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High and Intermediate Imperforate Anus
A study on children and their parents,
concerning aspects of care, psychosocial
function and experiences of treatment

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Institutet**

Stockholm 2007

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Published and printed by US-AB
Box 200, SE-171 77 Stockholm, Sweden
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ISBN 978-91-7357-282-8

To Lars, Emilia, Viktor, Oskar

ABSTRACT

The overall aim of this thesis was to examine how children with high and intermediate imperforate anus and their parents have been affected by the malformation.

Materials: For *Paper I* three patients with high imperforate anus and their families were interviewed. The patients were born before 1987 and at the time of the interview they were 12, 14, and 22 years old. *Papers II-V* are based on the same study sample. Twenty-five children with high or intermediate imperforate anus born 1987-1992 and their parents participated in the index group. Two groups of children similar in age and gender distributions, along with their parents, served as controls.

Methods: In *Paper I* an exploratory study was performed with interviews. An interview guide that consisted of open-ended and structured questions was used. The sessions covered many different aspects of the child's malformation, as well as treatment and psychosocial consequences of the defect. In *Papers II-V* individual questionnaires were used for the children and the parents. The questionnaires were developed on the basis of the previous interview study and covered psychosocial domains and physical domains. An additional questionnaire was used consisting of items about the child's experiences of the medical treatment in early age. To look for emotional or behavioral problems among children, the Child Behavior Checklist (CBCL) was used and filled in by the parents, while teachers filled in the Teacher's Report Form (TRF).

Results: In the analysis of the interviews four categories related to effects of the malformation emerged; they were physical, emotional, social, and family. The physical and emotional effects of the child's malformation influenced the family's experiences. Children with imperforate anus exhibited an overall positive attitude on the questionnaires; their psychosocial function seemed to be good even though the children had problems with their bowel functions as compared to the control groups. In the CBCL, the children with imperforate anus were assessed by their parents to have significantly more emotional/behavioral problems than were assessed by parents of the children in control groups. In the TRF, the teachers reported few symptoms for the index group children. Parents reported about extensive care of the child, saying that they had been very much involved in the follow-up treatment. The parents of children with imperforate anus were less satisfied with information received, compared with the parents in the two control groups. There was a gender difference on how responsibility for the child's care was divided between the mother and the father. The children had non-verbal memories of medical treatment early in life, according to their parents' reports.

Conclusions: Parents of children with imperforate anus had experienced suffering and many difficulties associated with the malformation. Children with imperforate anus in this study did not seem to have any psychosocial impairments, though their parents stated that they experienced leakage, constipation, and fecal odor. The children might have some psychosocial difficulties, according to parents, but not according to their teachers. Parents of children with imperforate anus experienced less satisfaction with medical care and reported that the support received had been insufficient. The mothers of children with imperforate anus seemed to be the main caregivers. Talking to your child about medical treatment at an early age seems to relate positively to the child's psychosocial function. The conclusions of this thesis may lead to an improved understanding of the care for the children with imperforate anus and their parents.

Keywords: imperforate anus, children, fecal incontinence, psychosocial function, care, gender perspective, experiences, non-verbal memories.

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- I. Nisell M., Öjmyr-Joelsson M., Frenckner B., Rydelius P-A., Christensson K.
How a family is affected when a child is born with anorectal malformation.
Interviews with three patients and their parents.
Journal of Pediatric Nursing, 2003,(18) 6,1423-32
- II. Öjmyr-Joelsson M., Nisell M., Frenckner B., Rydelius P-A., Christensson K.
High and intermediate imperforate anus: Psychosocial consequences among
school-aged children. *Journal of Pediatric Surgery*, 2006,41(7),1272-1278
- III. Öjmyr-Joelsson M., Nisell M., Frenckner B., Rydelius P-A., Christensson K.
Parental experiences: Care of children with high and intermediate
imperforate anus. *Clinical Nursing Research*, 2006,15(4):290-305
- IV. Öjmyr-Joelsson M., Nisell M., Frenckner B., Rydelius P-A., Christensson K.
A gender perspective on the extent to which mothers and fathers each take
responsibility for the care of a child with high and intermediate imperforate
anus. *Resubmitted to Journal of Pediatric Nursing after suggested revision.*
- V. Öjmyr-Joelsson M., Christensson K., Frenckner B., Nisell M., Lindholm T.
Children with high and intermediate imperforate anus: Remembering and
talking about medical treatment carried out early in life. *Resubmitted to
Applied Nursing Research after suggested revision.*

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LIST OF ABBREVIATIONS

CBCL	Child Behavior Check List
IA	Imperforate anus
JCA	Juvenile Chronic Arthritis
MACE	Malone's Antegrade Continence Enema
PSARP	Posterior Sagittal Anorectoplasty
TRF	Teacher's Report Form
VAS	Visual Analogue Scale

1 INTRODUCTION

The birth of a child with a congenital malformation is often a traumatic experience for the parents. The transition into parenthood becomes something other than what could be expected, and negative emotions and feelings of shock are common. A child's birth is one of the most important experiences in parents' lives. It is normally associated with feelings of joy, but when the newborn child is ill and needs hospital care the presumably positive birth event is transformed into another dimension (Hall, 2005b; Hopia, Tomlinson, Paavilainen, & Astedt-Kurki, 2005; Ludman & Spitz, 1996).

In our daily encounter with children with imperforate anus and their parents it has become obvious that the malformation is very complex. The surgical aspects of the malformation require highly qualified pediatric surgery, but besides this treatment there are several other very important aspects of this malformation that have to be considered. The imperforate anus malformation has not been described from a nursing perspective before, and aspects of caring for these children are still unexplored. The children with imperforate anus and their parents have a close and regular contact with the specialist team, including the bowel therapy unit. This connection is required, as the malformation involves several new demands on the parents on how to perform advanced medical care at home.

Imperforate anus is an uncommon congenital malformation of the anus and the affected child may have several difficulties at birth that derive from the lesion (Peña, 1993). Imperforate anus has been described in literature since ancient times, and various surgical interventions are mentioned as lifesaving measures (Stephens & Smith, 1971). The surgical method used to reconstruct the new anus is advanced nowadays, and together with the improved follow-up treatment makes it possible for a child with imperforate anus to have a fairly normal life.

Few studies on caring for a child with imperforate anus have previously been published. The psychosocial consequences of the malformation and how it affects the families' lives are also very seldom described. In our work at the pediatric surgery unit, a constantly recurring group of patients for follow-up visits are the children with imperforate anus. The support these families need may be underestimated since the care

of children with imperforate anus is demanding and time-consuming. As professionals we cannot always imagine how these parents strive to get their normal family lives to work. This study was aimed at investigating experiences of children with high and intermediate imperforate anus and their parents – how they have experienced care and treatment and what consequences the malformation involves.

2 BACKGROUND

2.1 IMPERFORATE ANUS

Imperforate anus is a malformation of the anorectum. Historically this malformation has been known since antiquity. One of the first surgeons mentioned who dealt with it was Paul of Aegina (A.D. 625-690). He performed an operation to open an orifice into the rectum, then kept it open with dilations (Stephens & Smith, 1971). The surgical technique improved and a second landmark was when Amussat developed the proctoplasty in 1835 (Stephens & Smith, 1971).

2.1.1 Incidence and classification

The incidence of imperforate anus is one in every 4-5000 children born. The malformation is somewhat more frequent among boys than girls. The most common defect among boys with imperforate anus is the rectourethral fistula and the most common defect among girls with imperforate anus is the rectovestibular fistula (Peña, 2000). Classifications of anorectal malformations differ in literature, though the severity can be divided into two categories – high and low lesions. In the high lesion, the development of the rectum has closed above the levator muscle. In the low lesions, the development of the rectum has closed below the levator muscle. There is also one type of imperforate anus that can be categorized as intermediate; it does not fit into either of the groups mentioned above. These intermediate malformations are generally treated as high lesions (Kiely, 1998).

An international classification of imperforate anus was proposed in 1970 (Stephens & Smith, 1971) and some years later the Wingspread Conference defined classification, identification, and assessment of surgical treatment of anorectal anomalies (Stephens & Durham Smith, 1986). Experienced surgeons Kiely (1998) and Peña (2000) have declared that neither of these classifications are useful in practice, and have proposed a more therapy-oriented classification of the malformation that makes a prognosis for the functional outcome (see below, 2.2 and 2.3). At the Krickenbeck conference on anorectal malformations in 2005 the Wingspread classification was still considered useful in choice of surgical approach. A modification of Peña's classification according to the type of fistula, including also rare variants of the malformation, was proposed.

The aim of this new classification was to make it easier to compare different follow-up studies on children with imperforate anus (Holschneider et al., 2005).

2.1.2 Associated malformations

The prevalence of associated congenital malformations is 50-60% in the children with imperforate anus. These associated malformations can include cardiovascular anomalies, gastrointestinal anomalies, vertebral anomalies, and genitourinary and skeletal defects (Kiely, 1998). The most frequently associated malformations are that of the urogenital system and that of the skeletal system (Stoll, Alembik, Dott, & Roth, 2007).

2.2 MALE DEFECTS

2.2.1 Cutaneous fistula

In the low defects the lowest part of the rectum is anteriorly mislocated and placed somewhere along the midline raphe, on the scrotum, or at the base of the penis. The anal fistula is often narrow (Peña, 2000).

2.2.2 Rectourethral fistula

This is the most common defect among male patients. In rectobulbar fistula the fistula is located in the lower part of the urethra. Rectoprostatic fistula is located in the prostatic portion of the urethra. The rectum and the urethra have a common wall above the fistula. A distension of the rectum is common and the most lateral part is surrounded by the levator muscle. The external sphincter muscle complex is located between the rectum and the perineal skin; when contracted it elevates the skin of the anal dimple. Children with low urethral fistulas, rectobulbar fistula, often have well-developed muscles, sacrum, midline, and an anal dimple. In children with higher urethral fistulas, rectoprostatic fistula, the risk is higher for poor muscles, sacral anomalies, and a flat perineum with absence of midline groove and anal dimple (Peña, 2000).

2.2.3 Rectovesical fistula

The rectum opens into the bladder neck with a fistula. Among boys with this condition the levator muscle and the external sphincter muscle complex are generally poorly

developed. The sacrum is often deformed. A flat bottom is often seen because of the poor muscle development (Peña, 2000).

2.2.4 Unusual male defects

Anorectal agenesis without a fistula and rectal atresia appear but are unusual defects.

2.3 FEMALE DEFECTS

2.3.1 Cutaneous fistula

The lowest portion of the rectum is located just in front of the normal place for the anus though the rest of the rectum has developed all the way through the muscle complex and is located within the sphincter mechanism. Rectum and the vagina are well separated (Kiely, 1998).

2.3.2 Rectovestibular fistula

The rectum opens in the vestibule of the female just behind the hymen. The vagina and the rectum are separated by a thin wall. These girls usually have well-developed muscles and a normal sacrum (Peña, 2000).

2.3.3 Rectovaginal fistula

This is a rare malformation that affects only 1% of all cases. The rectum opens into the vagina and the malformation can be low or high (Kiely, 1998).

2.3.4 Persistent cloaca

The definition of this defect is that the rectum, vagina, and urinary tract are fused together into a common channel. The length of this channel can vary but if the channel is longer than 3.5 cm the defect can be classified as a complex defect. It is not unusual to find varying degrees of septated or duplicated vagina and uterus (Peña, 2000). Associated urological defects appear in 90 % of girls with this high defect (Peña, 1990). Sacral defects and underdeveloped muscle complex are often seen.

2.3.5 Unusual female defects

Imperforate anus without a fistula is very rare in girls.

2.4 SURGICAL TREATMENT

A colostomy is generally done in a child with high or intermediate imperforate anus when the child is newborn to divert the fecal stream (Peña, Migotto-Krieger, & Levitt, 2006). The colostomy provides decompression of the bowel and acts as protection for the final repair of the malformation (Peña, 2000; Peña et al., 2006). The method of choice for construction of the new anus is generally the posterior sagittal anorectoplasty (PSARP). The surgical incision is made between the patient's buttocks. The patient is placed in a prone position with the pelvis elevated. The external sphincter is located with electrical stimulation. The incision is then made exactly in the midline through all of the posterior musculature. The rectal pouch is identified and the fistula is dissected and closed. The rectal pouch is mobilized and the different muscle layers are closed. The rectum is then placed with precision in the external sphincter and the skin is closed (Kiely, 1998; Peña, 1985, 2000). A laparoscopic approach to dissect the rectum can be used on very high malformations in order to avoid a laparotomy. The laparoscopic approach is then combined with the PSARP (Georgeson, Inge, & Albanese, 2000; Levitt & Peña, 2005).

The colostomy is kept for an additional two to three months while the reconstructed anus is dilated with a Hegar Dilator until the desired size is reached. The colostomy is then closed and the stools can pass through the neoanus.

2.4.1 MACE

Malone Antegrade Continence Enema (MACE) is a surgical method used as treatment for fecal incontinence. The appendix or an ileumconduit provide a continent catheterizable stoma and channel to the colon caecum. The laparoscopic surgical technique has also been found to be a simple method in creating an antegrade continence enema, as well as the use of a gastrostomy button to minimize the risk of a stomal stenosis (Antao, Ng, & Roberts, 2006; Kim, Beasley, & Maoate, 2006; Mousa et al., 2006; Yagmurlu, Harmon, & Georgeson, 2006). The patient takes a antegrade enema through the stoma daily in order to prevent constipation and fecal leakage and to keep clean (Malone, Ransley, & Kiely, 1990; Soylet, Yesildag, Besik, & Emir, 2006).

2.5 FUNCTIONAL OUTCOME

Constipation and fecal incontinence are common problems in patients with imperforate anus and the normal complex interplay between colonic motility, rectal properties, and sensation is affected or missing. Several follow-up studies on this issue have been

performed to evaluate the functional outcome after the surgical procedure. One study on children with high and intermediate imperforate anus showed that 35% of the patients had an excellent bowel functional outcome and 35% had a good bowel functional outcome (Rintala & Lindahl, 1995). Peña (2000) stated that of 1,192 patients, 75% had voluntary bowel movements and 37.5% were totally continent. Another study showed that 96% of children with high and intermediate imperforate anus had a good or fair anorectal function (Tsuji et al., 2002). Furthermore, a recently published study on adults with imperforate anus showed that 66% had good bowel function, and 33% of the patients with high and intermediate imperforate anus had occasional fecal soiling (Iwai et al., 2007). Besides continence, scoring an anorectal manometry to examine the rectoanal inhibitory reflex could give important information about the bowel function of patients with imperforate anus (Senel et al., 2007).

2.6 CARE OF A CHILD WITH A CHRONIC CONDITION

Caring for a child involves protecting the child from danger and providing protection and security. Caring for a child with a chronic condition also involves several other aspects. Hospital care with different treatments may be frightening and cause anxiety to the new parents, and their role as parents becomes something completely different from what they might have expected. The parents' possibility to establish control over the situation is often difficult when the child has a chronic condition. Children with anorectal malformations are a small but exposed group of children about which few attempts have been made to study care both from a perspective of parental care and nursing care. These children need a strict follow-up schedule after the operations, and as functional problems are common a regular treatment to reduce the consequences of the malformation is required. Caring for a child with a malformation or a disability often involves a lifelong responsibility for the child's well-being. It is a process of learning about something completely unknown (Nuutila & Salanterä, 2006). Parents' participation in care and sometimes in treatment both at home and at the hospital are substantial (Hall, 2005a; Lam, Chang, & Morrissey, 2005; Lawoko & Soares, 2004).

When the child is in need of hospital care the parents naturally want to stay at their child's bedside because of their commitment to care. It may not always be clear what parents can do and what the nurses should do and who "owns" the child in the hospital. Parents' feelings of love, protection, and responsibility and their desire to be involved in care make it obvious that they should stay with their children (Dudley & Carr, 2004).

The negotiation between parents and nurses may not always work out due to different opinions on who should do what and how capable the parents are. In a negotiation it is important that parents and nurses act as equal partners in caring for the child (Lee, 2007). Healthcare professionals often assume that parents really want to be engaged in the care of the child with malformations. Parents are therefore often obliged to act as nurses, performing complex clinical procedures on their children instead of being just mothers or fathers (Kirk, Glendinning, & Callery, 2005). Parents' daily care of their own children makes them specialists; they observe small changes in the child's health condition that we as healthcare professionals may fail to see. Multiple dimensions of who really is the expert in care of a child with a chronic condition – the parents or the nurse – have to be considered when the child is in need of hospital care (Ford & Turner, 2001).

Parents' care of a child includes several aspects of physical, social, emotional, intellectual, spiritual, and developmental health. Healthcare professionals' care of the child involves clinical care to ensure physical health. One goal parents and healthcare professionals have in common, though, is to return a sick child to health. This ambition can only be achieved by giving care from a family perspective, as has been described by Miceli and Clark (2005). A family-centered care implies seeing the family as a whole and adjusting care around all of the family members (Shields, 2007). Children staying at the hospital may be exposed to frightening treatments, and they need their parents to be with them all the time. Parents can decrease levels of anxiety as no one else can, and provide a shelter for the child's emotional status.

Parental satisfaction may be achieved if the parents get involved in their children's care, and this can also decrease their feelings of stress. For this reason an open communication between healthcare professionals and parents is needed (Simons, 2002; Youngblut, Brooten, & Kuluz, 2005). Sometimes communication between parents and healthcare professionals is somewhat limited, and a consequence of this might be that parents feel uninformed and insecure. Some studies have shown that nurses often underestimate parents' requests for information (Hall, 2005b; Sobo, 2004). In contrast, it has been found in a recently published study that parents of children with chronic conditions felt that they got more information than they could cope with (Hummelinck & Pollock, 2005).

2.7 PSYCHOSOCIAL ISSUES

Psychosocial function has different meanings and includes many different aspects. The social functioning of a child involves making friends, getting along with other children, assimilating in groups, and participating in activities. However, the psychosocial concept could also include the child's ability to adjust socially and emotionally (Adams, Streisand, Zawacki, & Joseph, 2002).

Imperforate anus and its consequences of constipation and fecal incontinence restrict a child's participation in social activities and may affect emotional adjustment. Parents' care of children during preschool years is presumably extensive, but as the child grows up and starts school problems like incontinence will become more obvious and different psychosocial problems may crop up. It has been found that among children with imperforate anus psychosocial problems have been underestimated (Diseth, Emblem, Solbraa, & Vandvik, 1994). Psychosocial problems such as secretive behavior among the children with imperforate anus to avoid teasing and bullying of classmates were described by Ludman and Spitz (1996).

Functional constipation and fecal incontinence is quite a common problem even in healthy children and may affect the children's quality of life (Loening-Baucke, 2007; Youssef, Langseder, Verga, Mones, & Rosh, 2005). The fecal incontinence among children with imperforate anus has other causes and differs from functional constipation. With the PSARP is not possible to reconstruct the anus and the anal canal exactly where nature has failed. Children with a very high malformation will have a poorer outcome of the function since the muscles often are not fully developed. This will give a poorer function and capability to evacuate the feces and to be continent. Problems with embarrassing odors are unpleasant and may affect the child's social contacts in school, but are not described thoroughly in the literature.

Depressive symptoms among children with a chronic condition have been found to affect the children's social relationships. Sandström and Schanberg (2004) and Pless and Nolan (1991) found that having a chronic illness will increase the risk for psychosocial dysfunction and these problems might affect the whole family. It has been found in an epidemiological study that childhood disability affects the entire family. In order to promote health and psychosocial functioning and to reduce stress symptoms, the family has to be seen as a whole (Warschburger, Buchholz, & Petermann, 2004; Witt, Riley, & Coiro, 2003).

2.8 PARENTAL STRESS

The parental stress of having a child with a congenital malformation or chronic condition is not always related to the severity of the child's problems (Hunfeld, Tempels, Passchier, Hazebroek, & Tibboel, 1999; Uzark & Jones, 2003). It may be other things concerning the child's health and daily care or the parents' own capability of fostering a child that cause parental stress. Furthermore, both lack of social support and financial issues may produce emotional strains among parents and cause conflicts in the marital relationship (Schor, 2003). Experiences of parental stress are shown to affect the attachment and the relationship between parent and child and this also might impede the child's psychosocial development (Pope, Tillman, & Snyder, 2005). Mothers in particular seem to show more stress and strain when the child has a chronic condition (Dellve, Samuelsson, Tallborn, Fasth, & Hallberg, 2006; Gerhardt et al., 2003). The parents' ability to handle the situation and to cope with the child's inconvenience is a challenge for the whole family, and it has been found that the parental functioning both as individuals and partners affect the child's psychosocial adjustment (Friedman, Holmbeck, Jandasek, Zukerman, & Abad, 2004). The knowledge of parental stress among parents of children with imperforate anus is limited.

2.9 MEMORIES

Infantile amnesia is the loss of memory of an event that occurs in early childhood. Before the age of four it is said that children do not have the ability to verbally tell about a memory (Carlson, Buskist, Brunswick, & Martin, 2000). Even so, salient events experienced before the age of four could be remembered. Invasive medical treatment at an early age is often stressful for the children, as they do not understand why the treatment is required. An unpleasant event like a medical procedure is often better recalled than other events but a small child cannot always give an accurate description of a stressful event (Brown et al., 1999). The small child has a limited ability to organize and make sense out of all the impressions he/she receives, and the child's current verbal and communicative skills at the time of the event determine whether he/she can tell about the memories (Fivush, 1998; Richardson & Hayne, 2007).

2.10 CLINICAL EXPERIENCES OF CARING FOR A CHILD WITH IA

Caring for a child with imperforate anus involves great effort for the parents, and both the child and the parents may be affected by the consequences of the malformation. The management of fecal incontinence is difficult, and its social consequences can be devastating. Our clinical work with the children and their parents has revealed the struggle the malformation involves for the family. Parents' anxiety and their worries about different aspects concerning the newborn child and the malformation have to be taken seriously. Our knowledge about these aspects has been limited and this is why this study was conducted.

3 AIMS

The overall aim of this thesis was to examine how children with high and intermediate imperforate anus and their parents have been affected by the malformation.

Specific aim of included papers:

- I. The aim was 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. (*Paper I*)
- II. The aim of the study was to evaluate the psychosocial effects of high and intermediate imperforate anus on school-aged children. (*Paper II*)
- III. The aim of this study was to evaluate parental experiences of care of children with high and intermediate imperforate anus. (*Paper III*)
- IV. The aim of this study was to evaluate a gender perspective on the extent to which mothers' and fathers' each take responsibility for the care of a child with high or intermediate imperforate anus. (*Paper IV*)
- V. The aim of the study was to examine if children with IA showed distressing memories of the treatment they underwent at an early age, and whether talking with a child about their experiences is related to the child's psychosocial functioning. (*Paper V*)

4 DESIGN

This study was conducted to increase our knowledge of children born with high and intermediate imperforate anus and of their parents. Our clinical experiences of working with the children and their parents indicated that there were several unexplored aspects of this malformation. The surgical aspects have been well described but there is a lack of studies on the care of these children and the psychological and the social aspects.

This research project is a product of a fully integrated cooperation between the pediatric surgery unit and the child and adolescent psychiatric unit at Astrid Lindgren Children's Hospital. Such a close cooperation is needed if all aspects of nursing care for patients with chronic conditions and their families are to be incorporated.

We choose to initiate the study by interviewing three selected patients with 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 observing and describing it (Polit & Beck, 2004). The extensive findings from these interviews (*Paper I*) created the basis for questionnaires for children with high and intermediate imperforate anus and their parents. These questionnaires were used as instruments throughout the study in *Papers II-V*.

Two control groups were selected and were used in this study (*Paper II-V*). The first control group consisted of children with a chronic condition, juvenile chronic arthritis (JCA), and their parents. The children with JCA had visited the hospital and gone through different medical treatments. The other control group contained children who had had a minor outpatient surgery and their parents. The children in the second control group had no chronic condition and were otherwise healthy. The use of control groups in this study was made in order to provide a context and to improve the understanding of our findings.

5 MATERIAL

5.1 PARTICIPANTS PAPER I

Three patients with high imperforate anus and their families were interviewed. The patients were 12, 14, and 22 years old at the time of the interviews. There were three mothers and two fathers participating in the study. One father had left the family early on and was therefore not asked to participate since he had not been involved in the child's growing up. All three patients suffered from a severe form of the malformation. Both the primary operation with a reconstruction of the anus and the primary follow-up treatment was done at St Göran Children's Hospital (Stockholm).

Inclusion criteria for participating in this study were: child born before 1987, high imperforate anus, problems with constipation and fecal incontinence, treatment with Malone's antegrade continence enema (MACE) and a willingness to discuss excretory problems they had experienced in childhood. Five children fulfilled the inclusion criteria and were contacted. Two of the patients did not want to take part in the study but three of them were willing to participate.

A stoma nurse first telephoned the families, briefly described the study, and asked them if they were willing to participate. After the children and parents gave their oral consent, they were sent written information about the study. Before the interviews started the family members got an information reminder about the purpose of the study to make them comfortable in talking about their experiences.

5.2 PARTICIPANTS PAPERS II - V

5.2.1 Children

Data in *Papers II-V* was obtained from 25 children with high or intermediate imperforate anus and their parents. The children had their definitive surgery for the anomaly with a PSARP performed at St. Göran Children's Hospital between 1987 and 1992. The original group consisted of 30 children; one of these patients had moved abroad and could not be located. The remaining 29 patients (9 boys and 20 girls) were asked to participate in this study together with their parents. They agreed but four families did not complete the study. The index group therefore consisted of 25 children

with high or intermediate imperforate anus (9 boys and 16 girls). The mean age in the index group at the time of the study was 10.5 years (range 8.0 to 13.6).

Table 1. Types of defects in the Index Group

	Females	Males
	n=16	n=9
Persistent cloaca	5	
Rectovaginal fistula	1	
Rectovestibular fistula	10	
Rectovesical fistula		2
Rectoprostatic fistula		4
Rectobulbar fistula		3

Additional information about the extent of the child's lesion (Table 1) and bowel function at the time of the study was excerpted from medical records and surgical reports. The majority of the children (19) had one or more additional congenital defects.

All of the children were operated with PSARP and fistula-saving technique (Husberg, Lindahl, Rintala, & Frenckner, 1992) by the same surgeon (Björn Frenckner) and the children had close and regular contact with the bowel therapist, the author of this thesis. The children had regular follow-up treatment that included daily dilations with Hegar Dilators until the anorectum reached adequate size, and closure of the colostomy at the age of 6-12 months. Eleven children underwent dilations under anesthesia after the age of 18 months because of anal strictures. Malone's antegrade continence enema (MACE) was performed on five children because of problems with fecal incontinence.

Two groups of children similar in age and gender distributions to those in the index group, along with their parents, served as controls. Control Group I consisted of 30 patients (5 boys and 25 girls) with Juvenile Chronic Arthritis (JCA) debuting before two years of age. The mean age was 10.6 years (range 7.8 to 13.6). Control Group II comprised 32 healthy (without chronic diseases) children (14 boys and 18 girls) who

were in the outpatient surgery unit for a minor procedure, e.g. to repair a hernia. The mean age was 10.9 years (range 8.3 to 13.9). In *Paper V* children in Control Group II were excluded since these children did not have a chronic condition and had received their diagnosis just recently, and therefore were not able to have memories of any medical treatment early in life due to their diagnoses.

5.2.2 Parents

Forty-five parents of children with high and intermediate imperforate anus participated in the index group. There were 25 mothers and 20 fathers in the group. Of the five missing fathers, one had died and the other four did not complete the study. Fifty-five parents of 30 children with juvenile chronic arthritis participated in Control Group I. There were 30 mothers and 25 fathers. Control Group II consisted of 52 parents of 32 healthy children. In this group there were 32 mothers and 20 fathers. The parents in Control Group II were excluded in *Paper V* since their children did not suffer from a chronic condition.

6 METHODS

6.1 INTERVIEWS, PAPER I

The interviews were conducted at the hospital by two of the authors of *Paper I*. Individual interviews were made by a registered nurse (Margret Nisell) and a child and adolescent psychiatrist (Per-Anders Rydelius.). Neither of them had been involved earlier in the children's treatment or follow-up. The interview guide consisted of open-ended, structured questions, and the sessions covered many different aspects of the child's malformation and treatment and of the defect's psychosocial consequences. The parents and the patients described their own perceptions of the situation. The interviews, between one and two hours long, were tape-recorded and transcribed.

6.1.1 Analyses of the interviews

The transcribed interviews were analyzed with a qualitative method, content analysis (Crabtree & Miller, 1999; Polit & Hungler, 1999). Two of the authors (Margret Nisell and the author of this thesis) read and reread the transcribed interviews. This was done to get a comprehensive impression of the text's contents. The two authors proceeded separately on the analyses with coding units, marked with different colors, according to content. These coding units, consisting of sentences or paragraphs, were grouped into categories identifying phenomena that emerged from the interviews (Morse & Field, 1998). This analysis was aimed at uncovering the meaning of the interview responses, and finally these responses could be broken down into four categories (physical, emotional, social, and family) based on the effects of the malformation.

6.2 QUESTIONNAIRES, PAPERS II-V

The instruments used to gather data were questionnaires developed on the basis of the previous interview study. The results of the interviews showed which questions were of concern to the children and their parents and therefore essential to creating the self-structured questionnaires. The children's and parents' questions were mostly congruent. The questionnaires were tested on both children and their parents in three subsequent pilot studies. Minor revisions of the questionnaires were made after each pilot study and eventually they were completed.

6.2.1 Children's questionnaire

The children's questionnaire consisted of 45 items that were divided into five psychosocial domains (school, sport activities, peer relationships, self-esteem, and emotional health) and one physical domain. The items were scored on a five-point Likert scale. The first part of the questionnaire was designed as a pictogram that featured five faces with various expressions. The answer alternatives were: very sad, rather sad, neither sad nor happy, rather happy, and very happy. This first section contained 24 questions. The final section consisted of 21 items with five alternative answers: always, almost always, sometimes, almost never, and never. These items were labeled with words to clarify the meaning of the scale points.

6.2.2 Parents' questionnaire

The parents' questionnaire consisted of 59 items covering several domains such as the child's hospital care, care at home, psychosocial issues concerning both the child and the parent, and finally the child's physical functioning associated with the malformation. The items were scored on a visual analogue scale (VAS). All the items had a VAS ranking from 0-100. A higher score indicated a more positive answer. Each question had two anchor words, i.e. negative - positive, never - always, very bad - very good.

6.2.3 Questions regarding children's memories

An additional questionnaire was used in the study. It consisted of seven questions about the children's memories of the medical treatment at an early age. The parents were asked to indicate whether they themselves or any other person in the family or among the nursing staff had talked to the child about the medical procedure that the child underwent at an early age. Moreover, the parents were asked whether their child had shown behaviors, emotional reactions, or other signs that the parent had interpreted as memories of the medical procedure. The parents were asked to give detailed descriptions of the child's presumed memory expressions.

6.2.4 Statistical analyses of the questionnaires

The data were analyzed using the Stata Statistical Software (StataCorp, 2001) program for statistical analysis.

6.2.4.1 Children's questionnaire

The children's questionnaires were analyzed with Pearson's X^2 -square test and Fisher's exact test for statistical intergroup differences. Ordered logistic regression was used to analyze the relationship between variables and the psychosocial outcome. The index group was compared with one control group at a time. A p -value of less than .05 was considered statistically significant.

6.2.4.2 Parents' questionnaire

Parents' questionnaires were analyzed with a t-test and the Wilcoxon Rank-Sum Test for statistical intergroup differences. Differences between mothers' and fathers' answers on the pair level were analyzed with the Wilcoxon Signed Rank Test. The index group was compared to one control group at a time. A p -value of less than .05 was considered statistically significant.

6.2.4.3 Questionnaire on memories

The questionnaire on memories was analyzed using Univariate or Multivariate Analysis of variance (ANOVA or MANOVA, respectively) for the continuous variables and Chi square (X^2) tests for categorical data. A p -value $\leq .05$ was considered statistically significant.

6.3 CHILD BEHAVIOR CHECKLIST

Swedish versions of the Child Behavior Checklist (CBCL/4-18) (Achenbach, 1991a) and the Teacher's Report Form (TRF) (Achenbach, 1991b) were used to measure children's emotional and behavioral problems as reported by parents and teachers. The CBCL presents descriptions of 120 emotional and behavioral problems that are likely to occur in children 4 to 18 years of age. The CBCL yields a total behavior problem score and two subscales referred to as internalizing (social, withdrawn, depression, anxiety) and externalizing (antisocial, aggressive, delinquency) symptom scales. The CBCL includes items about social competence, which measure the child's social ability and capacity in school and during extracurricular activities and hobbies.

6.3.1 Statistical analyses of CBCL and TRF

The data were analyzed using the Stata (StataCorp, 2001) program for statistical analysis. Data from the CBCL and TRF were analyzed according to instructions in the respective manuals (Achenbach, 1991a, 1991b).

6.4 ETHICAL CONSIDERATIONS

The local ethical committee at Karolinska Hospital approved these studies (Reference Number 99-202). The study was introduced to children and their parents both through a verbal presentation and by providing them with written information. Informed consent was received from all respondents.

7 SUMMARY OF RESULTS

7.1 PAPER I

Four categories of effects of the malformation emerged from the analyses of the interviews; these were physical, emotional, social, and family. The physical effects of the child's malformation influenced the family's experiences and permeated the interviews. The emotional effects of the malformation were the most frequently occurring effect in the data. The social effects described how the family's daily life was affected socially, and the family effects dealt with the how family members related to one another.

7.1.1 Physical effects of the malformation

The physical effects of imperforate anus were apparent from the very first hour of the child's life, in the delivery room. A colostomy had to be created during the first days, as the baby could not pass stools normally. A reconstruction of the anus was subsequently performed, and the parents had to dilate the anus to an optimal opening width for several months. To prevent constipation and fecal incontinence, the children were given enemas every other day for several years. The children had to wear diapers or other protection against soiling themselves until they reached preschool age. Incontinence also involved fecal odor. After several years of these difficulties, the children were offered a MACE operation.

7.1.2 Emotional effects of the malformation

Sorrow, pain, and disappointment were the most frequently described emotional effects of the malformation. These emotions were closely connected to anal manipulations (dilations and enemas) and incontinence. Parents felt that they had to force themselves on the child when they carried out the dilations, and that it became a fight. As the child grew older one parent had to hold him/her down, while the other parent performed the dilatation. All five parents stressed that they could never get used to it. The children themselves did not remember the dilations and how it had felt. The parents stated that prevention of constipation and incontinence was even worse than dilations. The children gave only very brief answers on this question. Disappointment and anger among the parents appeared when they talked about the reconstruction of the anus and

the child's bowel function. Toilet habits became very central in their lives for a long time. The parents were disappointed with information about the consequences of the malformation.

The emotional effect of sorrow was found when the parents reported that their children had experienced periods of depression. All three children acknowledged that they had been depressed. Feelings of shame were concealed, but recognizable when the parents talked about the fecal incontinence. Feelings of fear were apparent when parents discuss the children's fright and pain in reaction to anal dilations. The positive emotional effects of satisfaction and joy were visible when parents and children discussed the MACE operation, which had effected a positive change.

7.1.3 Social effects of the malformation

Procedures to prevent constipation and soiling interfered with the family's activities. Enemas and washouts were demanding and time-consuming and had to be planned with accuracy. Sports participation and the showers afterwards had been difficult. Bullying had occurred.

7.1.4 Family effects of the malformation

Relationships within families were strained from time to time because of the malformation. One mother told of quarrels over how to handle the treatment of the child. There were also disagreements about how to handle the enemas. The mothers said that they bore the main responsibility for everyday care; they did most of the dilations and enemas. The fathers and the children confirmed this.

7.2 PAPER II

Children with imperforate anus indicated an overall positive attitude and reported that they enjoyed school, had good relationships with schoolmates, had several friends, and participated in leisure activities. The children with imperforate anus said they were able to express their will and stated that they were generally in a good mood. Constipation occurred more frequently than in control groups. There were no correlations between the severity of the original malformation and psychosocial outcome, nor was any correlation found between MACE and psychosocial outcome. Overall, no differences were found specific to gender.

Parents of children with imperforate anus recorded more emotional and behavioral problems on the CBCL questionnaire than did parents of children in the control groups. Teachers recorded few emotional and behavioral problems among children with imperforate anus.

7.2.1 School

Comparing variables related to school activities between the children with imperforate anus and the children in the two control groups, a small number of statistically significant differences were found in favor of the index group. The children liked school more ($p<.05$) compared to children in Control Group I, and they reported having better relationships with their classmates ($p<.05$) compared with Control Group II. The parents confirmed these findings.

Children in the index group did not like using the school toilet for defecation and a statistically significant difference was found compared with Control Group I ($p<.05$). None of the children with imperforate anus reported being bullied, but they had been teased. School breaks were viewed positively by the children. Physical activity in school and showering afterwards were also considered positive.

7.2.2 Sports activities and hobbies

No statistically significant difference was found between groups regarding membership on a sports team. In the index group 84% of the children participated in some form of sport activity. In Control Group I 85% of the children participated in sports and in Control Group II 90% of children participated in sport activities. Children in all groups had one or more hobbies during leisure time.

7.2.3 Peer relationships

Children with imperforate anus reported that they had friends and that they had good relationship with their friends. They reported that they were more appreciated by their friends than did children in Control Group II ($p<.01$). They could also make decisions in a group just as often as children in the two control groups. No statistically significant difference was found between the groups in having a best friend and spending a lot of time with their best friend. Twenty (83%) of the children with imperforate anus, 28

(93%) of the children with JCA, and 22 (71%) of the healthy children reported that they had a best friend.

7.2.4 Emotional health

Children's emotional health and self-esteem scores did not differ between groups. The children with imperforate anus reported that they liked themselves and their bodies, and reported less frequent thoughts ($p<.05$) about how their bodies compared with those of their healthy peers. Children in all groups thought of their problems now and then but they estimated their parent's worries about their inconvenience as high. Both the index group and the two Control Groups stated that they were very much loved by their parents.

7.2.5 Physical function

Constipation was most frequently reported in the group of children with imperforate anus. Eighteen (72%) of the children with imperforate anus said that they were "always" or "almost always" constipated compared with four (13%) of the children with JCA, and four (13%) of the children in the healthy group. In the index group the mothers' assessments of the child's bowel function showed statistically significant differences on constipation, fecal leakage, and fecal odors compared with the control groups.

7.2.6 CBCL: parents' and teachers' report

On the CBCL total problem score scale a statistically significant difference was found between the index group and Control Group I ($p<.05$). However, subscales for internalizing and externalizing evinced no significant differences between groups. Teachers' reports on the TRF did not differ between groups.

7.3 PAPER III

7.3.1 Care

Statistically significant differences were found between parents in the index group and parents in the Control Groups I ($p<.01$) and II ($p<.05$) in mothers' reports on how they had been taken care of in the hospital. A statistically significant difference was found between the fathers in the index group and Control Group I ($p<.05$) on how they had been taken care of in the hospital. Both mothers and fathers in the index group were

less satisfied with how their children had been taken care of during their hospital stays, and a statistically significant difference was found between mothers and fathers of children with imperforate anus compared to mothers and fathers in Control Group I ($p < .01$).

The mothers in the index group reported less understanding and more negative experiences of the treatment, and statistically significant differences were found compared to both Control Groups. Fathers of children with imperforate anus also reported more negative experiences of their child's treatment and a statistically significant difference was found compared to fathers in Control Group II. The children's comprehensions of treatment, as reported by their parents, were lower in the index group compared with the two control groups.

7.3.2 Information

Satisfaction with information about the child's treatment was lower among fathers in the index group, and a statistically significant difference was found compared to fathers in Control Group II. Information to the child about treatment as reported by the mothers and the fathers in the index group was not satisfactory. A statistically significant difference was found compared to mothers in Control Group I and to fathers in Control Group II. Advice from the hospital about the child's difficulties was not sufficient as reported by the parents in the index group, and statistically significant differences were found compared to both mothers and fathers in Control Groups I and II.

7.3.3 Involvement

Mothers and fathers in the index group reported their involvement in the child's care to be high, and statistically significant differences were found between fathers in the index group compared to the fathers in Control Group I. Parents reported that children's involvement in the care, in all three groups, was low. Statistically significant differences between groups were found compared with mothers in Control Groups I and II and fathers in Control Group II.

7.3.4 Differences on the pair level

In the index group there were no statistically significant differences between the mothers' and fathers' answers on questions about care, information, and involvement on the pair level.

7.4 PAPER IV

7.4.1 Demographic data

The majority of children in the index group and in the two control groups lived with both their parents. The majority of children in all three groups lived in the city (including suburbs) or in a small town. Only four children lived in rural areas. The distribution of whether the parents' occupations required a university degree or not were the same in all three groups

7.4.2 Responsibility group-level

Mothers in the three groups reported taking much responsibility for the care of the child, but no differences could be found between groups. Fathers of children in Control Group II, the healthy children, reported taking much responsibility and a statistically significant difference was found in comparison to the index group ($p < .05$). Dissimilarities were found in mothers' and fathers' reports on responsibility, taken on a group level. There was a statistically significant difference between mothers and fathers in the index group ($p < .01$) and in Control Group I ($p < .02$).

7.4.3 Responsibility pair-level

Differences between mothers' and fathers' reported responsibility was found on the pair level, when the specific couple was compared to each other. Statistically significant differences were found between the mother and the father in the index group ($p < .01$) and in Control Group I ($p < .02$). Parents also assessed each others' extent of responsibility and this was compared with their own assessment of themselves. No statistically significant difference was found in any of the groups, but in the index group there was almost a significant difference between the parents' opinions on responsibility taken for care of the child's condition.

7.5 PAPER V

7.5.1 Non-verbal memories

Both the children with imperforate anus and the children with JCA had signs of non-verbal memories from the medical treatment at an early age. Nine of the 25 children participating in the study were reported to have non-verbal memories of the treatment. Parents of children with imperforate anus reported avoidance behavior on treatment needing anal manipulation as well as more general fears of seeing the doctor. The children with JCA also had distressing memories of the medical treatment. Fifteen out of the 30 children showed fears of injections and of seeing the doctor as well as more generalized fears.

7.5.2 Influences on behavioral problems and emotional feelings

Children with imperforate anus who had not been talked to about medical treatment at an early age had higher scores on the total behavior scale on CBCL, which implies that their parents had scored more behavior problem among these children. No statistical significance was found between groups. On the other subscales, internalizing and externalizing, no difference between groups could be seen.

The children who had been talked to tended to have less emotional problems than the children who not had been talked to, according to their parents. The children who had been talked to were reported to be generally in a good mood and a statistically significant difference was found $p < .05$. They were also less angry ($p < .01$) compared to the children who had not been talked to. They also cried less but no statistically significant difference was found between groups on this issue. They did not show depressive signs and were reported by their parents to be satisfied with life.

8 DISCUSSION

8.1 METHODOLOGICAL CONSIDERATIONS

The methodology of using both a qualitative and a quantitative approach in a study has advantages, and the different paradigms may bring us more in-depth knowledge. The interviews were aimed at exploring a phenomenon, in this case imperforate anus, and the nursing aspects of this malformation. The knowledge gained from the interviews was then used in the questionnaires for the children and the parents. With the questionnaires it is possible to investigate a larger sample and also to compare groups. This mix of methods with both a qualitative and a quantitative approach will give a more complete and better understanding of the phenomenon and also strengthen the reliability and validation of a study (Creswell, Fetters, & Ivankova, 2004; Nicholas & Keilty, 2007; Noorda et al., 2007).

The qualitative design using interviews gives respondents the opportunity to tell their stories with their own words, and these interviews can sometimes become more of a therapeutic conversation for the respondent. With open-ended and semi-structured questions the respondents have the opportunity to tell as much as they want about an issue, and the parents in this study sometimes gave long narrations about their experiences. The large amount of text has to be compressed into smaller parts, and this was done with a qualitative analysis (Kvale, 1996). In both qualitative and quantitative analysis the aim is to organize, provide structure for, and elicit meaning from research data. In the qualitative content analysis the researcher acts as an interpreter, reading through the data looking for meaningful segments and units and developing a categorization scheme (Polit & Beck, 2004; Polit-O'Hara, Hungler, & Polit, 1997). It is a challenge in the qualitative analysis for the researcher to make some sense of all the transcribed unstructured data as it consists of many pages of text. (Silverman, 2001) describes methods of strengthening the trustworthiness of a qualitative study. This can be improved by comparing the analysis of the same data by several authors, which was done with data in *Paper I*. The results from a qualitative study are always subjective and cannot be generalized to apply to other groups, although they bring a deeper understanding of a specific phenomenon (Polit & Beck, 2004).

The respondents in *Paper I* received copies of the interview guide after the interviews and were given the opportunity to give us comments on it. None of the respondents sent the interview guide back, and when we called them on the telephone none of them had anything to comment on or add from the interview guide.

Several questionnaires were used in the quantitative part of the study (*Papers II-V*); one questionnaire was used for the children, three questionnaires for the parents, and one questionnaire for the teachers. All data were collected at the same time and the study had a retrospective design comparing the index group with two control groups. In quantitative research when the phenomena of health or psychosocial functioning are studied, questionnaires are often used as an instrument (Polit & Beck, 2004; Polit & Hungler, 1999).

This sample of children is rather small, but still it was the total amount of children with high and intermediate imperforate anus born 1987-1992 and treated in Stockholm and its catchment areas. The children were all operated on by the same surgeon, Björn Frenckner, and with identical surgical technique. They had their follow-up treatment including anal dilations for 2-3 months. The families had regular meetings with the bowel therapist for help with a bowel management program to prevent constipation and fecal incontinence. The malformation is unusual and is only treated at four children's hospitals in Sweden. This sample from Stockholm was chosen since we knew that the children had been through the same medical treatment and follow-up. The children's ages at the time of the study were of importance. The children had to have been born 1987 or later since the surgical reconstruction of imperforate anus was changed that year. The children also had to be old enough to be able to answer a questionnaire. The small sample is a limitation that conceivably influenced the results, as we might have found additional dissimilarities among the groups with a larger sample size. A power analysis was not essential because the sample size was consistent with the inclusion criteria required. This modest sample makes it difficult to draw firm conclusions but could nevertheless give indications on how children with imperforate anus and their parents have experienced the malformation.

The children's questionnaire was designed to be easy for the children to answer. Children with a normal cognitive functioning are able to understand and answer a numeric rating scale and the Likert scale we used has been found to be easy for children

to complete (van Laerhoven, van der Zaag-Loonen, & Derkx, 2004). Children as young as five years old can give reliable answers on a Likert scale with faces if they receive assistance with reading the question. It has also been found that older children, from 8-12 years, easily can give answers on a Likert scale without faces (Howard & Freeman, 2007). Other researchers, though, have found that children younger than six years often give more extreme answers on questions of an emotional type (Chambers & Johnston, 2002; Rebok et al., 2001). Self-reported questionnaires used in studies on young children should be interpreted with respect to the child's developmental age since self-reported items may be less stable over time for young children and this may affect the reliability (Cremeens, Eiser, & Blades, 2007)

The children in our study were somewhat older, 7.8-13.9 years, and this might give strength to the children's answers. Rebok et al., (2001) also reported that children at the age of eight were able to report on virtually presented questions about their health. One additional thing that could give justification for developing a specific children's questionnaire is that all disease-specific quality of life instruments for patients with fecal incontinence are developed for adults and are inappropriate for use with children (Trajanovska & Catto-Smith, 2005).

The parents' questions were developed to correspond to the children's questions, in order to look for differences and similarities between parents and children. Parents, and most certainly mothers, are good at assessing their children's health and they are able to put it in relation to absence or presence of health problems (Monette, Seguin, Gauvin, & Nikiema, 2007). In studies on quality of life and health-related quality of life among children, self-reported questionnaires have been found valuable and if the children's questionnaire is combined with a parental proxy-version questionnaire a more complete result would be seen (Barr, Thibeault, Muntz, & de Serres, 2007; Sherman, Eisen, Burwinkle, & Varni, 2006; Varni, Burwinkle, Limbers, & Szer, 2007).

In the parents' questionnaire a visual analogue scale (VAS) was used. Answering questionnaires with many questions using the VAS has been found to appeal to the respondents, and they are easier fill in. The length of the VAS was 10 cm and this length of the visual analogue scale has been found to have the smallest measurement error (Seymour & Walton, 1982). VAS is often used to assess pain, but it has also been used in studies of health outcome (Lukacz et al., 2004). The VAS in the parents'

questionnaire in our study had two anchor words; this is preferable and makes it easier for the respondents to complete the questionnaire (Torrance, Feeny, & Furlong, 2001). The VAS was constructed with the most positive answer placed to the right. The seven questions about children's memories were constructed by an experienced memory researcher. The CBCL is a standardized instrument and is frequently used in research of children's behavior/emotional problems and in screening for these symptoms (Achenbach, 1991a, 1991b). It is a proxy version and it reflects the parent's assessment of the child's psychosocial function. CBCLs have well-validated scales for internalizing and externalizing problems and also a total problem score scale for emotional or behavioral problems (Dehon & Scheeringa, 2006). As mentioned above, this combination with the qualitative interviews and the quantitative approach with several questionnaires gives strength to the study and elucidates the aspects of the imperforate anus malformation.

8.2 REFLECTIONS ON THE FINDINGS

The main findings from this study involved the parent's distress over the child's malformation and the fact that they have been very engaged in their children and the consequences of the malformation. The parents' efforts may be the explanation for the good psychosocial functioning of the child. High and intermediate imperforate anus affects the child, the parents, and the family as a whole. Parents had been affected in several ways, especially emotionally, psychosocially and practically. The parents had to a great extent been involved in the children's care and follow-up treatment and the mothers seemed to have taken much responsibility for the children's care. However, the children appeared to handle their situations quite well as the malformation did not seem to have affected their psychosocial functioning. In addition, the children who had talked about their experiences of the medical treatment at an early age seemed to have a better psychosocial function. The children were reported to have non-verbal memories of the treatment at an early age. The malformation involves a considerable effort from the parents, and their capacity and strength is put to the test. For the child the long-term consequences of high and intermediate imperforate anus are multifaceted.

8.2.1 Surgical implication and medical treatment of the malformation

Since 1987 all children with imperforate anus at our clinic have been operated on with PSARP and the fistula-saving technique in order to preserve the anlage of the internal sphincter and to preserve a better sphincter function, although a normal function of the

internal sphincter is not possible (Husberg et al., 1992). The pediatric surgery experiences of this complex malformation have increased greatly and complications are mainly preventable, as reported by (Pena, Grasshoff, & Levitt, 2007). The children in this thesis had the PSARP when they were very young, about six months old, and the follow-up treatments with dilations were done before the age of one year. Follow-up studies done on children operated with the PSARP and the fistula-saving technique demonstrates that the patients seem to achieve an acceptable continence (Liu, Yuan, Geng, Wang, & Li, 2004; Rintala & Lindahl, 2001; Tsuji et al., 2002).

In *Paper II* the parents reported that the children had difficulties in controlling their bowel movements, and also had problems with constipation and fecal incontinence. These functional problems were treated with our bowel management program and the children and their parents had a close and regular contact with the bowel therapist. The odor that fecal incontinence entails is easily detected by others. Not being able to have control over your body could result in a socially isolated behavior among adolescents (van Kuyk et al., 2001).

Even among school-age children with a normal anal anatomy fecal leakage may occur, but contrary to the problems of the children with imperforate anus theirs will probably not last all their lives (Bongers, Tabbers, & Benninga, 2007; Brandt et al., 2007; Söderström, Hoelcke, Alenius, Söderling, & Hjern, 2004). Children with imperforate anus at our clinic are linked to the specialist team for imperforate anus immediately after birth. The bowel therapy clinic has an important task in supporting these families and preparing the children and their parents for problems associated with the malformation. The bowel management program for children with imperforate anus is decisive for preventing constipation and for keeping clean despite fecal incontinence, and most of the children have well-functioning bowels.

The transition into adulthood and the changeover to care in an adult clinic can be a big step and colorectal surgery units do not always have the same expertise and knowledge of what consequences this specific pediatric malformation entails (Hartman et al., 2004; Hassink, Rieu, Severijnen, Brugman-Boezeman, & Festen, 1996; Little et al., 2003). MACE has been one of the most successful treatments of fecal incontinence for patients born with imperforate anus and with this surgical method patients with fecal incontinence can achieve social continence (Aksnes et al., 2002; Curry, Osborne, &

Malone, 1998; Malone et al., 1990; Meier, Foster, Guzzetta, & Coln, 1998; Poirier, Abcarian, & Nelson, 2007) .

8.2.2 Impact of the malformation on the children

In this study we found that children had non-verbal memories of the medical treatment early in life (*Paper I*). Memories from this early age are not possible to present verbally as the child at the time of the event was too small to talk about it (Eacott & Crawley, 1999; Peterson, Grant, & Boland, 2005). Reminders of the invasive treatment at this early age may be seen when the child is exposed to some similar situation, such as a medical examination or medical treatment (Brown et al., 1999; Stoddard et al., 2006). The children with imperforate anus showed strong reactions against any anal manipulation. Talking with your child about experiences of medical treatments seemed to be related with the child's psychosocial function. In those families where an open dialogue was held on this subject, children seemed to find it easier to recall medical treatment early in life. The children who had been talked to seemed to be able to handle these memories and they also appeared to have a better psychosocial function.

The adolescents in the interview study (*Paper I*) admitted to being exposed to teasing and bullying. This malformation, with the inconveniences it brings, can stigmatize the child and make him/her an object of ridicule. However, the younger children admitted being teased, but denied being bullied. Children's ways of coping with difficult situations can involve denial both to themselves and to others. Escaping into dissociation and denial is a psychologically protective mechanism which may reduce the children's stress (Clayden & Wright, 2007; Klinnert, McQuaid, McCormick, Adinoff, & Bryant, 2000; Ludman & Spitz, 1996).

The younger children in this study (*Paper II*) did not report any psychosocial impairment even though they had some leakage and fecal odor, as reported by their parents. The children were positive about school and had friends, and the parents confirmed their children's satisfaction about school and social activities. These positive findings were in contrast to those of the adolescents who in the interviews admitted that they had sometimes felt depressed. An explanation for the difference in psychosocial impairment between the children in *Paper I* and in *Paper II* may be the children's ages. The consequences of the malformation become more obvious when a child gets older, as wearing sanitary napkins and having unpleasant odors are embarrassing. Another

explanation may be the difference in surgical method performed on these two groups of children, as earlier explained. The children in *Papers II-V* probably had better bowel functioning and this may be related to the children's psychosocial functioning. As the children get older a supportive network for children and adolescents with a chronic condition could be helpful. Discussing problems and sharing other children's experiences have been found to be important and could ease uncomfortable feelings about the condition (Kyngäs, 2004 Schmidt, 2003 #161; Schmidt, Petersen, & Bullinger, 2003).

The parents have recorded more emotional and behavioral problems on CBCL than did the control groups. However, the teachers seemed to think that children with imperforate anus did not differ from their classmates. This finding is in line with other studies on psychosocial functioning and quality of life among patients with imperforate anus and other chronic conditions, where no difference between them and healthy peers could be observed (Diseth, Bjornland, Novik, & Emblem, 1997; Ditesheim & Templeton, 1987; Hartman et al., 2004; Hassink, Rieu, Brugman, & Festen, 1994; Mackner & Crandall, 2005). The school environment seems to be important to children, and these children seem to have a lot of support from teachers and other adults to help them handle their problems.

The ages of the children are of concern in this thesis; in the quantitative part they were between 7-13 years old (mean age 10.5 years). At this age the parents are still the most important people in a child's life, handling the practical things like preparing their children for different events. Social relationships with friends and classmates are important for the child. As it seems, the parents have supported their children in a manner that helps them to avoid accidents with fecal incontinence and odor in the school situation. This malformation does not show on the outside but the consequences can become very obvious because of the odor. A higher risk for psychological morbidity including anxiety, embarrassment, depression and low self-esteem among children and adolescents with imperforate anus has been reported (Funakosi et al., 2005; Hamid, Holland, & Martin, 2007). The long-term consequences could also limit the children's and adolescents' quality of life (Davies, Creighton, & Wilcox, 2004; Hartman et al., 2007; Hartman et al., 2006; Stam, Hartman, Deurloo, Groothoff, & Grootenhuis, 2006)

This study on children with imperforate anus and their parents demonstrates that our knowledge about this group of children still is not sufficient. The exploratory study (*Paper I*) elucidates a multifaceted experience of the malformation – the parents’ suffering and the consequences described by the adolescents themselves. The younger children in the quantitative part of the study seemed to have good lives and in general a good psychosocial function even though they also had problems with their bowel functions. The children had non-verbal memories of the treatments they had undergone early in life and we found that talking with your child about memories influenced the child’s psychosocial function positively. Parents’ care of the child involves a substantial effort, and a gender difference was seen between parents’ responsibilities.

The complexity of this malformation as shown in this thesis reveals that several special competencies are needed in care of these children. The consequences it entails can be both embarrassing and shameful for the child. The burden the parents have to carry and the necessary treatment they have to perform, by exposing the children to invasive treatment for a long time is a difficult matter that cannot pass without leaving deep marks in their lives. A family-centered approach in care of children with this malformation is important in order to preserve family functioning that is as normal as possible.

We believe that the close relationships between the families and our specialist team with the bowel therapist and the pediatric surgeon at the clinic are related to “our” children’s abilities to function well. Even though all of the children have severe malformations, this close contact and support and regular follow-up made it possible for them to handle their situations quite well. The bowel therapist could give both medical support and psychosocial support, and when needed can be a liaison to the child and adolescent psychiatry unit. Such a liaison between the pediatric surgery unit and the child and the adolescent psychiatry unit should be a natural part of our work.

8.2.3 Impact of the malformation on the parents

In the exploratory interview study (*Paper I*) four categories – physical, emotional, social and family effects – emerged which reflected the parent’s reactions to the complexity of the malformation. The emotional impact of imperforate anus is most evident when parents describe their memories of caring for the child. Predominantly

negative emotions occur. Positive emotions like joy and satisfaction were mentioned in relation to the MACE operation.

The suffering and the extensive problems at home due to the malformation was new knowledge for us. It seemed as if the family had revolved entirely around this concern. The children with imperforate anus, though, seemed generally to be happy and unworried about their inconvenience (*Paper II*). The findings on CBCL of more emotional/behavioral problems among children with imperforate anus seem to be more of a reflection of the parents' views about the child's inconvenience. Our results could be confirmed by those of several other studies showing that the emotional impact, with mixed feelings of happiness, despair, and guilt is common among parents of children with chronic conditions (Erickson & Ray, 2004; Johansson & Ringsberg, 2004; Streisand, Swift, Wickmark, Chen, & Holmes, 2005; Uzark & Jones, 2003).

The concept of caring for a child with a chronic condition is comprised of several different aspects, including taking care of the child's health, activities, physical and emotional well-being, quality of life, social life and the parents' marital relationship. The gender perspective on parental responsibility for the child's care showed a more traditional distribution between parents, with the mother as the main caregiver (*Paper IV*). Even though fathers are very engaged in their children the mothers still seem to take the main responsibility in care when the child has a chronic condition (Hovey, 2005; Roach, Orsmond, & Barratt, 1999).

Parental satisfaction with care, involvement, and information was lower among parents of children with imperforate anus than in the control groups (*Paper III*). A possible explanation to our finding is that having a child with this malformation may provoke feelings of stress, and this has earlier been found by Ludman and Spitz (1996). The parental feelings may change over time as the child grows and new challenges to handle the consequences of the malformation appear. Similar parental experiences to those found in this thesis were seen in a study on children with imperforate anus and their parents made by a parents' association (Aminoff, La Sala, & Zaccara, 2006). Hassink et al. (1998) revealed that having a child with an anorectal malformation does not always involve problems, but that it is essential that the family and their well-being is seen as a whole and that the focus is not just on the child's malformation (Hopia et al., 2005; Kim, Keininger, Becker, & Crawley, 2005).

Parents experienced that they did not get enough information and this was more obvious in the group of children with imperforate anus (*Paper III*). Parents' needs of information may benefit their understanding and also enable them to talk with the child about the malformation and the treatment. This is consistent with findings in other studies on parents having a child with a chronic condition (Hummelinck & Pollock, 2006; Jackson et al., 2007). The parents may be overloaded with information when the child is diagnosed and it is not possible to take it all in. Internet information will provide parents with new facts, though it may not always give a better understanding (Aslam, Bowyer, Wainwright, Theologis, & Benson, 2005; Boston, Ruwe, Duggins, & Willging, 2005; Impicciatore, Pandolfini, Casella, & Bonati, 1997; Sim et al., 2007). Information has to be repeated and has to be adjusted to the parents' needs. A simply written information sheet and a simple drawing are often the best way for the surgeon to give information about an advanced surgical operation (Noll, Spitz, & Pierro, 2001). A specialist nurse could also be of assistance in presenting this important information.

Parents in this thesis seem to have been very much involved in their children's treatment and have had tough experiences of the child's malformation. Parents of children with a chronic condition may gain support by meeting other parents in the same situation. Parents' groups can increase knowledge about the condition and generate a better understanding about other families' concerns, and also be of important psychological help for the parents (Ireys, Chernoff, DeVet, & Kim, 2001; Young, McMenemy, & Perrin, 2001). Parents of healthy children could benefit from participating in a parents' group as well and this may improve the parental psychosocial health (Barlow, Coren, & Stewart-Brown, 2002). The use of Internet, apart from providing parents with information, can also serve as a parental support since parents meet other parents on different websites (Sarkadi & Bremberg, 2005).

The need of psychological support might have been underestimated among children with imperforate anus and their parents. To some extent support and advice about the malformation may be given by the bowel therapist and the pediatric surgeon but if problems change character to more psychopathology, more advanced psychiatric help may be required. Since the malformation concerns a very sensitive part of the body, extensive knowledge about the malformation is essential to avoid misunderstandings. The invasive anal dilations are often experienced by the parents as an act of abuse even

though they are well informed of the importance of the dilations for the child's future bowel function.

9 CONCLUSIONS

- Parents of children with high imperforate anus had experienced suffering and many difficulties associated with the malformation.
- Children with imperforate anus in this study seem to be psychosocially well adjusted, though their parents stated that they experienced leakage, constipation, and fecal odor.
- The children might have some psychosocial difficulties according to their parents, but not according to their teachers.
- Parents of children with imperforate anus experienced less satisfaction with medical care the children had received, and reported that support had been insufficient.
- The mothers of children with imperforate anus seemed to be the main care givers. Mothers and fathers seem to have different opinions on how much responsibility they take for the child's malformation.
- The children seemed to have non-verbal memories of the medical treatment they had undergone at an early age and parents' talking with the child about these memories seemed to be positively related to the child's psychosocial functioning.

Imperforate anus is an unusual malformation which requires advanced care. The care has several aspects, including the surgical aspect with the reconstruction of the anus and the follow-up treatment, the family aspect on caring for a child with this malformation, the aspect of the child's psychosocial function and the aspects of the child's experiences of the medical treatment due to the malformation. Our knowledge has earlier been limited but the conclusions of this thesis may lead to an improved understanding of caring for children with imperforate anus and their parents.

10 CLINICAL IMPLICATION AND FUTURE RESEARCH

The results from this thesis are valuable for our future work with children with imperforate anus and their parents. The support these parents need has been underestimated due to our previously limited knowledge. Support groups for parents with children with imperforate anus have been established at our clinic as a result of what we learned in this study. The bowel therapy unit has an important task in optimizing the child's bowel function and supporting the parents who handle the treatment. The bowel therapist establishes contact with these families immediately at the child's birth and will be involved in the children's treatment and care until they are 16 years old and leave the pediatric hospital. The knowledge gained from this thesis shows that this close contact between the families and the specialist team seems to be a good concept for the overall well-being of children with imperforate anus and their parents.

Future research from a nursing perspective on children with imperforate anus is important, since this malformation entails many different aspects. This thesis involves the children with high and intermediate imperforate anus but it would be of interest to conduct a similar study on children with low imperforate anus to investigate the psychosocial functioning in this group as well. A future follow-up study on the children participating in this thesis is absolutely essential to perform, when they have become young adults. This would increase our knowledge about this multifaceted malformation as the children get older and enter adolescence and puberty, and are still our pediatric patients.

11 ACKNOWLEDGEMENTS

I wish to express my sincere gratitude to everyone who helped and supported me in my work completing this thesis, particularly;

Professor Kyllike Christensson, my principal supervisor. Thank you for increasing my scientific knowledge, and for all your support and encouragement. You motivated me to go further and to complete this thesis; you have always given me very good advice and been very helpful when help was needed.

Professor Björn Frenckner, my co-supervisor, who initiated this study and also initiated the cooperation with the child and adolescent psychiatry research unit. Thank you for giving me the benefit of your great knowledge about imperforate anus, and for introducing me to research. Thanks also for always being supportive and giving me good advice. You encouraged me to finish my work after the licentiate degree, and to complete this thesis, for which I am very grateful.

Margret Nisell, my co-author and my very best friend. It has always been a pleasure to work with you. You have given me a lot of support and much good advice. Thank you for all the interesting discussions on research and the different aspects of this specific malformation. I hope that you also will finish your PhD soon, since we have struggled so hard with this study.

Professor Per-Anders Rydelius, my extra supervisor, who taught me about child and adolescent psychiatry, the importance of the child's development, and how important school is for children. Thank you for always being so generous and supportive and for letting me be a member of your research department.

Torun Lindholm, my extra supervisor in the part of the study focusing on aspects of memories. You have helped me to understand the children's experiences and how their memory works. Thank you for very good cooperation, for excellent advice, and for all your support.

Associate Professor Jan-Olov Larsson, thank you for help with statistical advice and for useful suggestions, as well as for all your support.

Gunilla Nilsson, thank you for all practical assistance. You have all the important dates in your mind. Thank you for all support and for being a good friend.

Laura Horowitz, thank you for being supportive with advice and with language revision. You are always helpful and encouraging. You have been my catcher in the rye.

Tobias Edbom, thank you for support and interesting discussions on nursing science. Thank you for help with IT support, and for all your humorous contributions and all the laughter.

Britt Husberg, who taught me about imperforate anus and who inspired and supported me to do research in this field.

Tina Granholm, who always asked about the progress of my study, who pushed me and believed in my study, thank you for all your support.

Johan Wallander and Lena Nordenson, thank you for all your support and for giving me the opportunity to complete this thesis.

Agneta Währner and Helena Wigander, my colleagues at the bowel therapy unit, who are doing excellent work with the children and their parents. I really thank you for all your support.

All colleagues at the “uro-, nefro- gastromottagningen” for support.

Hiie Allik, Bettina Friedrich, Kerstin Malmberg, Annelie Törnblom, and Elisabeth Rydshagen for supportive comments.

My sister Birgitta and your family, and my brother Hans and your family for all your support and your belief in me and in this study. My parents-in-law Gun and Rune Joelsson. My sister-in-law Lena, and Frida.

My father, Sture Öjmyr, who has always been a great support to me, and who worries that I work too hard. I know you look forward to the day I'm finished with my study. Thank you for all your care and concern.

Min pappa, Sture Öjmyr som alltid varit ett stort stöd och som oroat sig för att jag arbetar för mycket och som sett fram mot den dag då jag skulle bli klar med min studie. Tack för din omtanke.

My husband Lars, for love and understanding and always believing in me. Our children Emilia, Viktor, and Oskar. You are the always the best; this has been a tough time for you as well.

Thanks to all the children and parents participating in the study. We are grateful to the staff at the outpatient unit for juvenile arthritis, and to the day surgery unit at Astrid Lindgren Children's Hospital, for assistance and for letting us get in contact with your patients.

This study have been financially supported by Solstickan Foundation, the Foundation of Samariten, The first Mayflower Campaign, the Foundation of Ella Danelius, the Foundation of Kempe Carlgrenska, Crown Princess Lovisa Foundation, the Groschinsky Foundation, and the Jerring Foundation

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