The adolescent experience in Juvenile Idiopathic Arthritis: A narrative approach

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The adolescent experience in Juvenile Idiopathic Arthritis: A narrative approach

De ervaring van de adolescent met Juveniele Idiopatische Artritis: Een narratieve benadering

(met een samenvatting in het Nederlands)

Proefschrift

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Voor Bente, Tabe en Jet

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1

Introduction

> *Mandy (16 years) "I saw this self-investigation procedure as an opportunity to leave my illness behind, but now I realize that the arthritis is part of who I am. Having arthritis formed me and even helped me in becoming the strong individual that I am now".*

With the advance of medical knowledge and technology, the life expectancy of pediatric patients with serious illnesses increased impressively. Many diseases that once were life threatening have now become chronic conditions.[1] As a consequence of this development, health care's primary focus for these patients has now shifted from acute treatment to long-term regimens.

In the transition from childhood to adulthood, adolescents face the tasks of establishing a satisfying self-identity and developing interpersonal bonds beyond the family, acquiring a new status and moving toward a more autonomous stance with regard to the larger world.[2] The management of any chronic condition during this period forms an additional challenge for the teenager and his or her family. While many adolescents with a chronic illness manage their disease and treatment regimens rather well, reduced adherence to medical regimens and poor disease self-management are commonly reported.[3] During adolescence the responsibility for the management of the disease traditionally shifts from the parents and care-givers to the adolescents themselves. Adolescents are encouraged to develop a more active role in their treatment and to implement the consequences of their disease in everyday life. Clinicians and other caregivers are required to concurrently support the adolescent's emerging capacity for self-management.

It is current practice to stimulate a healthy and normal development of adolescents with a chronic illness. Positivism and strength of patients are strongly encouraged. However, several important questions have emerged from clinical practice. Since one of the developmental tasks of adolescents is to adapt psychosocially to their (peer-)environment, they will be eager not to focus on their illness or handicap, but try to live their lives just like their healthy peers. However, the question can be raised whether it is possible, and advisable, to maintain a one-sided positive and healthy identity in the transition to young adulthood? What are the (long-term) costs of possibly suppressing the more negative sides of dealing with a chronic illness? The developmental desire to belong to a peer group and to act 'normal', might reduce adherence with medication because of embarrassment or disobedience. The pediatrician faces the challenge of managing the medical treatment of adolescents with a chronic disease in a way that conforms to the young patients' perspective. Although nowadays the objective symptoms of disease like joint inflammation or fever are often treatable or manageable with medical intervention, more subjectively experienced symptoms like fatigue or chronic pain are frequently long lasting and more difficult to treat. How can such symptoms best be addressed, by the multidisciplinary team but also by the adolescents themselves?

Innovative approaches to self-management are needed to provide valuable insights into some of these important questions.[4] Not only the clinicians or the parents should provide the possible solutions to these challenges, but perhaps it should be considered of even greater importance to enable the adolescents themselves to successfully deal with the problems they face. This dissertation focuses on the emotional experience, self-positioning and self-narratives of adolescents growing up with juvenile idiopathic arthritis or chronic fatigue syndrome. The present chapter provides a description of the background of the dissertation and an outline of the studies that it contains.

Juvenile Idiopathic Arthritis

Juvenile Idiopathic Arthritis (JIA) is the most common chronic rheumatic disease in childhood. *Juvenile* in this context refers to an onset before the age of 16 years, *idiopathic* refers to a condition with no defined cause, and *arthritis* refers to inflammation of one or more joints. Results of various studies indicate that JIA affects 7 to 401 of every 100,000 children.[5,6] Reasons for differences in estimates are among others diagnostic difficulties, and definition of study population. Of these children, following the diagnostic criteria of the EULAR,[7] approximately 50% have oligoarticular arthritis (4 or fewer joints are affected in the first 6 months of disease), 40% have polyarticular arthritis (5 or more joints are affected in the first 6 months of disease) and 10% have systemic JIA (the most severe form of arthritis with fever, rash and inflammation of organs). The most common symptoms of all types of juvenile arthritis are persistent joint swelling with pain and stiffness that is typically worse in the morning, and prolonged fatigue. There are periods in which the symptoms of JIA improve, or even disappear (remissions), and times when symptoms flare up. JIA expresses itself differently in every individual and the prognosis of the disease is often uncertain. Patients with JIA generally need treatment by a multi-disciplinary team of professionals. The aims of treatment are to reduce pain and disease activity, to maintain the range of movement of the affected joints, to promote muscle strength, to stabilize affected joints, and to promote independence and overall well-being of the patient.

Psychosocial functioning of adolescents with JIA

Although it is commonly assumed that chronic illness can influence adolescent development and social interactions due to repeated hospitalizations, poor health status or changes in appearances,[8] research findings have been inconsistent. For adolescents with juvenile idiopathic arthritis (JIA) many cross-sectional studies found no significantly elevated risk for psychosocial difficulties compared to their healthy peers.[9,10] At the same time, a number of investigations identified difficulties in some specific domains of adolescent functioning, reporting raised levels of depression,[11] frustration,[12] anxiety, withdrawal, and other internalising problems.[13] Furthermore, young adults with arthritis report that JIA adversely affected employment,[14,15] body image,[16] social interactions,[17] mood,[11] and overall quality of life.[18,19]

Chronic Fatigue Syndrome

Chronic fatigue syndrome (CFS) is a syndrome of unknown origin. It is characterized by a disabling, persistent (>6 months) or relapsing, severe, unexplained fatigue that is not the result of ongoing exertion and cannot be alleviated by rest. This fatigue is commonly accompanied by other symptoms such as myalgia, headache, sleep disturbance, swollen lymph nodes and a difficulty to concentrate.[20] In adolescence, CFS often has a lengthy course that can lead to much absence from school and long-term detrimental effects on academic and social development.[21-24] Results of various studies indicate that CFS affects 111 to 1290 of every 100,000 adolescents.[25,26] Interventions for CFS attempt to reduce levels of fatigue, to increase activity levels, and to improve quality of life. At the moment, cognitive behavioral therapy (CBT) and graded exercise therapy (GET) emerge as the most promising treatments.[27] These approaches generally agree on the fact that illness-specific beliefs and behaviors perpetuate the suffering of these patients and that effective treatment needs to intervene in these perpetuating factors to facilitate improvement.[28]

Psychosocial functioning of adolescents with CFS

Several studies have reported a high prevalence of psychopathology in patients with CFS, predominantly depression, somatization disorder, anxiety and hypochondria.[29,30] Other studies have focused on personality disorders or personality traits,[31] reporting among others higher levels of neuroticism[32] and alexithymia[33] for adolescents with CFS. Frequently reported co-morbid problems for these adolescents are reduced self-esteem, low mood, learning problems, and difficulties with social interaction such as peer group problems.[34-36]

A need to look beyond the symptoms of disease

JIA and CFS share several similarities in physical symptoms, such as the high level of experienced fatigue, pain and chronicity. Functional impairment is a key aspect of both pediatric conditions, thereby affecting most areas of adolescent's lives. But, although physical condition is a well-recognized risk factor for childhood psychopathology,[37] the psychosocial problems and level of impairment seem considerably higher for adolescents with CFS than for adolescents with JIA.[38,39] So, although there are similarities in physical symptoms between the two pediatric conditions, the consequences for adolescent psychosocial functioning seem to differ considerably. A number of well-validated instruments have been developed for the assessment of functioning of adolescents with chronic illness (for example the Child Health Questionnaire (CHQ) or the Pediatric Quality of Life Inventory (PedsQL)). Such quantitative self-report measures are excellent to make research data comparable. But even well validated quantitative instruments cannot fully assess the individual adolescents' considerations, and the emotional impact of the difficulties a patient faces. Therefore, a search beyond the symptoms is needed to be able to understand the impact of CFS and JIA on adolescent life from the patients' own perspective.

Methodology

Exclusively relying on clinical signs and physician assessment of disease outcome is insufficient to understand the emotional impact of a chronic illness. The impact of chronic illness should be accompanied by studies using more qualitative methods to be able to discover new topics or processes that occur.

Personal narratives, emerging in adolescence, are constitutive of identity.[40] Through such life-stories, individuals understand social and physical events in their lives, and make sense of themselves and their experiences.[41] Personal narratives are not neutral descriptions of a succession of events and encounters, but are engaged stories of emotional experiences. The Self-confrontation Method (SCM) is an instrument to assess and alter personal narratives through self-investigation,[42] combining a focus on individuals' cognitive schemata with their underlying affective structures. One of the most important aspects of the SCM is that not only the researcher, but to a large extent the adolescents themselves discover and reveal the emotions and the central theme of their personal life-story.

It is during the adolescent years that the individual should begin to formulate a coherent sense of self that provides a sense of continuity with the past and a direction for the future.[43] During this developmental phase, the cognitive tools necessary for constructing global coherence in a life story and the social-motivational demands to construct a life-story develop.[44] Adolescence is a period of restructuring social relationships, finding a place in society, and making important choices for the future. The creation of a stable and coherent identity, or in the words of narrative psychologists – creating a unified self-story – is therefore seen as a central developmental task. In dialogical self theory, narrative identity is seen as emerging not through the narration of a single voice, but through the simultaneous narrations of a polyphony of voices.[45] Trying to structure and give voice to this polyphony of internal voices therefore seems to be one of the challenges of adolescence. Based on this theory, the Personal Position Repertoire (PPR), was developed as a method to study adolescents' self-positioning in their social contexts.

Purpose of this dissertation

The rationale for this dissertation comes from three questions that arose while working with adolescents with a chronic illness: a) Is it possible to gain insight into the personal narrative of the adolescent growing up with a chronic illness in a structured way that enables group-comparisons and analysis of different patient-populations, b) can this insight aid in counseling to improve the personal physical and psychosocial outcome of adolescents with a chronic illness? and, c) how do adolescents with a chronic illness present themselves in their social contexts?

The purpose of the dissertation is to generate a broader view on psychosocial functioning of adolescents with JIA and CFS by including their narratives, emotional experiences, and self-positioning. True participatory research is recognized to be essential in the adolescent health arena.[46] The SCM and the PPR have not been previously used in (adolescent) patient groups. This dissertation explores the feasibility of using a true adolescent perspective in clinical research. The study was conducted in The Netherlands, in the University Medical Center Utrecht in collaboration with two local high schools in Utrecht.

Outline

Chapter 2. In this chapter the use of the self-confrontation method (SCM) is introduced as a new instrument to explore the emotional experience of an adolescent with a chronic illness. The use of the SCM as a counseling method is illustrated with a case-example of an 18-year old student diagnosed with polyarticular arthritis.

Chapter 3. Based on a procedure developed from dialogical self-theory, the personal position repertoire (PPR) is described in this chapter. In a cross-sectional study, the self-positioning of adolescents with CFS, JIA and healthy adolescents is compared.

Chapter 4. The third chapter consists of a cross-sectional study in which the SCM is used to compare and analyze the emotional experience of adolescents with JIA, CFS and healthy adolescents. Furthermore, the health-related quality of life (HRQoL) of these adolescents is compared. When going beyond the symptoms of the chronic illnesses to the emotional experience, a more diversified story of the adolescents can be given.

Chapter 5. The fourth chapter investigates which adolescents with JIA benefit from psychological guidance with the SCM, and what is the best moment to offer guidance to these adolescents. The sample was split into a group with low and a group with high HRQoL to study the impact of the use of the SCM over a period of time.

Chapter 6. The use and effectiveness of the SCM as a self-management tool for adolescents with CFS is studied in this chapter. A description is given of the transformation of the emotional experience over time, combined with the changes in health of the participating adolescents.

Chapter 7. The final chapter consists of a general discussion in which the findings from the various studies are combined, and the main conclusions that can improve clinical practice are summarized. Finally, a view towards the future is given.

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2

Self-investigation to explore the impact of juvenile arthritis on adolescent life: a case-study

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ABSTRACT

Objective: To gain insight into the personal experience and feelings of an adolescent with a chronic disease.

Methods: We report on the application of the self-confrontation method (SCM), illustrated by a case-example of an adolescent with juvenile idiopathic arthritis.

Results: Although taken at face value she was not impeded by the arthritis, through self-assessment with the SCM this adolescent acknowledged and addressed the emotional struggle to keep the arthritis secret and to constantly test the physical limits of her body. After the process of self-reflection, the adolescent showed a better integration of her arthritis experiences into her life story.

Conclusion: With the SCM the adolescent could explore her own functioning and well-being on a *manifest*, as well as on an *emotional* and *motivational* level.

Practice implications: In future research, by studying the self investigations of a group of adolescents with chronic diseases, common risk factors for the development of a stable identity during adolescence might be identified. In clinical care, the SCM promotes self-knowledge, allowing for an intrinsic motivation to deal with the emotional impact of the disease.

INTRODUCTION

During adolescence, whilst independence and the creation of a stable identity are important developmental tasks, dealing with challenges caused by a chronic disease -such as arthritis- may constitute a risk for psychosocial development,[1,2] especially due to the lack of a definite cure, the pain and the unpredictable course of the disease. As yet, attempts to understand the impact of juvenile idiopathic arthritis (JIA) upon adolescent psychosocial functioning in a general sense seem inconclusive. Some studies demonstrate an increased risk of social and emotional problems,[3,4] while others show their functioning to be comparable to levels found in the general population.[5-7]

Laura, an 18-year old student, has been diagnosed with poly-articular arthritis. The arthritis went into remission at age 10. Control visits with her rheumatologist are on a yearly basis. Laura lives with both parents, she is finishing a secretarial course and she works in a shop. To study the value of the self-confrontation method in chronic illness, Laura volunteered to reflect on the role and the impact of arthritis in her life. Her case will be presented as an example of the use of the self-confrontation method to systematically study the self narrative of an adolescent with JIA, thereby expecting to a) gain insight in her psychosocial functioning and b) evaluate the process-promoting effect of her self-assessment.

METHODS

The self-confrontation method (SCM)

The SCM is developed to study individual experiences and their ordering into a meaning system through self-investigation, with special attention to affective organization.[8-10] Although the theoretical background and practical use of the SCM have been well evaluated, the use of the SCM with adolescents with chronic illness has not been previously described.

Phase 1 (session 1–3): Assessment

In the first two meetings, the therapist presents a set of open questions **Table 2.1**) in order to elicit the individual's life story. The adolescent formulates the most important units of meaning from the past, present and future into written sentences, so called "valuations". A self investigation with adolescents generally consists of 25 to 35 valuations.

After the formulation of valuations, a standard list of 24 affect terms (**Table 2.2**) is presented. Concentrating on a single valuation, the adolescent is asked to indicate on a 6 point scale to

Table 2.1 Questions of the Self-Confrontation Method

The past:
- Was there something in your past that has been of major importance or significance for your life and that still plays an important role today?
- Was there in the past, a person(s), an experience, or a circumstance that greatly influenced your life and still appreciable affects your present existence?

The present:
- Is there in your present life something that is of major importance for or of great influence on your existence?
- Is there in your present life a person(s) or a circumstance that exerts a significant influence on you?

The future:
- Do you foresee something that will be of great importance for or of major influence on your future life?
- Do you feel that a certain person(s) or circumstance will exert a great influence on your future life?
- Is there a future goal or object that you expect to play an important role in your life?

what extent each affect is experienced in connection with this particular valuation. This results in a profile representing the affective connotation of this valuation.

At the third meeting, the valuations are examined in relation with their affect connotations. Studying the *combination* of the different affects helps to make hidden and suppressed meaning aspects explicit which can serve as important material for a dialogue between therapist and adolescent. The 24 affects are subdivided into four scales (**Table 2.2**), consisting of the *sum of the separate affect terms*: *P*ositive (P) and *N*egative (N) affect dimensions are universally found in literature;[11] *S*elf (S) and *O*ther (O) scales are thought to reflect the "the striving for self-enhancement," and "longing for contact and union with the other".[12] The four scales are psychometrically sound and well suited for use in research and practice.[13] Two additional indices – a generalization (G) and an idealization (I) index – are derived. To derive the G index, the adolescent is asked to use the 24 affect terms to evaluate her response to the question "*how do you generally feel these days?*". The extent of generalization of any particular valuation with the system is found by computing the product-moment correlation between the pattern of affect belonging to that valuation with the pattern of affect representing how the adolescent generally feels. The more positive the correlation, the more that particular valuation generalizes within the system. To measure the extent of idealization of any particular valuation, the same procedure is followed in response to the question "*how would you like to feel?*".

Table 2.2 Affect terms used in the self-confrontation method

S	O
Self-confidence	Caring
Strength	Love
Self-esteem	Tenderness
Pride	Intimacy
P	**N**
Joy	Powerlessness
Happiness	Anxiety
Enjoyment	Shame
Trust	Self-alienation
Security	Guilt
Energy	Loneliness
Inner calm	Inferiority
Freedom	Anger

Note: S = affect reflects self-enhancement, O = affect reflects desire for contact with others, P = positive affect, N = negative affect.

The last step of the assessment-phase is to collect all the valuations that have a high product-moment correlation (r>.60) with the valuation that has the highest generalization index in order to search for similarities. The adolescent is asked to concentrate on the shared meaning of these valuations and to formulate a central theme.

Phase 2 (session 4–9): Process-promoting

In this phase the adolescent starts organizing and reorganizing the valuation system in order to develop a workable self-narrative. The central theme from the assessment-phase functions as the guiding principle of this new phase in which the adolescent is encouraged to gradually move from assessment to change.

Phase 3 (session 10–12): Evaluation

After nine weekly consults, an evaluation of the constancies and changes in the adolescent's valuation system takes place in the form of a second self-investigation.

Measures

The following questionnaires are used to objectively assess possible changes in well-being:

Health-related quality of life assessment

The Dutch version of the Child Health Questionnaire-Child Self-report 87 items (CHQ-CF87) was used as a generic self-administered instrument designed to capture physical, emotional, and social components of health status of children.[14]

Functional ability assessment

The Child Health Assessment Questionnaire (CHAQ) yields a disability index score and a discomfort index score. The reliability and validity of this instrument in children with JIA were previously established.[15,16]

Fatigue assessment

The Checklist Individual Strength (CIS-20), a 20-item self-report instrument of fatigue, concentration, motivation and physical activity.[17]

RESULTS

The questionnaires prior to Laura's self-investigation (t1) demonstrate that she does not experience physical limitations due to her arthritis, except occasionally an inability to participate in physical education at school (CHAQ, **Table 2.3**). The results of both the CHQ (**Table 2.4**) and the CIS-20 (**Table 2.5**) demonstrate that Laura also seems to function well; she reports to be happy with the functioning of her family, friendships and she feels self-confident, energetic and healthy.

Table 2.3 Laura's CHAQ-scores

Assessment of health (CHAQ)[a]	t1	t2
Disability index[b]	0.13	0
Evaluation of pain (VAS score; range 0–10 cm)[c]	3.11	5.19
Evaluation of overall well-being (VAS score; 0–10 cm)[c]	4.53	5.28

Note: t1 = before first self-investigation; t2 = after second self-investigation.

[a] Cronbach's alpha reliability coefficient was 0.94.

[b] 33 items (range 0-4), lower disability index indicate better functional ability.

[c] lower VAS score indicates less pain en more optimal experience of well-being.

Table 2.4 Laura's CHQ-scores

Assessment of physical and psychosocial functioning (CHQ-CF87)[a]	t1	t2
Physical functioning scale (9 items; 0–100)[b]	100.00	100.00
Role/ social emotional scale (3 items; 0–100)[b]	100.00	100.00
Role/ social behavioral scale (3 items; 0–100)[b]	100.00	100.00
Role/ social physical scale (3 items; 0–100)[b]	100.00	100.00
Bodily pain scale (2 items; 0–100)[b]	80.00	80.00
Behavior scale (16 items; 0–100)[b]	87.50	92.19
Mental health scale (16 items; 0–100)[b]	70.31	82.81
Self esteem scale (14 items; 0–100)[b]	78.57	73.21
General health perceptions scale (13 items; 0–100)[b]	59.17	66.67
Family activities scale (6 items; 0–100)[b]	100.00	100.00
Family cohesion scale (1 item; 0–100)[b]	85.00	85.00

Note: t1 = before first self-investigation; t2 = after second self-investigation.
[a] Cronbach's alpha reliability coefficients for the subscales were 0.69–0.92.
[b] Items scored on a Likert scale; higher scores indicate better physical or psychosocial well being.

Table 2.5 Laura's CIS-scores

Assessment of fatigue (CIS-20)[b]	t1	t2
CIS total (20 items; 20–140)[a]	50	41
Score subjective fatigue subscale (8 items; 8–56)[a]	12	16
Score concentration subscale (5 items; 5–35)[a]	29	16
Score motivation subscale (4 items; 4–28)[a]	5	5
Score physical activity subscale (3 items, 3–21)[a]	4	4

Note: t1 = before first self-investigation; t2 = after second self-investigation.
[a] Items scored on a Likert scale (score 1–7); a high score indicates a high level of subjective fatigue and concentration problems and a low level of motivation and physical activity.
[b] Cronbach's alpha reliability coefficient for the entire CIS-20 was 0.90, for the subscales 0.83–0.92.

Laura in phase 1: Assessment

Together with the therapist, Laura formulates 33 valuations for her past, present and future. At home, she relates all these valuations to the 24 affect terms. At the third meeting, Laura investigates her valuations in combination with their affect connotations. The following valuation has the highest generalization index (G): "*I notice I have less trust in others (for example*

Table 2.6 Valuations with the highest correlations with valuation 1 and their scores on S, O, P and N indexes in the first self-investigation

Valuations	S[a]	O[a]	P[a]	N[a]	r
1. I notice that I have less trust in others (for example when Bob says something nice to me).	1	2	0	14	1.00
2. I argue a lot with my mother because she thinks I have no consideration for her. She often tells me it's better if I would leave home.	0	3	0	10	.75
3. I became afraid of the blood withdrawal because it often went wrong. I remember hiding under my bed.	0	13	0	20	.72
4. I was hospitalized for 3 or 4 weeks, I had daily medical examinations.	0	13	0	20	.70
5. It was a difficult time; me and John broke up after 14 months. His family situation was too complicated.	3	9	4	17	.66
6. When I was 5, I dropped myself from the stairs because walking was too painful.	2	15	1	19	.63

Note: S = sum of affect scores reflecting self-enhancement, O = sum of affect scores reflecting desire for contact with others, P = sum of positive affect scores, N = sum of negative affect scores, r = product-moment correlation with valuation 1.

[a] Cronbach's alpha reliability coefficients for the subscales were 0.78–0.94.

when Bob says something nice to me)". **Table 2.6** shows the valuations that have the highest product-moment correlation (r>.60) with this valuation. These valuations have a *different* content, but show a *similar* affective profile. Laura is asked to concentrate on the similarity between the valuations in Table 2.6.

Thereafter, Laura summarizes the central theme she discovers in these valuations: "*I feel anxious and powerless when I'm not in control of a situation. Therefore I find it hard to fully trust other people, I'm afraid to get hurt*". Psychosocial counseling now focuses on this specific issue that Laura *herself* finds most important.

Laura in phase 2: Process-promoting

The central theme functions as the guiding principle to encourage Laura to gradually move from assessment to change. Laura starts observing the current situation by keeping a diary, focusing on situations in which she *does* and *does not* feel in control. Her arthritis soon becomes a central topic. Since her arthritis is in remission, Laura never tells people she meets of her disease. She is afraid people will treat her differently if they know about her arthritis. In her opinion this would result in a situation in which she feels little control (i.e. '*what would people think and say about me if they knew…?*'). In her diary she describes several situations in which this becomes clear to her; i.e. constantly testing her body by working 60 hours a week to feel in control of her arthritis.

Laura is encouraged by her therapist to initiate new ways of behavior. Laura feels strong enough to tell her closest friends about her disease, her hospital experiences from the past and her fears for a relapse of disease activity in the future. By experiencing positive reactions, Laura starts to feel more at ease with her newly required vulnerability.

By acknowledging the arthritis and by tolerating it in her life, Laura starts to be more realistic about living with arthritis. For example, Laura now stops working when she's in pain and she doesn't feel the urge to constantly test her body.

Laura in phase 3: Evaluation

In the last sessions, Laura reformulates her valuations. Some valuations have changed, others are no longer important to her and Laura formulates some new valuations. The most striking differences (compared to her first self-investigation in content of the valuation *or* its affect connotation) are reported in **Table 2.7**.

After her second self-investigation (t2), Laura completes the questionnaires again. Although no active arthritis has been observed by her rheumatologist, her subjective pain evaluation and overall well-being score a bit worse (t2 in Table 2.3). Laura explains that since she allows the arthritis in her life, she's more conscious of the pain she sometimes experiences. In Table 2.4 and 2.5 no substantial differences on these measures compared to the first results were found.

DISCUSSION AND CONCLUSION

Discussion

The self-confrontation method has been previously studied in diverse fields (e.g. depression,[18] value-crisis,[19] and visual impairment[20]) but not in the field of chronic illness. Much research has been conducted on the impact of JIA on adolescents' psychosocial functioning. Most studies[21] considered only one aspect of psychosocial functioning (e.g. disease duration, intrapersonal variables, life events) which might have resulted in an overly optimistic or pessimistic view.[22]

Table 2.7 Some notable valuations of Laura's first and second self-investigation

Valuations									
Investigation 1	S	O	P	N	Investigation 2	S	O	P	N
1.1 In the past, people in my surroundings knew me as the girl with arthritis.	3	2	4	15	2.1 *(Same valuation as at investigation 1, different affect score)*	7	7	14	7
1.2 For eight months I worked in a busy restaurant with an outside pavement, it was exhausting but I knew I could do it. I wanted to extend my limits and I did!	17	5	24	2	2.2 I now know that I can trust my body and that I also have to rest sometimes. I will now listen to my body when I'm tired or in pain. *(originated from valuation 1.2 and 1.3)*	14	12	30	5
1.3 I'm not going to listen to my body anymore; my body is now going to listen to me!	15	0	25	4	2.3 I feel confident about the fact that in the future there will be new medications for my pain. *(new valuation)*	11	7	27	6
---					2.4 Since my first self- investigation I have told several persons that I have arthritis. *(new valuation)*	15	8	26	3
---					2.5 I showed my valuations to my best friend and I've talked with mum about her memories of the time I was really ill. *(new valuation)*	12	10	30	1
My arthritis…[a]	3	17	5	26	My arthritis…[a]	15	13	29	11
My general feeling[a]	13	12	31	8	My general feeling[a]	13	11	30	3
My ideal feeling[a]	18	13	38	0	My ideal feeling[a]	19	16	38	0

Note: S = sum of affect scores reflecting self-enhancement, O = sum of affect scores reflecting desire for contact with others, P = sum of positive affect scores, N = sum of negative affect scores.

[a] Standard valuations offered by the therapist in order to form a general image of Laura's illness experience, her feelings in general and her ideal feelings.

In addition, most studies[23] used standardized questionnaires. A major weakness of this quantitative approach is that it decontextualizes human behavior in a way that it removes the event from its real world setting and ignores effects of variables that have not been included in the model. As psychosocial functioning is based upon subjective experiences, globally objectifying methods alone, such as questionnaires, can not provide the complete approach to the research of psychosocial functioning.[24]

The self-confrontation method was based upon Laura's own life story, which is less abstracting than standardized questionnaires since the personal context of her experiences was added.[25,26] Studying the patient's life story offers an opportunity to allay patients' fears and concerns and to support adaptation to chronic illness.[27,28]

Some side-notes of the SCM should be discussed. Other factors, like repeated contacts with the therapist and the expertise of the therapist to encourage the adolescent to engage in introspection, may have contributed to the favorable changes. Furthermore, the method requires a basic capacity for self-reflection, which is related to a certain developmental age and cognitive level of functioning.

Conclusion

Laura's case was presented as an example of the use of the self-confrontation method to systematically study the life story of an adolescent with JIA. The most important conclusions are that (a) in the SCM a more complete picture of Laura's current situation was obtained since her whole life was taken into account, not only in its temporality, but also its social and cultural context. Furthermore, (b) the SCM allowed Laura and the therapist to assess a deeper, affective level, behind the manifest level of the self-narrative. And finally, (c) equipped with this insight, Laura could gradually become receptive to other feelings and experiences which resulted in a reorganization of her self-narrative.

Practice implications

Because of the combination of qualitative and quantitative data, the SCM can be used for research purposes on both an individual and a group level.[18] Future research and patient care will benefit from more routinely including the adolescent's own perspective to yield a more complete picture of the psychosocial adjustment of adolescents to JIA. This would be valuable from several perspectives. Not only from the perspective of the adolescent, but also from the perspective of the clinician: the more insight the adolescent has in personal feelings and experiences, the better a treatment protocol can be adapted to personal expectations and needs. And, on a larger scale, by studying the self investigations of a group of adolescents with JIA, common risk factors for the development of a stable identity during adolescence might be identified, which can offer therapeutic possibilities for clinical counseling.

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The authors confirm that all patient identifiers have been removed or disguised so the patient described is not identifiable and cannot be identified through the details of the story.

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3

Health and identity: self-positioning in adolescent chronic fatigue syndrome & juvenile idiopathic arthritis

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ABSTRACT

The aim of this study is to gain more insight in basic aspects of identity, in relation to adolescent chronic fatigue syndrome (CFS) and juvenile idiopathic arthritis (JIA). In dialogical self theory, identity is regarded as incorporating multiple self-positions, such as 'I as tired', 'I as pessimistic', or 'I as decisive'. Physical and psychosocial impairment might alter the organization of these self-positions. The Personal Position Repertoire procedure, a quantitative method to analyze the prominence of self-positions, the Child Health Questionnaire, assessing health-related functioning, and the Checklist Individual Strength, measuring fatigue were completed by 42 adolescents with CFS, 37 adolescents with JIA and 23 healthy teenagers. Adolescents with JIA report impaired physical functioning and general health. However, they position themselves very similar to healthy teenagers – i.e. as strong and healthy. While this self-positioning approach might be adequate and sustainable in adolescence, it could prove too strenuous to maintain throughout adult life. Adolescents with CFS, besides indicating severe physical difficulties, also report more psychosocial problems. They position themselves as significantly less strong and more unwell. With this emphasis on positions relating to their illness, there seems to be little room left for stronger positions. It is regarded of clinical importance to address these issues in this crucial developmental period.

INTRODUCTION

A major challenge of adolescence is to attain a sense of self that is coherent, unique, and continuous, yet adaptable, over time.[1] In the transition from childhood to adult life, identity starts to mature and a new balance between autonomy and communion is sought. During adolescence, an exploration of different sides of the self is common, socially accepted and probably even necessary.[2] Prolonged sickness might substantially hinder this development.[3] Although this aspect of adolescent mental health is of clinical importance, as a well-balanced identity can support successful coping and adjustment to long-lasting illness,[4] it is not much studied in clinical settings.

This article focuses on adolescents with (a) juvenile idiopathic arthritis (JIA), and (b) adolescents with chronic fatigue syndrome (CFS). JIA is a rheumatic disease mainly characterized by unpredictable and repeated episodes of joint inflammation and pain, sometimes resulting in a permanent loss of function. In this study patients with three different types of JIA were included. Oligoarticular JIA is the most common form of JIA, and involves arthritis in four or fewer joints. Polyarticular Juvenile Idiopathic Arthritis is a less common subset of JIA, which involves arthritis in many (five or more) joints, often with finger involvement. Systemic JIA is the most severe form of arthritis with fever, rash, and inflammation of other organs.[5] In this study, as a result of the more severely affected population visiting our tertiary setting, 76% of the included patients have polyarticular or systemic JIA (Table 3.1).

Chronic fatigue syndrome (CFS) is a medically unexplained condition, mainly characterized by persistent and severe fatigue.[6] In this study patients were diagnosed with CFS following the diagnostic criteria established by the Centers for Disease Control and Prevention.[7] These include the presence of fatigue for more than 6 months associated with a diminution of functional activity and with somatic symptoms, and pain not attributable to a specific diagnosis or disease. Four of the following criteria need to be present: sore throat, impaired memory or cognition, unrefreshing sleep, postexertional fatigue, tender glands, aching stiff muscles, joint pain, and headaches.

Although the conditions of JIA and CFS differ in pathogenesis, they are both long-lasting, and may have an impact on self-identity. In previous research on CFS in relation to self-identity, patients with the syndrome have expressed the feeling of no longer having control over their own lives.[8] They describe a lack of physical and social activities, and a deprivation of the various emotions these activities entail, leading to a feeling that opportunities for true engagement with the world have been cut off.[9] Moreover, patients with CFS also report a prolonged struggle with their doctors' reluctance towards a somatic diagnosis of their symptoms.[10] Therefore, in addition to feeling severely ill, they experience blame, disbelief, and dismissal.[11,12] Furthermore, these patients indicate that certain unwanted sides of the self have gained a dominant role, seemingly impervious to change.[13] In the relatively few studies on the relation between self-identity and arthritis, patients with arthritis have described their search for an autonomous identity, their wish to achieve a meaningful and normal life, and their desire to participate fully in society.[14,15] In addition, they report a loss of social roles, and they express experiences of stigmatization and discrimination, leading to feelings of decreased self-worth.[16,17]

These studies provide valuable insight into self-identity in CFS and arthritis. Nonetheless, at present all research in this field has been qualitative. In addition, most investigations have been conducted without control groups. This makes it difficult to interpret the findings as relative to a specific condition, and to distinguish the outcomes from the general consequences of prolonged illness. Furthermore, very few studies have focused specifically on adolescence as a crucial period in identity formation. Especially in this developmental phase however, identity formation is not only a process that takes place within the individual, but also very much between individuals.[18,19]

Dialogical self theory is a relatively new current in identity research, incorporating quantitative methodology, and sensitive to the contextual dimensions of individuals' identity.[20,21] In recent years, personal life-stories emerging in adolescence, developing over time and emphasizing the integration of different aspects of an individual's subjective experience, have become regarded as a cornerstone of identity.[22,23] While acknowledging the human striving for a single and coherent life story, dialogical self theory at the same time points to the fact that (narrative) identity emerges from a plurality of positions.[24] The dialogical self is understood, not as one single and continuous 'I', but instead as a dynamic multiplicity of fluctuating, semi-autonomous self-positions.[25] Through a variety of internal and external circumstances some self-positions can become more present, than other self-positions. In dialogical self theory, a distinction is made between internal and external positions.[26,27] Internal positions are regarded as those positions that are relevant to the particular ways in which individual people organize their lives (e.g. I as adaptive, I as strong, I as unwell). From the perspective of these internal positions, those people in the social environment that are relevant, are referred to as external positions (e.g. my father, my teacher). At present in clinical research, dialogical self theory and methodology have mainly been used to study and conceptualize problems of identity in adult psychiatric disorder.[28-30] It has not yet been employed in the research into adolescent somatic, and unexplained medical conditions. Therefore, in this study, the self-positioning and health-related functioning of adolescents with JIA and adolescents with CFS are compared to the self-positioning and health-related functioning of healthy adolescents.

METHODS

Participants

The total group consisted of 42 adolescents with CFS, 37 adolescents with JIA, and 23 healthy adolescents (**Table 3.1**). The higher percentage of female participants in this study is in accordance with the general predominance of women in both (adolescent) CFS, and JIA.[31,32] All participating adolescents were asked by their paediatrician at the Department of Paediatrics (*n* = 79), or in two local high-schools (*n* = 23) to volunteer in this study. Study criteria dictated that *a)* each participant was between 14 and 19 years old, *b)* did not have a primary psychiatric or psychological diagnosis, *c)* the patients were diagnosed with CFS,[7] or JIA[33] by a paediatrician, and *d)* the healthy adolescents did not have a medical condition. The study was approved by the ethical committee of the university medical centre, and informed consent was obtained from all participating adolescents and their parents, or caregivers.

Child Health Questionnaire-Child Form (CHQ-CF87)

The CHQ-CF87 is an 87-item generic self-administered instrument measuring physical, emotional, and social components of health status in children and adolescents. It is divided over 10 multi-item scales, and has established reliability and discriminative validity in adolescent populations.[34]

Table 3.1 General characteristics

	CFS (*n* = 42)[1]	JIA (*n* = 37)[2]	Healthy (*n* = 23)	*p*
Age				
Mean in years (SD)	16.4 (1.1)	16.3 (1.5)	16.4 (0.8)	.92
Gender (% girls)	83	84	78	.84
Ethnicity *n* (%)				
Caucasian	42 (100)	33 (89)	20 (88)	
African	-	3 (8)	1 (4)	
Asian	-	-	1 (4)	
Latin-American	-	1 (3)	1 (4)	
Disease duration at survey				
Median in years (IQR)	1.6 (1.0–3.5)	7.5 (3.5–12.4)	-	.00
Type of JIA, *n* (%)				
Systemic[a]	-	3 (8)	-	
Polyarticular[b]	-	25 (68)	-	
Oligoarticular[c]	-	9 (24)	-	

Note. [1] Following diagnostic criteria of the US Centres of Disease Control[7] and [2] the EULAR.[33] [a] Most severe form of arthritis with fever, rash, and inflammation of other organs; [b] Affecting ≥ 5 joints; [c] Affecting < 5 joints.

Checklist Individual Strength (CIS-20)

The CIS-20 is a 20-item self report instrument, containing four subscales measuring the subjective experience of fatigue, concentration, motivation and physical activity. It is a validated assessment measure, commonly used in research into (adolescent) chronic illness.[35]

Personal Position Repertoire procedure (PPR)

The PPR is a method to investigate basic aspects of the relations between internal and external positions.[36] In the PPR, internal and external positions are combined into a matrix, in which the internal positions form the rows, and the external positions form the columns. A standardized list of a variety of internal positions – that are commonly regarded as important in the identity, and everyday experience of (chronically ill) adolescents[37-39] – was used to assess a broad array of positions (e.g. I as unwell, I as energetic, I as anxious).

In addition, each adolescent individually formulates the most important external positions in his or her life (e.g. my mother, my grand-father, my adversary, or my best friend). The external positions are divided into five categories relevant for adolescents:

1. Direct family (i.e. parents and siblings)
2. Indirect family (e.g. grandparents, aunts, or nephews)
3. School (e.g. teachers, or classmates)
4. Friends
5. Adversaries.

The complete list of internal positions is then combined with the teenager's list of external positions, to form the PPR-matrices. In completing such a matrix, individuals are asked to estimate on a 6-point Likert scale (ranging from 0 = not at all, to 5 = in a very strong degree), their answer to the question "*In which degree is this internal position (I as…) prominent in the contact with this external position (My…)?*". This is done for all internal positions, in relation to all external positions.

Design and statistical analysis

The PPR-data were analyzed with SPSS-GLM (general linear model) as a GROUP x DOMAIN "split plot design".[40] GROUP served as a between-subject factor with three categories (JIA, CFS, and Healthy), whereas DOMAIN was included as within-subject factor containing five categories (Direct Family, Indirect Family, School, Friends and Adversaries). With hierarchical cluster analysis and principal components analysis, extracting the maximum number of (orthogonal) components,[41] the internal positions were divided into discrete clusters to enable group comparisons. In order to analyze the data within a split plot design, these internal PPR-clusters were aggregated across five domains for every person. Global main and interaction effects were tested for. Subsequently, the differences in internal positioning, between the three groups, were analyzed. Data were now aggregated for every person, and simple one-way ANOVAs were performed, using post-hoc Bonferroni comparisons. All statistical analysis was performed using SPSS, version 15.0.

RESULTS

General health status, fatigue, physical and psychosocial functioning

On the CIS-20, adolescents with CFS had scores indicating among others more fatigue and less physical activity, than healthy teenagers and adolescents with JIA (**Table 3.2**). On the CHQ-CF87, adolescents with CFS and JIA scored lower than healthy teenagers on physical functioning, bodily pain and general health perception (**Table 3.3**). In addition, adolescents with CFS also scored lower than healthy teenagers and adolescents with JIA on self-esteem, family activities, mental health, general behaviour, and physical role functioning.

Clustering of internal positions

Through cluster analysis, the internal positions were divided into 6 discrete subgroups, in order to enable group comparisons:

Table 3.2 Scores Checklist Individual Strength (CIS-20)

	CFS	JIA	Healthy	*F* (2, 101)
CIS total (20 items; 20–140)	103.4 (15.9)^a	61.6 (25.3)^b	60.5 (18.5)^b	54.5**
Score subjective fatigue (8 items; 8–56)	48.1 (7.0)^a	29.0 (13.2)^b	26.7 (10.8)^b	46.7**
Score concentration (5 items; 5–35)	24.7 (7.6)^a	14.2 (7.7)^b	16.2 (6.1)^b	22.8**
Score motivation (4 items; 4–28)	15.5 (5.2)^a	9.6 (4.7)^b	10.0 (5.0)^b	16.6**
Score physical activity (3 items, 3–21)	15.1 (4.1)^a	8.8 (4.4)^b	7.6 (3.7)^b	34.58**

Note. Items scored on a Likert scale (score 1–7); a high score indicates a high level of subjective fatigue and concentration problems, and a low level of motivation and physical activity. Cronbach's alpha reliability coefficient for the entire CIS-20 was 0.90, for the subscales 0.83–0.92. ** $p < .001$. Means with different superscripts differ significantly at $p < .001$.

Cluster 1, '*strong*' (Cronbach's α = .86) consists of the internal positions: 'I as strong', 'I as healthy', 'I as energetic', 'I as self-confident', 'I as decisive', 'I as active', 'I as optimistic', and 'I as satisfied'.

Cluster 2, '*adaptive*' (Cronbach's α = .63) consists of the internal positions: 'I as adaptive', 'I as perfectionist', and 'I as caring'.

Cluster 3, '*vulnerable*' (Cronbach's α = .75) consists of the internal positions: 'I as vulnerable', 'I as anxious', and 'I as doubtful'.

Cluster 4, '*unwell*' (Cronbach's α = .81) consists of the internal positions: 'I as unwell', 'I as tired', 'I as victim', and 'I as dependent'.

Cluster 5, '*demanding*' (Cronbach's α = .66) consists of the internal positions: 'I as demanding', 'I attention-seeking', and 'I as jealous'.

Cluster 6, '*opposing*' (Cronbach's α = .70) consists of the internal positions: 'I as quarrelsome', 'I as dissatisfied', 'I as pessimistic', 'I as unmotivated', 'I as lazy', and 'I as avoiding'.

Table 3.3 Scores Child Health Questionnaire (CHQ-CF87)

	CFS	JIA	Healthy	*F* (2, 101)
Physical functioning (9 items; 0–100)	68.7 (17.0)^{a}	77.0 (25.4)^{a}	96.4 (4.7)^{b}	16.6**
Role functioning: Emotional (3 items; 0–100)	65.9 (33.2)	80.9 (26.8)	82.9 (18.2)	3.9
Role functioning: Behavioural (3 items; 0–100)	83.9 (26.8)	92.9 (19.2)	89.8 (18.2)	1.6
Role functioning: Physical (3 items; 0–100)	36.8 (23.7)^{a}	81.8 (26.3)^{b}	84.3 (22.0)^{b}	44.4**
Bodily pain (2 items; 0–100)	39.0 (23.7)^{a}	50.6 (25.2)^{a}	69.6 (20.5)^{b}	12.9**
General behaviour(16 items; 0–100)	74.1 (13.4)^{a}	82.1 (10.6)^{b}	80.3 (11.0)	4.8**
Mental health (16 items; 0–100)	56.9 (14.1)^{a}	71.8 (14.5)^{b}	70.7 (9.9)^{b}	14.4**
Self-esteem (14 items; 0–100)	57.4 (12.6)^{a}	68.8 (11.7)^{b}	71.7 (6.6)^{b}	16.0**
General health perceptions (13 items; 0–100)	38.8 (18.9)^{a}	57.0 (16.7)^{b}	76.3 (10.4)^{c}	40.3**
Family activities (6 items; 0–100)	68.8 (19.0)^{a}	85.3 (16.7)^{b}	85.4 (16.9)^{b}	10.8**
Family cohesion (1 item; 0–100)	66.5 (24.9)	69.2 (25.2)	66.9 (19.9)	0.1

Note. Cronbach's alpha reliability coefficients for the subscales were 0.69–0.92; Items scored on a Likert scale; higher scores indicate better physical or psychosocial well being. ** $p < .001$. Means with different superscripts differ significantly at $p < .05$.

Internal positioning in relation to external positions

The mean-scores of the internal positions for the three groups of adolescents in relation to their external positions are shown in **Figure 3.1** for the six different clusters. As can be seen in these six graphs, all three groups show a similar pattern for most of the clusters of internal positions. In other words, for all of the adolescents, whether healthy or ill, the same internal positions are prominent in the contact with the same group of external positions. In general, direct family and friends elicit the broadest range of internal positions. In contrast, indirect family, adversaries and school tend to evoke the least varied amount of internal positions.

Overall internal positioning

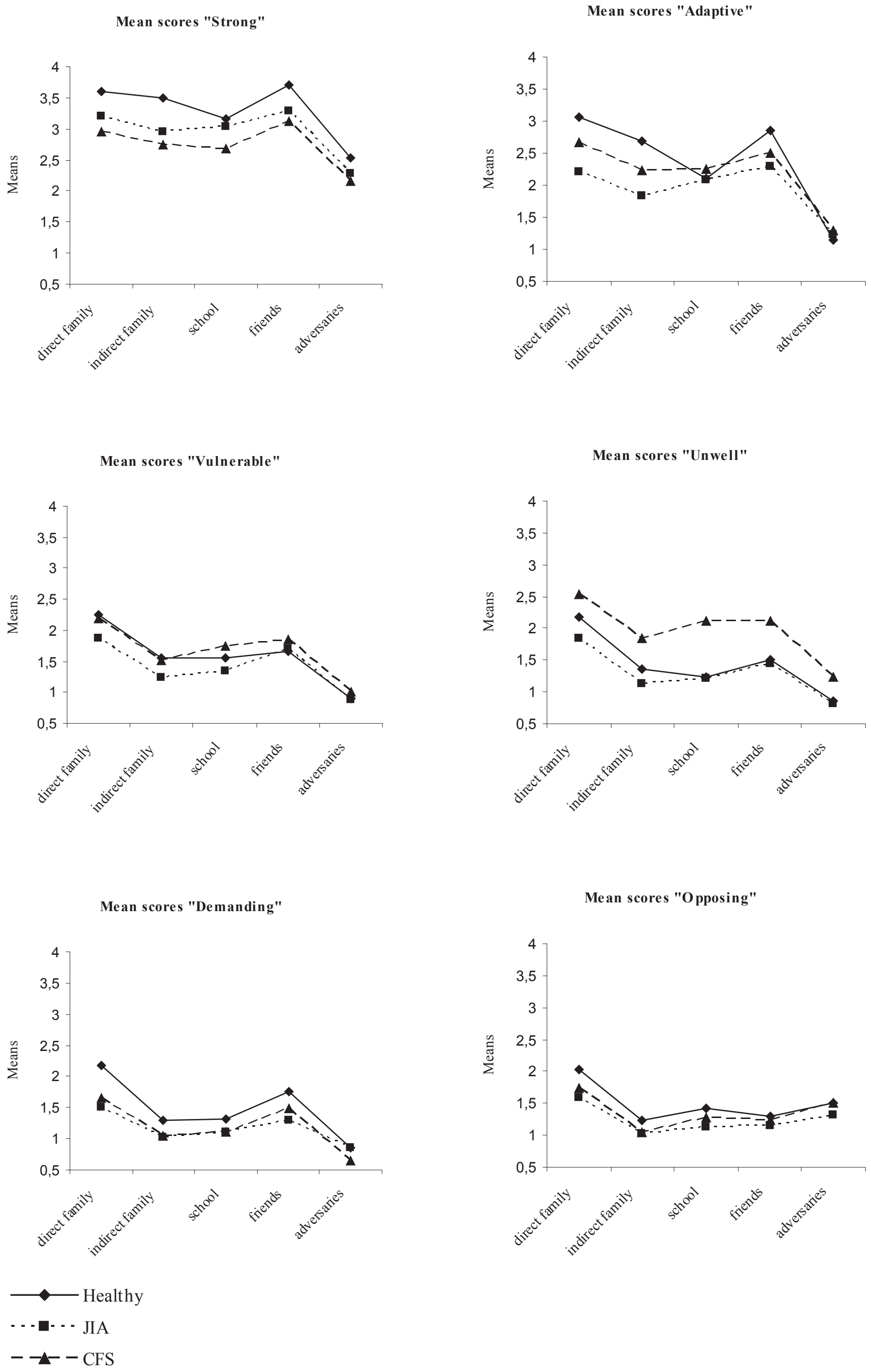
With one-way ANOVAs, significant differences on the mean scores of the six clusters of internal positions were found for the clusters '*adaptive*' (F (2, 101) = 3.56, p = .03), '*strong*' (F (2, 101) = 6.55, p = .002), and '*unwell*' (F (2, 101) = 9.56, p = .00). With Post-hoc Bonferroni tests these three clusters were further explored (**Table 3.4**).

First, the adolescents with JIA were compared with the healthy adolescents. Adolescents with JIA showed their '*adaptive*' internal positions less than their healthy peers (p = .031). Secondly, the adolescents with CFS were compared with the healthy adolescents. On two of the clusters the differences were significant. The adolescents with CFS showed their '*strong*' internal positions less than the healthy adolescents (p = .001), and they showed their '*unwell*' internal positions more (p = .008). Finally, the group of adolescents with JIA was compared with the group of adolescents with CFS. This too, resulted in a marked difference. The adolescents with CFS

Table 3.4 Means (*M*), standard deviations (*SD*) and *F*-values (*F*) for the six clusters of internal positions

Clusters	CFS *M* (*SD*)	JIA *M* (*SD*)	Healthy *M* (*SD*)	*F* (2, 101)
Strong	2.8 (0.8)^a	3.1 (0.8)	3.4 (0.4)^b	6.55**
Adaptive	2.3 (0.8)	2.0 (0.8)^a	2.6 (0.8)^b	3.56*
Vulnerable	1.8 (0.8)	1.5 (0.9)	1.7 (0.5)	0.79
Unwell	2.1 (0.6)^a	1.4 (0.8)^b	1.5 (0.7)^b	9.56**
Demanding	1.3 (0.8)	1.2 (0.8)	1.6 (0.7)	1.74
Opposing	1.4 (0.7)	1.7 (0.8)	1.5 (0.7)	0.90

Note. * p < .05. ** p < .005. Means with different superscripts differ significantly at p < .05.



Mean scores "Strong"

Mean scores "Adaptive"

Mean scores "Vulnerable"

Mean scores "Unwell"

Mean scores "Demanding"

Mean scores "Opposing"

Figure 3.1 Mean scores for the six clusters on five categories of external positions.

showed their ‘*unwell*’ positions more than the adolescents with JIA ($p = .000$). Contrary to the study’s first hypothesis, the adolescents with JIA did not differ from the healthy adolescents on this scale.

DISCUSSION AND CONCLUSION

In this study adolescents’ self-positioning in their social contexts was examined. Obvious similarities were found between the groups, in relation to the five categories of external positions. Although in varying degrees, for adolescents with or without a chronic disease, the same internal positions come forward in the contact with the same categories of external positions. ‘Direct family’ and ‘friends’ are confronted with the broadest array of internal positions. In these contacts, adolescents can freely experiment with different internal positions, resulting in situations in which opposing, as well as strong positions can alternate with unwell and vulnerable positions. With other external positions on the contrary, the relationship is typically characterized by a more selective positioning. Healthy adolescents, not dealing with the challenges caused by a chronic illness, prominently show their strong and adaptive positions. However, they seem to balance their position repertoires, by also tolerating vulnerable, demanding and opposing internal positions. Overall, healthy adolescents show the highest average mean scores for most of the clusters of internal positions.

The self-positioning of adolescents with JIA shows a striking resemblance to that of the healthy adolescents, the mean scores are similar for most clusters. One could conclude that adolescents with JIA have adapted themselves to their illness, in such a way that their self-positioning is not, or no longer, influenced by the consequences of their illness. In this regard, our findings on the identity level of self-positioning, seem to be in line with their scores on the psychosocial functioning scales of the CHQ-CF87, and with previous studies regarding the psychosocial functioning of adolescents with JIA. In general, they appear to cope relatively well with the consequences of their painful and potentially incapacitating illness, and there seem to be few differences between them and healthy teenagers.[42,43] For family, clinicians and researchers however, it is almost impossible to imagine that, for example, the possible weight gain, the sometimes transformed appearance, the joint inflammation and physical limitations in JIA, would not have social and behavioural consequences.[44] In fact, in the research literature on the outcome of JIA, it is pointed out that, in young adulthood, individuals who have (had) arthritis show higher unemployment rates and reduced physical health,[45] more psychosocial impairment,[46] and a lower quality of life.[47] This ‘transitioning problem’ might be (partially) due to a self-positioning approach that, while adequate and sustainable in adolescence, could prove too strenuous to maintain throughout adult life. Due to the particular challenges caused by JIA, such as impaired physical functioning, increased bodily pain and lower general health (see Table 3.3), it would seem reasonable to expect that their '*unwell*' internal positions, focusing on illness and dependence, would have a more prominent place in their self-positioning.

Parents, teachers, friends and physicians, in general encourage adolescents with arthritis to focus on their positive experiences, their skills, and their abilities.[48] From this point of view, it is understandable that adolescents with JIA nourish those internal positions that revolve around such themes in their self-identity. During the onset, or active periods of arthritis (concurring with an increase of inflammation, pain and distress) the inclination to respond to these challenges by focusing on strong and positive internal positions seems adequate to cope with the immediate burdens of medical treatment, and to overcome these difficult periods.[49] However, in times of disease remission or low arthritis activity, adolescents with JIA seem to hold on to such a self-positioning approach. Instead of also tolerating more negative subjects in their life story (that understandably go along with the consequences of growing up with a disabling and painful disease such as JIA), some adolescents seem to suppress or deny such themes.[50] As stated by Stanton, Revenson and Tennen, "*unbalanced attention to positive adjustment to chronic illness can also have untoward consequences. The expectation of the unfailingly 'strong' patient permits the ill person little latitude for having a bad day (or a bad year)*" (p. 568).[51] During adolescence, the mainly positive and strong position repertoires of these youngsters can possibly be maintained because of social support in this life-phase; protective parents,[52] an accommodating school with its continuity and safety,[53] encouraging peer relations,[54] and a paediatric hospital in which personal attention is central.[55] In this situation (in which the environment seems to mostly adapt itself to them) adolescents with JIA might have less need to adopt their '*adaptive*' positions. However, transition to adulthood may entail critical challenges for these adolescents, due to inherent changes in their environment; finding a job, living on one's own, committing to new relationships, and the transition to adult medical care, in which a protective approach is less obvious.

For adolescents with CFS, a very different situation seems to be at hand. On the CIS-20 and the CHQ-CF87, they differ from the healthy adolescents in levels of physical functioning, general health and pain. In addition, they score worse than healthy teenagers *and* adolescents with JIA on mental health, fatigue, general behaviour, physical activity and self-esteem. As a possible reflection of this, they show their unwell internal positions more than healthy teenagers, and adolescents with JIA. Furthermore, they also show their strong positions less, than their healthy peers. These results, with regard to identity on the level of self-positioning, are not only in line with these findings on the CIS-20 and the CHQ-CF87, but also with previous comparative research into illness behaviour and psychosocial functioning in adolescents with

CFS and JIA.[56-58] In these studies it was found that, in general, adolescents with CFS report more psychosocial impairment than adolescents with JIA. Severe fatigue (whatever its cause) entails a profound alteration of, and a withdrawal from adolescents' usual experiences.[59] In the prolonged absence of other self-positions to occupy and stories to tell, one position (or group of positions), 'I as unwell' for example, could become increasingly dominant in the adolescents' position repertoires.

Adolescents with CFS feel more unwell than healthy adolescents and adolescents with JIA. This is consistent with their low scores for physical functioning (CHQ-CF87). But another important aspect of CFS is that despite vigorous research into its possible causes, so far no aetiology for (adolescent) CFS has been found, and it remains a rather controversial diagnosis.[60] In a recent study into the attributions by medical personnel for example, physician assistants were presented with a case study of a patient with the symptoms of CFS. When this case was presented under the (somatic sounding) term neuroendocrine-immune dysfunction syndrome, it was rated as a more severe and disabling illness, than when it was presented as CFS.[61] This ambiguity of the status of CFS as a medical condition, and the experienced stigmatization by others,[62] might also have an influence on the way in which the positioning repertoires of the adolescents with CFS are organized. To overcome this perceived stigma, these patients might be inclined to focus on their unwell internal positions, in order to convince their surroundings of the severe and real impediments of their condition.

This theory-based study has been the first to quantitatively investigate adolescent CFS and JIA in relation to identity conceptualized on the level of self-positioning, while making methodologically sound comparisons between three different study groups. It should be noted however, that self-positioning is just one aspect of the dialogical self, and that the important notions of narrative and affect were not directly investigated in the present study. It must also be mentioned that while the participating adolescents completed a standardized version of the PPR, in a clinical setting it is probably more beneficial to have clients freely indicate and chose those internal positions that they recognize, and identify with. Moreover, although there is evidence that psychological difficulties are not related to disease activity,[63] it is imaginable that the quite severe illness symptoms and prolonged illness duration in our patient groups – related to the tertiary, academic hospital setting – could have had an effect on our findings. Future research might also want to focus on less severely affected patients, or patients at an earlier stage of their illness. In the analyses, we did not take into account the disease duration of the patients, nor the subtype of JIA. While we realise that these are important factors that might influence the self-positioning of the adolescents, our sample size was too modest for further differentiation.

The “self” described in this paper, is potentially under constant transformation, and has as one of its most crucial and distinguishing features that it is experienced, enacted, lived and told from a variety of unique and subjective stances. However, due to the cross-sectional design of this study, definite conclusions with regard to the flexibility or rigidity of the self-positioning of the participating adolescents cannot be drawn. Therefore, follow-up studies investigating longitudinal changes in adolescents’ self-positioning are of principal importance.

Conclusion

While most psychological studies into adolescent CFS and JIA have focused on psychosocial dysfunctioning and disorder, this study concentrated primarily on the everyday expression of adolescents’ identity in their social contexts. It was investigated whether adolescents with CFS and JIA would have a self-positioning approach, differing from that of healthy adolescents. There seems to be an indication that chronic illness in adolescence and the self, conceived on the identity level of self-positioning, are related. This seems especially obvious in CFS, although it remains unclear whether the way their self-positions are organized is caused by, or is an intrinsic part of the experience of severe fatigue in the syndrome.

These findings have implications for clinical practice. The organization of self-positions can not only be studied by researchers, it can also be scrutinized by the patients themselves in self-investigation.[30,64] For adolescent patients with JIA and CFS alike, it might be very helpful to investigate their different self-positions, to try to include and develop new positions, to form coalitions between existing positions, and to balance previously dominant positions with counter-positions in the experiences of everyday life. In this way, through the dialogue with their counsellor or therapist, communications between self-positions can be recovered, and new feelings, possibilities and perceptions (both positive and negative) can be opened.

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4

Beyond the symptoms: adolescents’ emotional experiences in chronic fatigue syndrome and juvenile idiopathic arthritis

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ABSTRACT

Objective: To compare the emotional experiences and health-related quality of life (HRQoL) of adolescents with juvenile idiopathic arthritis (JIA), chronic fatigue syndrome (CFS) and healthy controls.

Methods: The emotional experiences of 36 adolescents with juvenile idiopathic arthritis, 42 adolescents with chronic fatigue syndrome, and 25 healthy teenagers were studied with the Self-confrontation Method (SCM). The Self-confrontation Method is used to analyze the emotional level of personal narratives. Health-related quality of life was assessed with the Child Health Questionnaire (CHQ-CF87).

Results: Both adolescent patient groups reported less positive experiences of autonomy and success, compared to healthy adolescents. Furthermore, adolescents with CFS described more negative experiences of powerlessness, isolation and unfulfilled longing. In the Child Health Questionnaire results, both patient groups reported worse physical functioning than healthy peers. Adolescents with chronic fatigue syndrome also reported less mental health, self-esteem, and more role limitations due to physical functioning.

Conclusions: Without focusing on preliminary chosen topics such as disease symptoms, both adolescent patient groups report a lesser degree of positive experiences than their healthy peers. The CFS group reported more negative experiences and worse HRQoL than adolescents with arthritis and their healthy peers did. These findings indicate that therapeutic focus should not only concentrate on negative experiences, but most certainly also on enabling these patients to create stronger feelings of self-enhancement.

INTRODUCTION

There is widespread recognition that chronic illness not only has consequences on a biological, but also on a psychological and a social level.[1] Especially during adolescence, a chronic illness causes additional developmental challenges. Adolescence is a period of personal exploration and change. In the case of chronic illness, factors like chronic pain, severe prolonged fatigue, or uncertainties about improvement, have to be incorporated into the transition process to adulthood.

Research mostly focuses on the impact of chronic illness on biological and behavioral factors, whereas research on the emotional impact is more scarce.[2-4] Emotional problems have usually been considered to reflect negative affective states, and the amelioration of negative emotions has traditionally been the focus of psychotherapy. However, emotional experience might also become problematic when some positive affective states are absent.[3,5-7] Research on quality of life in arthritis, for example, has shown that patients with higher levels of pain and activity limitations reported less adaptive coping, associated with lower positive affect.[8]

One of the reasons for the paucity of studies into this field might be the lack of feasible, reliable, widely applicable and valid methods of eliciting, and measuring emotional experience of adolescents.[9,10] Standard methods, such as questionnaires, mostly focus on specific aspects of development or personality. The personal narratives of patients can provide a broader, integrated perspective. Study of the organization of these narratives might broaden our understanding of the various patterns of emotional responses to chronic illness.[11,12] More research about young people is needed as they tend to fall out of the remit of pediatric research and not fallen into the adult research arena.[13]

In this study, we applied the self-confrontation method (SCM) as an instrument to structurally assess and analyze the narratives of emotional experiences described by the adolescents themselves.[14] True participatory research is recognized to be essential in the adolescent health arena.[13] This study used the SCM to compare the emotional experience of adolescents with chronic fatigue syndrome (CFS) or juvenile idiopathic arthritis (JIA) with the emotional experience of healthy peers. CFS is a condition of unknown etiology, mainly characterized by seriously disabling fatigue.[15] JIA is an often severely impairing, chronic auto-immune disorder, to a large extent characterized by unpredictable episodes of joint inflammation.[16] We selected these two pediatric conditions to be compared since both CFS and JIA share fatigue, and other physical symptoms like pain, and chronicity. However for CFS, to date, no medically explained etiology has been identified. Therefore, JIA and CFS have many similar symptoms but differ in nosology. Health-related quality of life (HRQoL) was assessed to evaluate the reported impact of the disease on quality of life of the adolescents.

The objective of the present study was to examine HRQoL and the emotional experience, without preliminary focus on a specific topic, of adolescents with CFS and JIA. We expected these groups to differ from healthy adolescents, not only in their personal narratives and HRQoL, but also in their emotional experiences. We hypothesized that a) experiences with negative emotions are more prominent in the life stories of adolescents with CFS and JIA than in those of their healthy peers, that b) experiences with positive emotions are less prominent in the life stories of adolescents with CFS and JIA than in those of their healthy peers, and c) the patient groups will report lower HRQoL than the healthy controls.

METHODS

Participants

Participants were adolescents with JIA (*n* = 36), adolescents with CFS (*n* = 42), and healthy adolescents (*n* = 25). Their general characteristics are presented in **Table 4.1**. Study criteria dictated that a) each participant was between 14 and 19 years old, b) did not have a primary psychiatric or psychological diagnosis, c) the patients were diagnosed with CFS[19] or JIA[20] by a specialized pediatrician, and d) the healthy adolescents did not have a medical condition.

Table 4.1 General characteristics

	Healthy (*n* = 25)	JIA (*n* = 36)	CFS (*n* = 42)
Age			
Mean in years (SD)	16.5 (0.8)	16.3 (1.5)	16.4 (1.1)
Gender (% girls)[a]	80	83	83
Ethnicity *n* (%)			
Caucasian	20 (80)	32 (89)	42 (100)
African	1 (4)	3 (8)	-
Asian	3 (12)	-	-
Latin-American	1 (4)	1 (3)	-
Disease duration at survey			
Median in years (IQR)	-	7.7 (3.5–12.6)	1.6 (1.0–3.5)
Type of JIA, *n* (%)			
Systemic[b]	-	3 (8)	-
Polyarticular[c]	-	24 (67)	-
Oligoarticular[d]	-	9 (25)	-

Note. [a] The percentage of female participants in this study is in accordance with the general predominance of women in both CFS and JIA.[18,19] [b] Most severe form of arthritis with fever, rash, and inflammation of other organs; [c] Affecting ≥ 5 joints; [d] Affecting < 5 joints.

When inclusion criteria were met, adolescent patients were asked to volunteer in this study by their pediatrician in the University Medical Center ($n = 78$) during an outpatient visit. Healthy adolescents in two local High Schools ($n = 25$) were asked by the researchers to participate voluntarily. The ethical committee of the University Medical Center approved the study. Informed consent was obtained from participating adolescents and – if legally obliged – by their parents.

SCM procedure

The SCM is a well-established, evidence-based psychological assessment measure and clinical tool, developed to study personal narratives through self-investigation – with special attention to affective organization. Its theoretical background,[21] clinical use,[22-24] psychometric properties, validity and reliability,[25] and its use to compare groups,[26] have all been extensively described.

To elicit the personal narratives, the researcher presents three open questions, referring to the past, present and future (e.g. *'Was there something in your past that has been of major significance for your life and that still plays an important part today?'*). Subsequently, the adolescent formulates the most important experiences into written sentences. A self-investigation with adolescents generally consists of 20 to 35 sentences. The underlying narrative theme and emotions are implicit and can be more difficult to reveal. Therefore, after the formulation of sentences, a standard list of 24 affect terms is presented, subdivided into four psychometrically sound scales that are universally found in psychological literature:[27]

1. Positive (P): *joy, happiness, enjoyment, trust, security, and inner calm* (Cronbach's $\alpha = .93$).
2. Negative (N): *powerlessness, anxiety, shame, self-alienation, guilt, loneliness, inferiority, and anger* (Cronbach's $\alpha = .90$).
3. Self (S), reflecting 'the striving for self-enhancement': *self-confidence, strength, self-esteem, pride, energy, and freedom* (Cronbach's $\alpha = .91$).
4. Other (O), reflecting 'the longing for contact and union with the other': *caring, love, tenderness, and intimacy* (Cronbach's $\alpha = .86$).

For each particular sentence, the adolescent is asked to indicate on a 6 point Likert scale (0 = not at all, and 5 = very much) to what extent each affect is experienced. In **Table 4.2** as an example, ssome sentences formulated by patients are given, together with their affect scores.

Table 4.2 Examples of sentences formulated by 6 patients and their scores on S, O, P and N indexes

	S^a	O^a	P^a	N^a
Sentences of adolescents with CFS				
CFS 1:				
My headaches in elementary school were partly caused by problems I had in my class.	0.8	0.8	0.6	1.4
My brother had an appendicitis operation with complications and shortly after that he pulled a pot of boiling oil over himself.	0.8	2.0	0.8	1.9
I was surprised that I got a good grade for my language test.	4.0	0	2.0	0.3
CFS 2:				
I often feel afraid and I have the feeling that people can't help me with that. This feels like a part of me that I can't change.	1.3	1.0	0.5	3.5
My boyfriend has a large influence on my life. We fight and quarrel all the time.	2.3	3.8	1.4	3.1
My dream is to have a job and earn a lot of money, have a nice house with my partner and to do lots of fun stuff. I want kids and pets.	4.8	4.3	4.6	0
CFS 3:				
To my father, my brother was holy and I always got the blame for everything.	0	0	0	3.3
4 years ago my fatigue, pain and concentration problems started. From that time on everything that happened to me is a blur.	0	0	0	3.3
Three months ago I started riding a horse again.	4.0	4.0	4.3	0.3
Sentences of adolescents with JIA				
JIA 1:				
When I think of my past, I remember that I always had to be aware of my arthritis. When other kids did a physical activity, I could not participate.	0.8	0.8	0.1	1.6
When I'm 18, I will visit Colombia with my twin brother.	1.8	1.0	2.4	0.1
I am an example for many of my peers, both for good as well as bad things. They believe me and listen to me.	3.0	.75	2.9	0
JIA 2:				
I would rather not think about my future and arthritis.	1.5	0	0.4	1.5
I get along very well with my brother and we talk a lot.	3.3	1.3	3.8	0.1
In three years I see myself in a nursing education, because working with mentally disabled people seems impressive and important to me.	4.5	3.3	4.6	0
JIA 3:				
Especially if I cannot do certain things due to my arthritis, I really want to do them.	1.5	0	0.9	1.3
In the last 4 years I have gotten a very close group of friends with whom I do everything together.	4.5	4.3	4.8	0.1
During the holidays I had a fantastic time with all my friends.	5.0	4.3	4.6	0.1

Note: S = mean score of affect reflecting self-enhancement, O = mean score of affect reflecting desire for contact with others, P = mean score of positive affect, N = mean score of negative affect, ^{a} Six-point Likert scale 0–5, Cronbach's alpha reliability coefficients for the subscales were 0.86–0.93.

Finally, in order to get insight into the present, overall situation, the adolescent is asked to use the 24 affect terms to give a response to the question "*How do you generally feel these days?*", and "*How would you like to feel?*" In addition, the adolescents are asked to characterize the emotional experience of disease by rating the sentence: "*My CFS…*", "*My JIA…*", or "*When I'm ill…*".

SCM typology

From the combination of sentences with their affective organization, general types of emotional experiences can be discerned. The six most common, psychometrically validated types of experiences[26] are: '*Strength and Union*', '*Aggression and Anger*', '*Autonomy and Success*', '*Unfulfilled longing*', '*Unity and Love*', and '*Powerlessness and Isolation*'.

HRQoL assessment

The Child Health Questionnaire-Child Form (CHQ-CF87) was used to assess the health related quality of life (HRQoL) of patients and healthy adolescents.[28] The CHQ-CF87 is an 87-item generic self-administered instrument, which measures physical, psychological, and social components of health status in children and adolescents, independently of the underlying disease.

Statistical analyses

The data were analysed with one-way ANOVAs, focussing on three 'a priori contrasts': JIA vs. Healthy, CFS vs. Healthy, and CFS vs. JIA. In preparation for these comparisons measures were aggregated from the original SCM matrices ($n = 103$). First, every sentence was classified according to the abovementioned typology, using the affective profile. Subsequently, the percentages of the six types of emotional experiences were aggregated for every person. All statistical analyses were performed using SPSS, version 16.0.

RESULTS

Typology

The 103 adolescents formulated 2424 sentences ($M = 23.5$, $SD = 4.7$) concerning their past, present and future. A comparison was made of the proportion of the different types of emotional experiences within the total set of sentences, for each adolescent. Significant differences were found for the proportion of experiences of '*Autonomy and Success*', '*Unfulfilled longing*', and

'*Powerlessness and Isolation*' (**Figure 4.1**). Healthy adolescents formulated an average of 35.2% of experiences of '*Autonomy and Success*'. This percentage was significantly higher than the proportion of these experiences for adolescents with JIA (27.6%, t (100) = 1.697, $p < .05$), and for adolescents with CFS (24.1%, t (100) = 2.558, $p < .001$). In addition, adolescents with CFS had a higher proportion of experiences of '*Powerlessness and Isolation*' (15.4%) than their healthy peers (9.2%, t (100) = 2.003, $p < .05$); and also than the adolescents with JIA (10.5%, t (100) = 1.761, $p < .05$), who didn't differ from their healthy peers in the further distribution of the types of experiences. Finally, adolescents with CFS had a significantly higher proportion of experiences of '*Unfulfilled longing*' than the healthy adolescents (6.9% respectively 3.7%, t (100) = 1.639, $p = .05$).

Health-related quality of life (HRQoL)

The CHQ was completed by 100 of the 103 participating adolescents (**Table 4.3**). Adolescent patients scored significantly lower than the healthy controls on 'physical functioning', 'bodily pain', and 'general health perceptions'. The remaining scores for the health concepts of adolescents with JIA were comparable to the mean values of the healthy subjects. Adolescents with CFS showed worse values on *all* health concepts compared to both adolescents with JIA and healthy adolescents. These differences in HRQoL were statistically significant for: 'mental health', 'self-esteem', and 'role functioning: physical'.

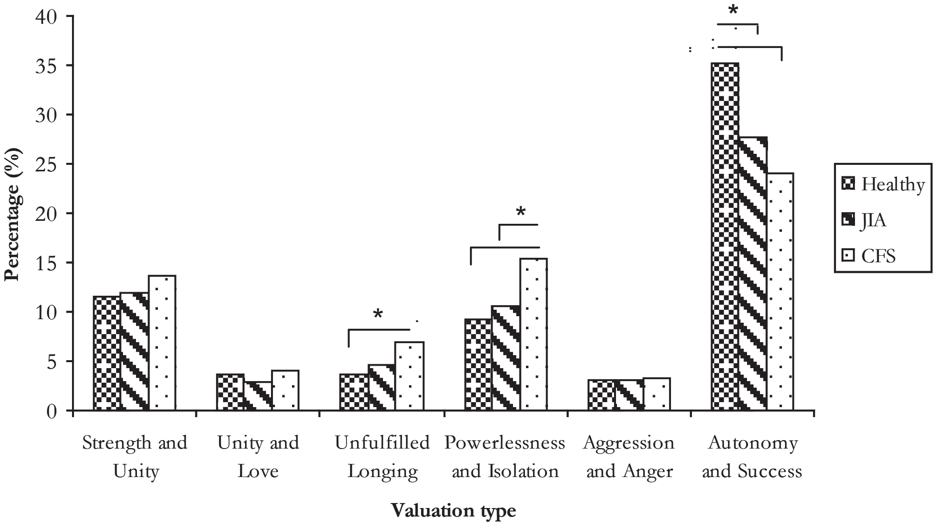

Figure 4.1 Proportion (%) of the six types of experiences within the total system of healthy adolescents, adolescents with JIA and adolescents with CFS. * $p < .05$. Of the total of 2424 sentences, 1415 sentences (58.4%) matched with the criteria of the 6 most common types of sentences.[27]

Table 4.3 Mean scores of health-related quality of life (CHQ) for adolescents with CFS, JIA and healthy controls

	CFS (*n* = 42)	JIA (*n* = 35)	Healthy (*n* = 23)	*F* (2, 97)
Physical functioning (9 items; 0–100)	68.7 (17.0)^a	76.5 (25.7)^a	96.4 (4.8)^b	16.0**
Role functioning: Emotional (3 items; 0–100)	65.9 (33.2)	80.6 (27.1)	82.1 (18.3)	3.6
Role functioning: Behavioral (3 items; 0–100)	83.9 (26.8)	92.7 (19.4)	89.4 (18.5)	1.5
Role functioning: Physical (3 items; 0–100)	36.8 (23.7)^a	81.3 (26.5)^b	83.6 (22.2)^b	42.2**
Bodily pain (2 items; 0–100)	39.0 (23.7)^a	51.1 (25.6)^a	68.3 (19.9)^b	11.4**
General behavior (16 items; 0–100)	74.1 (13.4)^a	82.5 (10.5)^b	79.5 (10.6)	5.0*
Mental health (16 items; 0–100)	56.9 (14.1)^a	72.6 (14.0)^b	70.4 (10.0)^b	15.4**
Self-esteem (14 items; 0–100)	57.4 (12.6)^a	68.8 (11.9)^b	71.7 (6.8)^b	15.5**
General health perceptions (13 items; 0–100)	38.8 (18.9)^a	57.6 (16.6)^b	76.3 (10.6)^c	39.4**

Note. Items scored on a Likert scale; higher scores indicate better physical or psychosocial well-being. Cronbach's alpha reliability coefficients for the subscales were 0.69–0.92. * $p < .05$. ** $p < .001$. Means with different superscripts differ significantly at $p < .05$.

DISCUSSION

Each personal narrative is unique, in experiences as such, as well as in the emotions attached to these experiences. This makes narratives notoriously difficult to analyze and compare quantitatively. In this study, we compared the emotional experiences, without preliminary focus on a specific topic, of three groups of adolescents. The emotional experiences of all adolescents showed apparent similarities. The most obvious finding of this study is that the personal narratives of adolescents dealing with the challenges involved in CFS or JIA contained a significantly smaller proportion of these experiences of 'autonomy and success'. Integrating these specific experiences into a personal narrative is generally needed to develop an adult identity, and enhanced independence from caregivers, two of the main tasks of adolescence.[29] Chronic illness, especially in adolescence, can limit the possibilities to develop strong feelings of self-enhancement by increased dependence on parents due to physical limitations. Furthermore, parents easily come to aid or support their child with a chronic illness, sometimes thereby limiting their possibilities to discover their own abilities. Chronic illness may cause a more negative body image, and may disrupt peer relationships and school performances due to absenteeism.

Contrary to our expectation, adolescents with JIA did not report significantly more experiences of a negative affect type. However, compared to adolescents with JIA and their healthy peers, adolescents with CFS reported a significantly higher proportion of experiences referring to 'powerlessness and isolation' in their personal narratives. This is often related to situations in which the individual feels that there is no way out.[14] Feelings of numbness, hopelessness and helplessness can be related to this type of experiences. In addition, compared to their healthy peers, adolescents with CFS had significantly more experiences of 'unfulfilled longing'. This type of experience is directed to the adoption or maintenance of a loving orientation toward another person (or object) that is, or seems, unreachable.[14]

In addition, adolescents with CFS reported a greater impact of their condition in their health related quality of life than adolescents with JIA or healthy adolescents. All adolescent patients reported their physical functioning, bodily pain, and general health perception to be worse than their healthy peers. In everyday life however, especially the patients with CFS reported to be limited by physical impairment, with more role limitations in their physical functioning, and significantly worse scores on mental health, and self-esteem. CFS and JIA have quite a different nosological status. Whereas JIA is considered a clearly somatic (although idiopathic) condition, CFS is regarded as a functional somatic syndrome. Patients with CFS often report to feel stigmatized by others who tend to doubt the status of CFS as a medical condition.[30] The unknown etiology of CFS may influence the emotional experience of the adolescents, resulting in more negative experiences and worse HRQoL compared to the adolescents with JIA. The direction of the relation between HRQoL and emotional experience cannot be revealed in this cross-sectional study but adolescents reporting poor HRQoL also report less positive, and more negative experiences.

It should be noted, that the SCM requires the adolescents to have a basic capacity for self-reflection, which is related to developmental stage, and the cognitive, and emotional level of functioning. Furthermore, the quite severe illness symptoms and prolonged illness duration of our patient groups – related to the tertiary, academic hospital setting – could have had an effect on our findings. These factors, along with disease activity, were not specifically taken into account. In this study, there was a considerable variability in duration of illness. Adolescents with JIA were seen later in the course of their illness than adolescents with CFS. It is difficult to predict how this might have influenced our results. Patients with a longer period of illness might experience less emotional distress with time. On the other hand, when these adolescents experience ongoing symptoms, they might exhibit greater levels of emotional distress.

The SCM enabled us to explore the personal experiences of the adolescents without focusing on preliminary chosen topics. From our findings, we suggest that psychological interventions should focus on the integration of *positive*, as well as negative emotions. Direct reflection upon emotional experience should be considered to be an important focus of psychological treatment as it is closely associated with beneficial therapeutic change.[31,32] The most important clinical implication of our study is that, while the presence of negative emotions plays a role in our CFS group, the lesser degree of positive emotions appears to be equally important in the emotional experience of both the JIA group, as well as the CFS group. By studying the organization of the narratives of adolescent patients, our understanding of the various emotional responses to chronic illness is broadened. Adolescents with CFS and JIA report less experiences of autonomy and success than their healthy peers. Such emotional experiences should be a major focus of therapeutic interventions.

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5

Self-investigation in adolescent chronic fatigue syndrome: Narrative changes and health improvement

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ABSTRACT

Objective: A small-scale intervention study into narrative self-investigation in adolescent chronic fatigue syndrome (CFS).

Method: The Self-confrontation Method (SCM) is an instrument to assess and change personal life stories. Forty-two adolescents diagnosed with CFS were included and randomly assigned to either 6 or 12 sessions with the SCM. Twenty-five healthy adolescents were assigned to 6 sessions. Outcome was measured directly after the self-investigation procedure at 4 months. Follow-up measurements were made 10 months later. The Checklist Individual Strength and the Child Health Questionnaire were used to measure changes in fatigue, physical and psychosocial functioning.

Results: Self-investigation resulted in significant changes in participants' narratives. Moreover, after self-investigation there was a significant improvement in fatigue, physical and psychosocial functioning for the adolescents with CFS. The patients who completed 12 sessions improved most. At follow-up, the positive effects were maintained.

Conclusion: Self-investigation enables a move beyond the symptoms of CFS in an individualized, patient centered way. Narrative transformation seems to contribute to improved physical and psychosocial outcome in adolescent CFS.

Practice implications: The SCM allows adolescents to discover (for themselves) factors that might cause or perpetuate their fatigue. The results suggest that self-investigation is a useful instrument in the management of adolescent CFS.

INTRODUCTION

Adolescent chronic fatigue syndrome (CFS) is a heterogeneous, medically unexplained condition.[1,2] The main symptom of adolescent CFS is a disabling fatigue that severely affects patients' lives.[3] Although there is growing support for a positive effect of cognitive behavior therapy (CBT),[4,5] a substantial proportion of adolescents with CFS remain severely fatigued and functionally impaired for extended periods of time.[6,7] Therefore, further research into the management of (adolescent) CFS and the customization of its treatment has been urged.[8,9]

Recently, there has been an increasing interest in self-management interventions, in which patients are actively involved in their own care.[10,11] These approaches aim at shared expertise and responsibility of patient and professional.[12] Such strategies could prove especially relevant for medically unexplained symptoms, as it is generally agreed that in these conditions treatments actively involving the patients are most effective.[13,14] It has therefore been suggested that the therapeutic focus should go beyond the unexplained symptoms and also take the life-stories of the patients into account.[15-17]

Through personal life-stories people try to create coherence in their own history and give their lives meaning and direction.[18] From adolescence onward these narratives are considered to be the building-blocks of identity formation, and help individuals to understand and integrate a variety of positive and negative life-experiences.[19] The forming and transforming of these stories have been shown to bring about improvements in both mental and physical health.[20] Paying systematic attention to the narrative context of adolescent CFS might therefore not only lead to a better understanding of patients' experiences, and provide a more acceptable and effective framework for the management of adolescents' problems based on their own perspectives, but may also entail several health benefits.

The Self-confrontation Method (SCM) is an instrument to assess and change individuals' life-stories, through narrative self-investigation.[21] Personal narratives – far from being neutral descriptions of a succession of events and encounters – are engaged stories of emotionally laden experiences. Therefore, the SCM combines a focus on individuals' verbal reports, with an analysis of the underlying affective and motivational structures. The method's theoretical background,[22,23] its clinical use in (adolescent) somatic and psychiatric conditions,[24,25] its use to compare groups,[26] and its psychometric validity and reliability,[27,28] have all been extensively reported. In this small-scale intervention study, the use of the SCM as a self-management tool in adolescent CFS was investigated. It was hypothesized that adolescents' self-investigation of their personal stories would lead to narrative changes and health improvement.

METHODS

Participants

Forty-two consecutively referred adolescents with CFS, who had previously participated in a cross-sectional study,[29] were included in this study. These adolescents were all assessed for CFS and the diagnosis, in accordance with the CDC-1994 criteria,[30] was either made or confirmed by a pediatrician in the University Medical Center Utrecht, The Netherlands. From a local high-school, twenty-five adolescents without any medical, psychiatric or psychological condition volunteered to participate as a control group. The study was approved by the ethical committee of the University Medical Center, and informed consent was obtained from all participating adolescents and their parents.

Self-investigation procedure

Phase 1, first self-investigation

In order to elicit their life story, adolescents reflect upon three open questions, referring to their past, present and future. In dialogue with a counselor, adolescents formulate their life-stories' most important experiences into written sentences. A self-investigation with an adolescent generally produces 20 to 35 sentences. After the formulation of these sentences, a standard list of 24 affect terms is presented. These affects are subdivided into four scales that are reliable, validated and internally consistent;[27] [1] Positive (P): *joy, happiness, enjoyment, trust, security, and inner calm* (Cronbach's $\alpha = .93$), [2] Negative (N): *powerlessness, anxiety, shame, self-alienation, guilt, loneliness, inferiority, and anger* (Cronbach's $\alpha = .90$), [3] Self (S): *self-confidence, strength, self-esteem, pride, energy, and freedom* (Cronbach's $\alpha = .91$), and [4] Other (O): *caring, love, tenderness, and intimacy* (Cronbach's $\alpha = .86$). At home on their own, the adolescents indicate on a 6 point Likert-scale (0 = not at all, and 5 = very much) to what extent each affect is related to each particular sentence. Furthermore, in order to get insight into their present overall situation, the adolescents are asked to relate the 24 affect terms to the questions "*How do you generally feel these days?*", and "*How would you like to feel?*".

Subsequently, the 20–35 sentences that describe meaningful experiences are examined in relation to these affective connotations. Sentences that appear different on a verbal level may show similarity on an emotional level. By computing product-moment correlations between the affective profiles of every sentence, similarities on this implicit level of the personal narrative can be found. In order to find a theme in teenagers' personal narratives, the standardized sentence representing how the adolescents generally feel is used as pivotal sentence. Concentrating on the resemblances between the sentences with a positive correlation ($r > .60$) with the sentence concerning their general feeling, adolescents are asked to summarize the central theme in their personal narratives. In this way, the underlying thematic organization of the life-story is elicited. (In Table 5.1, an abbreviated example of this process is presented for a 16 year old girl with CFS.) The first phase is completed in 3 individual sessions of approximately one hour.

Phase 2, process-promoting

The central theme emerging from the first self-investigation serves as the guiding principle in this phase. Guided by the counselor, the adolescents start assessing in which daily situations their theme is present or absent, and how they cope with these situations. Subsequently, a gradual move from assessment to change is made. The adolescents begin to create and engage novel situations in order to develop and re-organize their personal narratives. This phase consists of 6 individual sessions of approximately one hour.

Table 5.1 Abbreviated case example: Establishing a theme in an adolescent's personal narrative

	S[†]	O[†]	P[†]	N[†]	*r*[‡]
General feeling at first self-investigation	1.8	1.0	0.4	4.4	1.00
Ideal feeling at first self-investigation	4.5	4.8	5.0	0.4	-0.83
Self-described sentences					
1) I always feel as if people cannot help me with any of my problems. This makes me anxious and nervous.	1.3	1.0	1.0	4.0	0.95
2) My concentration is really bad. I feel dumb asking questions to adults.	2.0	1.8	0.4	3.9	0.88
3) I often dream that a murderer is chasing me. I hide, but he still finds me.	1.0	0.8	0.8	3.9	0.84
4) I am very jealous and I am always afraid my boyfriend will like other girls better.	2.3	3.5	0.9	4.2	0.72
Self-described resemblance between sentences (1-4)					
(1-2) In my head, I am often quite stressed. I am afraid to look stupid.					
(1-3) I'm in a situation where I'm afraid and alone. I feel helpless and cannot do anything.					
(1-4) Jealousy is a part of me. I'm powerless to change that, just like my CFS.					
Self-described theme					
I feel powerless, frightened and alone. I don't know what to do and I do not think people can help me. It's always very busy in my head and I find it difficult to relax. Therefore I don't sleep well and I can't concentrate. It makes me very tired.					

Note: [†] S: mean score of self-directed affect; O: mean score of other-directed affect; P: mean score of positive affect; N: mean score of negative affect. Mean scores range from 0–5. [‡] *r*: product-moment correlation with the sentence regarding general feeling.

Phase 3, second self-investigation

The constancies and changes in the adolescents' personal narratives are evaluated in the form of a second self-investigation. This phase is also completed in 3 individual sessions of approximately one hour.

SCM typology

From the sentences, in combination with their affective organization, six common, psychometrically validated types of experiences, based on the four different affect dimensions (Self, Other, Positive and Negative) can be discerned: '*Strength and Union*', '*Aggression and Anger*', '*Autonomy and Success*', "*Unfulfilled Longing*', '*Unity and Love*', and '*Powerlessness and Isolation*'.[28] With these six types at hand it is possible not only to study individual matrices of sentences and affects, but in addition a comparison of group results on the level of emotional experience can be made.

Design

In order to explore two different modes of self-investigation with the SCM in adolescent CFS, the patients were randomly assigned to either 12 or 6 weekly sessions (see Figure 5.1). In the first condition, patients went through all three phases of the SCM under the guidance of a counselor. In the latter condition, the patients only did a first and second self-investigation (phase 1&3) under the guidance of a counselor. For them, between phase 1 and 3 there was a period of approximately six weeks in which they did not have contact with a counselor. In this condition therefore, the adolescents were expected to take more individual responsibility to work with the insights gained from their first self-investigation. All healthy adolescents did a first and second self-investigation (phase 1&3) under the guidance of a counselor. For them as well, between phase 1 and 3 there was a period of approximately six weeks in which they did not have contact with a counselor. Patients agreed not to have other treatment for their CFS during the self-investigation procedure. Questionnaires were used to assess fatigue, physical impairment and psychosocial variables. They were filled in at inclusion, directly after the second self-investigation (at a mean of 4 months), and again at follow-up (at a mean of 10 months after the second self-investigation).

	CFS: 12 sessions	CFS: 6 sessions	Healthy controls
Week 1 *(Phase 1)*	X	X	X
Week 2 *(--)*	X	X	X
Week 3 *(--)*	X	X	X
Week 4 *(Phase 2)*	X	--	--
Week 5 *(--)*	X	--	--
Week 6 *(--)*	X	--	--
Week 7 *(--)*	X	--	--
Week 8 *(--)*	X	--	--
Week 9 *(--)*	X	--	--
Week 10 *(Phase 3)*	X	X	X
Week 11 *(--)*	X	X	X
Week 12 *(--)*	X	X	X

Figure 5.1 Design and time course. Phase 1: first self-investigation, Phase 2: process-promoting, Phase 3: second self-investigation. X: 1 session, --: no session. Although the aim was to complete the self-investigation procedure in 12 weeks, the mean duration was 4 months.

Outcome measures

Checklist Individual Strength (CIS-20)

Fatigue was assessed with the subjective fatigue subscale of the CIS-20.[31] This is a reliable, validated assessment measure, with good internal consistency (Cronbach's α of 0.93) that has previously been used in research of adolescent CFS.[4,32]

Child Health Questionnaire (CHQ-CF87)

Functional impairment, psychosocial and health related variables were measured with the CHQ-CF87.[33] This is a reliable, validated assessment measure, with good internal consistency and has also previously been used in research of adolescent CFS.[34] The physical role functioning subscale, measuring limitations in school work and daily activities as a result of physical health (Cronbach's α of 0.86), the emotional role functioning subscale, measuring limitations in school work and daily activities as a result of emotional problems (Cronbach's α of 0.90), the behavioral role functioning subscale, measuring limitations in school work and daily activities as a result of behavioral problems (Cronbach's α of 0.71), the bodily pain subscale, measuring severity and frequency of bodily pain (Cronbach's α of 0.85), the general behavior subscale, measuring the exhibition of aggressive, delinquent and immature behavior (Cronbach's α of 0.79), the mental health subscale, measuring a diversity of positive and negative feelings (Cronbach's α of 0.86), the self-esteem subscale, measuring satisfaction with abilities, looks, family/ peer relations and life overall (Cronbach's α of 0.89), and the general health perception subscale, measuring believes concerning health (Cronbach's α of 0.77) were employed.[35]

Statistical analyses

In all statistical analyses SPSS (version 16.0) was used. On the inclusion and outcome variables, group means and standard deviations were calculated. One-way ANOVAs, using post-hoc Bonferroni comparisons, were employed to investigate significant differences between groups. Changes on outcome variables were determined with paired samples T-tests. Potential risk factors for non-improvement were quantified through linear regression with fatigue and physical impairment as dependent variables. The significance level on all analyses was set at $p < .05$ (two-tailed tests).

RESULTS

Thirty-five adolescents with CFS completed the procedure. At baseline their mean age was 16.5 (SD = 1.2) years, 85.7% was female, they were all Caucasian, and the mean disease duration was 2.4 years (SD = 1.8) years. Sixteen healthy adolescents completed the procedure. At baseline their mean age was 16.3 (SD = 0.7) years, all were female, and 93.8% was Caucasian. There were no significant differences in these two groups with regard to age, ethnicity and gender. Seven adolescents with CFS withdrew from the self-investigation procedure. The primary reason for withdrawal was the belief that the self-investigation procedure would not aid in their recovery. At baseline, there were no significant differences between the adolescents with CFS who completed the procedure and those who did not. Nine healthy adolescents withdrew from the self-investigation procedure. The primary reason for withdrawal was that they found the procedure too time-consuming. The healthy adolescents who did complete the procedure were all female and had a significantly higher behavioral and physical functioning score on the CHQ-CF87 ($p < .05$) than those who did not. Five adolescents with CFS did not return follow-up questionnaires. At baseline and at 4 months, there were no significant differences between those participants who returned the follow-up questionnaires and those who did not.

In Table 5.2, the percentages of the different types of experiences in the adolescents' life-stories for those participants that completed the self-investigation procedure are presented. For the most frequently reported types, variables had a normal to near-normal distribution. Both at the

Table 5.2 Proportion (%) of types of experiences within adolescents' total narrative systems

	Scores, Mean (SD)		Change, Mean (SD)
	0 months	4 months	0–4 months
Strength and union[1]			
CFS: 6 sessions (*n* = 17)	13.0 (13.6)	10.9 (14.5)	-2.0 (13.3)
CFS: 12 sessions (*n* = 18)	13.2 (12.2)	16.2 (18.5)	3.0 (15.7)
Healthy controls (*n* = 16)	8.4 (11.5)	16.8 (19.7)	8.4 (12.8)*
Autonomy and succes[2]			
CFS: 6 sessions	26.3 (18.7)	30.2 (19.6)	3.9 (14.7)
CFS: 12 sessions	24.4 (10.9)	43.8 (17.5)	19.3 (14.4)*
Healthy controls	32.2 (15.4)	32.7 (18.0)	0.5 (17.0)
Unity and love[3]			
CFS: 6 sessions	2.2 (4.0)	2.9 (4.7)	0.7 (3.7)
CFS: 12 sessions	5.4 (8.2)	1.2 (2.6)	-4.2 (6.6)*
Healthy controls	3.3 (4.9)	1.6 (2.5)	-1.6 (4.2)
Powerlessness and isolation[4]			
CFS: 6 sessions	17.0 (15.2)	10.9 (13.2)	-6.1 (7.7)*
CFS: 12 sessions	13.4 (6.8)	6.8 (7.8)	-6.5 (11.4)*
Healthy controls	12.0 (12.2)	10.5 (11.6)	-1.5 (10.2)
Aggression and anger[5]			
CFS: 6 sessions	3.1 (4.9)	2.3 (4.2)	-0.8 (6.1)
CFS: 12 sessions	4.3 (8.4)	3.3 (6.5)	-1.0 (7.9)
Healthy controls	3.3 (6.0)	1.2 (2.2)	-2.0 (5.0)
Unfulfilled longing[6]			
CFS: 6 sessions	5.7 (9.7)	4.0 (8.9)	-1.7 (3.8)
CFS: 12 sessions	9.1 (10.3)	4.1 (5.0)	-5.0 (7.1)*
Healthy controls	3.5 (4.4)	1.6 (3.3)	-1.8 (4.0)

Note. See Van Geel & De Mey[28] for psychometric data regarding types of experiences. [1] High self-directed affect (S), high other-directed affect (O), high positive affect (P), low negative affect (N). [2] High S, low O, high P, low N. [3] Low S, high O, high P, low N. [4] Low S, low O, low P, high N. [5] High S, low O, low P, high N. [6] Low S, high O, low P, high N. * Indicates significant change on the *p* < .05 level.

first and the second self-investigation there were no significant differences in the proportions of the types of experiences between the CFS/ 6-sessions group, the CFS/ 12-sessions group and the healthy controls. However, in the period between the two self-investigations there were several changes within the three groups. At 4 months, both the CFS/ 6-sessions group and the CFS/ 12-sessions group reported significantly fewer experiences of powerlessness and isolation. In addition, the CFS/ 12-sessions group also reported significantly fewer experiences of unfulfilled longing and unity and love, and described significantly more experiences of autonomy and success at the second self-investigation. At 4 months the healthy adolescents reported significantly more experiences of strength and union.

In Table 5.3, the scores on the measures of fatigue, physical and psychosocial functioning are presented. Variables had a normal to near-normal distribution. At baseline, the two CFS groups did not differ from each other, but had significantly worse fatigue, physical functioning, bodily

Table 5.3 Fatigue, physical and psychosocial functioning

	Scores, Mean (SD)			Change, Mean (SD)
	0 months[†]	4 months[†]	14 months[‡]	0–4 months
Subjective fatigue[1]				
CFS: 6 sessions	48.4 (7.5)[a]	36.0 (12.2)[a]	34.7 (12.6)[a]	-12.4 (9.4)[*]
CFS: 12 sessions	47.4 (7.6)[a]	29.0 (12.4)	29.2 (12.0)	-18.4 (12.6)[*]
Healthy controls	26.1 (10.9)[b]	20.7 (10.9)[b]	21.3 (11.2)[b]	-5.4 (7.1)[*]
Role functioning: Physical[2]				
CFS: 6 sessions	34.6 (24.8)[a]	59.5 (29.1)[a]	69.2 (33.4)[a]	24.8 (32.5)[*]
CFS: 12 sessions	36.3 (24.3)[a]	79.3 (25.5)	81.7 (18.4)	43.0 (29.7)[*]
Healthy controls	91.1 (17.4)[b]	96.3 (10.0)[b]	97.0 (8.9)[b]	5.2 (19.2)
Role functioning: Emotional[2]				
CFS: 6 sessions	68.0 (33.5)	81.0 (25.7)	75.2 (25.3)	13.1 (28.9)
CFS: 12 sessions	60.0 (40.5)	85.9 (19.5)	88.2 (14.4)	25.9 (38.4)[*]
Healthy controls	86.7 (17.4)	92.6 (10.0)	87.4 (25.8)	5.9 (15.1)
Role functioning: Behavioral[2]				
CFS: 6 sessions	85.0 (24.2)	94.8 (13.1)	92.3 (12.3)	9.8 (25.4)
CFS: 12 sessions	78.5 (34.7)	94.1 (10.2)	93.5 (13.1)	15.6 (33.3)
Healthy controls	97.0 (6.6)	98.5 (3.9)	92.6 (25.8)	1.5 (7.1)
Bodily pain[2]				
CFS: 6 sessions	41.8 (26.0)[a]	53.5 (21.2)[a]	52.3 (32.2)[a]	11.8 (28.1)
CFS: 12 sessions	35.3 (22.3)[a]	58.0 (22.4)	55.9 (28.7)[a]	22.7 (22.5)[*]
Healthy controls	72.7 (21.2)[b]	76.7 (20.6)[b]	83.3 (15.0)[b]	4.0 (13.5)
Mental health[2]				
CFS: 6 sessions	58.8 (16.1)[a]	65.2 (10.0)	66.3 (20.8)	6.3 (11.8)[*]
CFS: 12 sessions	54.5 (14.5)[a]	67.3 (11.3)	67.4 (13.5)	12.8 (15.0)[*]
Healthy controls	71.7 (10.4)[b]	74.4 (12.5)	72.0 (16.7)	2.7 (6.6)
Self esteem[2]				
CFS: 6 sessions	55.4 (12.0)[a]	61.7 (13.2)[a]	64.8 (19.1)	6.3 (15.0)
CFS: 12 sessions	58.3 (12.6)[a]	72.3 (12.5)	67.8 (12.8)	14.1 (12.0)[*]
Healthy controls	71.1 (8.0)[b]	76.3 (11.7)[b]	71.4 (15.1)	5.2 (8.7)[*]
General health perception[2]				
CFS: 6 sessions	42.9 (15.8)[a]	51.3 (12.8)[a]	53.0 (19.2)[a]	8.4 (12.0) [*]
CFS: 12 sessions	36.5 (18.8)[a]	50.7 (22.0)[a]	49.2 (22.1)[a]	14.2 (20.2)[*]
Healthy controls	77.7 (10.9)[b]	79.7 (13.5)[b]	78.2 (12.6)[b]	2.0 (9.8)

Note. [†] CFS: 6 sessions (n = 17), CFS: 12 sessions (n = 18), Healthy controls (n = 16). [‡] CFS: 6 sessions (n = 13), CFS: 12 sessions (n = 17), Healthy controls (n = 16). There were no significant changes during the follow-up period, so these change-scores are not indicated in the table. [1] Subjective fatigue subscale (CIS-20): score ranges from 8–56; a high score indicates a high level of fatigue. [2] CHQ-87 scores range from 0–100; higher scores indicate better functioning. [a,b] In the columns, different superscripts indicate significant group differences on the $p < .05$ level. * In the rows, indicates significant change on the $p < .05$ level.

pain, mental health, self-esteem and general health perception scores in comparison to the healthy adolescents. At 4 months, both CFS groups showed a significant improvement on the fatigue, physical functioning, mental health and general health perception scales. (For a general illustration of these effects; see Figure 5.2 & 5.3.) In addition, the CFS/ 12-sessions group also showed a significant improvement on the emotional role functioning, bodily pain and self-esteem scales. Furthermore, except on general health perception, after the second self-investigation the CFS/

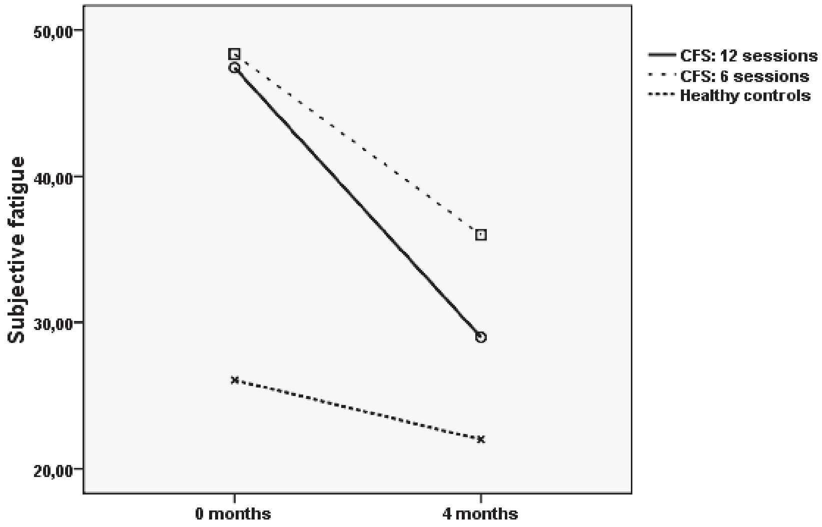

Figure 5.2 Subjective fatigue.

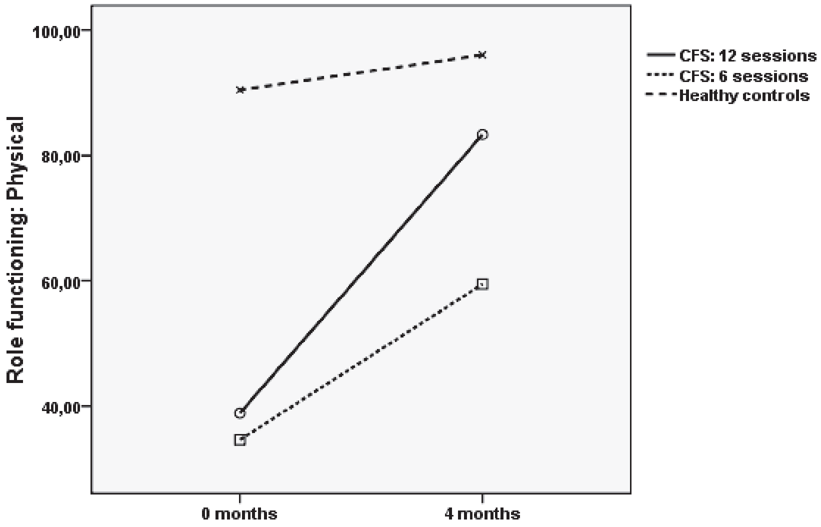

Figure 5.3 Role functioning: Physical.

12-sessions group no longer differed from the healthy controls on any of the scales. In contrast, while the CFS/ 6-sessions group showed several significant improvements, at 4 months they continued to differ from the healthy controls on the fatigue, physical functioning, bodily pain, self-esteem and general health perception scales. After the second self-investigation the healthy adolescents showed a small, but significant improvement on the fatigue and self-esteem scores. At follow-up 10 months after the second self-investigation, there had been no significant changes on any of the scores and the positive effects were maintained.

Linear regression analysis indicated that only a high fatigue score at baseline was a risk factor for a high fatigue score ($t = 3.22$; $p = .01$), and a low physical functioning score ($t = 2.16$; $p = .05$) at 4 months.

DISCUSSION AND CONCLUSION

Discussion

The results seem to indicate that, in adolescent CFS, significant improvement in health and psychosocial variables is accompanied by a mechanism of narrative transformation. In SCM-theory it is generally assumed that positive change is initiated by a reduction of experiences of powerlessness and isolation.[36] This process can be witnessed in both CFS groups. Further improvement is usually in the direction of a predominance of experiences of autonomy and success. In the course of counseling, this development towards more autonomy and activity often gets (temporary) priority over relation-oriented experiences, such as unity and love. This specific kind of process seems to underlie the major improvements in the CFS/ 12-sessions group.

Somewhat to our surprise we did not find (initial) differences in the types of experiences in the life-stories of adolescents with CFS and healthy controls. Furthermore, because of the infrequent reporting of some narrative types (e.g. unity and love & aggression and anger), a normal distribution of these variables was not guaranteed. We believe this to be due to the modest size of the sample described. Moreover, even though Bonferroni corrections for the ANOVA analyses were made, a rather large number of statistical tests was done, increasing the risk of type 1 errors. Finally, as this was a small-scale study, we did not include a waiting list control group. Though patients had been symptomatic for a long time, it cannot be completely ruled out that improvements were partly spontaneous and/ or the result of the attention of a health care professional. The results therefore require reproduction in a larger-scale randomized controlled trial.

This is the first study reporting on the use of narrative self-investigation in adolescent CFS.[37,38] The female to male ratio was in accordance with research findings, the mean age of participants did not have a wide range, and only adolescents diagnosed with CFS according to the CDC-1994 criteria were eligible for the study. Like in previous studies, few risk factors for continued fatigue and physical impairment were identified.[39] The percentage of participants who withdrew from the study is similar to other studies into the management of adolescent CFS with CBT.[4] Both CBT and narrative self-investigation show positive effects in the self-reported health status of adolescents with CFS. The difference between both methods is primarily one of focus. While CBT addresses the maintenance of symptoms through a change of cognitive processes,[40] the SCM intends a move beyond the symptoms by eliciting emotions and establishing a personal narrative theme.

Conclusion

Personal narratives are unique. By incorporating the broader context of patients' affective life-stories into the management of adolescent CFS the therapeutic focus is sharpened. Furthermore, self-investigation offers adolescent patients the opportunity to explore those elements of their problems – which might not exclusively be fatigue or physical impairment – that they themselves find most important and feel they can influence. A continuing dialogue with a counselor during this process seems to stimulate a comfortable transition from assessment to change.

This investigation of their own experience and the changes in their life-stories is reflected in significant improvements in fatigue, physical and psychosocial functioning in both CFS groups. At follow-up, this positive effect was maintained. At 4 months as well as at 14 months, there was a significant difference in improvement between the two CFS groups. Those patients who had 12 sessions in the self-investigation procedure showed considerably more narrative changes and improvement on outcome measures than those who had only 6 sessions. A more intensive counseling procedure is therefore advisable.

Practice implications

The SCM is fundamentally a collaborative process between the adolescents and their counselor, through which the patients are challenged and empowered to be experts of their own life and experiences. The dialogue supporting the process of self-investigation enables the counselor to move beyond the symptoms of CFS, and to address previously unexpressed problems in an individualized, patient centered approach. It allows the adolescents to discover for themselves factors that might cause or perpetuate their fatigue. The results are positive and suggest that enabling adolescent patients with CFS to participate in their own treatment stimulates narrative transformation and health improvement.

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6

Psychological counseling for adolescents with Juvenile Idiopathic Arthritis: for whom and when?

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J Rheumatol, conditionally accepted

ABSTRACT

Objective: To study which adolescents with juvenile idiopathic arthritis (JIA) benefit from psychological guidance, and what is the best moment for it.

Methods: In 3 months, 28 adolescents with JIA and, as a control-group, 14 healthy adolescents received psychological counseling. The adolescents with JIA were split into a group with low Health-related Quality of Life (HRQoL) and high HRQoL. The Child Health Questionnaire, Checklist Individual Strength and Childhood Health Assessment Questionnaire were used to measure fatigue, physical and psychosocial functioning at baseline, and at 3 months and 9 months after baseline.

Results: Adolescents with JIA and low HRQoL at baseline reported less fatigue and better HRQoL after psychological guidance. These changes could not be explained by changes in disease activity. Low HRQoL at baseline was associated with a more recent onset of JIA, higher levels of pain, more severe physical disability, and higher levels of fatigue.

Conclusion: Two-thirds of adolescents with JIA function well before and after psychological guidance. One-third of adolescents with JIA reporting low HRQoL at baseline benefit from guided self-reflections and should be the focus of psychological counseling. The most effective moment for psychological guidance is when the adolescent him- or herself reports difficulties in HRQoL.

INTRODUCTION

Adolescence is a period of personal exploration and change. The increase of autonomous behavior, the development of multiple peer relationships, experimentation with identity, and the development of emotional skills are important tasks of adolescence. Although it is commonly assumed that chronic illness influences adolescent development and social interactions due to repeated hospitalizations, poor health status or changes in appearances,[1] the findings in this field have been inconsistent. Many studies found little evidence for psychosocial deficits in adolescents with JIA compared with normative or healthy control samples.[2,3] At the same time, a number of investigations identified difficulties in some specific domains of adolescent functioning, reporting higher levels of depression,[4] frustration,[5] anxiety, withdrawal, fatigue and sleep disturbances,[6] and other internalizing problems.[7] Furthermore, young adults with arthritis report that JIA adversely affected employment,[8,9] body image,[10] social interactions,[11] mood,[4] and overall quality of life.[12,13] In most pediatric rheumatology settings, youngsters are referred to a mental health professional at some time during their treatment. But which patients benefit most from psychological counseling and what is the best moment for referral?

Most adolescents with JIA don’t need acute psychological aid for a specific problem. Instead the reason for referral to the psychologist is mostly broader e.g. stagnation of certain developmental tasks, coping with chronic disease or non-compliance to long-term regimens. As a psychologist, to address these broad issues, it has been suggested that the therapeutic focus should go beyond the symptoms and take the broader life-stories of the patients into account.[14,15] Personal narratives, emerging in adolescence, are constitutive of identity.[16] Through such life-stories, individuals understand events, and make sense of themselves and their experiences.[17] It remains unclear how adolescents integrate a chronic disease such as JIA into their personal narrative. Therefore, in this study the Self-confrontation Method (SCM) is used as a method for psychological counseling. The SCM is an evidence-based psychological assessment measure[18] and clinical tool. This method guides narrative self-reflections with special attention to the affective organization.[19] The method’s theoretical background,[20,21] its clinical use,[22,23] its psychometric properties, validity and reliability,[24] and its use to compare groups,[25] have all been extensively reported. Furthermore, the use of the SCM with children,[26] and with a group of adolescents with a chronic illness[27] has been described.

To the pediatrician, the management of adolescents in daily clinical practice can be challenging, because the time to evaluate their overall well-being is limited. Adolescents generally do not easily disclose their psychosocial functioning. Therefore, short health-related quality of life questionnaires measuring physical, psychological and social functioning might be used as a general instrument to understand the impact of disease on daily life.[28]

The aim of this study is to discover which adolescents with JIA benefit from psychological counseling. The divergent findings of previous studies on adolescent psychosocial functioning, might imply that the psychosocial development of not all, but just a subgroup of adolescents, is negatively influenced by JIA. This subgroup might be detected by their self-reported HRQoL. We hypothesized that 1) adolescents with JIA and low HRQoL would benefit more from psychological counseling than adolescents with JIA and high HRQoL and healthy adolescents. Furthermore, we hypothesized that 2) adolescents with JIA and high HRQoL have the same psychosocial functioning as their healthy peers. And finally, our hypothesis is that 3) those adolescents with JIA receiving more sessions of psychological counseling would benefit most.

METHOD

Participants and study design

Adolescents with JIA (Table 6.1) were asked by their pediatrician in a University Medical Center to participate in this study. As a control group healthy adolescents were included. They

Table 6.1 General characteristics at baseline

	Healthy (*n* = 14)	JIA low HRQoL[4] (*n* = 9)	JIA high HRQoL[4] (*n* = 19)
Age			
Mean in years (*SD*)	16.4 (0.7)	16.1 (1.7)	16.1 (1.4)
Girls[1] (*n*)	14	8	17
Intensively coached (*n*) (12-sessions)	--	6	8
Ethnicity (*n*)			
Caucasian	13	8	18
African	1	1	-
Other	-	-	1
Disease duration at survey			
Median in years (IQR[2])	-	1.8 (1.0–12.3)	10.5 (5.7–13.6)
Age at diagnosis			
Median in years (IQR)	-	12.0 (3.0–16.5)	6.2 (2.9–10.0)
Type of JIA[3] (*n*)			
Systemic[a]	-	0	2
Polyarticular[b]	-	8	10
Oligoarticular[c]	-	1	7

Note. [1] The percentage of female participants in this study is in accordance with the general predominance of women in JIA[29]; [2] IQR = interquartile range; [3] Following diagnostic criteria of the EULAR[30]; [4] The clusters will be explained in the Results-section; [a] Most severe form of arthritis with fever, rash, and inflammation of other organs; [b] Affecting ≥ 5 joints; [c] Affecting < 5 joints.

were asked by the researcher in two local high-schools to volunteer in this study. Study criteria dictated that each participant was between 14 and 19 years old at inclusion, and the patients were diagnosed with JIA[29] by a pediatric rheumatologist. Excluded from participation were adolescents that had a primary psychiatric or psychological diagnosis, or healthy adolescents with a medical condition. 37 adolescents with JIA and 25 healthy adolescents received an invitation, of which 28 and 14 adolescents respectively volunteered in the study. The ethical committee of the University Medical Center approved the study. Informed consent was obtained from all participating adolescents and – if legally obliged – by their parents. For the study design and time course: see Figure 6.1.

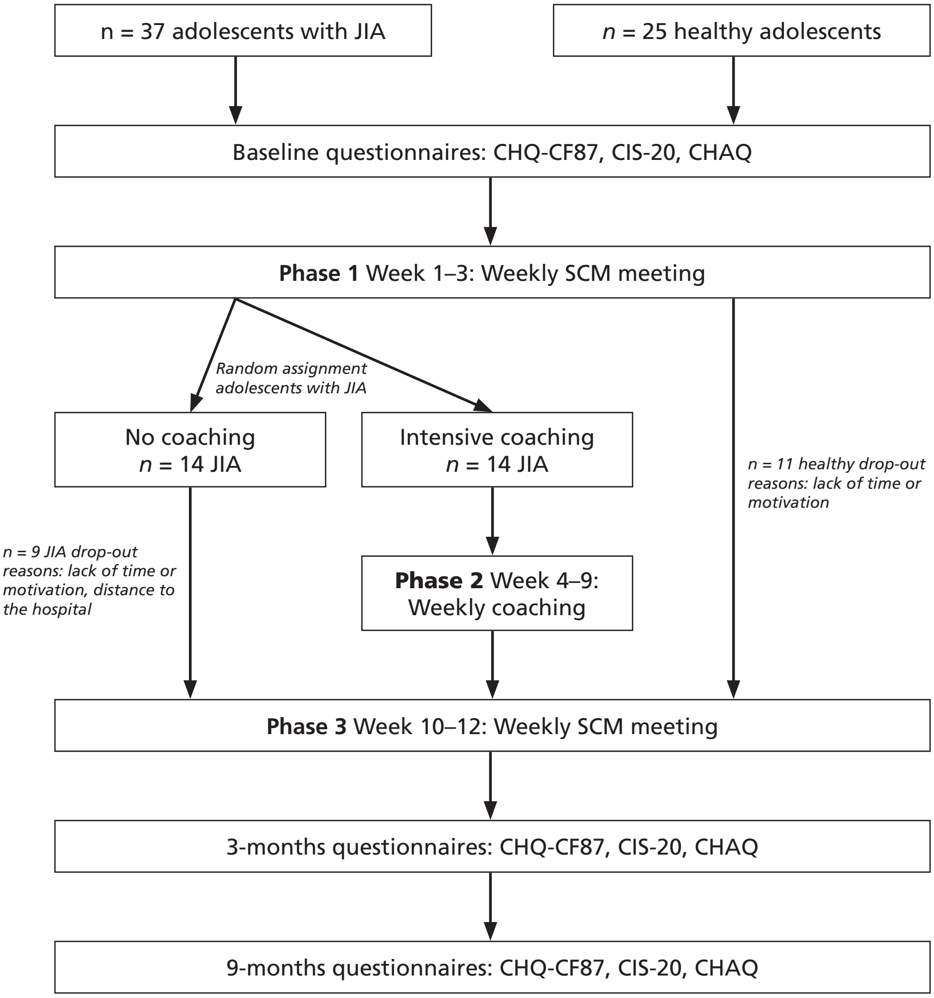

Figure 6.1 Design and time-course.

Psychological counseling

The SCM[27,31] was used as a psychological counseling method. The counselors were specially trained in the use of the SCM.

Phase 1, first self-investigation

In order to elicit personal narratives, three open questions are presented, referring to their past, present and future (e.g. '*Was there something in your past that has been of major significance for your life and that still plays an important part today?*'). In dialogue with the counselor, adolescents formulate their life-stories' most important experiences into written sentences. After the formulation of these experiences into sentences, a standard list of 24 affect terms (such as happiness, anxiety, pride, love…) is presented. For each particular sentence, the adolescent is asked to indicate on a 6 point Likert scale (0 = not at all, and 5 = very much) to what extent each affect is experienced. This affective profile helps the adolescent to find the central theme in their personal narrative. In SCM theory, the process of self-confrontation by itself is seen as having an activating function. As a consequence of the reflection upon the personal narrative, the person is stimulated and has gained direction to gradually change by making new connections between experiences and emotions.[31] A full description of the use of the SCM with an adolescent with JIA is given in a published case-study.[32]

Phase 2, process-promoting

The self-formulated central theme emerging from the first self-investigation serves as a guiding principle in this phase that is only completed by the adolescents randomly assigned to the 12-session group. Adolescents assess daily situations, and the way they cope with these situations. Subsequently, guided by the counselor, the adolescents are encouraged to gradually move from assessment to change. The adolescents are stimulated to create, and engage in novel situations, in order to develop and re-organize their personal narratives. This phase consists of 6 weekly individual sessions of approximately one hour.

Phase 3, second self-investigation

For all adolescents, the consistencies and changes in their personal narratives are evaluated in a second self-investigation (in 3 weekly individual sessions of approximately one hour).

Health related quality of life (HRQoL) assessment

The Child Health Questionnaire-Child Form (CHQ-CF87) was used to assess the HRQoL.[33] The CHQ-CF87 is an 87-item generic self-administered instrument which measures physical, emotional, and social components of health status in children and adolescents, independently of the underlying disease.[34] The following scales were used: the physical role functioning subscale, measuring limitations in school work and daily activities as a result of physical health (Cronbach's alpha of 0.86), the emotional role functioning subscale, measuring limitations in school work and daily activities as a result of emotional problems (Cronbach's alpha of 0.90), the behavioral role functioning subscale, measuring limitations in school work and daily activities as a result of behavioral problems (Cronbach's alpha of 0.71), the bodily pain subscale, measuring severity and frequency of bodily pain (Cronbach's alpha of 0.85), the general behavior subscale, measuring the exhibition of aggressive, delinquent and immature behavior (Cronbach's alpha of 0.79), the mental health subscale, measuring a diversity of positive and negative feelings (Cronbach's alpha of 0.86), the self-esteem subscale, measuring satisfaction with abilities, looks, family/peer relations and life overall (Cronbach's alpha of 0.89), and the general health perception subscale, measuring believes concerning health (Cronbach's alpha of 0.77).

Fatigue assessment

The Checklist Individual Strength (CIS-20) – a 20-item self-report instrument – was used to measure the subjective experience of fatigue. This is a reliable, validated assessment measure, with good internal consistency (Cronbach's alpha of 0.93) commonly used in research into pediatric chronic illness.[35]

Functional ability assessment

The Child Health Assessment Questionnaire (CHAQ) was completed by each adolescent with JIA.[33] The CHAQ measures the patients' ability to perform functions included in eight areas of daily living, which are averaged to calculate the CHAQ disability index. Two horizontal 10-cm visual analogue scales (VAS) are incorporated to assess the adolescent's overall well-being and the intensity of the adolescent's pain. Cronbach's alpha reliability coefficient of the total CHAQ is 0.94. Pediatric rheumatologists checked the disease activity at regular outpatient visits.

Clustering of adolescents with JIA

The hypothesis of this study is that HRQoL may predict which subgroup benefits most from psychological counseling. Therefore, the sample of adolescents with JIA was split into two clusters using hierarchical cluster analysis (Ward's method, Squared Euclidean distances) on the standardized CHQ-CF87-scales at baseline.[36] For ascertaining the stability of the solution two non-hierarchical clusterings were performed: one with centroids from the hierarchical analysis as seeds and one with random seeds. Agreement between the two solutions was obtained by calculating Cohen's kappa, which was 0.94, indicating almost perfect agreement. The sample was split into a) adolescents with JIA and low HRQoL ($n = 10$) and b) adolescents with JIA and high HRQoL ($n = 21$).

Statistical analysis

Descriptive statistics were reported as means and standard deviations for the continuous variables and in terms of absolute frequencies for the categorical variables. To study whether psychological counseling improves psychosocial well-being of the sample, we used repeated measurement analysis with general linear models. In this study, we used the following outcome parameters for psychosocial well-being: HRQoL, fatigue, functional ability, overall well-being and pain. Statistical tests included one-way analysis of variance with post-hoc Bonferroni corrections. To report the strength of differences partial η^2 was calculated, providing a measure of effect size. For partial η^2 the traditional cut-offs of small, medium, and large effects of respectively .01, .06, and .14, are useful, although they might underestimate the effect size.[37] All statistical analysis was performed using SPSS software, version 19 (SPSS, Chicago, IL).

RESULTS

Baseline

Thirty-seven adolescents with JIA and twenty-five healthy adolescents were invited to participate in this study. Nine adolescents with JIA (24.3%) and eleven healthy adolescents (44%) withdrew from the SCM-procedure (Figure 6.1). The primary reason for withdrawal was a lack of time or motivation. Three adolescents with JIA and two healthy adolescents did not return follow-up questionnaires.

Adolescents with JIA and low HRQoL had a higher mean age at onset (t (26) = 2.31, $p < .05$), and shorter disease duration at survey (t (26) = 2.21, $p < .05$) (Table 6.1). In the low HRQoL group, 8 out of 9 adolescents had the more severe polyarticular arthritis, and only 1 out of 9 had milder oligoarticular arthritis. In the high HRQoL group, 12 out of 19 adolescents had polyarticular or systemic arthritis, and 7 out of 19 had oligoarticular arthritis. Most of the adolescents were in a situation of stable disease activity and stable treatment of the JIA.

Adolescents were randomly assigned in phase 2, resulting in 6 out of 9 adolescents with JIA and low HRQoL, and 8 out of 19 adolescents with JIA and high HRQoL being assigned to the intensive coaching trajectory.

Adolescents with JIA and high HRQoL reported almost similar HRQoL as their healthy peers (Table 6.2). The two clusters of adolescents with JIA differed at baseline in their functional ability (Table 6.3), with adolescents with low HRQoL reporting worse scores on: 'CHAQ score' (t (26) = -3.43, $p < .05$), 'VAS pain' (t (20) = -4.64, $p < .001$), and 'VAS overall well-being' (t (20) = -3.92, $p < .05$). Adolescents with JIA and low HRQoL reported worse 'Subjective fatigue' (F (2, 40) = 19.28, $p < .05$, partial $\eta^2 = .50$) than adolescents with JIA and high HRQoL and healthy adolescents (Table 6.2).

Effect of intensive coaching

After analysis of the results at 3 months of the two different groups – intensively coached in phase 2 or not –, no differences were observed in the outcome parameters (functional ability, fatigue nor pain). Therefore, since the total sample size was modest, the separation was left out from further analysis.

After psychological counseling and follow-up

Over time, no significant improvements at $p < .05$ could be observed for both groups in their physical functioning. At follow-up, adolescents with JIA and low HRQoL at baseline still had worse physical functioning, which is reflected in their higher CHAQ score (t (26) = -3.10, $p < .05$) (Table 6.3). The VAS scores of adolescents with low HRQoL at baseline improved. At 3-months follow-up, adolescents with JIA and low HRQoL at baseline did no longer report worse 'VAS-well-being' and 'VAS-pain' scores, compared to the adolescents with high HRQoL at baseline. Clinically the arthritis of the adolescents was stable, but the more subjective well-being and pain improved.

The subjective fatigue (Table 6.2), over time, improved for all three groups of adolescents, with a significant interaction (time X cluster) for the adolescents with low HRQoL at baseline: 'CIS subjective fatigue' (Wilks' Lambda= .66, F (4, 66) = 3.77, $p < .05$, partial $\eta^2 = .19$).

Table 6.2 Changes over time for health-related quality of life (CHQ-CF87†) and fatigue (CIS-20‡)*

	Healthy	JIA low HRQoL	JIA high HRQoL	*p*
Physical Role functioning (3 items; 0–100)				
Baseline score (*SD*)	90.5^{a} (17.9)	50.6^{b} (24.3)	96.5^{a} (8.3)	< .001
3 months	5.5^{a}	27.2^{b}	2.9^{a}	< .001
9 months	6.3^{a}	33.4^{b}	1.2^{a}	< .05
Emotional Role functioning (3 items; 0–100)				
Baseline score (*SD*)	85.7 (17.7)	66.7^{a} (34.2)	91.8^{b} (16.9)	< .05
3 months	6.4	15.8	-4.4	.50
9 months	0.8	22.2	5.3	.22
Behavioral Role functioning (3 items; 0–100)				
Baseline score (*SD*)	96.8 (6.8)	80.2^{a} (34.2)	98.8^{b} (5.1)	< .05
3 months	1.6	12.9	-5.0	.52
9 months	-4.7	12.4	-0.6	.54
Bodily pain (2 items; 0–100)				
Baseline score (*SD*)	70.7^{a} (20.6)	32.2^{b} (20.5)	65.3^{a} (19.8)	< .001
3 months	4.3^{a}	16.6^{b}	5.3	< .05
9 months	11.4^{a}	15.6^{b}	2.6	< .05
General behavior (16 items; 0–100)				
Baseline score (*SD*)	81.3 (12.1)	73.4^{a} (6.8)	87.2^{b} (8.4)	< .05
3 months	-0.1	4.9	-1.6	.24
9 months	-2.2	7.2	0.4	.18
Mental health (16 items; 0–100)				
Baseline score (*SD*)	71.2^{a} (10.7)	60.6^{b} (11.6)	80.4^{c} (9.7)	< .001
3 months	1.6	3.4^{a}	0.2^{b}	< .05
9 months	0^{a}	6.2^{a}	2.7^{b}	< .05
Self-esteem (14 items; 0–100)				
Baseline score (*SD*)	71.1^{a} (8.4)	58.4^{b} (10.0)	74.4^{a} (9.2)	< .001
3 months	4.1	7.9	0.6	.20
9 months	-0.5	9.5	2.4	.13
General health perceptions (13 items; 0–100)				
Baseline score (*SD*)	77.9^{a} (11.4)	50.3^{b} (17.9)	65.0^{c} (12.2)	< .001
3 months	0.3^{a}	0.4^{b}	1.2	< .05
9 months	-0.8^{a}	2.2^{b}	6.8^{a}	< .05
CIS subjective fatigue (8 items; 8–56)				
Baseline score (*SD*)	26.0^{a} (11.3)	43.3^{b} (7.4)	19.6^{a} (8.6)	< .05
3 months	-4.0^{a}	-6.4^{b}	-1.7^{a}	< .05
9 months	-3.9	-10.5^{a}	1.4^{b}	< .05

Note.* Values are the mean changes from baseline except for the baseline values; all statistical analyses were performed on the raw data. All were items scored on a Likert scale. Means or mean changes with different superscripts differ significantly at $p < .05$.

† Higher scores indicate better physical or psychosocial well-being.‡ Higher score indicates a higher level of subjective fatigue.

Table 6.3 CHAQ scores of adolescents with JIA subdivided in two clusters

	JIA low HRQoL	JIA high HRQoL	*p*
CHAQ score (range 0–3)[a]			
Baseline mean (SD)	1.5 (0.7)	0.6 (0.7)	< .05
3 months	-0.3	0	.09
9 months	-0.2	0	< .05
VAS score overall well-being (0–10 cm)[b]			
Baseline mean cm (SD)	5.1 (2.5)	1.5 (1.8)	< .05
3 months	-3.0	0	.50
9 months	-1.9	-0.3	< .05
VAS score pain (0–10 cm)[b]			
Baseline mean cm (SD)	6.3 (2.5)	1.9 (1.9)	< .0001
3 months	-3.2	-0.1	.21
9 months	-3.1	-0.1	.18

Note. VAS = visual analogue scale; [a] 33 items (range 0–3), lower disability index indicate better functional ability; [b] lower VAS score indicates more optimal experience of well-being and less pain.

The adolescents with low HRQoL at baseline, improved significantly more compared to the healthy adolescents and the adolescents with JIA and high HRQoL at baseline, on the following subscales: 'Physical Role functioning' measuring limitations in school work and daily activities as a result of physical health (Wilks' Lambda= .58, F (4, 72) = 5.68, $p < .001$, partial $\eta^2 = .24$), and 'Emotional Role functioning' measuring limitations in school work and daily activities as a result of emotional problems (Wilks' Lambda= .75, F (4, 70) = 2.69, $p < .05$, partial $\eta^2 = .13$).

DISCUSSION AND CONCLUSION

Discussion

The aim of this study was to discover which adolescents growing up with JIA benefit from psychological intervention. Therefore, we conducted a study offering psychological counseling to a group of adolescents with JIA and a group of healthy adolescents. The choice was made to offer the adolescents psychological guidance with the SCM, a method that combines the personal narrative with the affective structure. While a psychological method like cognitive behavioural therapy (CBT) addresses the reduction of symptoms through a change of cognitive processes, the SCM intends a move beyond the symptoms by eliciting emotions and establishing a personal narrative change.

Since literature on the impact of JIA is inconclusive, probably not all, but just a subgroup of adolescents with JIA experiences difficulties in their psychosocial functioning. In our study, two-thirds of adolescents with JIA reported similar psychosocial functioning as their healthy peers. However, one-third of the adolescents with JIA reported low HRQoL at baseline. Like in other studies, low HRQoL at baseline was associated with a more recent onset of JIA, higher levels of pain, more severe physical disability, and higher levels of fatigue.[5,38,39] Adolescents with low HRQoL were older when diagnosed with JIA. Being diagnosed with such a severe illness during the already tumultuous period of adolescence is imaginably challenging for these individuals. Chronic illness, especially in adolescence, can – amongst others – limit the possibilities to develop strong feelings of self-enhancement by increased dependence on adults due to physical limitations. During psychological counseling with the SCM adolescents with low HRQoL reported fewer experiences of autonomy and success in their narratives (unpublished observations). Integrating this specific type of experience into a personal narrative is generally needed to develop an adult identity, and enhanced independence from caregivers, two of the main tasks of adolescence.[40]

After psychological guidance, adolescents with JIA and low HRQoL at baseline improved more than adolescents with JIA and high HRQoL at baseline and the healthy peers. This improvement could not be explained by changes in disease activity since most of the participating adolescents were in stable situations with no significant functional changes during the study period. But, although their functional ability and disease activity status were stable, the subjective experience of pain and well-being ameliorated in the group with low HRQoL. The reported subjective fatigue ameliorated most for the adolescents with low HRQoL. After psychological counseling, the adolescents with JIA and low HRQoL at baseline reported a significant improvement in their HRQoL. The intensity of the counseling (6 or 12 sessions in 3 month) did not influence the outcome, indicating that adolescents with JIA already benefit from modest guidance. These improvements in functioning may cause changes in daily life, such as full participation at school after long periods of absence due to fatigue and pain.

Defining the level of well-being from the adolescent's perspective may be an important strategy for improving daily clinical management of these patients. Especially in the context of the adolescent's development, repeated measurements of their well-being can be valuable to detect problems early so that psychological counseling can be offered before psychosocial problems increase. The CHQ-CF87 is simple and quick to administer. It can be routinely used in clinical practice, in particular for adolescents in the high-risk group (adolescents with a more recent onset of JIA, higher levels of pain, more severe physical disability, and higher levels of fatigue), to identify psychological adjustment problems. Appropriate psychosocial interventions, like the

SCM, can then be targeted to support these adolescents.[38] A few adolescents gave high scores for their HRQoL, whereas clinical observations didn't match with these reported high scores. However, during psychological counseling these few adolescents held on to their 'positive coping strategy', leaving no room for change. For the adolescents in our study, the most effective moment for psychological guidance was when the adolescents *themselves* reported that they experience difficulties in their psychosocial functioning.

Some caveats of our study should be mentioned. The method requires adolescents to have a basic capacity for self-reflection, which is related to a certain developmental stage and cognitive level of functioning, thereby selecting a specific subgroup of adolescents. In addition, the rather large improvements in health, functioning, and emotional experience in the group of adolescents with JIA and low HRQoL could be partly due to a floor effect in the other two groups, who already functioned well with little room for improvement. Finally, for this explorative study, the total sample size was modest. In future research, it would be appealing to study the emotional experience of adolescents with JIA in a larger cohort.

Conclusions

Adolescents with JIA already benefit from a relatively modest level of guidance and self-reflection. The current findings underline that two-thirds of the adolescents with JIA have similar emotional experiences as their healthy peers. Therefore, broad-based interventions to address psychosocial outcome are not necessary for *all* adolescents with JIA, instead the focus should be on those at risk. This study underlines the value of HRQoL patient questionnaires in clinical practice as they provide important data from the patient perspective. Careful screening for adolescents that report the highest negative effect of their disease on their quality of life, helps to identify youngsters who benefit most from psychological counseling.

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7

Summary and general discussion

Introduction

Advances in pediatric medicine have resulted in an increasing number of adolescents with chronic pediatric conditions growing into adulthood.[1] With success in extending the lives of children with chronic conditions, new challenges emerged. A chronic health condition in adolescence can have a major influence on the normal development of the youngster. Initial stresses associated with diagnosis, ongoing stresses from treatments and disruption of regular life, stigma and marginalization, and changes in plans and expectations about the future can be substantial challenges to social and emotional wellbeing. Adolescents with chronic health conditions and their families are responsible for managing the multi component treatment regimens that often include the management of medication, dietary prescriptions, lifestyle modifications, and attendance at routine clinic appointments with a variety of health care professionals.[2] The importance of understanding and promoting self-management in the care of pediatric chronic conditions is becoming increasingly apparent.

In pediatric settings, patients present themselves with a great number of different symptoms. Many of these patients show disease-specific, demonstrable abnormalities of structure or function that can be translated into a medical diagnosis. Treatment for these patients will focus on curing the disease, managing the symptoms, improving quality of life, and helping the patient and family cope with the impact of the condition. Other patients will be characterized more by their reported symptoms, suffering and disability, for which no clear medical explanation will be found. These medically unexplained physical symptoms, generally referred to as functional somatic symptoms, are common and often chronic in nature. The most frequently reported functional somatic symptoms are headache, musculoskeletal pain, fatigue, gastrointestinal distress, and sleep difficulties, thereby covering almost all medical specialisms.[3] Adolescents with these symptoms will often report significant functional impairment and emotional difficulties in their daily lives. This group of patients with functional somatic symptoms, place a heavy burden on the healthcare system due to frequent utilization of health resources and hospitalizations, specialist consultations, or unnecessary treatments.

In the way described above, it sounds like an almost perfect distinction between these two groups of patients; one presenting themselves with somatic symptoms with a clear pathology, the other with medically unexplained symptoms – often referred to as psychosomatic. In clinical practice however, this split is more arbitrary. For example, the severe fatigue of a patient diagnosed with the chronic fatigue syndrome (CFS) can initially have started with an Epstein-Barr virus infection. On the other hand, a patient with juvenile idiopathic arthritis (JIA) can report fatigue or pain that cannot be attributed to disease activity. Regardless of disease activity, in the case of JIA, pain, fatigue and sleep disturbance are prevalent among most patients[4,5] and patients with comparable disease severities differ widely in the reported impact of these subjectively experienced symptoms. In adult patients, the statistical correlation between pain and objective measures of disease severity is only modest, ranging between 0.23 and 0.52,[6] and successful treatment of the disease process is not necessarily accompanied by improvement in symptoms.[7] In medical care, the more subjectively experienced symptoms often challenge health care providers. For example, should the pediatrician accept or treat the reported fatigue and emotional distress of a patient with JIA in remission? And, if treatment is the choice, should the focus lay on medication, arthritis education, physical exercise or psychological counseling?

Subjectively experienced, but medically unexplained symptoms are common within a large number of pediatric patients. Although there is growing support for a positive effect of cognitive behavior therapy (CBT) in the treatment of medically unexplained symptoms,[8,9] the effect sizes remain limited and only a proportion of patients benefit from this approach.[10-12] CBT mainly focuses on reducing the symptoms by altering dysfunctional cognitive processes and behavior. By focusing on symptomatology, a true understanding of the meaning of these medically unexplained symptoms for the individual patient will remain undiscovered. To come to long-term management and to achieve optimal health outcomes, health care providers should not only treat the symptoms, but also be aware of the more subjective emotional experiences of the patients dealing with the medical conditions.[13-15]

Our goal was to provide an in-depth understanding of the adolescents' experiences in growing up with a chronic health condition. Not the professionals or caregivers point of view on this subject, but understanding that of the adolescent him- or herself was our primary ambition. In this final chapter, the findings of the different articles of this dissertation will be combined to a general discussion concerning the emotional experience of adolescents facing a chronic health condition.

Beyond the symptoms

In this dissertation, we focused on the subjective experience of being ill and the way in which adolescents organize these experiences through self-narratives. A growing number of researchers and theorists in the humanities, and social- and medical sciences, conceive human identity in narrative terms.[16-18] The theory of narrative identity assumes that individuals form an identity by integrating their life experiences and interpretations of those experiences into an internalized, evolving story, which provides the individual with a sense of unity and purpose in life.[16] The stories individuals tell about themselves and their social worlds form key aspects of their identity. However, the narrative is not only seen as a new way of conceptualizing human experience and identity, but also as a useful clinical tool to help individuals understand how they behave, and organize their lives in certain ways, and to aid them in retelling and reorganizing their life-story.[19] A coherent narrative is first acquired during adolescence.[20] Adolescents' development and creation of a life-story is an integral part of the process of forming an adult identity. However, constructing a contextualized and intelligible self-narrative is an ongoing process since circumstances change, new possibilities arise and limitations in our life-story are discovered.[21] Therefore, editing and revising one's life-story is an ongoing task that takes place throughout the whole life span.

Self-narratives are constructed as we live in a social world and are accordingly always entangled with other people's stories. Therefore, narrative identity can best be studied when placed in a context. Dialogical self theory[22,23] is a relatively new current in identity research, incorporating quantitative methodology, and sensitive to the contextual dimensions of individuals' identity. While acknowledging the human striving for a single and coherent life-story, dialogical self theory at the same time points to the fact that identity emerges from a plurality of positions.[24] Dialogical self theory considers the self as a multiplicity of positions (voices, characters, parts) that have the potential of entertaining dialogical relationships with each other. The different positions of the self are not only involved in communicative interchange, but are also subjected to relative dominance, with some positions being more powerful or speaking with a louder voice than other positions.[25] These different parts of the self are able to entertain dialogical relationships, both within and between persons. The self, as studied in this thesis, is therefore seen as multivoiced and dialogical.[26]

As an example of a multivoiced and dialogical self, let us consider the case of Mia, an 18-year old girl, diagnosed with JIA at the age of 9. The disease is currently in remission, but while at high school, she had frequent disease flares. Her classmates knew of her illness and were familiar with the symptoms. This made Mia feel at ease and confident enough to show different sides of her self. Mia possesses a broad position repertoire, containing, among others, positions like '*I-as-cheerful*', '*I-as-creative*' but also '*I-as-tired*', and '*I-as-dependent-on-my-parents*'. Mia recently started law school. In this new environment, Mia at first chose to present herself with healthy and strong positions like '*I-as-self-secure*', and '*I-as-independent*' to gain confidence. But at home and in the weekends, she meets her old friends. At these encounters, she narrates her experiences from more vulnerable positions like '*I-as-insecure*' or '*I-as-lonely*'. Mia maintains a healthy narrative organization since she can flexibly move from one position in the story to the other.

In order to respond adequately to changing personal and social demands, a person needs to be able to employ an emotionally varied repertoire of positions to narrate the life-story.[25] Certain life experiences are relatively easy to narrate and fit smoothly into the individual's ongoing stories of their self. Events such as starting graduate school, failing an important exam or making a career choice, are experiences that occur in many individual's lives and most adolescents are familiar with common cultural stories about these types of situations. By contrast, other experiences are less common and pose a challenge to successfully be integrated into one's narrative identity.[27] Having a chronic illness during adolescence is an example of a particularly challenging experience, for this experience is unplanned, physically and emotionally difficult, and typically not a part of the youngster's ideal future script. The adolescent must significantly revise his or her existing life-story and create new positions from which the story can be told. This task is an important narrative challenge, which will be completed differently by every adolescent.

Summary and main study findings

Although every narrative is unique, the self-confrontation method (SCM) (28) and the personal position repertoire (PPR)[29] enabled us to structurally analyze and compare the different stories told. The most notable findings of the different studies of this thesis will be summarized.

In order to understand the intensive SCM procedure, an extensive report of its application is given in **Chapter 2**, illustrated by a case-example. The SCM is an instrument to assess and change individuals' self-experience, through narrative self-investigation. It combines a focus on individuals' verbal reports, with an analysis of the underlying affective and motivational structures. The most important conclusions were that (a) with the SCM a more complete picture of the patients' current situation could be obtained since their whole life is taken into account, not only in its temporality, but also its social and cultural context. Furthermore, (b) the SCM allowed adolescent and therapist to access and assess a deeper, affective level, behind the manifest level of the self-narrative. And finally, (c) equipped with this insight, the adolescent could gradually become receptive to other feelings and experiences, which results in a reorganization of the self-narrative.

In **Chapter 3**, the theoretical assumptions of the dialogical self theory are introduced. The PPR is a method to gain insight into the plurality of self-positions and to investigate basic aspects of the relations of the self-positions in their social context. In this chapter, some of the baseline measures are presented. The self-positioning of 42 adolescents with CFS, 37 adolescents with JIA and 23 healthy teenagers are presented. It was found that adolescents with CFS presented themselves from the stance of their 'unwell-I-positions', focusing on illness and being dependent. With these dominating positions relating to their illness, there seemed to be little room left for stronger positions. In contrast, a different outcome was found for adolescents with arthritis. Adolescents with JIA presented a position repertoire very similar to that of their healthy peers, mostly dominated by their healthy and strong self-positions. This healthy and strong presentation might be adequate and sustainable in adolescence, but could prove too strenuous to maintain throughout adult life.

A further investigation of these findings, addressing the adolescents' life stories, underlying the self-positioning as reported in the previous chapter, is described in **Chapter 4**. The SCM was used to analyze the emotional level of the personal narratives of the participating adolescents. It was found that the personal narratives of the adolescents with CFS were in accordance with their self-positioning. Namely, the themes of these self-narratives revolved around negative experiences of powerlessness, isolation and unfulfilled longing. Furthermore, their self-narratives contained fewer experiences in which they felt autonomous and successful. For adolescents with JIA, their strong and healthy presentation was not in accordance with the emotional experience they reported in the SCM-procedure. Behind their overly positive and healthy presentation, a self-narrative with fewer experiences of autonomy and success, compared to the experiences of their healthy peers, was found.

Chapter 5 presents the study into the use of the SCM in adolescent CFS, and the longitudinal changes in patients' self-narratives. The patients with CFS described in chapter 3 and 4 were randomly assigned to either 6 or 12 sessions with the SCM. The healthy controls were assigned to 6 sessions. Self-investigation resulted in significant changes in participants' narratives. Moreover, after self-investigation there was a significant improvement in fatigue, physical and psychosocial functioning for the adolescents with CFS. The patients who completed 12 sessions improved most. At follow-up, the positive effects were maintained. The SCM allowed the adolescents to discover – for themselves – factors that might cause or perpetuate their fatigue. The results suggested that self-investigation is a useful instrument in the management of adolescent CFS.

To study which adolescents with JIA benefited from narrative counseling with the SCM, the longitudinal results of these patients are presented in **Chapter 6**. The patients with JIA described in Chapter 3 and 4, received psychological guidance with the SCM. Two-thirds of adolescents with JIA functioned well before and after self-investigation. The remaining adolescents with JIA reported low health-related quality of life at baseline, and this subgroup benefited from guided self-reflections. The most effective moment for psychological guidance of adolescents with JIA was when the adolescent him- or herself reports difficulties in health-related quality of life.

The patients' narrative: case-examples

It seems evident that all adolescents with a chronic illness are affected by this experience in their own particular way. In the total study-period, more than hundred adolescents shared their personal, and ever fascinating life-stories with the counselors. To stay with the uniqueness of some of the patients we saw, we present three adolescents from the study in Box 7.1, to illustrate the different types of adolescents that we identified. As can be seen in these case-examples, an important ingredient for a positive outcome of narrative counseling is that the adolescent him- or herself can realistically acknowledge the impact of the illness on their quality of life

General conclusions

This dissertation focused on the self-experience of adolescents with JIA and CFS. Although the etiology and nosology of JIA and CFS are fundamentally different, some commonalities in the emotional experience of adolescents dealing with these chronic illnesses have been found. The most striking observation is that the self-narratives of both patient groups contained fewer experiences in which they felt autonomous and successful compared to a healthy control group. Especially during adolescence, this type of experiences is essential to develop an adult identity and enhanced independence from caregivers.[31] Although emotional problems have usually been thought of as reflecting the presence of negative affective states, and the amelioration of negative emotions has traditionally been a focus of psychological counseling, emotional experience might also become problematic when certain positive affective states are absent.[32-34] While the presence of negative emotions certainly seems to play a significant role in our CFS group, the absence of certain positive emotions appears to be of importance in the emotional experience of both our JIA group, as well as our CFS group. Therapeutic focus should therefore lie on the integration and utilization of *positive*, as well as negative emotions, and the importance of variety and flexibility in the experience and voicing of these emotions.[35-37]

Although communality is found in a diminished experience of autonomy and success, adolescents with JIA and CFS position themselves differently in their social context. To generalize the results of this thesis, adolescents with CFS position themselves in accordance with their emotional experience of less positive and more negative experiences. Unwell-positions, focusing on illness and being dependent, are dominating most of the self-narratives of adolescents with CFS. For many adolescents with JIA, while experiencing less autonomy and success then their healthy peers, they position themselves as their healthy peers: healthy and strong. The different etiology of the two illnesses might, to an extent, have caused this difference in personal positioning. Although JIA is a serious somatic illness, which disrupts the adolescents'

Box 7.1 Three case-examples

a) Amy is a 15-year old girl, diagnosed with polyarticular arthritis at age 6. The arthritis is in remission since two years. At school she performs well, she has several close friends, and does currently have a boyfriend. She has told her classmates of the arthritis. She stopped with gymnastics because of the arthritis but now started with drama classes, which she really likes.

In narrative counseling:
Amy integrated the experience of having arthritis into a broad life-story. She was able to adapt herself to the unfortunate consequences of the arthritis, by choosing an activity that suited her abilities. She was able to take different personal positions, by also showing her vulnerable side of being ill to her classmates. Amy's self-narrative contained different emotional experiences, she was able to shift between self-positions and she scored her health-related quality of life high. After narrative counseling, no major changes were observed in social, physical or emotional outcome parameters, but she indicated that the insights she obtained from the counseling, were valuable to her.

b) Jane, diagnosed with CFS six months prior, is now 16 years old. She's the only child of parents that worry a lot about the cause of the ongoing fatigue of their daughter. Jane doesn't share these worries about the cause of her medical complaints, but feels sad she can't do the things she used to do. Especially riding her horse is an activity she misses a lot. She's capable of going to school for two hours a day, and spends the rest of her day at home with her mother.

In narrative counseling:
Jane's self-narrative was dominated by experiences of helplessness, hopelessness, powerlessness and isolation. She experienced her chronic fatigue as so extremely disabling that her illness determined her, and her parents, complete life. As a result, Jane alienated from most other self-positions she could adopt before being ill. Her narrative coherence stopped to be a continuous process of ongoing dialogue and interaction, but instead became rigid and monological. Together with the counselor, Jane discovered that one of the central themes in her self-narrative was that Jane worried a lot about her mother's problems (her mother lost her job and was feeling depressive). In narrative counseling, Jane was stimulated to rediscover old, but also new personal positions. Especially, more autonomous internal self-positions were introduced. Jane was stimulated to talk with her mother about her worries, to start with a drawing class, and to try to renew peer relationships. In this way, through external dialogue, communication between self-positions could be recovered and new feelings, possibilities and perceptions were opened (30). Jane's fatigue ameliorated and her health-related quality of life improved after counseling.

c) William is 17 years old. He has systemic JIA since the age of 9. At the age of 11 he had a stem cell transplant. Although it was successful, his functional ability remained limited due to the prior arthritis. William has got several good friends. He never talks about the arthritis with others than his family members and pediatrician. He wants to go to medical school, although his fingers are severely affected by the arthritis.

In narrative counseling:
William presented himself as a cheerful, positive and charming adolescent. Although walking with crutches to support his deformed legs, having a short stature and painful fingers, he gave high scores for his health-related quality of life. His self-narrative was dominated by strong and healthy self-positions. He was convinced he could fulfill his dream of becoming a surgeon. The central theme in his self-narrative was that 'with a lot of perseverance, everything can be possible'. In counseling, William held on to his strong and healthy position repertoire, leaving no room for new self-positions to be introduced. After narrative counseling, few changes were observed in social, physical or emotional outcome parameters. With the knowledge generated from the outcome of our studies, we would now wait for counseling till William himself would report a reduced, but thereby more realistic, health related quality of life. At that moment, an intrinsic motivation for change will arise. Another option in trying to support William is by creating external situations in which he can experience feeling successful and autonomous in order to validate his strong presentation, and in addition, in which he can also safely acknowledge his (physical) limitations. In this way, he can create room for new positions in his self-narrative.

sense of self and impairs their capacity for social participation, one of the main themes in their experience is a striving for normality. Adolescents with JIA generally don't want to be socially labeled as sick or disabled.[14] In trying to be more acceptable, adolescents adapt their behaviour sometimes even to the detriment of their illness, e.g. by non-adherence to therapy responsible for changing their appearance,[38] or by pushing themselves beyond their physical limits to be able to fully participate. CFS on the other hand, is a medically unexplained illness. The stigma associated with a functional somatic illness often makes patients feel that the legitimacy of their illness is being discounted which may make them cling more assiduously to a biomedical explanation of their symptoms,[39] and present themselves as seriously ill in their social context.

We expected that counseling with the SCM would enable these adolescents to develop their selves and their stories and adapt to the challenges of life through a continuous renewal and shifting in the organization of their self-narrative. CFS or JIA often altered the self-experiences of the adolescents and diminished their position repertoire, seemingly resulting in a more rigid and monological self-narrative. Their life-stories are lived from a more one-sided perspective, which makes it more difficult to integrate new experiences. Psychological counseling might be effective, not through directly modifying negative or maladaptive cognitive content, but rather by changing patients' relationships to their dysfunctional thoughts and feelings, through the creation of new personal meaning.[40,41] In our studies, the crucial ingredient for a positive outcome of narrative counseling was that the adolescent him- or herself acknowledged the negative impact of the illness on their quality of life. Health-related quality of life questionnaires proved to be a good measure to identify those adolescents that might be receptive to therapeutic help. Almost all adolescents with CFS benefitted from narrative counseling by giving voice to the more suppressed sides and regaining flexibility in their self-narrative. The adolescents with JIA were less unanimous. Three distinctive types seemed to emerge within the total sample: a) the first type is the adolescent that has properly integrated the arthritis experience into his or her life-story, giving voice to the strong but also the more unwell sides of their experience and reporting a generally good health related quality of life, b) the second is the type of adolescent with JIA that narrates the story from the unwell and ill positions, not being able to experience or give voice to more strong sides of their story, reporting low health-related quality of life, and c) the last type has a strong and healthy monological self-narrative, not giving room to the weaker sides of the self, reporting high health-related quality of life. The first type doesn't really need guidance since they already accomplished a flexible and dialogical self. The adolescents belonging to the second type resemble the adolescents with CFS and benefited from the narrative counseling method by regaining flexibility in their self-narratives. Adolescents with JIA of the last type, narrated their self-story from mainly healthy and strong self-positions and scored their quality of life accordingly, but clinical observations didn't match with the image the adolescent created of him- or herself. However, during psychological counseling most of these adolescents held on to their 'positive coping strategy', leaving no room for change. For these adolescents, our suggestion is to wait for counseling until the adolescent him- or herself would report a more realistic view which would be seen in clinical practice and in reduced reported health related quality of life. Another idea is to reinforce the strong positions by letting them experience situations in which they can really feel successful and autonomous. This will create more ground, or a 'safety net' for the negative emotions. In line with this, the University Medical Center Utrecht currently implemented preventive group-programs ('Skills Lab') to promote self-management skills and physical consciousness, specially developed for youngsters with a chronic illness.

Future directions

These explorative studies into the self-experience of adolescents with chronic illness, gave us interesting and promising results that hopefully will lead to future research on this topic. Our understanding of the impact of chronic illness on adolescent life will be further improved by considering the following reflections.

The adolescents with CFS in our study have shorter disease duration than the adolescents with JIA. It is possible that the adolescents with CFS benefitted more from counseling, because there is greater latitude for emotional, behavioral and cognitive change early in the course of illness when coping styles have not yet become wholly rigidified. Furthermore, the age at diagnosis might be an important factor in determining how the illness is incorporated into the personal narrative. Before adolescence, the sense of self is not yet coherent and the self-story is not unified. When diagnosed during those younger years, the illness experience might be differently integrated into the life-story than when diagnosed during adolescence, when, to some extent, a coherent self-narrative is often already being developed. Furthermore, being diagnosed with a chronic illness during adolescence, a tumultuous phase after all, might also be of importance for how the illness experience is integrated into the ongoing self-narrative.

Adolescents with JIA appeared to cope relatively well with the consequences of their painful and potentially incapacitating illness, and there seemed to be few differences between them and healthy teenagers. This view on psychosocial functioning of adolescents with JIA is also found in literature on this topic.[42,43] But, young adults with arthritis report that JIA adversely affected, among others, work employment,[44,45] body image,[46] social interactions,[46] mood,[47] and overall quality of life.[48,49] Therefore, the strong and healthy presentation of the adolescents with JIA might be too strenuous to be maintained into young adulthood. It seems important to study the long-term effects on identity formation of the integration of the arthritis experience into the life-story in different developmental stages.

Implications for daily practice

- Health care professionals should look beyond the symptoms and try to include the personal experience of the adolescent into disease-management.
- Short health-related quality of life questionnaires measuring physical, psychological and social functioning can be used as a general instrument to select those adolescents with diminished health-related quality of life that will potentially benefit from psychological guidance.
- If adolescents with a chronic illness experience diminished health-related quality of life, regaining flexibility in their life-narrative improves physical, social and emotional well-being.
- From a clinical point of view, the SCM proved a) to be a valuable method for adolescents that are hesitant to seek professional mental-health care, because focus lays on the complete life-story instead of a specific focus on potential problems, b) to be a method in which the adolescent him- or herself has a leading role in the search for underlying narrative themes, which is in line with the developmental tasks of an increased autonomy and more self-management, and c) to create an opening for change if the adolescents are able to confront themselves realistically with the challenges they face due to their chronic condition.

Iris (17 years) "I can now let go of my mask, I don't need to convince everybody I'm ok. They just have to take me how I am."

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Samenvatting (Summary in Dutch)

Vooruitgang in de pediatrische geneeskunde heeft geresulteerd in een toenemend aantal adolescenten dat met een chronische pediatrische aandoening volwassen wordt. Met dit succesvol verlengen van de levens van kinderen met chronische aandoeningen, zijn nieuwe vraagstukken ontstaan. Een chronische ziekte in de adolescentie kan een grote invloed hebben op de ontwikkeling van de jongere. Onzekerheid bij de initiële diagnose, spanning door de intensieve behandelingen, een verstoring van het dagelijks leven, stigmatisering en marginalisering, en zorgen over veranderingen in de plannen en verwachtingen voor de toekomst kunnen allen aanzienlijke invloed hebben op het sociale en emotionele welzijn van de jongere. Adolescenten met een chronische aandoening en hun families worden verantwoordelijk voor het opvolgen van strenge behandelingsvoorschriften, het volgen van diëten, het aanpassen van de levensstijl en het regelmatig nakomen van afspraken bij verschillende professionals in de gezondheidszorg. De nadruk in de zorg voor jongeren met een chronische ziekte komt steeds meer te liggen op het ondersteunen en bekrachtigen van het zelfmanagement van de jongeren.

In pediatrische instellingen presenteren jongeren zich met een verscheidenheid aan klachten. Veel patiënten hebben duidelijk aantoonbare medische symptomen die samen leiden tot een medische diagnose. Een andere groep patiënten kenmerkt zich meer door gerapporteerde klachten en beperkingen waarvoor geen duidelijke medische oorzaak gevonden kan worden. Zoals hierboven beschreven, lijken dit twee duidelijk te onderscheiden groepen patiënten, de een met somatische symptomen met een duidelijke pathologie, de andere met medisch onverklaarde, psychosomatische klachten. Maar in de klinische praktijk is deze verdeling niet zo zwart-wit. Zo kunnen de vermoeidheidsklachten van een patiënt met CVS oorspronkelijk begonnen zijn met een Epstein-Barr virusinfectie. Of kan een patiënt met juveniele idiopathische artritis (JIA) vermoeidheids- en pijnklachten rapporteren die niet kunnen worden toegeschreven aan ziekte-activiteit. Subjectief ervaren, maar medisch onbegrepen symptomen komen veel voor bij pediatrische patiënten. In de behandeling van medisch onbegrepen klachten is er veel steun voor cognitieve gedragstherapie (CGT), maar de effecten zijn soms beperkt en alleen een deel van de patiënten heeft baat bij deze aanpak. CGT focust voornamelijk op het verminderen van de symptomen door disfunctionele gedachten en gedragingen te beïnvloeden. Echter, door vooral te focussen op de symptomen, wordt er voorbij gegaan aan de *betekenis* van de medisch onbegrepen klachten. Om tot lange-termijnverbetering te komen, zullen professionals in de gezondheidszorg zich meer bewust moeten zijn van de subjectieve, emotionele ervaringen van de patiënten.

Het doel van deze dissertatie was om een diepgaand inzicht te verkrijgen in de ervaringen van adolescenten die opgroeien met een medische aandoening, niet vanuit het gezichtspunt van de professional of verzorgers, maar vanuit de jongeren zelf. Voorts is bestudeerd of begeleiding op basis van dit inzicht zou leiden tot een verbetering van hun welbevinden.

Meer dan symptomen

In deze dissertatie ligt de focus op de subjectieve ervaring van de jongeren met een chronische ziekte en de manier waarop ze hun ervaringen organiseren in een 'zelfverhaal'. Een groot aantal onderzoekers en theoretici in de sociale, medische en geesteswetenschappen bestuderen de menselijke identiteit in narratieve termen. De theorie van de narratieve identiteit veronderstelt dat individuen een identiteit vormen door hun (interpretatie van) levenservaringen te integreren tot een geïnternaliseerd, zich ontwikkelend verhaal. De verhalen die mensen over zichzelf en de wereld om hen heen vertellen, vormen belangrijke aspecten van hun identiteit. Het narratief wordt echter niet alleen gezien als een nieuwe manier om ervaringen en identiteit te conceptualiseren, maar ook als een klinisch middel om individuen inzicht te geven in zichzelf en de manier waarop ze hun leven organiseren, en om ze hun levensverhaal te laten formuleren en te herorganiseren. Een zelfverhaal wordt voor het eerst gevormd tijdens de adolescentie. Om tot een volwassen identiteit te komen is de creatie en de ontwikkeling van een coherent zelfverhaal een onmisbaar onderdeel. Maar het creëren van een begrijpelijk, in de context passend, zelfverhaal is een voortdurend proces aangezien omstandigheden veranderen, nieuwe mogelijkheden ontstaan en beperkingen van het verhaal worden ontdekt. Daarom is het aanpassen en herzien van het zelfverhaal een levenslange taak.

Zelfverhalen worden geconstrueerd in een sociale wereld en zijn daardoor ook onlosmakelijk verbonden met andermans verhalen. De narratieve identiteit kan dan ook het best bestudeerd worden in de sociale context. De 'dialogische zelf'-theorie is een relatief nieuwe stroming in het onderzoek naar identiteit. Deze theorie integreert een kwantitatieve methodologie en is gevoelig voor contextuele dimensies van de identiteit. Terwijl het streven naar een enkel, coherent zelfverhaal erkend wordt, neemt de 'dialogische zelf'-theorie aan dat identiteit ontstaat vanuit meerdere posities. Het zelf wordt hierin gezien als een veelheid aan posities (stemmen, karakters, delen) die met elkaar dialogische relaties onderhouden. Deze verschillende posities van het zelf zijn onderhevig aan hiërarchie, waarin sommige posities sterker zijn of met een luidere stem spreken dan andere posities. Deze verschillende delen van het zelf kunnen dialogen voeren zowel binnen het zelf als naar buiten toe met andere personen. Het zelf dat bestudeerd wordt in deze dissertatie, wordt dan ook gezien als meerstemmig en dialogisch.

Om adequaat te reageren op de veranderende persoonlijke en sociale eisen, moet een persoon in staat zijn om vanuit een emotioneel gevarieerd repertoire aan posities zijn levensverhaal te kunnen vertellen. Bepaalde levenservaringen zijn relatief gemakkelijk om te vertellen en passen soepel in de doorlopende verhalen van het zelf. Gebeurtenissen zoals het starten van voortgezet onderwijs, een onvoldoende halen voor een belangrijke toets, of een beroepskeuze maken, zijn ervaringen waar de meeste jongeren mee geconfronteerd worden en de meeste adolescenten zijn bekend met culturele verhalen over dergelijke ervaringen. Andere ervaringen daarentegen zijn minder gangbaar en daardoor moeilijker te integreren in de narratieve identiteit van een persoon. Het hebben van een chronische ziekte tijdens de adolescentie is een voorbeeld van een bijzonder lastige ervaring om te integreren in het zelfverhaal, aangezien deze ervaring ongepland is, fysiek en emotioneel lastig is, en zeker niet onderdeel is van het ideale toekomstscript van de jongere. De adolescent moet zijn bestaande levensverhaal aanzienlijk aanpassen en nieuwe posities creëren om het verhaal te kunnen vertellen. Deze lastige taak zal door elke adolescent op een andere manier volbracht worden.

Samenvatting en belangrijkste bevindingen

Ook al is elk narratief uniek, de zelfkonfrontatiemethode (ZKM) en het persoonlijk positie repertoire (PPR) maken het mogelijk om op een gestructureerde manier levensverhalen te analyseren en te vergelijken. De resultaten van de verschillende studies uit dit proefschrift worden hieronder samengevat.

Om inzicht te krijgen in de intensieve procedure van de ZKM, wordt in **Hoofdstuk 2** een uitgebreid verslag van het gebruik van de ZKM aan de hand van een voorbeeld gegeven. De ZKM is een instrument om verhalen die personen over zichzelf vertellen te onderzoeken en eventueel te veranderen. Het combineert daarbij een focus op verbale uitingen met een analyse van de onderliggende emotionele en motivationele structuren. De meest belangrijke conclusies zijn dat a) in de ZKM een compleet beeld van de actuele situatie van de patiënt kon worden geschetst, omdat het hele leven, met niet alleen de temporele maar ook de sociale en culturele context werd onderzocht. Verder, b) konden dankzij de ZKM de patiënt en de behandelaar verder dan het manifeste niveau van het zelfverhaal, ook een dieper, affectief niveau onderzoeken. En tenslotte, c) dankzij dit inzicht kon de adolescent stap voor stap meer open staan voor andere gevoelens en ervaringen, hetgeen resulteerde in een herorganisatie van het zelfverhaal.

In **Hoofdstuk 3** worden de theoretische hoofdaannames met betrekking tot de 'dialogische zelf'-theorie geïntroduceerd. Hieruit voortkomend is het PPR ontwikkeld, een methode die inzicht kan bieden in de pluraliteit van de zelfposities van een persoon. De methode kan daarnaast ook basale aspecten van de relaties van de zelfposities in de sociale context in beeld brengen. In dit hoofdstuk wordt een aantal van de uitgangskarakteristieken gegeven van de deelnemers die voor de studies van deze dissertatie geïncludeerd werden. De zelfpositionering van 42 adolescenten met CVS, 37 adolescenten met JIA en 23 gezonde jongeren wordt gepresenteerd. De patiënten met CVS presenteerden zichzelf vanuit de ziekte-gerelateerde zelfposities, met een focus op ziekte en afhankelijkheid. Met deze dominantie voor de ziekte-gerelateerde zelfposities, lijkt er weinig ruimte voor sterkere zelfposities. Voor de jongeren met JIA werd een ander beeld gevonden. Adolescenten met JIA presenteerden een persoonlijk positie repertoire dat erg overeen kwam met dat van hun gezonde leeftijdsgenoten, waarin de gezonde en sterke zelfposities domineerden. Deze gezonde en sterke presentatie zal waarschijnlijk tijdens de adolescentie vol te houden zijn, maar is mogelijk te inspannend om als volwassene vol te houden.

Een verdere uitwerking van de resultaten, de zelfverhalen die achter de persoonlijke positionering van de adolescenten uit het vorige hoofdstuk schuilgaan, wordt gepresenteerd in **Hoofdstuk 4**. De ZKM is gebruikt om het affectieve niveau van de persoonlijke verhalen van de deelnemende adolescenten te analyseren. De zelfverhalen van de adolescenten met CVS kwamen overeen met hun zelfpositionering. De thema's in hun zelfverhalen focusten namelijk op negatieve ervaringen van machteloosheid, isolement en onvervuld verlangen. Bovendien bevatten de zelfverhalen van adolescenten met CVS minder ervaringen van autonomie en succes. Bij adolescenten met JIA kwam de affectieve ervaring van hun zelfverhalen niet overeen met hun sterke en gezonde presentatie. Bij deze adolescenten kwamen, achter de voornamelijk positieve presentatie, in hun zelfverhalen aanzienlijk minder ervaringen van autonomie en succes voor dan in de zelfverhalen van gezonde leeftijdsgenoten.

Hoofdstuk 5 beschrijft de resultaten van de studie naar de begeleiding van de adolescenten met CVS door middel van zelfonderzoek, en de longitudinale veranderingen in hun zelfverhalen. De adolescenten met CVS die werden beschreven in Hoofdstuk 3 en 4, zijn gerandomiseerd over 6 dan wel 12 sessies met de ZKM. De gezonde jongeren kregen allen 6 sessies. Zelfonderzoek leidde tot significante veranderingen in de zelfverhalen van deelnemers uit de verschillende groepen. Bij de jongeren met CVS bleek er tevens een significante verbetering te zijn in hun vermoeidheid, en hun fysieke en psychosociale functioneren. De patiënten die 12 sessies toegewezen hadden gekregen verbeterden het meest. Bij follow-up bleken de positieve resultaten te zijn behouden. Er werd dan ook geconcludeerd dat zelfonderzoek niet alleen leidt tot een verandering in de zelfervaring van de adolescente patiënt met CVS, maar ook tot gezondheidsverbetering. De ZKM maakt het voor deze jongeren mogelijk om zelf factoren te ontdekken die bijdragen aan hun vermoeidheid, of deze zelfs veroorzaken. De resultaten wijzen erop dat zelfonderzoek een nuttig instrument is in de behandeling van CVS.

In **Hoofdstuk 6** worden de longitudinale resultaten gepresenteerd van de adolescenten met JIA. De patiënten met JIA die in Hoofdstuk 3 en 4 zijn beschreven, ondergingen psychologische begeleiding aan de hand van de ZKM. Tweederde van deze groep adolescenten functioneerde op psychosociaal vlak goed voor en na het zelfonderzoek. De rest van de adolescenten met

JIA rapporteerde op baseline lage scores voor hun gezondheidgerelateerde kwaliteit van leven, en deze subgroep verbeterde aanzienlijk door begeleiding met de ZKM. Het meest effectieve moment voor psychologische begeleiding aan de hand van de ZKM voor adolescenten met JIA was als de adolescent zelf aangaf problemen te ervaren in zijn gezondheidgerelateerde kwaliteit van leven.

Algemene conclusies

In deze dissertatie zijn de zelfervaringen van adolescenten met JIA en CVS bestudeerd. Alhoewel de etiologie en de nosologie van deze ziektes fundamenteel anders zijn, kunnen er toch een aantal overeenkomsten gegeven worden in de emotionele ervaring van adolescenten die met deze ziektes opgroeien. De meest opvallende bevinding was dat de zelfverhalen van de adolescente patiënten minder ervaringen bevatten waarin ze zich autonoom en succesvol voelen, vergeleken met de zelfverhalen van de gezonde jongeren. Zeker tijdens de adolescentiefase is juist dit type ervaringen essentieel om onafhankelijk te worden en een volwassen identiteit te ontwikkelen. Ook al is vaak gedacht dat emotionele problemen worden veroorzaakt door de aanwezigheid van negatieve gevoelens, en ligt in traditionele psychologische behandeling de focus vaak op het verminderen van negatieve emoties, de emotionele ervaring lijkt juist ook problematisch te worden bij het ontbreken van bepaalde positieve gevoelens en ervaringen. Terwijl bij adolescenten met CVS de aanwezigheid van negatieve emoties zeker een rol lijkt te spelen, is bij zowel de adolescenten met JIA als met CVS de afwezigheid van bepaalde positieve emoties ook mogelijk van invloed op hun welbevinden. De therapeutische focus zal zich daarom moeten richten op de integratie en het gebruik van zowel *positieve* als negatieve emoties, en het flexibel kunnen gebruiken en ervaren van deze emoties.

Ook al is er een overeenkomst gevonden in het hebben van minder ervaringen van autonomie en succes in beide patiëntengroepen, de jongeren met CVS en JIA positioneren zich verschillend in hun sociale context. Generaliserend positioneren adolescenten met CVS zich overeenkomstig hun emotionele ervaring van meer negatieve en minder positieve ervaringen, namelijk posities met betrekking tot ziekte en afhankelijkheid domineren de meeste zelfverhalen van jongeren met CVS. Veel adolescenten met JIA daarentegen geven aan minder ervaringen van autonomie en succes te hebben, maar ze positioneren zich even sterk en gezond als hun gezonde leeftijdsgenoten. Tot op zekere hoogte zou het verschil in etiologie een verklarende factor kunnen zijn voor dit verschil in positioneren in de sociale context. Ook al is JIA een ernstige somatische aandoening die vaak van grote invloed is op het zelfgevoel en de sociale participatie van de jongeren, is een van de centrale thema's van jongeren met JIA, net als dat van gezonde adolescenten, het nastreven van normaliteit. Adolescenten met JIA willen over het algemeen niet gelabeld worden als ‘ziek’ of ‘beperkt’. Ze streven sociale acceptatie dusdanig na dat het zelfs ten koste kan gaan van hun ziekte, bijvoorbeeld door medicatie te weigeren die van invloed is op hun uiterlijk of door voorbij te gaan aan hun fysieke grenzen om volledig mee te kunnen doen. CVS aan de andere kant is een medisch onbegrepen ziekte. Het stigma dat geassocieerd wordt met een functioneel somatische aandoening kan ervoor zorgen dat patiënten het gevoel hebben dat de legitimiteit van hun ziekte wordt betwist, waardoor ze zich meer zullen vastklampen aan een biomedische verklaring voor hun symptomen en ze zich ook als ernstig ziek zullen presenteren in hun sociale context.

Bij aanvang van de studie verwachtten we dat begeleiding met de ZKM de adolescenten de mogelijkheid zou geven om de organisatie van hun zelfverhaal te veranderen waardoor ze zich makkelijker zouden kunnen aanpassen aan de moeilijkheden waar ze in hun dagelijks leven tegenaan lopen. CVS of JIA bleek vaak de zelfervaringen van de jongeren te veranderen en het aantal zelfposities in hun persoonlijk positie repertoire te verminderen, hetgeen kan leiden tot een meer rigide en monologisch zelfverhaal. Deze zelfverhalen werden beleefd vanuit een meer enkelzijdig perspectief waardoor het moeilijker leek te worden om nieuwe ervaringen in te passen. Psychologische begeleiding zal wellicht effectief zijn, niet door direct in te grijpen op negatieve of verstoorde cognitieve gedachtes, maar door de relatie van de patiënt met dysfunctionele gedachtes en gevoelens te veranderen door er een nieuwe persoonlijke betekenis aan te koppelen. In onze studies bleek een essentieel ingrediënt voor een positieve uitkomst van de begeleiding te zijn dat de jongere zelf onderkende een negatieve invloed van de ziekte te ervaren op zijn kwaliteit van leven. Vragenlijsten naar gezondheidgerelateerde kwaliteit van leven bleken een goed instrument te zijn om die adolescenten te identificeren die ontvankelijk zouden kunnen zijn voor psychische behandeling. Vrijwel alle adolescenten met CVS hadden baat bij narratieve begeleiding, waarin onderdrukte kanten van het zelfverhaal naar voren kwamen en de flexibiliteit in het zelfverhaal terug kon worden gebracht. Adolescenten met JIA waren meer verdeeld. Binnen de totale groep adolescenten met JIA lijken drie verschillende types naar voren te zijn gekomen: a) het eerste type is de adolescent die de artritiservaring op een adequate manier in zijn of haar levensverhaal heeft geïntegreerd, de sterke en de meer zwakke kanten van ervaringen stem geeft en over het algemeen een goede score geeft voor gezondheidgerelateerde kwaliteit van leven, b) het tweede type is de adolescent met JIA die zijn levensverhaal vanuit de zieke-zelfposities vertelt en daarbij niet meer de sterkere kanten van het verhaal kan ervaren, en een lage score geeft voor zijn of haar gezondheidgerelateerde kwaliteit van leven, en c) het laatste type heeft een sterk en gezond monologisch zelfverhaal, met weinig tot geen ruimte voor zwakkere kanten van het zelf en een hoge score voor gezondheidgerelateerde kwaliteit van leven. Het eerste type heeft geen begeleiding nodig aangezien deze adolescenten al beschikken over een flexibel en dialogisch zelf. De adolescenten die bij het tweede type horen, hebben veel overeenkomsten met de adolescenten met CVS en zij hadden baat bij de narratieve begeleiding door flexibiliteit in hun zelfverhaal te hervinden. Adolescenten met JIA van het laatste type vertelden hun zelfverhaal vanuit met name sterke en gezonde zelfposities en gaven een overeenkomstig positieve score voor hun kwaliteit van leven, alleen kwamen de klinische observaties niet overeen met het beeld dat de jongeren van zichzelf creëerden. Tijdens psychische begeleiding echter, hielden deze jongeren vast aan hun 'positieve copingstijl' en was er geen ruimte voor verandering. Wij zouden willen adviseren om bij deze jongeren te wachten met het aanbieden van psychische begeleiding tot de adolescent zelf een realistischer beeld van zijn of haar situatie heeft, hetgeen aan het licht zal komen in de klinische praktijk of doordat de jongere een verminderde kwaliteit van leven rapporteert. Een andere mogelijkheid zou kunnen zijn om de sterke zelfposities te ondersteunen door deze jongeren situaties te laten ervaren waarin ze zich werkelijk succesvol en autonoom voelen. Zodoende kan er ruimte geschept worden om ook negatieve emoties toe te laten. In lijn hiermee worden er momenteel in het Universitair Medisch Centrum Utrecht preventieve groepsprogramma's ('Skills Lab') aangeboden aan jongeren met een chronische ziekte om zelfmanagement en fysieke bewustwording te bevorderen, en positieve ervaringen op te doen.

Toekomst

Deze exploratieve studies naar de zelfervaring van adolescenten met een chronische aandoening geven nieuwe, veelbelovende resultaten die hopelijk leiden tot meer onderzoek naar dit onderwerp. Ons inzicht in de invloed van chronische ziekte op het leven van de adolescent zal nog verder ontwikkeld kunnen worden door verder in te gaan op de volgende overdenkingen.

De adolescenten met CVS in onze studie hadden een kortere ziekteduur in vergelijking met de ziekteduur van de jongeren met JIA. Het is mogelijk dat de adolescenten met CVS meer baat hebben gehad bij de begeleiding omdat er in het begin van het ziekteproces nog meer veranderingsmogelijkheden zijn op emotioneel, gedragsmatig en cognitief vlak omdat copingstijlen nog niet geheel rigide zijn. Verder kan de leeftijd ten tijde van de diagnose ook een belangrijke factor zijn voor hoe de ziekte geïntegreerd wordt in het levensverhaal. In de jongere jaren, voor de adolescentie, is het zelfgevoel nog niet geheel coherent en het zelfverhaal onsamenhangend. Bij een diagnose op kinderleeftijd, zal de ziekte-ervaring wellicht op een andere manier geïntegreerd worden in het zelfverhaal dan wanneer de diagnose gesteld wordt in de adolescentie, als het zelfverhaal meestal al verder ontwikkeld is. Bovendien zal het krijgen van een diagnose van een chronische ziekte tijdens de adolescentiefase, een toch al roerige tijd, van invloed kunnen zijn op hoe de ervaring wordt geïntegreerd in het doorlopende zelfverhaal. Een verdere exploratie van de plek van de ziekte in het zelfverhaal in verschillende ontwikkelingsfasen is belangrijk om goed aan te kunnen sluiten bij de ervaring van deze jongeren.

Adolescenten met JIA lijken redelijk goed te kunnen omgaan met de gevolgen van de pijnlijke en mogelijk invaliderende ziekte, aangezien er ogenschijnlijk weinig verschillen bestaan tussen adolescenten met JIA en gezonde leeftijdsgenoten. Dit beeld van het psychosociaal functioneren van adolescenten met JIA wordt ook terug gevonden in de literatuur over dit onderwerp. Maar jong-volwassenen met artritis rapporteren dat JIA wel een negatieve invloed had op onder andere werkgelegenheid, lichaamsbeeld, sociale contacten, stemming en kwaliteit van leven in het algemeen. Wellicht is de sterke en gezonde presentatie van de adolescenten met JIA te belastend om vol te houden in de volwassenheid. Het lijkt van belang om de langetermijngevolgen te bestuderen van de integratie van de artritiservaring in het levensverhaal.

Implicaties voor de praktijk

- Professionals in de gezondheidszorg moeten verder dan de symptomen kijken en de persoonlijke ervaring van de adolescent proberen te integreren in de behandeling.
- Korte gezondheidgerelateerde kwaliteit-van-leven-vragenlijsten die inzicht geven in fysiek, psychisch en sociaal functioneren kunnen als een algemeen screeningsinstrument gebruikt worden om adolescenten met een verminderde gezondheidgerelateerde kwaliteit van leven te signaleren, aangezien deze groep mogelijk baat heeft bij een psychologische begeleiding.
- Als adolescenten met een chronische ziekte een verminderde gezondheidgerelateerde kwaliteit van leven ervaren, kan het herwinnen van flexibiliteit in hun levensverhaal het fysiek, sociaal en emotioneel welbevinden verbeteren.
- Vanuit klinisch oogpunt blijkt de ZKM a) een waardevolle methode te zijn voor adolescenten die huiverig zijn voor psychologische begeleiding omdat de focus op het gehele levensverhaal ligt en niet specifiek op de problemen, b) een methode te zijn waarin de adolescent zelf een leidende rol heeft in het vinden van de narratieve thema's, hetgeen past bij de ontwikkelingstaak van het krijgen van meer autonomie en zelfstandigheid, en c) een start te kunnen zijn voor verandering als de adolescent een realistische blik heeft op de gevolgen van de chronische ziekte in zijn of haar leven.

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'The universe is made of stories, not of atoms.'
Rukeyser (1968)

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Coralie Fuchs
Haarlem, december 2012

About the author

CURRICULUM VITAE

Coralie Elisabeth Fuchs is op 6 mei 1975 geboren in Heerlen als middelste van drie dochters. Toen ze 8 jaar oud was verhuisde ze met haar gezin naar Parijs waar ze tot haar 13^{e} naar het Lycée International ging voor tweetalig onderwijs (Frans/Nederlands). Ze behaalde in 1994 haar VWO-diploma aan het Sint Maartenscollege in Maastricht en ging een jaar naar Siena (Italië) om Italiaanse taal en cultuur te studeren aan de Università per Stranieri. Ze is daarna Orthopedagogiek aan de Universiteit Utrecht gaan studeren.

Na haar studie (2001) werkte ze in de crisishulpverlening met multi-problem-gezinnen bij Sac-Amstelstad Jeugdzorg (nu Spirit genaamd) in Amsterdam Zuidoost.

In 2004 startte ze in het Wilhelmina Kinderziekenhuis (WKZ) van het Universitair Medisch Centrum Utrecht (UMCU) met het onderzoek dat tot dit proefschrift heeft geleid. Dit deed zij onder begeleiding van prof.dr. W. Kuis, prof.dr. G. Sinnema en em.prof.dr. H.J.M. Hermans.

Sinds 2009 is ze werkzaam op de afdeling Medische Psychologie en Maatschappelijk Werk van het Wilhelmina Kinderziekenhuis. In 2012 behaalde ze haar registratie als gezondheidszorgpsycholoog.

Coralie is getrouwd met Pim Kist en samen hebben ze drie kinderen, Bente (2009), Tabe (2010) en Jet (2012).

LIST OF PUBLICATIONS AND SUBMITTED MANUSCRIPTS

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Submitted manuscript

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