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Swedish Parents of Children with Down Syndrome

A study on the initial information and support, and the subsequent daily life

BY

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ABSTRACT

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In this study 165 Swedish parents of young children with Downs's syndrome (DS) were investigated regarding their perception of the quality of the first information and support received after the birth of the child. The parents' opinions were compared with clinical routines at the paediatric clinics regarding these issues. Strong clinical ambitions fell short, however, since 70 % of the parents felt insufficiently informed; 56 % felt unsupported, and the timing of the disclosure varied between 0 hour to >5 days. On the basis of a grounded theory analysis the parents' written narratives regarding the quality of the first information and support were analysed to better understand the reasons underlying the parental dissatisfaction. Criticisms were raised by the parents concerning: the low communication skills by professionals; the lack of privacy; too much negative information; and an unmet desire to early meet other DS parents.

The implications of being DS parents regarding their daily life were examined by measuring parental health, stress, sense of coherence, employment and sick leave rates. Results were compared with those in a randomly selected group of parents of healthy age-matched children. The similarities between the DS and control parents were more pronounced than the differences regarding divorce rates, siblings in the family, time spent on child care, employment and sick leave rates, and their self-perceived health, stress, and sense of coherence. However, self-perceived health of the DS mothers was impaired and stress was increased. A small group of DS parents (5 mothers and 1 father) had an extremely high rate of sick leave and no such group was seen in the control parents. In addition, the DS mothers stayed at home because of the child's sickness most frequently and the DS fathers stayed at home for this reason more than control mothers.

Conclusions: Existing guidelines for optimal first information and support of new parents of children with DS are not always followed in Sweden. Qualitative clinical improvements from the parents' perspective are proposed. Most DS parents live an ordinary family life in respect to the measured parameters, but the risk for health deterioration, particularly in DS mothers, might need attention.

Keywords: Down syndrome; parents; first information; support; disclosure; health; stress; sickness absence; sense of coherence.

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LIST OF PUBLICATIONS

This thesis is based on the following original publications, which will be referred to in the text by their Roman numerals:

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II. Hedov G, Annerén G, Wikblad K. Swedish Parents of Children with Down Syndrome: Parental Stress and Sense of Coherence in Relation to Employment Rate and Time Spent in Child Care. Scand J Caring Sci 2002. In press.

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IV. Hedov G, Wikblad K, Annerén G. Sickness absence in Swedish parents of children with Down syndrome: Relation to self-perceived health, stress (Manuscript)

V. Hedov G, Annerén G, Wikblad K. A qualitative analysis of the first information and support as experienced by Swedish parents of children with Down syndrome (Manuscript).

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GENERAL INTRODUCTION

Among children with mental retardation (MR), one-third are classified as having severe mental retardation (SMR) (IQ<50), and children with Down syndrome (DS) represent one-third of those with SMR. The diagnosis of DS is in most cases obvious immediately after birth, as with other syndromes and malformations such as neural tube defects, skeletal dysplasia, and congenital syndromes such as Cri du Chat and Cornelia de Lange syndromes. The fact that in most cases the diagnosis is evident immediately at birth makes the situation in DS different from that in the other two-thirds of the children born with mental retardation. In the latter group of children the intellectual disability will become increasingly evident with increasing age of the child, and the parents, who are aware that something is wrong, will be waiting for the answer as to whether the child has MR; in some families the diagnosis will come as confirmation of their suspicions (1).

The situation of parents of children with DS (referred to in the following as DS parents) has been extensively investigated (2, 3) and has served as a model for families of children with other disabilities diagnosed immediately after birth. To tell parents that their child is not "normal" and is suffering from a congenital life-long disability such as DS is one of the most difficult tasks in clinical practice (4). Inconsiderate comments by professionals, however trivial, may have deep long-lasting effects on the weak beginning of a new balance for the family (5). The postnatal period, including the first information is, an event, which is never forgotten by the parents (6, 7). They also have a great need for more details and support (8). It has also been shown that a discrepancy exists between what professionals consider to be the best way to disclose the diagnosis and what parents believe to be the best approach (6, 7). There is a lack of evidence-based guidelines and overall agreement about the first information and support provided to parents (10) and about what effect the quality of the first information and support has on the parents. To become parents of a child with a chronic handicap such as DS might result in disruption of the normal family regulatory processes. Routines have to be developed for managing the disability, roles need to be reallocated, and rules have to be altered (11, 12, 13).

AIMS OF THE STUDY

This study was carried out with the following aims:

1. to examine the quality of the first information and support from the view of the professionals, i.e. the clinical goals regarding the initial information and support at the Swedish departments of paediatrics;
2. to examine the quality of the first information and support from the parents' perspective;
3. to investigate the daily life of parents in respect regarding divorce rates, siblings in the family, time spent on child care, employment and sick-leave rates, and their self-perceived health, stress, and sense of coherence;
4. to formulate evidence based suggestions concerning clinical improvements of the first information and support.

BACKGROUND

DOWN SYNDROME

DS is the most common disorder associated with mental retardation. The incidence of DS in Sweden is about 1/800 births (14), which means that 110-120 children with DS are born annually. Individuals with DS do not suffer solely from a mental handicap, but also from medical problems affecting several organs, e.g. congenital heart malformations (50%), growth retardation (80%), thyroid disorders (50%), susceptibility to infections (50%), childhood leukaemia (2%), visual and hearing impairment (70%), and early dementia (>50%)]. An accepted medical care programme for individuals with DS has been available in Sweden since 1990 (15). This programme, however, pays only minor attention to the initial information and support given to the parents.

THE FIRST INFORMATION

The disclosing process and support offered to parents challenge the health care providers' competence in communication, and a close professional relationship with parents at hospitals is rich in intense emotions (16).

Many parents have unresolved inner conflicts that may persist for a long time after the disclosure. There appear to be two different types of reactions. Firstly, there are parents who attempt to deal with the strong long-lasting feelings about the disabled child – sadness, loss of

what might have been, why it happened and so on. Some research has been focused on this topic of chronic feelings of sorrow and grief (17, 18, 19). Other parents are critical of the way in which they were treated initially at the hospital at the time of the disclosure. Many parents have a therapeutic need to retell the story with a hope of seeking confirmation that they were badly treated (10).

In a study in Finland it was found that parents of disabled children who initially were insufficiently informed and supported experienced feelings of insecurity and helplessness and were poorly prepared to take care of their child at home (20).

Pelchat and co-workers showed that in unselected families of children with DS, between six and 18 months of age, the parents experienced increased levels of stress in comparison with those who participated in an early family intervention programme (21). The parents who took part in that early programme (including the first information) perceived gains in several domains, for example they experienced lower levels of stress, anxiety and emotional distress, compared with those who did not participate. They were also more confident in their own resources and had more positive attitudes towards the disability of the child and the parental situation.

In Sweden there have been only minor attempts to improve the situation for the parents over the years. As early as in 1977 clinical guidelines on this topic were formulated with the aim of providing better information and support to parents of children born with mental retardation (22). Those attempts were not based on systematically collected research data, however, but on clinical experiences and observations. Later studies showed that the parents of children with DS in Sweden were less satisfied with the initial support compared with parents from the other Nordic countries (23). It has been shown that the information routines and the parental support at paediatric hospitals differ from place to place in Sweden (23, 24). Thus, parents of children with DS will be exposed to different amounts and qualities of information and support depending on where the child is born.

It is not inevitable, however, that parents of DS children are dissatisfied with the initial disclosing process. Their satisfaction depends on the quality of the information and support received, such as whether they are given some positive information about the child and are given enough time (5, 25, 26, 27, 28, 29).

Guidelines concerning the first information and support given to parents of a newborn child with DS have been formulated previously (5, 25, 30). Such guidelines have often been strongly linked to the commitments of certain health care providers and in times of personnel reshuffling the guidelines may fall into oblivion (personal communication, Cunningham 1998). It has also been stated that the parents' wants and needs must be considered if improvements in this field are to take place (31).

THEORIES ON PARENTAL REACTIONS TO THE FIRST INFORMATION

The present study focuses on how the parents experienced the quality of the initial information and support and does not concern primarily the parent's reactions to the disclosure in terms of crisis and sorrow. However, several theoretical models to explain the reactions of parents of a child with DS or other congenital defect have been developed with the aim of better understanding the parents' situations. These models have derived from experiences in trauma and grief as first studied by Lindemann (32). Over the years the models have undergone refinements, and concepts of crisis, stress and coping have been adopted (33, 34, 35). In Sweden the theoretical model most frequent used in describing the initial situation of parents of children with DS and their reactions of sorrow and grief is the psychodynamic approach proposed by Cullberg (36). He has described the traumatic crisis theoretically as an unexpected emotional situation experienced by individuals – in the current study the DS parents – as threats, in the form of existential threats, threats against the person's identity and social security, or threats against profound individual possibilities of leading a pleasant life. Cullberg used the stage model, describing the crisis in four different stages: the shock, the reaction, the rearrangement, and the new orientation phases. Models following this stage theory have been used by others Gustavsson (1985) and Kollberg 1997 (23, 37) and fig. 1 shows theoretically how parents might adjust after the birth of a disabled child.

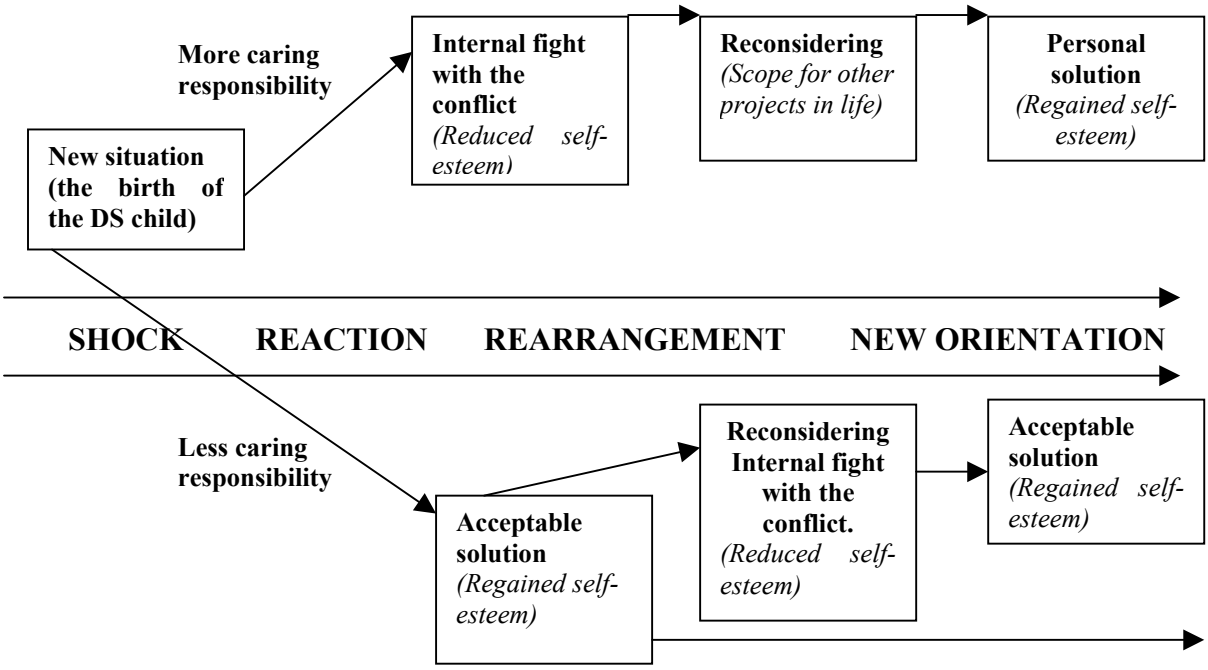

Figure 1. The parents' adaptation and mastering of the conflict between the responsibility of having the disabled child and other own projects in life. (Modified from Gustavsson 1985 and Kollberg 1997.)

Another theoretical principle is that parent adjustment is a process of cognitive re-construction. The birth of a child with DS mostly has the effect of turning the expected, but imaginary child, into countless questions. The majority of the parents experience a sense of loss or grief, and uncertainty about their own feelings for the child and for the future. The parents need sensitive and well-balanced information and support in order to understand and make sense of what has happened. The overriding aim during the initial period is to help them to start thinking about what this event means to them personally (10). The cognitive adaptation theory of Taylor (1983) describes the parents' situation, and his model contains three key elements (38):

1. A search for a meaning in the experience – what caused it? What does it mean for my life now?
2. The need to regain mastery over the event, and control one's life generally – what can I do to manage the situation?
3. An effort to enhance one's self-esteem, usually through social comparison with others.

Theories have been developed from different approaches and disciplines and Ferguson and Ferguson (39) categorise the different approaches and the authors grouped areas of changes in families of children with chronic conditions into (A) Feelings and attitudes of the parents and (B) Activities in daily living in the families. The nature of the changes was divided into law-conformable changes, i.e. (a) Psychodynamic (crisis and grieving theories) and (b) Functionalistic (focus on parental roles and distribution of tasks) and situation-based changes, i.e. (c) Psychosocial (theories of adaptation, shock, stress and coping) and (d) Interactional (focus on interaction between the family and the world around).

THE DAILY LIFE OF PARENTS OF CHILDREN WITH DS

In Sweden some empirical studies regarding the overall situation of parents with DS have been conducted (23, 37, 40, 41). The exploratory study by Gustavsson (1985) dealt with the new situation experienced by the parents of children with a disability, and the associated new unsuspected and unwanted demands placed on them (37). The study addressed the parents' problems in adjusting personal goals in life and the way in which they learned to live with their disabled child. In a retrospective interview study of five parents of children with DS, Borggren (1990) investigated their possibilities of raising a child with DS and their early reactions to the child, and examined the adjustments made by the family as a group (40). Westbom (1992) studied the impact of chronic illness in children on the parental daily life, in a population–based study, which did not include parents of children with DS (42). Bränholm and co-workers (1992), in a study in a northern region of Sweden, included only parents of children with DS, and addressed the life satisfaction and activity preferences of the DS parents (41). The Kollberg study (1997) "Children with Downs syndrome: the parents' perspective" described the parents' situation in Nordic countries (23).

PARENTAL STRESS

It has been stated that DS parents as a group are exposed to more stress and distress in parenthood and in relation to their child compared with parents of healthy children (21, 43, 44, 45). In the above-mentioned study by Cunningham (1995), it was found that one-third of the families of DS children experienced difficulties in the form of greater stress and reduced satisfaction with life in general (10). In a recent Greek study it was observed that mothers of children with DS experienced more stress, as a result of greater total time demands, than mothers of non-disabled children (46).

PARENTAL SENSE OF COHERENCE

The orientation to life questionnaire was developed by Antonovsky in 1987 in an attempt to understand why some of the individuals in a certain affected population remained healthy and some did not (47). It has been claimed that individuals with higher sense of coherence (SOC) scores are better able to deal with increased stress than those with lower SOC scores. It has also been reported that there is a link between SOC and the quality of life in families after diagnosis of illness (48). Studies exclusively measuring SOC in parents of disabled children such as DS have been sparse. However, in a recent Swedish study of a group of parents of children with developmental disabilities, the SOC levels of these parents were found to be lower than those of control parents of healthy children (49). The authors concluded that the salutogenic theory is valuable for understanding psychological adaptation dissimilarities in parents of disabled children.

PARENTAL HEALTH

The occurrence of serious illness in a family, or for example the birth of a child with a serious disability, such as DS, may increase the vulnerability of the family, leading, in turn, to health problems in family members (50, 51).

Studies restricted to the health of parents of disabled children are rare. If the health concept is widened to include parental well-being, parental functioning, coping and adjustment, then some studies have been performed (21, 52, 53, 54). It has been shown that DS parents are subjected to more chronic sorrow (55), depression (56, 57) and marital dissatisfaction (58) than parents of healthy children.

Although both parents of a child with DS are adversely affected, it seems that the mothers are the targets of most of the negative effects (59). Similarly, Westbom reported that the situation of parents of children with physical and mental disabilities differed compared to randomly selected parents of non-disabled children and that particularly the mothers experienced more health problems and had a higher sickness absence rate than the control mothers (42). Roach and co-workers also found that DS mothers experienced more health problems and depression symptoms in comparison with mothers of healthy children (45).

In contrast, however, Gath reported few differences in mental or physical health between parents of disabled children and those of healthy children (60). Concerning daily life

satisfaction and activity preferences, an earlier Swedish study did not reveal any differences between DS and control parents (41). In that study health parameters were not studied. In an extensive UK cohort of DS parents (n=203 families) investigated between 1973 and 1980, it was found that the majority (60-70 %) of those parents had no psychological or health problems and that the overall impression of the families was one of normality (2). In addition, Ryde-Brandt (1988) reported that depression was uncommon in DS mothers (61).

PARENTAL SICKNESS ABSENCE

In the general population, women are more often absent from work because of sickness than are men (62). In a recent Norwegian study, the number of sick-leave days of Norwegian employees was 1.65 times higher in women than in men (63). Traditionally, mothers in general stayed at home and took greater responsibility for the over-all child care (64), especially the care of children with special needs (65, 66).

PARTICIPANTS OF THE PRESENT STUDY

SWEDISH PARENTS OF CHILDREN WITH DOWN SYNDROME (DS PARENTS) (STUDIES I-V)

DS parents (207 parents of 105 families) living in 10 randomly selected geographical areas in Sweden were asked to participate. The areas were selected so that all parts of Sweden were represented. All parents of the children with DS born during the defined period were invited to participate in the study. The mean ages±SD of the responding DS mothers and DS fathers were 37.6±years5.5 and 39.6±years5.9 respectively. The children with DS were born between January 1, 1989 and December 31, 1993 and were between 3.5 and 7 years old at the time of the study (mean age 4.7 years). Parents whose children had severe congenital heart malformation and parents who did not fully understand the written Swedish language were not included in the study. During the period 1989-1993, 762 children with DS were born in Sweden. The current study thus comprised 14 % (105/762) of the children with DS born during that period. Since more than 50 % of children with DS have a congenital heart malformation, and such children were excluded, the study covered almost 30 % of the children with DS born in Sweden. Data from 165 parents from 86 families of children with DS, 86 mothers (DSM) and 79 fathers (DSF) were collected, which meant a response rate of 80 per cent. For additional demographic data, see Table 1.

CONTROL PARENTS OF HEALTHY CHILDREN (STUDIES II AND IV)

"Healthy children" were defined as children born during gestational week 38-42 with a birth weight > 2,500 g and without any previous or current medical disorders at the time of the study. The Centre for Epidemiology of the Swedish National Board of Health and Welfare randomly selected 1% of all families with healthy children from three of the ten geographical areas of the study. These three areas were selected so that the northern, central and southern parts of Sweden were represented. The children in the control group were between 4 and 6 years of age. Control parents (237 parents from 120 families) were invited to participate in the study and 169 parents (87 mothers and 82 fathers) from 87 families of healthy children answered the questionnaire, giving a response rate of 71 %. The mean ages ±SD of the responding control mothers and fathers were 35.2 years± 4.9 and 38.3±years 5.8 respectively, and the mean age of the control children was 4.7 years. No significant differences were found between the DS parents and control parents concerning age, education, socio-economic status or marital status. Non-responders constituted 29% of the control group. The mean age of the healthy children in the non-responding group was 4.8 years. No differences in dropout rate were found between the different geographical areas in the control group. The possibility of other differences between responders and non-responders among either the DS or the control parents could not be ruled out. Parents in the study group and the control group were not matched on individual rank, but only on the basis of group characteristics. For additional demographic data, see Table 1.

Table 1. Socio-demographic data for Swedish families of children with Down syndrome and control parents of healthy children.

	(Studies I-V) DS parents n=165	**(Studies II and IV) Control parents** n=173
Mothers (n)	85	89
(mean ages±SD)	37±5.4	35.2±4.9
Fathers (n)	80	84
(mean ages±SD)	39.6±5.9	38.3±5.8
Marital status (families)		
Married/cohabitant n (%)	77 (90)	75(84)
Divorced n (%)	9 (10)	14 (16)
Parental education		
Compulsory school n (%)	23 (14)	22 (13)
More than 9 years n (%)	142 (86)	146 (87)
Gainful employment		
Mothers n (%)	74 (87)	80 (90)
Fathers n (%)	77 (96)	84 (100)
Part-time employment		
Mothers * n (%)	55 of 74 (74)	40 of 80 (50)
Fathers n (%)	5 of 77 (7)	6 of 84 (7)
Child's age (range)	3.5-7	3.5-5.5
(mean)	4.7	4.7
Siblings n (%)	80(94)	80(90)

*χ^2=6.25; df=1; p<0.01

REFERENCE PARENTS OF HEALTHY CHILDREN (STUDY I)

A sample of mothers (n=100) and fathers (n=100) randomly selected from the official SF-36 database of the Swedish norm population served as reference parents in the current study.

This official database consists of 8,930 individuals from whom data had been collected by mailed SF-36 questionnaires. In this population the numbers of mothers and fathers between 25 and 45 years of age and with children below the age of 18 years were 1,550 and 1,120 respectively. Their mean ages (±SD) were 36.2±5.6 and 36±5.6 years. It was from this

population that the present reference group was selected. Other demographic data for the control group besides age and the fact that they had children below the age of 18 were not available. The Health Care Research Unit of the Department of Internal Medicine, Sahlgrenska University Hospital, Göteborg, Sweden, undertook the random selection of this control group of parents.

SWEDISH PAEDIATRIC CLINICS (STUDY III)

In 1992 - 93 a national survey was conducted with the aim of determining how physicians of all departments of paediatrics in Sweden provided the first information and support to parents of children with DS. All departments of paediatrics in Sweden (n=51) completed a questionnaire focused on the way in which they informed and supported parents of children with DS. Ten of the above-mentioned 51 clinics were thereafter selected, so that all parts of Sweden were represented, to participate in the future study concerning the parents' perceptions of how they were initially informed and supported.

There were no differences in the clinical guidelines regarding the first information between the 10 selected departments and the total 51 departments. The 10 randomised departments were therefore considered to be representative (Table 4).

METHODS

In Table 2 the five separate studies are presented due to methods and topics.

Table 2. Overview of methods used in the five separate studies.

Topic	Study	Method
Self-perceived health	I	Questionnaire (SF-36)
Parental stress and Sense of coherence	II	Questionnaire (PPI and SOC)
Quality of the first information	III	Questionnaire* (psychometric)
Sickness absence	IV	Official (RFV) statistics
Quality of the first information (The parents' perspective)	V	Qualitative study (Grounded theory inspired secondary analysis of written narratives)

*Questionnaires distributed both to the paediatric clinics and to the DS parents

ASSESSMENT OF THE FIRST INFORMATION AND SUPPORT (STUDIES III AND V)

The questionnaire sent to the departments of paediatrics in Sweden in 1992 included questions concerning the timing of the first information to DS parents, who was to give the information, how often the parents were to be offered information, and whether the mothers and fathers were to be told together. In addition, there were questions about the opportunities for the fathers to stay overnight at the maternity ward and whether the fathers were offered a possibility of being put on the sick-list.

The questionnaire sent to the parents in 1996, to collect retrospective data concerning the first information and support, was more extensive. It included questions on the same aspects of the first information as were asked about in the questionnaire sent to the departments of paediatrics. In addition, the DS parents were asked about the content, quality and amount of information received (was the information understandable, sufficient and given with confidence?) and who gave the first information (doctor on duty or specialist). Questions were

also included as to whether the parents felt sufficiently informed and supported and had been allocated sufficient time by the physician.

The questionnaires were developed in order to cover the aspects previously reported to be of importance concerning the quality of the first information and support provided to parents of a newborn child with DS (67, 68). A pilot study of the questionnaire was performed on 26 parents of children with DS before the current study (69). The parents in the pilot study answered more than 90 % of the questions and the questions were therefore considered relevant. Some questions were subsequently rephrased or excluded and others were added. The questionnaire reached a Cronbach's alpha of 0.74. Copies of the questionnaires are available from the author.

Questions to be answered with use of the visual analogue scale (VAS) technique (70, 71) were included. Parents were asked to rank the answers to certain questions on a scale from 0 to 10. For example, "Did you think the information you received was insufficient/sufficient?" "Did you think the information you received was difficult to understand/easy to understand?" Low marks on the scales were consistently negative and high marks were positive. A result of 8 or more was interpreted as true positive and results of less than 5 as true negative. Answers between 5 and 8 on the VAS scale were regarded as neither negative nor positive. If the parents put a mark at 8 or higher in answer to the question "Do you think the information was difficult to understand (0) or easy to understand (10)?" this was considered to mean that the parents understood most of the information beyond reasonable doubt. On the other hand, if they placed a mark at 5 or lower on the VAS, this could mean that they had understood at least some of the information. In that case some information, perhaps essential, was too difficult for them to understand.

Among open-ended questions, one concerned whether the parents had important unmet needs regarding the first information. They were asked to state freely whether they thought that anything important was lacking regarding the information and support. (See also study V.)

The questionnaires were distributed separately to the fathers and mothers. Before the questionnaires were sent to the parents, their local paediatricians asked them if they agreed to participate in the study. Only small individual differences between mothers and fathers regarding how they experienced the quality of the first information and support were observed in the current study, and the results are therefore presented for both parents together.

QUALITATIVE ANALYSIS OF NARRATIVES WRITTEN BY THE PARENTS (STUDY V)

In 1996 the parents in the current study completed a questionnaire regarding how they were first told about the diagnosis of DS in their newborn child and how they were initially supported. Mothers and fathers were asked to fill in the questionnaire separately from each other. The parents' ages were 25 to 52 years and the age of their child with DS was between 3.5 and 7 years at the time of the study.

The questionnaire was developed to obtain knowledge about the parents' experiences of the quality of the first information and support. The questions were formulated from both clinical and theoretical points of view. The clinical aspects are based on experience of one of the authors (GA) of giving DS parents information and support. The theoretical points of view were mostly based on studies by Cunningham and co-workers and Cooley (5, 10, 25, 26).

Besides the clinical and theoretical aspects mentioned above, we wanted to elucidate the parents' own experiences of the initial information and support. For that reason the parents were requested to put in writing items regarding the first information and support that they thought we might have missed in the questionnaire. They were given the opportunity to write narratives in other places in the questionnaire, as additional answers to specific questions. They were asked to describe in writing needs that they felt remained unmet regarding the first information and support. All written narratives made by the parents were rewritten verbatim (as word documents) and a secondary analysis with a grounded theory-inspired approach was performed (72, 73) with the aim of better understanding the reasons underlying the parental criticisms.

Theoretical sampling of the narratives

Of the 165 parents, 109 (65 mothers and 44 fathers) contributed with written narratives. A sample of 33 questionnaires was drawn from the 109 with written narratives. These 33 were considered representative concerning social classes and were both from parents who were satisfied with the first information and from those who claimed that they were dissatisfied, and these questionnaires included the most detailed narratives from both fathers and mothers. When these 33 narratives were analysed we read through the remaining 76 narratives to confirm that no further categories of topics (see below, The analytical procedure) were included and that no new information was found that would add to the understanding of the categories.

Of the 33 parents (20 mothers and 13 fathers) who had written the narratives, two of the mothers and 2 fathers had completed nine years of compulsory education, and the remainder

all had higher education. Four of the mothers were not employed, while all fathers were gainfully employed. Fourteen of the mothers and 5 of the fathers considered the first information to be insufficient and the rest of the parents felt that they had been sufficiently informed. Two of the mothers and 3 fathers felt that they had received the support they needed, but the majority of the parents (18 mothers, 10 fathers) did not consider that they had been sufficiently supported.

The analytical procedure

Study V was focused on the experiences of Swedish parents of children with DS regarding the way in which they were first informed about the diagnosis and the support they received at that time. The analysis started with all three authors reading through the narratives in order to get a holistic picture of the material. Two of the authors (GH and KW) then carried out open coding by reading each narrative sentence by sentence to identify major categories and subcategories of topics. In the next step (axial coding) the categories were specified in terms of how they were related to the context, the actions carried out and the consequences of the actions. The method of constant comparison was used for refining the categories. This gave rise to two central phenomena and four categories commented on in all the narratives (Fig. 3).

ASSESSMENT OF PARENTS' DAILY LIFE (SELF-PERCEIVED HEALTH, STUDY I)

The Swedish version of the SF-36 was used for assessing different aspects of parental self-evaluated health (74). The SF-36 is a generic test of self-perceived health and measures health outcomes in eight separate health domains, namely: Physical Functioning, Role-Physical, Bodily Pain, General Health, Vitality, Social Functioning, Role-Emotional, and Mental Health. Each domain comprises two to ten questions. The items in the SF-36 are scored such that a higher score indicates superior perceived health, as shown in Table 3. Table 3 was also used as a tool in interpreting the results. The questionnaire included an additional question on how many hours per day the parents spent in caring for the child with DS.

The SF-36 has been reported to have high validity and high internal consistency, as is summarised elsewhere (74, 75). As in these earlier studies, a high level of internal consistency was found in the present study.

Table 3. Definitions of the SF-36 health domains (during the past 4 weeks) and the meaning of low and high scores

	Meaning of Scores	
Health domain	Low	High
Physical functioning	Limitations in performing all physical activities, including bathing and dressing.	Performs all types of activities, including the most trying ones, without limitations.
Role-physical	Problems with work or other daily activities as a result of impaired physical health.	No problems with work or other daily activities as a result of impaired physical health.
Bodily pain	Very severe and extremely limiting pain.	No pain or limitations due to pain.
General health	Believes personal health is poor and likely to get worse.	Believes personal health is excellent.
Vitality	Feels tired and worn out all of the time.	Feels full of pep and energy all of the time.
Social functioning	Extreme and frequent interference with normal social activities by physical or emotional problems.	Carries out normal social activities without interference by physical and emotional problems.
Role-emotional	Problems with work or other daily activities as a result of emotional problems.	No problems with work or other daily activities as a result of emotional problems.
Mental health	Feelings of nervousness and depression all the time.	Feels peaceful, happy, and calm all the time.

ASSESSMENT OF PARENTAL STRESS (STUDY II)

In study II an instrument for measuring parental stress developed in the US by Hymovich, called the "Parental Perception Inventory" (PPI), was used (76). This inventory was originally produced to evaluate stress in parents of children with a variety of chronic disorders as a direct result of a child's chronic condition, e.g. osteogenesis imperfecta, cystic fibrosis, juvenile diabetes mellitus, biliary atresia, inherited metabolic disorder, spina bifida and haematological disorders (76). The PPI was therefore chosen as appropriate in evaluating stress in DS parents. Jennings and Rogers consider it of great importance to select instruments that are not only psychometrically reliable but also relevant to the population of interest, so that the correlation between construct and empirical perspectives will be enhanced (77). The PPI in its entirety consists of six independent units, namely concerns, beliefs, feelings, coping, general information about the families, and the siblings. The original version of the PPI also included questions on concerns of and coping by the spouse. Each unit of the PPI can be used separately. The concern unit was used in the current study, and is hereafter referred to as parental stress. Comparisons between the DS parents and the control parents were based on 20 items.

The parents were asked to answer questions about how often (during the past 3 months) they had perceived different factors as stressors. Examples of stressors in the PPI are extra time demand, feeling worn out, the child's future, getting enough sleep, own health, enough time for relaxation/recreational activities and enough time alone with the spouse/partner. Each concern was presented on a 3-point Likert scale: never (1), sometimes (2) or often (3). The mean PPI scores were used for comparing parental stress between the study and control group. The PPI score could range between 20 and 60, the higher the score the greater the experienced stress. The level of internal consistency of the concern unit in the PPI was calculated with Cronbach's alpha coefficient and reached 0.89.

The translation of the PPI from English into Swedish was made by a previously tested translation-back translation procedure (IQOLA PROJECT). One of the authors (GH) made a forward translation into Swedish and an independent translator performed the back translation into English again. A new forward translation into Swedish was performed and the first and the second Swedish versions of the PPI were compared. The originator of the PPI, Professor Debra Hymovich, Charlotte, North Carolina, US was initially contacted and permission to translate and use the PPI in Sweden was received. Professor Hymovich was available via e-

mail and letters and she was interested and supportive. The first version was pilot-tested in 26 parents of DS children from a local cohort (69).

ASSESSMENT OF PARENTAL SENSE OF COHERENCE SOC (STUDY II)

The short form (13 items) of the Orientation to Life questionnaire developed by the late Professor Antonovsky in 1987 was used in this study (47). The choice of this instrument for measurements in study II was motivated by its universal usefulness and its ability to cut across lines of gender, social classes, religion and cultures, as stated by Antonovsky (78). Professor Antonovsky was contacted by mail in 1993 and was supportive to our studies, but he had no experiences of his own in using the SOC in parents of disabled children. The SOC questionnaire was initially developed in an attempt to understand why some of the individuals in a certain affected population remained healthy and some did not. The 13 items refer to a wide range of situations related to the three SOC components comprehensibility (5 questions), manageability (4 questions) and meaningfulness (4 questions). The 13 items are universal statements answered on Likert scales, with seven possible positions. For example: "When you talk to people, do you have the feeling that they don't understand you?" (comprehensibility), "Do you have the feeling that you don't really care about what goes on around you?" (meaningfulness), and "Do you have the feeling that you have been treated unfairly?" (manageability). Cronbach's alpha measure of internal consistency in the SOC has ranged from 0.74 to 0.91 in previous studies (79). The current study showed an alpha level of 0.88.

ASSESSMENT OF PARENTS' SICKNESS ABSENCE AND PARENTS' TEMPORARY CASH BENEFIT (STUDY IV)

In Sweden there are two different forms of sick leave, namely that due to one's own sickness (sickness benefit) and that due to sickness of a child (parent's temporary cash benefit). The parent's sick-leave rates (own sickness) and temporary cash benefit rates (care of sick children) in both groups (DS and control parents) were obtain from the National Swedish Social Insurance Board (RFV) and the data covered four years from January 1 1997 to December 31 2000. The sickness benefit and the temporary cash benefit can be paid at full- or part- time rates (three-quarters, half or one-quarter), depending on the extent of the loss of working capacity. In study IV, days with partial payments were consistently recalculated as full days.

ETHICS

The Ethics Committee at Uppsala University approved all parts of the study. In addition, legal representatives at RFV made a thorough inspection of our application for access to the sickness absence data (study IV). They considered our aims as trustworthy and the study was judged to be appropriate.

STATISTICS

Study I

The instructions for scoring of the responses in the health questionnaire recommended in the scoring manual for the SF-36 were followed. A mean score for each of the eight health domains is computed. The mean score for each domain can range from 0 to 100, with a higher score indicating superior health. For group comparisons of the mean scores of the SF-36 health domains, Student's unpaired two-tailed t-tests were used. Differences obtained in three group comparisons were evaluated for significance, namely differences between DS mothers and DS fathers, differences between mothers and fathers of the control group, and differences between DS parents and parents of the control group. Additionally, the Bonferroni test of significance was used (α=0.05/32 or 0.002). This test is a more conservative post hoc method for avoiding the phenomenon of mass significance, since multiple comparisons was accomplished. Accordingly, a p value of <0.002 was adopted as significant and higher p values were regarded as tendencies. Differences between the mothers and fathers in the study group regarding the amount of time devoted to caring for the DS child were tested with the Chi-square test.

Study II

The results obtained with the PPI are presented as a percentage distribution describing how many of the parents had perceived each particular item as a source of stress during the past 3 months, and how often. The Chi-square test was used to detect differences in the frequency distribution between the parents. A mean PPI value was also used for comparing the level of stress between the parents. Student's t-test was applied for testing the significance of differences in mean PPI values between the DS and control parents. Pearson's correlation was used to describe the relationship between parental SOC and PPI values.

The scores for the 13 SOC items were summarised into a total SOC score. The total SOC scores were categorised as 75-87 (strong SOC), 61-74 (moderate SOC) and 27-60 (weak

SOC), as proposed by Langius and Bjorvell (80). Comparison of mean PPI stress values in parents with weak, moderate and strong SOC was made by use of ANOVA.
Differences between the mothers and fathers of the study group regarding the amount of time devoted to caring for the DS child were tested with the Chi-square test. This test was also used when comparing the parents' degree of gainful employment.

Study III

To test the significance of differences regarding how long after the birth the parents received the first information on DS, Student's paired t-test was used. The content of the answers to the open-ended questions, like most of the results, was analysed descriptively.

Study IV

The Chi-square test was used for descriptive statistics and distribution comparisons. Mean values between the groups were compared with Students' paired t-test. Correlations were tested with Pearson's product moment correlation.

RESULTS

THE QUALITY OF THE FIRST INFORMATION AND SUPPORT (STUDIES III AND V)

The first information and support were studied in different ways. Firstly, a questionnaire with questions based on the literature and clinical experiences was sent to all paediatric departments. Secondly, a questionnaire with the same questions regarding the first information and support was sent to parents of children with DS at 10 of the paediatric clinics. The questionnaire sent to the parents also included space for writing narratives about observations concerning their experiences of the quality of the information and support received. The written narratives were analysed with a qualitative grounded theory-inspired approach aiming to achieve better understanding of parental criticisms.

The departments of paediatrics (the views of professionals):

The clinical goals regarding the first information and support provided to the parents of children with DS are presented in Table 4. As this table also shows, there was a high degree of similarity between the clinical goals of all paediatric clinics and those of the ten included in the study. The ten selected clinics where the children with DS were born were therefore considered representative.

Table 4. Description of clinical goals of all Swedish paediatric departments and of the 10 studied ones regarding the provision of the first information and support to parents of children with Down syndrome and the reported experience of 165 such parents with respect to this information and support.

	Clinical goals of all 51 Swedish paediatric departments	**Clinical goals of the 10 studied paediatric departments**	**Reported experiences of DS parents**
Immediate information if DS clinically suspected	63%	80%	52%[1]
Information given by paediatrician on duty	53%	60%	85%
Give parents daily information	42%	40%	14%[2]
Parents always told together	96%	100%	79%
Always offer the fathers placement on the sick-list	58%	50%	65%
Always offer the fathers opportunity to join the mothers and stay overnight at the maternity ward	65%	60%	65%
Establish contact with the public habilitation activity during the neonatal period	69%	70%	46%[3]

[1]=within 10 hours after the delivery
[2]=daily information or when the parents asked for it
[3]=within 1 month after the delivery

The parents' view of the first information and support (semi-structured questionnaire):

The DS parents were initially informed that their child had DS within a wide range of times after the birth. Sixty-one families were informed about the diagnosis within 24 hours. Ten families received the first information three days or later after the birth (Table 5). Three different groups were defined on the basis of whether the parents perceived the timing of the information as acceptable or not: the "too late" group, the "acceptable" group, and the "too early" group. The mean times of disclosure in the three groups are presented in Figure 2. Thirty-seven of the families were satisfied with the timing of the disclosure of the diagnosis, irrespective of when the information was given to them. More families were dissatisfied with the timing on the grounds that they received the information too late, while a smaller group of dissatisfied families felt that they were informed too early.

Figure 2

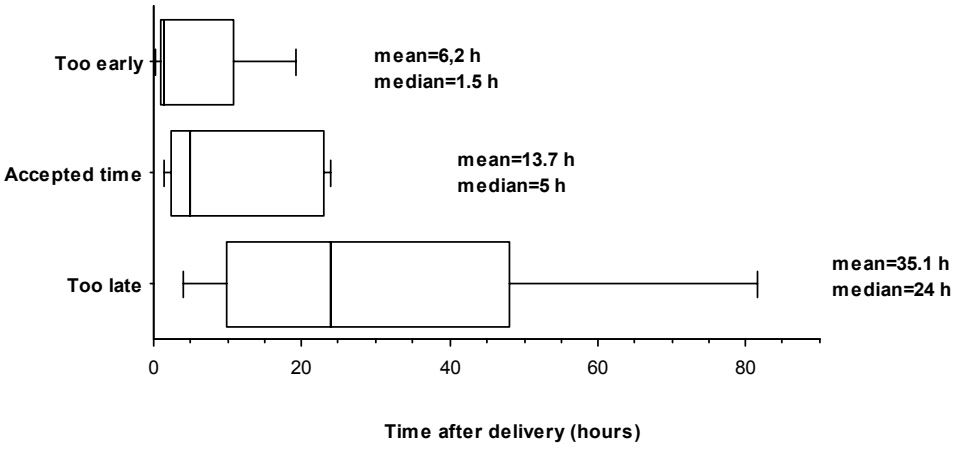

Figure 2. Timing (in hours after the delivery) of the first information about Down syndrome in Swedish families, divided into the groups "too early" (n=7) "acceptable timing" (n=39) and "too late" (n=31).

A significant difference in the mean time of receiving the first information was found between the "acceptable" and the "too late" groups ($t=2.8$; degrees of freedom $df=30$; $p<0.001$), but not between the "acceptable" group and the small group of families (n=7) who considered that they were informed "too early".

Table 5. Time after the delivery of a child with Down syndrome when the families received the first information about the diagnosis, and number of families who were satisfied with the timing.

Time after delivery	Families informed n	Families satisfied with the timing n
≤ 2 hours	18	12
3-5 hours	13	10
6-10 hours	11	4
11-24 hours	19	8
1-2 days	10	2
3-5 days	8	3
> 5 days	2	0
	81	37

In 79 % of the families (61/77) the mother and father received the initial information about their child's condition together, and in 21 per cent (16/77) the mothers were told alone.
Sixty-five per cent of the parents (107/165) were interviewed in undisturbed conditions on two or fewer occasions. Nineteen per cent (15/79) of the fathers were never offered any opportunity to be present when the diagnosis was disclosed.
Eighty-five per cent of the families (73/86) were told about the diagnosis by the paediatrician on duty.

The parents' experiences of the quality of the first information and support, described in the questionnaire, are summarised in Tables 4 and 6. Fourteen per cent of the parents had received information daily or when they asked for it. Seventy two per cent of the parents would have liked more undisturbed interviews with information about their child's condition. The parents who had such interviews on two or fewer occasions would have liked more information.

Table 6. The experiences of Swedish parents of children with Down syndrome regarding the first information and support.

Answers to question	Parents (n=165)
Yes, the information was insufficient	70 % (n=116)
We did not understand the information	46 % (n=76)
The informer did not give us enough time	44 % (n=72)
The informer was not confident	47 % (n=77)
We felt insufficiently supported	56 % (n=93)

The father's situation

Sixty-five per cent (51/79) of the fathers were offered an opportunity to stay overnight at the maternity ward together with the mothers, and 65 % (51/79) of the fathers were also asked whether they wanted to be put on the sick-list. Not all fathers accepted these offers. Sixty-nine per cent (35/51) accepted the offer to stay overnight and 78 % (40/51) agreed to be put on the sick-list. As seen in Table 4, there was relatively high concordance between the clinical goals and the way in which the fathers were actually treated.

The qualitative analysis using, a grounded theory-inspired approach, of the parents' written narratives

The results are presented as two central phenomena, the context in which the information and support were given, and the message i.e. disclosure of the diagnosis. These phenomena consisted in turn of four core categories: the messenger, the content of the information, the situation, and the parents' reaction to the news (fig. 3). The parental perceptions of the quality of the initial information and support included normal reactions of parents to a highly stressful situation, due both to the fact that their child had got DS and to inconvenience in some of the other three categories (the messenger, the content of the information and the situation). In the following some examples of the parents' narratives are given.

The messenger:

-Nobody really took time to listen to our despair. It was obvious they wanted to end the conversation and move on (P84)

The content of the information:

-I longed for someone who saw the child, the child's character and the family in a holistic perspective (M116)

The situation:

-….. I was given no information or support. The doctor did not speak to me, but he spoke to the nurse and then he left me on my own. I rushed to my room and everything felt upside-down, a terrifying moment (M14)

Parent's reaction to the disclosure:

- *…This came as a direct shock to me. The worst thing I had ever experienced* (P103)

Since it was not the principal aim of the present study to analyse the parents' reactions to having a child with DS, the questions were not formulated to elucidate this issue. Nevertheless, many parents spontaneously stated that the experience was so strong that they felt that the world had collapsed, or described it as the most horrifying event ever.

THE CONTEXT

THE MESSAGE

THE SITUATION	THE CONTENT OF THE INFORMATION
THE MESSENGER	PARENTAL PERCEPTIONS

Figure 3. Shows the four categories regarding the context and the message of the diagnosis (first information and support of parents of children with Down syndrome).

THE PARENTS' DAILY LIFE WITH THEIR CHILDREN WITH DS

The self-perceived health of DS mothers versus DS fathers

The mean score in the Vitality domain differed significantly ($p<0.0005$) between DS mothers and DS fathers. The General Health, Social Functioning and Role-Emotional domains displayed tendencies towards lower, less favourable mean values in the DS mothers. These results indicate that DS mothers considered themselves to be tired all the time. Furthermore, compared to DS fathers, the DS mothers showed a tendency to rate their health as poorer and more likely to continue to deteriorate. They also had more difficulties in carrying out normal social activities, work and other daily activities on account of emotional and physical problems.

The self-perceived health of control mothers versus control fathers

No significant difference was observed between control mothers and control fathers in any of the studied domains.

The self-perceived health of DS mothers versus control mothers

In two of the eight SF-36 domains, significant differences ($p<0.001$) were found between DS mothers and control mothers. The DS mothers had significantly lower mean scores in the Vitality and Mental Health domains than the control mothers. These results implied that DS mothers more often experienced fatigue, and that they felt nervous and depressed most of the time. In addition, the DS mothers had lower scores for another four SF-36 domains, namely the Role-Physical, General Health, Social Functioning and Role-Emotional domains. Thus the DS mothers tended to have more frequent problems with work or other daily activities as a result of impaired physical and emotional health, to evaluate their health as poor and likely to continue to worsen, and to find that their physical and emotional problems caused extreme and frequent interference with normal social activities.

The self-perceived health of DS fathers versus control fathers

Compared to the control fathers, the DS fathers were significantly more affected in the Mental Health domain ($p<0.002$). The mean score in the Vitality domain was also lower in the DS fathers. This meant that the DS fathers experienced nervousness and depression most of the time and that they also tended to feel tired and worn out more often.

The differences between DS mothers and DS fathers were more pronounced than those between the mothers and fathers of the control group.

PARENTAL STRESS AND SENSE OF COHERENCE

Parental stress, DS mothers vs. control mothers

Sixty-seven per cent of the DS mothers were stressed by feeling extra demands on their time, compared with 46% of the control mothers (χ^2=8.114; df=1; p<0.01). Feeling worn out was another item that differed between DS mothers (44%) and control mothers (27%) (χ^2=5.969; df=1; p<0.025). To experience limitations in getting time alone with their spouse was perceived as stressful by 44 % of the DS mothers and 27 % of the control mothers (χ^2=5.969; df=1; p<0.025). Experience of limitations in doing activities together as a family was reported to be stressful by 27 % of the DS mothers and 12 % of the control mothers (χ^2=6.516; df=1; p<0.025). The above-mentioned differences between the DS mothers and the control mothers were all seen in areas perceived as time demanding in parenthood (Table 2). The item regarding the child's future was perceived as stressful by 44% of the DS mothers, compared with 11% of the control mothers (χ^2=23.056; df=1; p<0.0001). Furthermore, 31 % of the DS mothers and 10 % of the control mothers considered it stressful not to get enough sleep (χ^2=11.630; df=1; p<0.001).

Parental stress, DS fathers vs. control fathers

In three of the 20 stress items significant differences were found between DS fathers and control fathers. Fifty-seven per cent of the DS fathers and 35 % of the control fathers felt extra demands on their time (χ^2=7.556; df=1; p<0.01). Forty-eight per cent of the DS fathers felt stressfulness when thinking of the child's future, compared with 13% of the control fathers (χ^2=24.790; df=1; p<0.0005). To find someone to stay with the child was perceived as stressful by 23 % of the DS fathers and 8 % of the control fathers (χ^2=6.227; df=1; p<0.025).
The 18 DS fathers who spent less than 1 hour daily in direct child care perceived significantly more stress (mean PPI score 41) than the 18 control fathers (mean PPI score 35) with the same amount of daily child care (t=2.27; df=17; p<0.025).

Parental stress, DS mothers vs. DS fathers

In two of the 20 stress items significant differences were found between DS mothers and DS fathers. Forty-four per cent of the DS mothers felt worn out, compared with 27 % of the DS fathers (χ^2=5.555; df=1; p<0.025). On the other hand, concern about the spouse's health was

significantly more common (χ^2=4.463; df=1; p<0.05) in the DS fathers (18%) than in the DS mothers (7%).

The same pattern was observed among the control parents. Eleven per cent of the control fathers felt concerned about their spouse's health, compared with only 2% of the control mothers (χ^2=5.223; df=1; p<0.025). This was the only significant difference observed between control mothers and control fathers. There was no significant difference in parental stress between the DS mothers and DS fathers who spent less than 1 hour daily on child care.

Parental sense of coherence

In this study no significant differences in mean parental SOC scores were found between the DS mothers and DS fathers or between the DS parents and control parents (Table 3). The percentage distribution of the SOC categories weak, moderate and strong between the groups of DS mothers and fathers (DSM and DSF) and control mothers and fathers (CM and CF) was as follows: Weak (DSM = 27%, CM = 25%, DSF = 15% and CF = 19%), Moderate (DSM = 42%, CM = 46%, DSF = 53% and CF = 50%) and Strong (DSM = 31%, CM = 29%, DSF = 32% and CF = 31%).

Relationship between parental stress and sense of coherence

The correlation between the mean SOC and mean PPI scores was significant both in DS parents (r=-0.456; p<0.0001) and in control parents (r=-0.543; p<0.0001). Compared to parents with weak and moderate SOC those with strong SOC showed significantly lower mean PPI stress values (DS parents: F=17.25, df=2/160, p<0.0001; control parents: F=24.31, df=2/163, p<0.0001) (fig. 4).

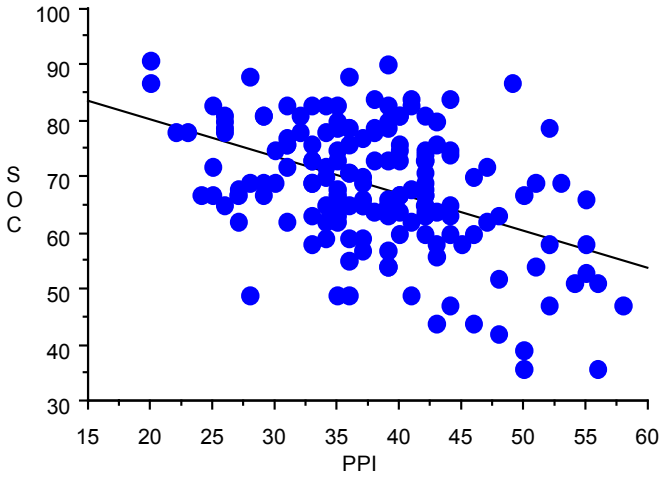

Figure 4. The correlation between the mean PPI score and mean SOC score in the combined group of parents of children with DS and control parents of healthy children.

Parents' gainful employment

Seventy-four (86%) of the DS mothers and 80 (92%) of the control mothers were gainfully employed. The corresponding figures for the DS fathers and control fathers were 96 % and 100 % respectively. Thus, no difference in total employment rate between the DS and control parents was found. On the other hand the DS mothers more commonly worked part-time than the control mothers (55 out of 74 and 44 out of 80 respectively) (χ^2=6.25; df=1; p<0.01).

Amount of time spent on child care by the parents

Seventy per cent of the DS mothers spent 3 hours or more each day on direct care of the DS child, compared with 30 % of the DS fathers (χ^2=25.56; df=1; p<0.0001). In the control families the corresponding proportions were 66 % and 26 % respectively (χ^2=18.13; df=1; p<0.0001).

The eight DS mothers who spent more than 8 hours daily in child care perceived their stress as significantly greater (mean PPI score 44) than the seven who spent less than 1 hour in child care each day (mean PPI score 37) (t=1.79; df=1; p<0.05). In addition, the eight DS mothers with a greater amount of daily child care reported significantly greater self-perceived stress (mean PPI score 44) than the nine control mothers (mean PPI score 35) who reported a similar amount of daily child care (t=1.84; df=1; p<0.05).

Sickness absences of DS and control parents (Study IV)

Own sickness

About 70 % of the DS parents and control mothers and 80 % of the control fathers had no days on sick leave for their own sickness during 1997 – 2000.

Five of the DS mothers and one DS father had >400 days on sick leave, and this number constituted more than 50 % of all reported days of sick leave in the DS group (Table 7). When those six parents were excluded, no difference between the DS and the control parents was seen concerning the number of days on sick leave due to their own sickness. Those six parents did not differ from the other DS parents with regard to age, education, employment rates, marital status or number of siblings. The parents of this small group experienced worse self-perceived health and seemed to be more stressed and stayed home from work because of a child's sickness more days than the rest of the parents. None of the control parents had >400 days of sickness absence during the study period.

Table 7. Mean days of sickness absence in Swedish parents (n=159) of children with Down syndrome and control parents (n=173) of a healthy children and a group of DS parents with an extremely high rate of sick leave days during the period 1997 – 2000.

Number of days (mean)	**DS parents >400 days** (n=6)	**DS parents** (n=159)	**Control parents** (n=173)
Mothers	628	19	25
Fathers	449*	14	9
Total	598	17	17

*=only one father

Temporary cash benefit (the care of a sick child)

In 95 % of the DS and 81 % of the control families the parents stayed at home because of illness in a child during the study period (Table 8). Eighty-five per cent of the DS mothers and 71 % of the control mothers took care of a sick child, and the corresponding figures for the DS and control fathers were 70 % and 50 % respectively. The DS mothers stayed at home because of a child's illness 3.0 times as many days as the control mothers and the DS fathers

stayed at home 3.5 times as many days as the control fathers and 2.3 times as many days as the control mothers.

Table 8. Parents' temporary cash benefit, (care of sick child) in families of children with DS, and in control families with healthy children during a period of 4 years (1997-2000).

	DS parents		Control parents
Number of parents			
Mothers	72	*	63
	*		**
Fathers	56	**	42
Total	127	**	105
Number of days			
Mothers	1827	*	606
	ns		ns
Fathers	1436	*	406
Total	3264	*	1015

* p<0.05, ** p<0.01

In DS parents, mean sickness absence showed almost no correlation with parental self-perceived health, stress, SOC (Table 9) regardless of whether the DS parents with a high rate of sick leave were excluded from the calculation or not.

Table 9. Correlations between sickness absence and parental self – perceived health (SF-36), and stress (PPI) in Swedish parents of children with Down syndrome and control parents of healthy children.

	Correlation	p-value
DS parents		
SF-36	0.17	<0.05
PPI	0.08	n.s.
Control parents		
PPI	0.03	n.s.

DISCUSSION

This study has given information on the way in which parents of a child with Down syndrome perceived the quality of the first information and support after the birth of the child, and the degree of agreement between the parents' perception of this information and support and on the clinical practice. The study has also provided an insight into the daily lives of Swedish parents of children with DS regarding the amount of time they spend on child care, and the parents' self perceived health, stress, and sense of coherence, employment and sickness rates, marital status, and number of siblings.

PARENT'S VIEWS OF THE FIRST INFORMATION AND SUPPORT

The routines of providing information, including disclosure of the diagnosis to new parents of a child with DS and offering support, at the paediatric clinics in Sweden vary. Relatively strong efforts to inform and support the parents properly are made at the clinics, but clearly the efforts fall short in many cases, since a large proportion of the parents expressed criticisms of the manner of professionals and stated that they did not feel sufficiently well informed and supported. The reason for this might be a lack of uniform guidelines within this area, in spite of the fact that today we have so much empirical knowledge about what these parents need initially. The situation is so crucial for the parents that improvisations by professionals should be avoided and well-considered guidelines should be followed. On the other hand, the negative experiences described by the parents regarding the first information and support might be overstated, since so many children with DS grow up in families with warm attachment to the child (2).

In this investigation the question of the first information and support to DS parents was addressed in two ways. Firstly, the parents answered directly formulated questions in a questionnaire, concerning, for example, the timing of the information, where the parents received the information, the amount of information, how they were supported, whether they understood the information, whether the information was given to the parents together, whether the informer was confident, whether sufficient time was allocated to them, and whether help was given to the fathers to encourage their involvements. This questionnaire was sent both to the departments of paediatrics and to the DS parents. The questions were derived from the literature in this field as well as from experience in clinical practice in informing and supporting parents of DS children. The results from the current study showed that 70 % of the

parents thought that the information was insufficient, 56 % felt unsupported, 46 % did not understand the information, and 44 % felt that they were not given enough time. The parents also expressed an unmet need to meet other DS parents.

Secondly, written narratives by the parents were interpreted as expressing their view of how they were initially informed and supported, and served as a complement to the questions in the semi-structured questionnaire. The parents' criticisms regarding this initial information and support process at the hospital concerned to a great extent the manner and attitudes of the health care professionals.

The communication between the DS parents and the staff at the hospital who received them after the birth of their child with DS was in many cases obviously not satisfactory. In an attempt to explain the results, two communication theories in the area of medicine were used for clarifications.

COMMUNICATION THEORIES WITHIN MEDICINE

With the aim of increasing satisfaction with medical information in parents of sick children, Barbara Korsch and co-workers, as early as in 1968, developed an affective communication model 81, 82 83). They collected data from 800 consultations at the paediatric clinic of the children's hospital in Los Angeles. The purpose of this was to decrease the phenomenon of one-way communication that is mostly used in medical care: that is, the health care provider decides what the parents need to know and then communicates this information. The parents' degree of satisfaction with the information was related to three affective behaviour ratings in the messenger. Parents were more satisfied with the information if the messenger act sympathetic, seeming to understand their situation, and if the messenger was a good communicator. The Korsch model deals with affection and social interaction and is based on the idea that if those vital components are missing in the interaction between the two parties, the rate of satisfaction of the parents will decrease.

In contrast to this affective social model, a cognitive model was developed by Ley and colleagues (84). Ley argues that satisfaction of the parents with given information is dependent both upon the parental understanding of and upon their memory of the information. Parents are likely to be more satisfied and willing to act in accordance with the information if they understand and remember it. Ley suggests three interconnected reasons why parents do

not understand received medical information: the information given to them is too difficult to understand, the parents lack basic technical medical know-how, and the parents have mistaken beliefs, which make understanding more difficult. In summary, if parents have problems in understanding given information, they will not remember it and will be more likely to be less satisfied with the information. Ley considers that the model is valuable both for theory and for practice. He described it schematically as follows (fig. 5):

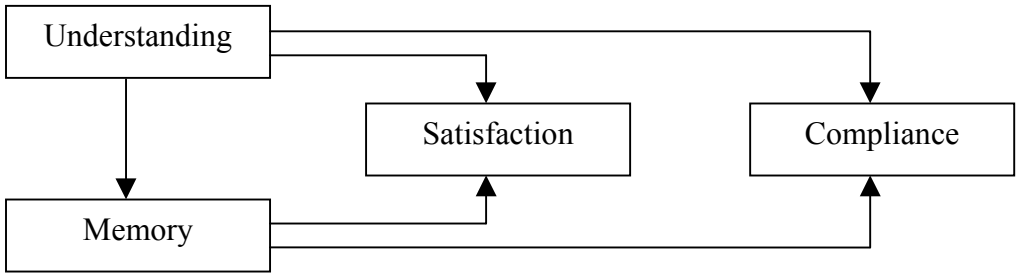

Figure 5. A model of satisfaction and compliance in communication in medicine (Ley 1982)

The current study offers support to both of the theories above. Since most of the criticisms raised by the parents dealt with the manner of the health care personnel, Korsch's affective and social interaction-based theory seems valuable. In addition, the parents' written narratives support this theory. Nevertheless, nearly half of the parents mentioned that they did not understand the information, and the written narratives by the parents describing the language used by the doctors as too technical also give support to Ley's cognitive communication theory.

THE FIRST INFORMATION AND PARENTAL SATISFACTION

As seen in (Table 10) the proportions of parents who have been found in different studies to be satisfied with the first information have varied between 25 and 61 per cent. The corresponding percentage in the current study was thus in accordance with the results of others. Among unselected populations of parents of DS children, about 50 % are usually dissatisfied with the first information and support, which seems to be a relatively constant figure over time.

Table 10. Parental satisfaction with the first information concerning the diagnosis of Down syndrome .

	Parents satisfied (%)	N	Year
Drillien & Wilkinson	60	71	1964
Berg et.al.	54	95	1969
Phillips & Smith	61	54	1976
Lucas & Lucas	52	79	1980
Cunningham et.al.	42	62	1984
Murdoch	50	117	1991
Kollberg*	37	104	1992
Rastelli & Pothos	25	40	2000
Current study	**45**	**165**	**2002**

*The sample of Swedish DS parents in the Kollberg Nordic study.

It has been shown, however, that it is not inevitable that the parents of children with DS are dissatisfied with the first information and support. Cunningham and co-workers recruited 11 families of children with DS and gave them the greatest possible amount of information and support at the birth, with continuous support for at least six months. The information included, for example, maximal oral and written information about DS, and information about how the societal support was elaborated, the parents were assigned a contact person at the hospital initially, and they were given a telephone number to call whenever they needed (25).

In Sweden the initial situation at the hospital described in the interview study by Gustavsson (1985) regarding professionals at the delivery, maternity and paediatric departments seemed to be different from that described by the parents in the current study (37). Gustavsson described a “new era” in the health care of disabled children in 1985 and he wrote within the concept “principles of normalisation”, which meant a new possibility for disabled children to live at home with their parents. Even at that time many professionals were still suggesting parents of disabled children that they should place their child in an institution. Today in Sweden it goes without saying that a child with Down syndrome shall live at home with his or her parents. No parents in the present investigation wrote that initially they had talked to any

professionals about alternative forms of placement and the topic never came up for discussion. The parents in the study by Gustavsson describe the atmosphere at the paediatric and maternity departments in terms of supporting the parents in their "new" roles as parents of a disabled child and of helping them to be able to take care of the child at home by themselves. The atmosphere was influenced by this "new era" and the manner of professionals at that time seemed to encourage the parents. In so far as parents in the study by Gustavsson described their reactions at the birth of the DS child as traumatic and with strong feelings, few critical voices against the manner of the messenger or other staff were raised.

WHAT PARENTS ARE USUALLY DISSATISFIED WITH?

Criticisms expressed by parents in the current study included the following: insufficient information and support, too little time allocated, and difficulty in understanding the information. In addition, one of five mothers stated that they alone were told the diagnosis, and other reasons for dissatisfaction were the timing, the place where the information was given, and the manner of the professionals when informing and supporting the parents. Some parents also stated that clinical routines were lacking regarding this situation. The DS parents in the current study were given information about the disability in their child in a context that was too negative for them and they also expressed an unmet need to meet other DS parents.

Cunningham found that the criticisms expressed by parents were usually similar and fell into three broad categories (10). Firstly: The manner of the person giving the diagnosis; for example it was too insensitive and cold or too businesslike. The information was difficult to understand and sometimes given in medical jargon. Secondly: Problems with information; for example the information was insufficient, there was a lack of information on what could be done, or the information was highly negative, misleading, contradictory or not updated. Thirdly: Organisational aspects; for example delay in giving information, and difficulty for the parents to get access to help, lack of privacy and lack of co-ordination between actions (10).

THE TIMING OF THE FIRST INFORMATION

It was more usual for parents who were told the diagnosis late to be dissatisfied with the timing, but a few parents (n=7) felt dissatisfied that they were told too early. The qualitative analyses of the parents' written narratives also revealed criticisms about the timing, but 37 of the families were satisfied with the timing of the diagnosis, irrespective of whether they were

told the diagnosis early or late. The results show that the parents who were satisfied with the timing of the initial information received it within a wide range of time. Overlaps in both the ”too early” and ”too late” groups were observed (Fig. 2). In the “too early” group the mean length of time after delivery when the first information was received was slightly more than 6 hours. It seems that some parents of children with DS might feel that they want a period by themselves after the delivery before being given information about the diagnosis.

The timing of the disclosure has been considered by some authors not to be a critical factor. In some situations delay in giving the first information may be appropriate, for example if the birth is difficult or if other medical complications set in, and also, such delay might be acceptable by the parents, after a Caesarean section when they often need time in privacy to recover.

Table 11 presents an overview of changes and differences in the timing of telling parents about a diagnosis of DS. The timing to tell the diagnosis to the parents has changed over time. Before 1959 techniques for chromosomal analysis were very slow and clinicians had to rely on clinical stigmata. Today the principle seems to be to tell the parents about DS as early as possible. This routine is based partly on the wish to avoid complaints that professionals have withheld information from the parents.

Table 11. The time after delivery when families have been first told about the diagnosis of Down syndrome, as reported from different studies

STUDY	Studied period	Number of families	Per cent told within 1 week
Berg et al. (1969)	1929-48	44	11.4
Berg et al. (1969)	1949-68	51	47.1
Drillien & Wikinson (1964)	1950-56	71	22.5
Carr (1970)	1963-64	46	41.3
Gayton & Walker (1974)	1946-73	85	59
Cunningham & Sloper (1977)	1974	30	66.6
Shiono & Kadawahi (1979)	1965-79	137	27.7
Murdoch (1983)	1971-81	123	88
Current study	**1989-93**	**86**	**97.5**

THE DAILY LIFE OF THE PARENTS IN RESPECT TO THEIR HEALTH, STRESS AND WORKING CAPACITY

In this investigation the studies of the daily life of Swedish parents of children with DS concerned their health, stress, sense of coherence, marital status, employment rates, sickness absence rates and time spent on child care.

The overall impression was that most of the DS parents lived an ordinary family life, with only few differences compared to parents of healthy children. This finding supports the concept expressed as “ordinary families with special children” by Seligman & Darling, who emphasised the normality of these parents rather than just looking at the problems these parents may experience (85).

Regarding self-perceived health, no significant differences were found between DS and control mothers in six of the eight measured health dimensions, and similar scores were found between DS and control fathers in seven of the eight health dimensions. Concerning parental stress, more than 2/3 out of 20 hypothetical stress items showed identical levels in the DS and control mothers. In the fathers 17 of the 20 hypothetical stress items did not differ. No significant differences were seen in the mean sense of coherence scores between the parents in the two groups.

The present study showed a significant negative correlation between the overall parental stress (PPI) and the sense of coherence (SOC), as seen in Figure 4. This correlation was found in both DS parents and control parents, implying that parents who experienced lower stress in parenthood also had a somewhat stronger sense of coherence. This similarity between the two groups supports the previously mentioned indications of normality within the DS families.

No difference regarding the amount of time spent on child care was found between the DS and control families. This might be a result of the improved support given by the society to families with children with disabilities such as DS. The aim of this support is to give all parents the opportunity to live a normal family life. In 94 % of the 86 DS families the DS child had one or more siblings. In the control families of healthy children this figure was 90%. Regarding the marital status, 90 % of the DS parents were cohabitants, and the corresponding figure in the control families was 84 %. No significant difference in the total gainful employment rates was seen between the two groups of parents. Almost 70 % of the DS parents had no days on sick leave during the studied four-year period (1997-2000), a figure close to that of the control parents, which was 73 %.

All these findings together show that most of the DS parents live a life fairly comparable to the life lived by the parents of healthy children. Van Riper stated that although the birth of a child with DS involves a change of plans for families, it cannot be taken for granted that the family or individual members are “at risk” for psychological and health problems just because of DS in the child (3). This is in line with the report by Cunningham, who found that the majority (60-70%) of a cohort of 165 families of children with Down syndrome in the UK was harmonious, with high levels of family cohesion as well as perceived satisfaction with life, and relatively normal level of stress (10).

DIFFERENCES BETWEEN DS AND CONTROL PARENTS

Some significant differences were found, however, between the DS parents and control parents, especially between the DS and control mothers.

Regarding the self-perceived health, the DS and control mothers differed significantly in two health dimensions, namely in Vitality and the Mental Health dimensions. The DS mothers more often felt fatigue and were nervous and depressed. The DS and control fathers differed significantly in only one health dimension, namely the Mental Health dimension. The difference in self-perceived health, between the DS spouses was more pronounced in comparison to the control spouses and no significant difference was noted between control mothers and fathers in any of the studied health domains of the SF-36.

Parental stress was measured with a self-administered questionnaire (PPI) that was developed particularly for use in parents of children with chronic disorders. Between the DS and control mothers significant differences were found in 30 % of the 20 stress items asked about, and between the DS and control fathers the scores for 15 % of the items differed significantly. The differences were all in a negative direction for the DS parents and were found mostly in time-restricted items. The DS parents more often felt increased demands on their time as parents, especially the DS mothers. No differences in time spent on direct child care were found, however, between the groups, but the DS mothers who spent more than 8 hours daily on child care had the highest mean parental stress score. For those DS mothers this might imply a constant feeling of "having a bad conscience", with an attempt to combine the care of the child with a professional career.

Only one time-related parental stress item differed between DS mothers and DS fathers in the present study, namely the feeling of being worn out, which was experienced more often by the DS mothers than the DS fathers. Dyson did not find any differences in parental stress between mothers and fathers of school-age children with developmental disabilities (43). It has previously been found, however, that the fathers of children with developmental delays experience less parental stress than the mothers (86, 87).

These results are in accordance with those of Padeliadu, who found that mothers of children with DS experienced the increasing total time demands associated with parenthood as more

stressful than did mothers of healthy children (45). In addition, the DS parents were more concerned about their child's future compared with the control parents.

This study showed no difference in total employment rate between the two groups, but the DS mothers worked part-time significantly more often than the control mothers. Generally, in Sweden today, it is more common that mothers work part-time when the children are small. The proportion of Swedish mothers in general who work part-time gradually decreases when the children grow up, but when the children are 11-16 years old 42 per cent of the mothers still work part-time (88). Earlier studies have shown that mothers of children with DS are more sensitive to their disabled children than are the fathers and are far more likely to sacrifice their professional activities than the fathers (89). Barnett and Boyce found that mothers of children with DS reduced their time in paid work by about 7 hours per week and increased their child care time by 9 hours per week (89).

Regarding sickness absence, a small sub-group of DS parents (five mothers and one father), however, had an extremely large number of days on sick leave. In fact they accounted for half of all days of sickness absence in the whole group of DS parents. The reasons for the high rates in these six DS parents could not be determined, since the medical diagnoses were not obtainable. It is possible that the DS parents in this small sub-group were more vulnerable or that the study coincided with severe, chronic disorders in these persons. No sub-group with such high rates of sick leave was found, however, among the control parents. When this sub-group were excluded the DS parents as a group had no larger number of days of sick leave due to their own illness compared to the control parents. This indicated that the majority of the DS parents do well and live a normal life in respect to their own sickness.

Among the DS parents of the present study it was the mother who stayed at home most of the days if the child was sick. Mothers in general have for various reasons stayed at home and taken greater responsibility for the over-all child care (64). Earlier studies have shown that it is the mother of the disabled child who mostly performs the extra tasks when needed (64, 65, 90). It has also been suggested that gender disproportions in the responsibility for child care could function as a complicating factor in the effort for health equality between mothers and fathers (58). The DS fathers, however, assumed significantly greater responsibility for the care of their sick child compared with both the control fathers and control mothers. In fact the DS fathers stayed at home and took care of the sick child more than twice as many days

compared to the control mothers, and 3.5 times as many days as the control fathers. The present study shows that the child with DS might be a negative factor for both the parents regarding their professional careers.

The sick-leave rate for Swedish women was 15.5 days and for men 8.7 days during the study period (Official statistics from the National Social Insurance Board), and it is well known that women in the general population are more often absent from work because of illness than are men (62). In a recent Norwegian study, the number of sick leave days of Norwegian employees was found to be 1.65 times higher among women than among men (63).

In DS parents, mean sickness absence showed almost no correlation with parental self-perceived health, stress, SOC (Table 9) regardless of whether the DS parents with a high rate of sick leave were excluded from the calculation or not. Although they had worse self-perceived health, compared with the control parents, they did not have more days on sick leave. This meant that deteriorations in self-perceived health and stress seldom resulted in increased sick leave in either the DS or the control parents. This was somewhat remarkable and indicates that self-reported health and stress measures used in affected populations, such as DS parents, do not necessarily reflect the sick leave situation. In contrast, it might be possible for individuals to experience increased stress, poorer self-perceived health or deteriorations in other psychological well-being parameters without being on sick leave. The SF-36 and the PPI reflect parental self-perceived health and stress over a shorter time span, whereas the sickness rates reflect a period of four years.

METHODOLOGICAL CONSIDERATIONS

In the comparisons between DS parents and parents of healthy children in these studies the parents of healthy children represented the norm. There is a risk of interpreting the results through the "misery perspective", since DS parents as a group might have less favourable scores for health, stress or other health-related measures. In the "misery perspective" the parents of children with DS are described and judged in terms of what they seem to regret compared to parents of healthy children. It is important to point out that in the current studies the similarities between the parents of the two groups were more pronounced than the differences.

In the letter accompanying the questionnaire the parents in both the study group and the control group were urged to reply to the questions separately. No other measures to ensure that the parents answered the questionnaires apart from each other were taken. In such situations it cannot be relied upon that the parents comply with the request.

The ability of parents to remember how they first received information about a disability in their child is strongly questioned. Is it possible to rely on parents' statements about actions of professionals in the early postnatal period, some years ago when the parents were in shock and most vulnerable? Do they actually remember what happened if they were emotionally affected? Are their statements trustworthy many years later? The parents' recollection has been considered to be coloured by the shock and confusion experienced after the birth, and later recall of what happened is influenced by later events and rehearsal. Thus their criticisms may be unreliable. Cunningham wrote that it is certainly true that many parents find it difficult to remember all the information provided early after the birth. In fact a major criticism is that they are given insufficient time to assimilate the information (10). The parents' memories of this particular event, however, are mostly characterised by vivid details and strong emotions. This is close to the term of "flashbulb" memories used in the research on individual memory of major emotional events. Rubin and Kozin concluded that emotions and life impacts are better predictors of clarity of memory than rehearsal (91). In conclusion, parental perceptions of how they initially were informed and supported should be considered to have some degree of reliability.

Multiple comparisons were performed with Student's unpaired two-tailed t-tests (Study I) and there was a risk of getting mass significance. Accordingly, Bonferroni's correction for multiple comparisons was additionally used. This statistical method is a more conservative post hoc method for dealing with the phenomenon of mass significance.

CONCLUDING REMARKS:

Present study has shown that

1. There are strong clinical goals regarding the first information and support at the Swedish departments of paediatrics, but those seem to fall short.

2. Seventy percent of the parents felt insufficiently informed, 56 % felt unsupported, and 46 % did not understand the information. The parents of children with DS raised critical voices regarding the first information and support in respect to the content of the information, organisational aspects and the manner of the professionals who informed and supported them initially. The criticisms did not primarily concern the medical information given, but rather the over-all treatment of the new fragile family.

3. Most DS parents live an ordinary family life compared with parents of healthy children in respect to divorce rates, siblings in the family, time spent on child care, employment and sick-leave rates, and their self-perceived health, stress, and sense of coherence. But significant differences were seen in a small group of DS parents particularly DS mothers, who were more frequently employed on a part-time basis, perceived deterioration in their health, and were more stressed. A small group of DS parents (5 mothers and 1 father) had an extremely high rate of sick leave.

4. There is a need for improvements in the initial information and support of new DS parents, since about 50 % expressed criticisms. In this care and informing process, experienced and well-trained professionals should be assigned, as well as a contact person among the staff. Routines should be developed for a “parents supporting parents” programme.

On the basis of the present results and the considerations discussed above, the following clinical guidelines are proposed:

PROPOSITIONS TO IMPROVE THE INITIAL PROCESS OF INFORMATION AND SUPPORT FOR PARENTS OF CHILDREN WITH DOWN SYNDROME (UPGRADE)

Inform the parents together.

Give much information, both oral and if possible written and on several occasions.

Avoid too much negative information, and allow enough time so that questions can be asked.

Develop a parents-to-parent support programme.

Assign early a contact person among the staff.

Be responsive.

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