki, 2005).

In *Papers II-V*, parents' suffering was not expressed as openly. We did not ask the parents straightforwardly about their own psychological well-being; they answered primarily questions about their children's psychosocial function in the questionnaire. Few questions addressed the parents' psychological well-being, though there might be an indication that the mothers in the Index Group were more distressed about their children's disabilities (*Paper III*); they expressed that their social relationships were more affected by the child's inconvenience, and more affected in respect to their child's disability (*Paper V*).

On the other hand, several mothers of children with imperforate anus reported positive experiences in connection with their child's condition in *Paper V*. How the parents perceive and cope with their child and their child's condition is of significance for the child. Parenting has the potential to promote the health and well-being of children (Gage, Everett, & Bullock, 2006). This is further revealed in a study on adolescents with IBD; mothers who were less positive had children who were more depressed and had more difficulties in school and social functioning (Tojek, Lumley, Corlis, Ondersma, & Tolia, 2002).

### **6.1.3 Psychosocial Consequences on Interaction within the Family**

The results of this study indicate that family functioning and communication have been influenced when a child has imperforate anus, including the psychosocial consequences

found on the child and the parents. Parents' functions play a vital role in children's adjustment to chronic illness (Harwood & Eyberg, 2006; McClellan & Cohen, 2007; Wagner et al., 2003). In a study by Hopia and Paavilainen (2005) it was found that caring for a child with chronic illness generates a relationship transformation between family members and the outer world. It appears that the malformation and the unpleasant but necessary treatments of imperforate anus had an impact on some parts of family interaction. However, the basic necessary parent-child bond seemed to be intact; the children with imperforate anus reported being loved by their parents (*Paper II*) and the parents seemed very involved and concerned about the child (*Papers III, IV, V*).

Family function also plays a role for the child's further interaction with other children. The children in our study seemed to have good relationships with friends (*Paper II*), which may originate in their good relationships with their parents. In a review study (Hay, Payne, & Chadwick, 2004) the underlying skills needed for interaction with peers are studied, and there were significant links found to the parent-child relationship. The opposite has been identified in the role of parenting styles in children's problem behavior; high levels of psychological control and high affection predicted increase in problems among the children (Aunola & Nurmi, 2005).

Unfortunately we did not ask straightforwardly about the influence of the child's condition on the relationships within the family, although we found that the relationship was close between mother and child in *Paper I* and there were also strained relationships between the parents. There was one less discrepancy in reports from mothers and children in the Index Group and in Comparison Group I than in Comparison Group II (*Paper III*). This may possibly be an indication of a closer relationship between children with chronic conditions and their mothers. The well-being and functional capacities of the parents may have both direct and indirect implications for the situation of the chronically ill child (Goldbeck & Melches, 2005). It has been proposed that children who require help from their parents with their treatment regime (e.g. enemas) might become dependant on the parents (Charron-Prochownik, 2002).

There is a specific item/issue of significance that can be related to communication and interaction between child and parent, and that is will. Both the parents and the child were asked questions on will in general and not specifically (in relation to a specific incidence). Mothers of children with imperforate anus assessed their child to express

their will to a lesser extent than the children with no chronic condition were assessed by their mothers (*Paper IV*). Furthermore, the mothers of children with imperforate anus answered that they did not respect their child's will as much as the mothers of children with juvenile chronic arthritis reported that they did in relation to their child (*Paper V*). The children were asked if they expressed what they wanted in general. When analyzing data at first (*Paper II*), there was a tendency for the children with imperforate anus not expressing their will as much as the children with no chronic condition. When this item was analyzed in *Paper VI* (focusing on questionnaire psychometric properties) these discrepancies could be discerned as well.

It seems as if expression of will of children with imperforate anus in relation to their parents (mothers) is a delicate issue. Will is closely connected to autonomy. *Autonomy* refers to the ability to make decisions for oneself without being controlled by others. The 'autonomy' concept covers two aspects: the right to self-determination and to independence (Funkquist, Carlsson, & Nyqvist, 2005). The parents of children with imperforate anus have to abuse their child's bodily integrity during the child's first years. The invasive treatments, both the dilations and the enemas, that the parents of the Index Group children had been responsible for might cause a direct impact on the parent-child relationship, in regard to the expression of the child's will and the development of the child's autonomy. These parents may have to defy the child's will (and autonomy) by giving the enemas regularly for many years. This is a period when the child takes control of bodily functions related to bladder and stool. Parental feelings of stress during this period can influence the way they set limits for the child (Hägglöf, 1999). The enemas also involve a procedure where the parents (as long as the child is immature) have to check and be constantly aware of the child's bowels movements and to interfere with more private and personal matters.

In general, parents want to encourage their child to become independent and self-determined as soon as possible, and to stress the child's ability to become autonomous. According to a study on children with diabetes the children have to be involved in their treatment so they can take charge of their own health care decisions (Alderson, Sutcliffe, & Curtis, 2006). An important issue for successful progress towards adult care as well as for well-being is that young people are allowed to make their own decisions and take responsibility for their own treatment (Berntsson, Berg, Brydolf, & Hellström, 2007).

In *Paper III* we examined more closely the differences between mothers' and children's answers, which may also be one aspect of parent-child interaction. An interesting finding is that the mothers in the three groups scored higher/more of the item measured, (anger, happiness, sadness) than their child, irrespective of question. This might show the mothers' concerns and emotional involvement in their children's lives, and also the difficulties of proxy ratings. Contradictions in answers may be a result of different reasoning and different response styles rather than interpretation of the items (Davis et al., 2007). Instead of seeing discrepancies of opinions as faults, these different views may be important windows to the functioning of the child or family system (Holmbeck, Li, Schurman, Friedman, & Coakley, 2002).

One of the two specific discrepancies found between mothers' and children's answers in the imperforate anus group (*Paper III*) was the mothers' reporting higher self-confidence for the child than the child her/himself reported. This was a surprising answer, but it may be explained and supported by the findings in *Paper V*, where many mothers of children with imperforate anus reported positive experiences, and they highly valued their child's capacity and functioning. Factors associated with disagreement are different, depending on the direction of disagreement, and may have different explanations; parental well-being should be taken into account (White-Koning et al., 2007).

In spite of the hardship of having a child with a chronic condition, there were optimistic findings revealed on the matters of family interaction. In *Paper V* the parents and above all the mothers of children with imperforate anus answered that the child's condition had strengthened family unity. Maybe this finding is not so unique, though more seldom described than the difficulties experienced. In a study on children with Hirschsprung's disease and their parents, good family functioning was found (Athanasakos et al., 2006). A more positive attitude towards family and stronger bonds were found in a review on positive consequences of childhood cancer (Mattsson, Lindgren, & Von Essen, 2008). In a study on families of children with chronic conditions these families functioned as well or better compared with families with healthy children (Rodrigues & Patterson, 2007). Another indicator of family function might be the divorce rate; just as many or in other words no more parents of children with imperforate anus were divorced (see background variables in Table 3) than in the families with a healthy child.

In *Paper V*, 12 parents (7 mothers and 5 fathers) of children with imperforate anus answered that they had received psychological help. We do not know if the parents received the help jointly or individually. Furthermore, it was not found that psychological support was related to positive outcome. What kind of psychosocial help can be of help should be examined more closely. Psychosocial help should not be given to just one member of the family; family support, structure, and organization are associated with better adaptation among young people with medical disorders (Moos, 2002). Optimizing family and parent functioning would have a positive impact on the psychosocial adjustment of the child with a chronic condition. Mothers of critically ill children who received a special parent empowerment program experienced enhanced maternal function that gave positive effects on child adjustment (Melnyk, Crean, Feinstein, Fairbanks, & Alpert-Gillis, 2007). Even though interventions should be family-focused, children have to be active participants in the management of their conditions (Mitchell et al., 2007). Positive impact on children with different chronic conditions appeared after a psycho-educational intervention (Last, Stam, Onland-van Nieuwenhuizen, & Grootenhuis, 2007).

## **6.2 METHODOLOGICAL CONSIDERATIONS**

Both quantitative (*Papers II-V*) and qualitative methods (*Papers I and V*) have been used in this thesis. *Paper I* is considered exploratory, using interviews to collect data. A qualitative approach was also implemented on parts of the data in *Paper V*. Qualitative research responds to the “how” issue, and could give deeper understanding of human experiences and of why people do as they do (Rowan & Huston, 1997; Wolf, 2003). It is worthwhile to combine methods of analysis because each method supplies different information, and by integrating qualitative and quantitative methods a fuller understanding of variables will be obtained (Mitchell et al., 2007). The approach of combining methods may improve the clinical and ethical utility of survey findings, which in turn will increase patient care (Flemming, Adamson, & Atkin, 2008).

The informants in *Paper I* were chosen because of specific motives; one of these was to gather information about the problems the family had experienced that were connected with the malformation. A good interviewee has to be knowledgeable, cooperative, and informative (Kvale, 1996). A natural question is whether the sample of eight informants to interview was big enough. It would have been preferable to have more informants.

However, there were in fact no more informants that fulfilled the sample criteria to interview. The researchers chose a method for collecting data to get the information needed for the study (Patton, 2002). The researcher decides on the sample and on what informants to include – a so-called strategic sample. A strategic sample demands careful consideration of the parameters of interest (Silverman, 2001).

The method for analysis of the interviews in *Paper I* was “editing analysis style”, resembling content analysis. When the work is inductive, the researcher looks for prominent patterns in data (Silverman, 2001). In *Paper V*, manifest content analysis was used in the study-specific questionnaire to analyze the parents’ comments on their positive experiences. The manifest content analysis comprises both numeric and qualitative aspects. Analyzing for the purpose of understanding appearance of a particular content in textual material may be referred to as manifest content analysis (Hsieh & Shannon, 2005).

Questionnaires were used (*Papers II-V*) as being a recognized way to collect data from a group of respondents in quantitative research (Polit & Beck, 2004). Various informants of the child’s function were included, primarily reports from mothers and fathers who each answered an identical study-specific questionnaire along with the CBCL. Parents are known to be raters of their children’s health status in pediatric settings. Mothers’ perceptions of their (young) children corresponded with the actual health status of the children, as reflected by the presence or absence of selected health problems (Monette, Seguin, Gauvin, & Nikiema, 2007). Even though child/patient self-report is advocated, there remains a fundamental role for parent proxy report in pediatric research. Parents are viewed as appropriate proxy respondents to provide information concerning the child’s illness and quality of life (Barr, Thibeault, Muntz, & de Serres, 2007; Sherifali & Pinelli, 2007; Varni, Limbers, & Burwinkle, 2007b). Both the child and parent perspectives must be taken into account to understand the impact on the child, as well as for nurses to understand the physical, emotional, and social consequences (Noyes, 2007). Proxy versions are useful, though the child should be considered the first informant of his/her HRQoL (Hutchings et al., 2008). The value of including mothers and specifically fathers in childhood is underlined (Kerr, Lunkenheimer, & Olson, 2007).

Few discrepancies were found on a pair-level between mothers' and fathers' responses within groups in our study. One reason for this might be that the parents filled in the questionnaires together at home. There seems to be variations on how mothers and fathers usually report on their child's function. Fathers' and mothers' reports of child behavior on the Strengths and Difficulties Questionnaire correlated, and there was high agreement on normal behaviors but lower on abnormal behaviors (Dave, Nazareth, Senior, & Sherr, 2008). Another recent study on Norwegian families showed moderately high correlation between fathers' and mothers' reports on their schoolchildren (Jozefiak, Larsson, Wichstrom, Matthejat, & Ravens-Sieberer, 2008).

The study design included two Comparison Groups. The reasons are that this is a project with clinical implications as the subjects are children with a chronic condition, and it is reasonable to assume that children with chronic conditions coming to a hospital will have different needs than will children with non-chronic disorders. Due to this two Comparison Groups were included.

One Comparison Group consisted of children with another chronic condition and their parents. The correctness of choosing and comparing children with juvenile chronic arthritis and their parents with the Index Group can be argued. Caring for a child with juvenile chronic arthritis is different from caring for a child with imperforate anus. Juvenile chronic arthritis is an illness that may affect the child in a special way; pain is often a penalty (Ross et al., 1993) and it is an unpredictable disease that can cause long-term disability (Peterson, Mason, Nelson, O'Fallon, & Gabriel, 1997). However, both groups of parents had to manage and cope with a condition that started very early in their child's life and the children in both groups had been subjected to invasive treatment, probably including pain and emotional stress.

To compare children with chronic illness to healthy children from the general population (without experiences from hospital care) could be argued to be irrelevant in this case. Average, normal children may show symptoms of stress and psychosocial problems of different kinds, for various reasons. As the project also has clinical aims, children's opinions on the quality of treatment and care are essential to assess. A Comparison Group recruited from the average population would therefore not be a commensurable group. Due to this, children and parents in Comparison Group II were

recruited from the outpatient surgery unit where they were treated for minor surgery problems. Otherwise they were healthy children with no chronic disorder. Such a procedure can of course be criticized, but is relevant as children with chronic and minor medical problems and in need of hospital care are of interest when improving care and treatment.

Finally, the sample size should be commented upon. The number in the Index Group is small, but the group is representative of the patients with high and intermediate imperforate anus, treated according to the current routines. They constituted all patients, born between 1987 and 1992, treated in the Stockholm area and affiliated regions. Twenty-five out of 30 available patients, one had moved abroad, represents about one quarter of all patients in the country.

The final *study* in the thesis (*Paper VI*) contains strictly methodological aspects. In order to have a more elaborate evaluation of the children's own opinions, the exploratory interviews with the children and the parents (*Paper I*) were used to develop a questionnaire for the children themselves. Using the traditional way (the Chronbach's alpha) to assess the psychometric properties, the internal consistency of the psychosocial domains was found to be sufficient (*Paper III and Paper IV*). As this project should be looked upon as a pioneering study to include opinions of children with imperforate anus and to develop a questionnaire for clinical use and for further research, a Rasch analysis was performed. By using the Rasch analysis, an indication was provided on how well each (psychosocial) item measured the latent construct across levels of the construct in focus (Lambert et al., 2003). The results of *Paper VI* support the previous assessments of the questionnaire and its usefulness but also indicate the need of further revisions.

### **6.2.1 The Study Measurements**

*Papers II-V* are considered retrospective cross-sectional studies with comparison of groups, and study-specific questionnaires were used to collect data, as well as the CBCL and the TRF. Because of a lack of available tools to assess psychosocial consequences of imperforate anus on children, we designed a questionnaire specifically for the children with imperforate anus, and one for their parents. Many Quality of Life

instruments have restricted specificity to one illness group, and there are QoL questionnaires focusing on other chronic conditions such as asthma or juvenile chronic arthritis (Norrby, Nordholm, & Fasth, 2003; Reichenberg & Broberg, 2001).

Our main focus was on children with imperforate anus and their parents, although our ambition was to create a measurement instrument that could be used by, and be suitable for Comparison Groups as well, including the group of “healthy” children. The first questionnaire attempt was followed by three testing processes on the questionnaire performed on somewhat younger children and their parents, comprising the same groups of children and parents as in the future main study. Even these younger children had no problems answering the questions. Revisions were made primarily for the group of children with no chronic condition, and after minor linguistic changes the questionnaires were completed

The questionnaires seemed to have reached *content validity*. Content validity is similar to face validity in that it relies on the judgment of the researchers. Expert opinions, literature searches, and pre-test open-ended questions helped to establish content validity to determine if a measure provides adequate coverage of a topic (Frost, Reeve, Liepa, Stauffer, & Hays, 2007). In the children’s questionnaire construct validity seemed to be recognized on the psychological dimension, as presented in *Paper VI*.

Validity is crucial when questionnaires are performed and evaluated. Validation involves collecting “evidence” to understand required data. How different forms of validity are labeled depends on the authors (Kazdin, 1998) and there are no different types of validity other than validity evidence (Standards, 1999). Even when a standardized questionnaire is used, validity should be reassessed. Scores are not valid or invalid, but involve more or less validity to strengthen the interpretation proposed (Waters, Stewart-Brown, & Fitzpatrick, 2003).

The Swedish version of the Child Behaviour Checklist (CBCL) was available to measure children’s psychosocial problems (Larsson & Frisk, 1999). CBCL assesses parents’ ratings of their children’s emotional behavioral/emotional problems and competence. The CBCL is a well-used, well-known, and well-evaluated questionnaire

(Crone, Vogels, Hoekstra, Treffers, & Reijneveld, 2008; Saxe et al., 2003). However the problem scales in CBCL are “deficit-based” and focus on children’s problems; the competence scales are more focused on the children’s strengths. The use of this instrument for children with chronic illness has been discussed among researchers, citing its considerable limitations (Harris, Canning, & Kelleher, 1996) and its limited assessment of psychosocial adjustment (Mackner & Crandall, 2006). On the other hand these instruments are very well-established (Bilenberg, Petersen, Hoerder, & Gillberg, 2005; Najman et al., 2007). CBCL is one of the best-studied instruments for the evaluation of children’s and adolescent’s psychopathology (Roessner, Becker, Rothenberger, Rohde, & Banaschewski, 2007).

### **6.2.2 Scales**

In the parents’ questionnaire we used the Visual Analogue Scale (VAS). It represents a free choice on a continuum including anchor words at both ends. VAS has been pointed out to be a risky scale because it is hard to calculate, but with the help of statistical programs it is more manageable (Clark & Watson, 1995). VAS has been criticized for being problematic, especially if the anchor words are not well-defined. It is, however, helpful for introducing health statuses and for defining ordinal preferences (Torrance, Feeny, & Furlong, 2001). When the VAS scale with words and the Likert scale with numbers were used to measure subjective phenomena such as pain and anxiety, VAS showed the best validity (Ponce de Leon, Lara-Munoz, Feinstein, & Wells, 2004). When a modification of the VAS scales were used to describe pain, no differences were found between children and adults (Hamunen, Maunuksela, & Olkkola, 2008). Internal reliability and construct validity was demonstrated when a VAS instrument was used for children/patient self-report and parent proxy report (Sherman, Eisen, Burwinkle, & Varni, 2006).

In the children’s questionnaires, Likert scales were used. A Likert scale consists of a number of items that articulate a standpoint on a topic. These scales are commonly used with distinct numerical steps from four to nine. A good indicator of preference is how easy an instrument is to use; irrespective of age, children favored the Likert scale and found it the simplest to complete (van Laerhoven, van der Zaag-Loonen, & Derkx, 2004). Facial expression scales are recommended when asking children questions on social items such as friends and family (Cremeens, Eiser, & Blades, 2007).

There have been discussions of what age children must have reached in order to respond reliably to questions in questionnaires. Our impression from this study is that the children responded easily to the items. We do not know, however, whether the parents assisted in answering the questionnaire, except for one mother who admitted that she helped her daughter to fill in the questionnaire. It was revealed that schoolchildren at the age of eight had the ability to reply to a five-point response format on all aspects of their health (Rebok et al., 2001). In another study where children answered questions about their respiratory health experiences, it was argued that children at age ten and above were considered to be as valid informants as their parents (Yu & Wong, 2004). However, five-year old children can report their HRQoL with an age-appropriate instrument, taking into account validity and reliability (Cremeens et al., 2007). Children aged 5-16 answering HRQoL instruments interpreted the items in a similar manner, regardless of their respective ages (Limbers, Newman, & Varni, 2007).

The different scales used in child and parent questionnaire – Likert and VAS – made comparison a bit more complicated. It would probably have been easier to use Likert scales for both children and for parents. Anyway, it has been confirmed that correlation was found to be good enough between the Likert and the VAS formats of a questionnaire (Lukacz et al., 2004).

### **6.3 THE NON-RESPONDENTS**

There were a number of families who did not complete the study. The majority (36%) of non-respondents were represented in the second Comparison Group, the group of children with no chronic condition and their parents. Beside the participants, eighteen families in this group agreed to participate in the study, but never returned their questionnaires. Two reminders, including a telephone reminder, were sent to those families who did not return their questionnaires. In the group of families with a child with no chronic condition, the non-responding families might have been less concerned about participating than the other two groups who had children with chronic conditions. In the group of families of a child with JCA, 32% families did not complete the study. In the Index Group, the number of non-participants was the lowest; 5 out of 30 families (17%) did not complete the study. We do not know the

exact reasons for non-responding (in any of the groups). In two other studies, the most important determinants for non-participation were cultural, social demographic factors, and lifestyle behaviors (Goldberg et al., 2001; Goss, Rubenfeld, Ramsey, & Aitken, 2006). It was found that clinical variables such as disease and disease severity did not seem to influence response activity (Hartman et al., 2007).

The teachers were hard to reach and unfortunately only a few (altogether 36) teachers answered the TRF. The main reason for the low number of participants among the teachers is that several parents in the three groups involved did not permit us to contact the teachers. For this reason a number of teachers were never asked for participation. i.e. 10 teachers in the Index Group, 15 in Comparison Group I and 17 teachers in Comparison Group II.

## 7 STRENGTHS AND LIMITATIONS

This study will provide additional knowledge on a rare disease. So far, the psychosocial aspects of imperforate anus have not been sufficiently studied and described. This project commenced with an exploratory study, and this is considered its core strength. The exploratory study gave us the foundation; it is the basis of the following studies. With the knowledge from the informants participating in the exploratory study we got invaluable information about the imperforate anus phenomenon. The interviews may have comprised too many questions (88) for the parents, though they were necessary to obtain the informants' experiences. This information made it possible to create new pertinent questions for the study-specific questionnaires.

The different perspectives on the child's function are considered to be another strong point. Reports from various sources are represented – those from two Comparison Groups and the completion and use of all three of the child, parent, and teacher reports. In detecting psychosocial problems, a combination of parent and child self-reports should be applied. A more inclusive outcome would be seen if children and parent-proxy questionnaires are combined, and it is recommended to use more than one source in preventive child health care (Raat, Mohangoo, & Grootenhuis, 2006; Sherman et al., 2006; Vogels et al., 2003). The value of multi-informant assessment is supported, and specifically the inclusion of fathers in childhood research (Kerr et al., 2007).

Moreover, the qualitative approach in *Paper V* gives a deeper and additional understanding of what it means to have a child with imperforate anus. This is a significant contribution to make to the evidence base for nursing that would not have been possible by simply using quantitative methods.

There are limitations with this study; the core restriction is the small sample size, which reduced the possibility to draw broad conclusions from the study. However, as pointed out earlier, all available children in the region were included, and this could be considered a strength. Other main limits are the not fully validated study-specific questionnaires used for the children and the parents. Overall, it is not recommended that researchers develop their own questionnaires; this should be used only as a last alternative (Switzer, Wisniewski, Belle, Dew, & Schultz, 1999). At the time of the

study no relevant questionnaires existed, so these study-specific questionnaires were created. There can be arguments for new scales when respondents are special (Spector, 1992). When special conditions and treatments are to be evaluated, an illness-specific questionnaire can be suitable (Eiser & Morse, 2001b). On the other hand, the internal consistency was found to be sufficient in child and parent questionnaire (*Papers III and IV*). Additionally, the evaluation with Rasch analysis on the psychosocial domains in the child questionnaire showed reasonable psychometric properties (*Paper VI*). This result also indicated credibility for further psychometric testing and is considered a strong point. The questionnaire has the potential to become a measure for psychosocial functioning of children with imperforate anus.

## **8 CLINICAL IMPLICATIONS**

The findings and the knowledge obtained from this study indicate the importance of psychosocial issues. Psychosocial aspects should be implemented in everyday nursing. Psychological support and counseling may be contributing factors to help parents manage and understand their child's condition. Providing nursing care to optimize family and parent functioning would have a positive impact on the psychosocial adjustment of the child. A health care plan will be developed from the findings in this thesis.

From the very beginning nurses and other health care personnel need to be aware of the psychosocial aspects. There is a need to be delicate and create optimism around the time of birth and diagnosis when a child is born with impairment and/or when a child is diagnosed with a chronic condition during child's first years. Nursing interventions include assessing family perceptions and coping abilities and preventing strategies; they have a vital mission to increase parents' progress into adjustment, promising an optimistic perspective that will benefit the child. Parent support groups could be a key intervention for the family who has a child with chronic illness.

What kind of help is required and when it should be performed has to be further explored. Support and psychological help may be necessary; it should be individualized, most probably by those who are knowledgeable about the child's condition. Professionals should recognize and be aware of how a child's specific condition impinges on the entire family. As mothers (still) seem to be the main care provider, fathers' involvement needs to be underlined.

Child autonomy was found to be a delicate issue and could be considered in clinical care. It is of importance to encourage the child to become independent and self-determined but also to support the parents in this process, to advance the children's coping skills, and to encourage their attainment of an active role in making decisions for their health care. Overall, it is important that the child's strengths are observed and acknowledged.

Imperforate anus necessitates an understanding of the nature of the biological issues facing the child, as it impacts on and interacts with the course of the child's life, and on

his/her family, depending on the society they live in. Understanding the psychosocial consequences of this special malformation can help in forming a collaborative relationship between professionals who care for the child so that biomedical and psychosocial interventions are integrated. Usually nurses and doctors in the pediatric wards and outpatient clinics supply sufficient psychological care, and the specialist nurses are highly qualified to take care of these children and their parents. When extra resources are needed, it is of importance that the psychiatric health professionals are informed about the child's specific malformation and the treatments being used.

## 9 FUTURE RESEARCH

More learning in the field of children's chronic conditions is required and the limited quantity of studies on the influence of imperforate anus, also including our results, indicates the necessity of more knowledge on this topic. There is still much to be learned about the short- and long-term consequences of imperforate anus, and there is a need for additional studies on these children and their parents, preferably within an expanded group that could also include children with low imperforate anus. It would then be possible to focus on potential gender differences between the reactions of mothers and fathers. In such a study the children's and parents' personal attributes and cognitive and emotional resources should be more fully described to better understand coping mechanisms for implementing the proper interventions.

A longitudinal study is suggested; the children participating in this study need to be followed up. It would be of vital interest to study how the children participating in this study attain psychosocial adaptation during the transition to adolescence, and how they manage to cope with the disability and also with autonomy. During the teenage years, sexual identity and intimate relations including sexual function is usually initiated and established and that would be of interest for further study. The implications of age would then be studied as well as to what extent the child's malformation has an impact on the parents.

An additional and interesting view could be to include the siblings of children with anorectal malformation. The siblings could give another supplementary perspective on the influence of the malformation on family function, in addition to that provided by the children with imperforate anus and their parents.

School issues seemed to have a favorable valor for the children with imperforate anus and specifically for their parents (fathers). School issues could be further examined, to better understand the importance and positive impact the school context may have, including the significance of peer relations.

Tool development is another field to for research. The findings indicated that the child self-report questionnaire is good enough for future use and may likely become a measure for psychosocial functioning of children with imperforate anus. The

questionnaire has to be reformulated to some extent, involving removal of unnecessary items and making the items more “difficult”. The additional and unexamined domains (the physical aspect and the experiences of care) in the child questionnaire as well as the parent questionnaire also need to be examined and psychometrically evaluated.

To evaluate the study-specific questionnaire in a follow up would be a challenge; it was developed and evaluated among children between the age of 8 and 13 years. It is presumable that the experiences of imperforate anus among adolescents/teenagers may differ from those of younger children.

The last but not the least, and maybe the most vital field for nursing care, is to do psychosocial interventions, to find out the most effective support for the families of children with imperforate anus, both for the children and for their parents. One proposal for the children is to let them meet in mixed age groups to find mentors for the younger children, as well as implement psycho-educative involvement. The other urgent concern would be to find the appropriate interventions for parents of children with imperforate anus to reduce their distress. One potential application is that parents can meet in parent groups, exchange experiences, and support each other. Such a group could include professional psycho-educational interventions as well.

## 10 CONCLUSIONS

- Parents of children with high imperforate anus have to go through difficult experiences associated with the malformation.
- According to self-report questionnaires, the children with imperforate anus seem to be psychosocially well adjusted, though their parents state that they experience leakage and fecal odor.
- The children with imperforate anus might have some psychosocial difficulties, though informants do not agree. It is vital to assess the children's psychosocial function from more than a single informant.
- Parents of children with imperforate anus seem to be psychosocially influenced by their children's condition even though positive experiences may also ensue.
- Further research is needed on and about children with imperforate anus and their families. Aspects related to child autonomy need more consideration, as do school issues. There is a need for development of measurement tools and a special need for clinical nursing interventions.

Psychosocial issues are essential for both children with imperforate anus and their parents. Qualified individualized advice and assistance should be a vital part of their continuing care, and psychosocial interventions are needed. Collaboration with expertise from child and adolescent psychiatry may be required.

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