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**PSYCHOSOCIAL CONSEQUENCES
OF HIGH AND INTERMEDIATE
IMPERFORATE ANUS**

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Institutet**

Stockholm 2005

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Published and printed by Karolinska University Press

Box 200, SE-171 77 Stockholm, Sweden

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ISBN 91-7140-532-1

To Sven, Miriam, Elias and Hannah

ABSTRACT

Imperforate anus including lack of a normal anorectum is an uncommon malformation. The parents of children born with high and intermediate imperforate anus have to take a big responsibility for the follow-up treatment. It has been revealed that children with imperforate anus have psychosocial problems, though knowledge of this is limited.

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

Methods: In *Paper I*, the purpose was to illuminate the children's and parents' experiences of imperforate anus and to create a baseline for further research. Interviews were done with three patients with high imperforate anus who had had severe problems with the malformation, and with their parents (5). Answers to the open-ended questions were analyzed with a qualitative method, "editing analysis style". In *Paper II* and *Paper III*, 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 59 items for the parents and 45 items for the children, covering psychosocial domains, one physical domain and one domain on experiences of hospital care. The parents filled in the Child Behavior Checklist (CBCL), and the children's teachers filled in the Teacher's Report Form (TRF). Two groups of children along with their parents served as comparison groups. They included 30 children with juvenile chronic arthritis and 32 healthy children.

Results: *Paper I:* The parents were outspoken in the interviews, and the children's answers were meager. Four categories of effects of the malformation were revealed: physical, emotional, social, and family. The emotional effects permeated the interviews; the parents' suffering was overwhelming. In *Paper II*, the children with imperforate anus displayed an overall positive attitude; they reported that they were in a good mood, that they liked school, and that they had good relationships with peers. Furthermore, the children had fecal incontinence and constipation, according to parents. In the CBCL, the children in the index group were assessed to have significantly more emotional/behavioral problems than the children in Comparison Group I. In the TRF, the teachers reported few symptoms for the index group children. *Paper III* showed disagreement on psychosocial variables from children with imperforate anus and their mothers (on a pair level). Differences were found in particular on psychological variables, i.e. the child's emotional life. The competence scales in the CBCL revealed no differences between the index group and the other two groups. In the TRF, the teachers assessed the children with imperforate anus to be less adapted and adjusted in school than the children in comparison groups.

Conclusions: Parents of children with high imperforate anus have to go through difficult experiences associated with the malformation. Children with imperforate anus in this study seem to be well adjusted psychosocially, despite fecal incontinence and/or constipation. The children 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. Psychosocial issues are crucial to children with imperforate anus and to their parents, and qualified advice and assistance should be a central part of their continuing care. Perhaps collaboration with expertise from child and adolescent psychiatry may be required.

Keywords: imperforate anus, psychosocial, children, parents, fecal incontinence, multiple informant assessment.

LIST OF PUBLICATIONS

- 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.*
18(6), 423-432, (2003).

- 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. In press.*

- III. Nisell M., Öjmyr-Joelsson M., Frenckner B., Rydelius P-A., Christensson K.
Views on psychosocial functioning; Responses from imperforate anus
children, their parents, and their teachers. *Submitted.*

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LIST OF ABBREVIATIONS

CBCL	Child Behavior Checklist
IA	Imperforate Anus
IBD	Inflammatory Bowel Disease
JCA	Juvenile Chronic Arthritis
MACE	Malone's Antegrade Continence Enema
PSARP	Posterior Sagittal Anorectoplasty
QoL	Quality of Life
TRF	Teacher's Report Form
VAS	Visual Analogue Scale

1 INTRODUCTION

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. 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 staff, the child's physical well-being is primarily and naturally in focus. To get a more complete approach to the family, an integration of different viewpoints, including child and adolescent psychiatry, allows a better assessment of the child's functioning. This might be essential, since it has been found that any kind of chronic pediatric illness is a psychological risk factor for children (Glazebrook, Hollis, Heussler, Goodman, & Coates, 2003).

When children with chronic conditions are in need of extra psychosocial interventions, they could get help through consultation and liaison work between the child psychiatric department and the pediatric disciplines. Psychosocial support for the children with anorectal malformations and their mothers may be necessary, taking into account the age of the child (Funakosi et al., 2005).

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

2 BACKGROUND

2.1 CHILDREN WITH CHRONIC CONDITIONS

Children with chronic conditions are known to be at increased risk for psychosocial adjustment problems. It has been estimated that about 10% of all children have some type of chronic illness and that these children have twice the risk of psychosocial maladjustment (Pless & Nolan, 1991).

One can assume that a child's Quality of Life (QoL) could be affected by having a chronic condition. This is confirmed in a recent study where it was found that aspects of health-related QoL in children are negatively affected by chronic illness (Epstein, Stinson, & Stevens, 2005).

It has been argued that chronic conditions might have certain commonalities; in a study of children with asthma and diabetes, no disease-specific effects were shown after controlling for family factors and childhood illness variables (Holden, Chmielewski, Nelson, Kager, & Foltz, 1997). No obvious differences were found between groups of children with varying chronic conditions (Meijer, Sinnema, Bijstra, Mellenbergh, & Wolters, 2000). However, the kind of disability the child has, influences the outcome (Boekaerts & Roder, 1999).

When children with chronic physical conditions become distressed, it may be more difficult to recognize the psychosocial factors that could be contributing to the symptoms (Knapp & Harris, 1998). An example is abdominal pain, a common sign among children that is often unassociated with organic pathology, and the psychosocial factors play an important role in this condition (Boey & Goh, 2002).

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).

2.2 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 affected, and when the patient is a child, special concerns can be distinguished. There are more demands on parents in raising a child with a chronic condition than there are with a healthy child (Ray, 2002).

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 among their parents (Bauman, Drotar, Leventhal, Perrin, & Pless, 1997). Both children with a chronic condition and their mothers are at increased risk for psychosocial adjustment difficulties, as compared to peers (Wallander & Varni, 1998).

The family's/parents' role in psychosocial adjustment of a child with a chronic condition appears to be most influential. 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).

It is crucial to assess children's views of their 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, 2001).

The examination of psychosocial functioning in children with a chronic condition is an important area of study, and the results are of great value as well in evaluating and understanding the parents' reaction to illness (Krener & Wasserman, 1994).

2.3 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 grade of severity varies from the most severe, the high form, to the intermediate and the low form.

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 three months old. After reconstruction, the new anus must be dilated to attain normal width. The follow-up treatment requires daily dilations of the newly constructed anus. The parents have to learn this follow-up treatment and invasive procedure, and have the responsibility of continuing it for two to three months (Diseth, Egeland, & Emblem, 1998; Pena, 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. Although the surgical procedures have been successful, many children with imperforate anus have functional difficulties including constipation and fecal incontinence. Regular enemas are often given as treatment and prevention for several years, and the parents are in charge of giving their children the enemas. However, even with these treatments the children can continue to suffer constipation as well as fecal incontinence.

When a child is born with 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. The family needs support throughout the child's development, because of the huge impact of the malformation (Hassink et al., 1998).

The impact of the disability has been discussed and psychosocial consequences of imperforate anus have been explored in a small number of studies. Psychiatric symptoms, such as anxiety, low self-esteem, and depression may develop in adolescents with high imperforate anus. 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 et al., 2000). The physical consequences of the malformation have been shown to cause psychological problems; fecal incontinence is socially unacceptable, and clinical experience has indicated a link between physical and emotional difficulties (Ludman, Spitz, & Kiely, 1994). Furthermore, the same authors pointed out that certain characteristics of a child, and of his or her parents or family, may have positive or negative effects on the child's psychological adjustment.

Psychosocial consequences of imperforate anus have not been fully investigated. In the course of the collaboration between the pediatric surgery unit and the child and adolescent psychiatry unit, we were interested in further examination of the psychosocial consequences of the malformation so the children with imperforate anus and their parents will receive the nursing care and support they need.

2.4 DEFINITION OF PSYCHOSOCIAL

Childhood is a flexible time when family and social relations have a big impact on personal development. In a well-known theory about the child's development, psychosocial issues are included (Erikson, 1950). This theory is based on a multifaceted perspective, where development is based on biological inheritance and on psychosocial factors.

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 a chronic condition (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).

Additionally, 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 severely interfere with everyday functioning (Brugman, Reijneveld, Verhulst, & Verloove-Vanhorick, 2001).

Some may argue that psychosocial functioning and Quality of Life are interchangeable terms and if not identical, they seem to be closely related. 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).

Information related to factors of emotional life, family relations, school, friends, and activities were chosen as relevant psychosocial variables for this study.

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 3 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 -based on imperforate anus children's, their parents', and their teachers' responses to questionnaires.

4 STUDY DESIGN, MATERIAL, AND METHODS

This study is a product of a fully integrated cooperation between the pediatric surgery unit and the 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 et al., 2003).

In an interview study with healthcare 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, 1999).

This collaboration was developed as a common base for nursing care for patients at the Children's Hospital. We decided to focus on patients with imperforate anus because this malformation represents features that could be attached to 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 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 phenomena, rather than simply observes and describes it (Polit & Beck, 2004).

The extensive findings from these interviews (*Paper I*) was the basis for questionnaires for children with high and intermediate imperforate anus and their parents. These questionnaires were used as study instruments in *Paper II* and *Paper III*.

4.1 PAPER I

In *Paper I*, interviews were done 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 that had been treated at St. Görans's 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 (5).

Individual interviews were conducted at the hospital to collect data. The questions were constructed by specialists in the area of child 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 (parents) and an additional 9 open-ended questions (children) were used as data.

4.1.1 Analyses

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 (M.N & M.Ö-J.). The next step was to get data coded out of 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. In addition, data was analyzed to uncover the meaning of the interview responses. The interview responses could be broken down into four categories, based on the effects of the malformation: physical, emotional, social, and family.

4.2 PAPER II AND PAPER III

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 conclude the study. The children and the parents were recruited from the gastro outpatient clinic at Astrid Lindgren Children's Hospital. 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 were included because the surgical method was modified and refined in 1987. 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. Members of Group I had a chronic disorder that started early in life, and they had extensive contact with the hospital because they needed treatment. The children in 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 that had pain-alleviating injections earlier than four years of age, were pertinent for the study. Forty-four families were contacted by the team at 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 conclude 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 Group II were admitted to the hospital with comparatively harmless disorders. The children were recruited from the day care 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 that were born 1987-1992 were asked by staff at the day care surgery unit if they were interested in getting more information about this ongoing study. If they agreed, they got

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.

All respondents answered questions in questionnaires. A study-specific questionnaire was used for all participating parents, containing statements that fell into these domains: three psychosocial, one physical, and one experiential (i.e., hospital-related care). The questionnaire included 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.

The children's questionnaire included five psychosocial domains, one physical and one experiential (i.e., hospital-related care) domain. The questionnaire consisted of 44 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 (24) —or responses were written, i.e., always, almost always, sometimes, almost never, and never (20). One response was written “yes or no”.

4.2.1 Analyses

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 X^2 -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*, 16 congruent psychosocial variables were chosen in children's and parent's questionnaires, to obtain comparable data. The variables were further categorized as psychological and social. Parents' and children's answers were compared on the pair level and Wilcoxon signed ranks test was used to analyze data.

4.3 CBCL AND TRF

Both parents were asked to fill in the Child Behavior Checklist (CBCL). CBCL is a proven, well-evaluated questionnaire (Saxe et al., 2003). CBCL measures children's emotional/behavioral problems and the children's competence.

On the CBCL, parents rate 118 items pertaining to emotional and behavioral problems shown by their children during the last six months on a three-point scale. This part of CBCL was used in *Paper II*. The competence scales in CBCL could be considered the "healthy reports" while the problem scales focus on problems and symptoms. The social competence scales include activity, social, school, and total competence. 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 the activeness in organization or club participation (social scale) and academic performance in school (school) (*Paper III*).

The children's teachers filled in the Teacher's Report Form (TRF) to examine the experiences of the teachers. TRF is an instrument that measures the child's emotional/behavioral problems in school. To get in touch with the teachers, the parents were asked for permission to allow their children's teachers to participate in the study. Thirty-six teachers participated: 12 teachers of the index group children, 12 teachers of Comparison Group I, and 12 teachers of Comparison Group II (*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 III*).

Data from the CBCL and the TRF were analyzed according to instructions in the respective manuals (Achenbach, 1991a, 1991b). We used t-test and compared the index group with one comparison group at a time.

4.4 ETHICAL ASPECTS

The study was approved by the Ethical Committee at Karolinska Hospital. All participants could contact the researchers for concern if they wanted to.

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. They can tell more than their intention was from the beginning, which might entail feelings of having said too much. However, the interviewers were experienced in the field of child and adolescent psychiatry and were used to talking about sensitive issues. The informants were offered the possibility of contacting the interviewers if they were concerned.

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

The parents of children with imperforate anus and parents of children with juvenile chronic arthritis could also be reminded of trying experiences connected to the child's disability. The Child Behavior Checklist comprised questions that could be understood as sensitive and personal, though no one made any comment on that.

5 FINDINGS

Findings from the interviews in the qualitative study and results from the questionnaires in the two quantitative studies are presented. The informants participating in *Paper I* were not the same as the subjects taking part in the two following studies (*Papers II-III*).

In *Paper I*, three patients with imperforate anus and five of their parents were interviewed. The parents were very outspoken when telling their experiences, but the children's answers were meager. 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 disability" 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; they were optimistic and there were no signs of depression among the participating children. The children were in good moods just as the children in the comparison groups, according to the children themselves. The children with imperforate anus did not 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, and this was confirmed by their parents. The children in the index group were more enthusiastic about school than were the children in the comparison groups, and they did not report being bullied. They enjoyed being with friends and spent much time with their best friends.

The children in the index group reported being more constipated than children in the comparison groups. Their parents reported their children to have more fecal leakage, constipation, and odors than parents of the children in both control groups reported for their children.

No correlation was found between the severity of the malformation and psychosocial outcome.

In the CBCL the parents in the index group assessed their children to have statistically significant more total emotional/behavioral problems than the parents in Comparison Group I reported for their children. Teachers of children with imperforate anus reported few problems for the children with imperforate anus in the TRF. No differences were shown between groups.

Paper III showed discrepancies between children's and mothers' responses in all groups and above all on the psychological variables. There were less discrepancies between children's and mothers' responses in the index group and in the group with juvenile chronic arthritis than in the "healthy" control group. 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 the CBCL on the 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. Moreover, the children performed in school as well as the children in comparison groups.

In the TRF, the teachers of children with imperforate anus stated that the children did not work as hard as did the children in the comparison groups. The children in the index group scored statistically lower than children in Group I on the "total adaptive scale". There were no statistically significant differences among groups on academic performance.

6 DISCUSSION

There seem to be certain psychosocial consequences of high and intermediate imperforate anus among the children and the parents who participated in this study. There also seems to be a heavy emotional burden on the parents, seen in *Paper I*, and psychosocial consequences on the children, found in reports specifically from the CBCL and the TRF (*Papers II -III*). However, the children with imperforate anus seem to be psychosocially competent (*Papers II-III*).

The results tend to confirm that the psychosocial consequences seen in this study are linked with high and intermediate imperforate anus. In *Paper I*, the negative emotional consequences of the malformation were obvious 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. These negative emotional consequences were not seen in *Paper II*. However, the index group children confirmed that they were more constipated than the comparison groups. According to their parents, the children in the index group had more fecal incontinence than the children in both comparison groups had.

One explanation for these diverse findings might be the *severity* of the malformation's physical consequences and an underlying assumption that psychosocial consequences depend on illness characteristics. Supposedly the children in *Paper II-III* had less 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 with a different technique, than used today. After 1987, children with imperforate anus at Astrid Lindgren Children's hospital were operated with the Posterior Sagittal Anorectoplasty (PSARP) (Pena, 1985), and a fistula saving technique (Husberg, Lindahl, Rintala, & Frenckner, 1992).

It is difficult to say if the psychosocial function of a child with chronic illness is caused by the illness itself or if 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). Nevertheless it has been

confirmed that patients with anorectal malformations experience psychosocial problems because of the malformation (Hartman et al., 2005).

Moreover, psychosocial outcome was not related to the severity of the malformation (*Paper II*). Disease severity has not been found to be constantly associated with adjustment; children with Inflammatory Bowel Disease (IBD) appeared to have difficulties similar to those of children with other chronic conditions (Mackner & Crandall, 2005b). 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). In addition, surgically corrected hypospadias was not a risk factor for psychosocial maladjustment among boys in middle childhood (Sandberg et al., 2001). Moreover, a notable finding in a study on children with end-stage renal disease was that illness severity was not a significant predictor of adjustment (Madden, Hastings, & V'Ant Hoff, 2002).

Another cause for the positive results in *Paper II* might be that social attitudes in society toward fecal incontinence have improved in recent years. Nowadays it is easier to talk about this subject, and the taboo has more or less disappeared (Kamm, 2003). 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. 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).

However, 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 healthcare staff. Few parents voice concerns about psychosocial problems, and therefore healthcare personnel need to ask both the parents and the children about these issues (Wildman, Kinsman, & Smucker, 2000). Another reason that children may not receive needed psychological care is if their hospital stays are short, in which case physical care is prioritized. It has been reported that nurses had limited time available, and therefore could fulfill only the more immediate needs of ill patients (Ford & Turner, 2001).

It was not surprising that the families had revealed difficulties in *Paper I*. However, the intensity of the parents' suffering was greater than we expected. According to the parents, they had never been asked before about their psychosocial experiences associated with the child's malformations and they had a need to voice their concerns. 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-III*, 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-III*, parents' suffering was not expressed. 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 questionnaires. Few questions addressed the parents' psychological well-being and there might be an indication that the mothers in the index group were more distressed about their children's disabilities (*Paper III*) than were the mothers in comparison groups.

To have a child with a chronic condition influences the relationship between mother and child. The invasive treatment that the parents of the index group children had been responsible for might have had an impact on the mother-child relationship; closeness

was shown between mothers and their children in *Paper I*. The fact that there were less discrepancies in reports from mothers and children in the index group and in Comparison Group I (*Paper III*) could also be a sign of a closer relationship between children with chronic conditions and their mothers.

Closer relationships might stem from concern about the child's anomaly and the associated treatment. It has been stated that the relationship between a child with a chronic illness and his/her mother is dependant on various aspects, and the parents' distress is an important factor in child outcomes (Bonner et al., 2005). 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).

In *Paper III* we examined more closely the differences between mothers' and children's reports on the pair level, within the family. Instead of seeing discrepancies of opinions as faults, these different views may be an important window to the functioning of the child or family system (Holmbeck, Li, Schurman, Friedman, & Coakley, 2002).

It might not be sufficient to look only at the mother-child relationship for explanations; it might be necessary to put the focus more on the whole family. It seems as if research concerning the whole family is necessary to understand families experiencing extraordinary circumstances. 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).

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).

Social function (the ability to participate in school, social and daily living activities) seemed to work fine for the children participating in *Papers II-III*. This was a pleasant and unexpected surprise. The parents agreed that the children enjoyed school and the activities associated with school. In *Paper III*, there were fewer discrepancies found between mothers' and children's responses on variables related to school.

Friends play important roles for all children. For children with a chronic disability the support of classmates and friends might even more central to psychosocial adaptation. In a study on children with limb insufficiencies, lower levels of depressive symptoms and higher self-esteem was associated with higher classmate support (Varni, Setoguchi, Rappaport, & Talbot, 1992). There is evidence in studies that children having 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 stigmatized disability (La Greca, Bearman, & Moore, 2002). At least 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.

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 teacher about the child's disability. It is not fully understood when to disclose information about a child's medical condition to classmates (La Greca & Bearman, 2000). It may have a positive or a negative influence on the other children's attitudes, depending on the visibility of the condition.

As for the children participating in this study, 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 close friends. In *Papers II-III*, the children with imperforate anus reported that they have both close friends and peers, according to themselves and to their parents. 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).

In a review study (Hay, Payne, & Chadwick, 2004) the underlying skills needed for interaction with peers are studied, and there were significant links found, for instance, to the parent-child relationship. We can presume that the parents of the children participating in this study had a positive effect on their children. The opposite was

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).

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 III*). In a study on psychosocial adaptation in adolescents with spina bifida, the authors emphasize the importance of psychosocial interventions in the classroom (Coakley, Holmbeck, & Bryant, 2005).

We did not find any differences in psychosocial outcome in relation to age either in *Paper II* or in *Paper III*. The oldest child participating in *Papers II-III* was under the age of 14. Age may be an important factor; especially later in childhood (*Paper I*). 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 et al., 2005). Children with imperforate anus may experience risk factors unique to this special malformation, given the potentially embarrassing symptoms, that are comparable to those of children with IBD (Mackner & Crandall, 2005b). It would be of vital interest to further study how these children attain psychosocial adaptation during the transition to adolescence, and how they manage to cope with the disability.

The results from the CBCL and the TRF might seem contradictory. The children with imperforate anus showed more internalizing syndromes than the children in Comparison Group I on the problem scales in the CBCL (*Paper II*), but were assessed as being as competent as the other children (*Paper III*). The teachers of children in the index group anus reported no more problems on the problem scales (*Paper II*), though these children were reported to be less adapted and adjusted in school in comparison to children in the comparison groups. This result may indicate that the children with imperforate anus do have some psychosocial problems, although the mean scores fell within the normal range in a Swedish youth population (Larsson & Frisk, 1999).

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. Additionally, the use of this instrument for children with chronic illness has been discussed among researchers;

for its considerable limitations (Harris, Canning, & Kelleher, 1996) and for its limited assessment of psychosocial adjustment (Mackner & Crandall, 2005a).

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 (Ludman & Spitz, 1995). The prevalence of emotional problems was higher than those in normative populations, though incontinent children were not less well-adjusted than those with bowel control.

The influence of having the spotlight more on “healthy” than on “disease” factors has penetrated research in this area in recent years. It has shifted the focus to how the child actually gets along and lives with the illness, rather than assessing a set of objective or subjective symptoms. It has been a shift from “deficit-based” evaluations (e.g. counting symptoms) toward a more positive approach focused on development and adaptation (Knapp & Harris, 1998).

The differences in reports from parents and teachers of children with imperforate anus may possibly explain the difficulties healthcare staff have in observing and noting psychosocial difficulties. Usually the meetings are short and are focused on the more physical issues. This is confirmed in a study on identification of psychosocial problems; 27.5% of children with one or more problems were identified (Horwitz, Leaf, & Leventhal, 1998). Additionally, differences were found in perceptions of children's well-being between healthcare staff and parents of children with chronic conditions (Janse, Sinnema, Uiterwaal, Kimpen, & Gemke, 2005).

The results of these three Papers indicate that children with chronic conditions and their parents need psychosocial support, and that families with children with imperforate anus might need extra support. 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 recourses are needed, it is of importance that the psychiatric health professionals are informed about the child's specific malformation and the treatments being used.

Chronic illnesses differ in etiology and disease characteristics, which will make differences in the psychosocial impact of the conditions on both the children/patients and their families. Imperforate anus requires an understanding of the condition, as it impacts on and interacts with the course of the child's life, on his/her family, and on the society in which they live. Understanding the psychosocial consequences of this special malformation can help to form a collaborative relationship between professionals who care for the child so that biomedical and psychosocial interventions are integrated.

6.1 METHODOLOGICAL CONSIDERATIONS

6.1.1 Qualitative methods

In this study both qualitative and quantitative methods have been used. It is worthwhile to combine methods because each method supplies different information. Qualitative research responds to the “how” issue, and could give deeper understanding of human experiences and why people do as they do (Wolf, 2003; Rowan & Huston, 1997).

The informants 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 to interview that fulfilled the sample criteria. The researchers choose 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 of analysis used was “editing analysis style”, resembling content analysis. When the work is inductive, the researcher looks for prominent patterns in data (Silverman, 2001). The first step is to read the text several times and to find a manageable way to structure data. Afterwards, data should be identified, coded, and categorized. If more than one person is responsible for data, each of them should read the text individually, in order to discuss similarities and differences (Patton, 2002).

The aim of qualitative research is to give knowledge that is valid, i.e. an analysis that contains truth, captures experiences, and deepens our understanding (Kazdin, 1998).

When evaluating qualitative research, a term such as trustworthiness is used (Rowan & Huston, 1997). When trustworthiness is discussed in qualitative research, one has to consider that a text gives different estimations that entail a certain interpretation (Graneheim & Lundman, 2004).

6.1.2 Developing the questionnaires

Because of a lack of available instruments to assess psychosocial consequences of imperforate anus on children, we designed a questionnaire specifically for children with high and intermediate imperforate anus. Many Quality of Life instruments have restricted specificity to one illness group, and there were QoL questionnaires focusing on other chronic conditions such as asthma or juvenile chronic arthritis (Norrby, Nordholm & Fasth, 2003; Reichenberg & Broberg, 2001).

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). However, there can be arguments for new scales when respondents are special (Spector, 1992). When special conditions and treatments are to be evaluated, it can be suitable with an illness-specific questionnaire (Eiser & Morse, 2001b).

Professionals in the area of surgery and child and adolescent psychiatry created the interview questions. This could be regarded as face validity (Polit & Beck, 2004). The questions were structured, semi-structured, and open; there were 88 for the parents and 34 for the children. The questions were considered to cover the child's and parent's experiences in relation to the malformation and its consequences. After analyzing the findings in *Paper I*, we identified variables of interest that were operationalized to formulate questions for the questionnaires. 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 subsequent pilot studies that were performed on somewhat younger children and their parents in the three groups comprised. Even somewhat younger children had no problems answering the questions.

The comments we received from the parents in the pilot studies helped us to increase validity. Revisions were made primarily for the “healthy group”, and after minor linguistic changes the questionnaires were completed.

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). There are no different types of validity, but 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).

6.1.2.1 Scales

In the parents’ questionnaire we used the Visual Analogue Scale (VAS). It represents a free choice on a continuum including anchor words in both ends. VAS is usually used to measure subjective experiences, fatigue, nausea, and pain (Polit & Beck, 2004).

VAS has been pointed out to be risky scale because it is hard to calculate, but with the help of computers 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 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).

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).

There have been discussions of what age children must have reached in order to be reliably able to respond to questions in questionnaires. In a 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, another study 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).

7 CONCLUSIONS

- Parents of children with high imperforate anus have to go through difficult experiences associated with the malformation.
- Children with imperforate anus in this study seem to be psychosocially well adjusted, though their parents state that they experienced leakage and fecal odor.
- The children 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. The presence and directionality of disagreement can give valuable information for future care and where to put in extra effort.

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

8 ACKNOWLEDGEMENTS

Professor Kyllike Christensson, main supervisor. Thank you for taking me as your research student. During these years, you have given me support and good advice, and helped to control the direction of the study. You have given me help when needed, and shared your experience and great knowledge of science.

Professor Per-Anders Rydelius, co-supervisor. Thank you for support and good advice and for having confidence in my ability. You introduced me to the world of research, gave me the responsibility of conducting this study, and facilitated its implementation.

Maria Öjmyr-Joelsson, my research-partner. If it had not been for your cooperation, I would not have completed this study. Our teamwork has been stimulating, unflinching, funny, and fruitful. Coming from diverse units, we now think alike and eventually have become close friends. Thank you for sharing your knowledge and letting me take part of your dedicated work with children with anorectal malformations. You have the qualifications to become an excellent researcher, and you have to go on with this urgent work.

Professor Björn Frenckner, my extra supervisor. You have been supportive and encouraging and an accurate and fast reader. Special credits to you, for being one of the initiators of this study of psychosocial character.

Associate Professor Jan-Olov Larsson. Thank you for statistical assistance, practical support, and a constant interest in our study. You stay calm and patient, even when interrupted by strange questions about analysis, calculations, and hidden books.

Tobias Edbom. Thank you for invaluable help with installing technology, computers, and programs. Thank you, too, for guidance into the road of nursing science, the scientific discussions, and last but not least, for the humor that you inject into work.

Gunilla Nilsson. Thank you for practical assistance and vital discussions. You are the best language reviser and a rescuer when time is short. We could soon start the “red pencil office”!

Anneli Törnblom and Hie Allik, colleagues at the unit for child and adolescent psychiatry, for exchanging interesting thoughts and discussions.

Elisabeth R. Thank you for giving fresh and supportive comments.

Colleagues at the Red Cross High School. Thanks for interest and support, and special thanks to Christina Andersson, who facilitated the fulfillment of this study.

My close friends Ann, Annica, Eva, Krystyna, Maria, for being interested in my work and being concerned about me.

My parents Aina and Ove and my siblings Vivianne, Ralph and Cecilia and their families for all your support and your belief in me.

My close family. My husband Sven for love and support, and my clever, beautiful children Miriam, Elias and Hannah, for being you.

And finally, thanks to all participating children and parents who helped this study become a reality. We are grateful to the staff at the outpatient unit for children with juvenile arthritis and to the day care surgery unit at Astrid Lindgren Children's Hospital, for assistance and for helping us get in contact with "your" patients.

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