A longitudinal follow-up study of physical and psychosocial health in young adults with chronic childhood arthritis

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Abstract

Objective

The aim was to describe physical and psychosocial health status in a second follow-up of a cohort of patients with chronic childhood arthritis, to compare results from the present study with the first follow-up, and to explore the course of physical and psychosocial functioning from baseline.

Methods

At a median of 18.3 years after symptom onset 55 patients answered the self-administered questionnaires Health Assessment Questionnaire Disability Index (HAQ-DI), Visual Analogue Scales (VAS) of pain, fatigue and illness, and General Health Questionnaire (GHQ) 30-item version. Results from the current study were compared to first follow-up median 8.7 years after symptom onset, and the course of physical and psychosocial function from baseline was discussed.

Results

At second follow-up, 38% reported HAQ-DI above 0 indicating physical disability, 22% had a GHQ-30 score in the clinical range indicating psychiatric distress, and fatigue seemed to be an overarching aspect of the health status. Pain was an important correlate of physical disability at first and second follow-up. At second follow-up psychiatric distress was a significant correlate of pain and fatigue, indicating a relation to disease severity. The association between psychosocial functioning and chronic family difficulties observed at first follow-up is not evident at second follow-up.

Conclusions

The favourable physical and psychosocial outcome reported at first follow-up seems to persist. However, arthritis-related ill-health is still evident in a considerable proportion of the patients, indicating a constant impact of the disease on every-day life of the individual.

Key words

Follow-up study, chronic childhood arthritis, physical health, psychosocial health.

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Introduction

Chronic childhood arthritis is a heterogeneous group of diseases characterised by joint inflammation, stiffness and pain, and about 60% are girls (1, 2). The disease can be defined as arthritis of unknown aetiology that begins before the 16th birthday and persists for at least 6 weeks and where other known conditions are excluded (3). Treatment is aimed at controlling pain, improving range of motion and muscle strength, and facilitating normal growth and development (1).

There is an increased heterogeneity in the clinical picture of childhood arthritis and a geographical and ethnic diversity in the occurrence rates as studies have started to be published from all over the world (4). A review study reported an annual incidence of 10–19.2 per 100 000 children less than 16 years old based on European and American studies with similar criteria and methodology (5). Recent Nordic studies show an annual incidence of 15 and a prevalence of 130 per 100.000 children less than 16 years old (6, 7).

Overall, an estimated 40% of patients with childhood arthritis have active disease in adulthood, although estimates in the different subtypes of the disease vary (8, 9). Continued active disease and disabling residua into adulthood are among the problems faced by a considerable proportion of the patients, although the proportion of patients with severe disability has decreased (10-12).

The increasing number of studies conducted in recent years reflects an increasing interest in outcome of childhood arthritis regarding disease activity and physical function (9, 13, 14), but also regarding psychological and socioeconomic outcomes and health-related quality of life (8, 11, 12, 15-18). However, disease duration and age at follow-up differ as well as study design and methods. Moreover, so far, studies of outcomes in adulthood report controversial results with respect to quality of life and psychosocial functions as comparable or not to those in controls (8, 15). Thus, further studies in this field are required.

The present study is a second follow-up of a Norwegian cohort of patients with childhood arthritis first admitted in 1985-86 to the Center for Rheumatic diseases at the National Hospital of Norway which has the only paediatric rheumatology clinic in Norway and serves the whole country (4.5 million inhabitants). At first admission to hospital, high rates of physical and psychosocial distress were reported (19, 20). At first follow-up Aasland and Flatö (21, 7) found that the outcome was favourable for the majority of the patients investigated as 60% were in remission, 60% had no physical disability and 75% had not developed joint erosions. Mental health and psychosocial functioning were comparable with that of the normal population. Unfavourable psychosocial outcome was predicted by chronic family difficulties, but was not closely related to disease severity.

The purpose of the present study was to describe physical and psychosocial health status in a cohort of patients with childhood arthritis, to compare results from the present study with results from the first follow-up, and to explore variations and changes in physical and psychosocial functioning during the disease course. The time span investigated include childhood, adolescence and early adulthood in one cohort and may illuminate the disease process and impact on physical and psychosocial functioning over time.

Disease classification

In the first follow-up study (7, 23) all the patients were re-assessed with the diagnoses of juvenile rheumatoid arthritis (JRA) including pauciarticular and polyarticular arthritis, and juvenile spondyloarthropathy (JSpA) including syndrome of seronegative enthesopathy and arthropathy (SEA), juvenile ankylosing spondylitis (JAS), arthritis in inflammatory bowel disease (IBD), and juvenile psoriatic arthritis (JPsA) (24). Due to comparisons in this study we prefer to keep the terms of disease subtypes (JRA and JSpA) utilised in the first follow-up study.

Patients and methods

Design

The present study has a longitudinal perspective of a representative Norwegian

cohort of patients with chronic childhood arthritis investigated three times since first admission: baseline (1985/-86) was the first-time-hospitalisation with suspected or definite rheumatic disease (20), first follow-up (1994-95) was at a mean (SD) of 8.5 (0.5) years after first admission (21), and second follow-up (2004) at a mean (SD) of 18.2 (0.6) years after first admission (the present study).

Patients

The study sample includes 55 young adults of in all 84 children with arthritis first admitted from September 1985 to September 1986 (20). In a first follow-up study median (min-max) 8.7 (7.2–19.9) years after symptom onset, 72 (85.7%) patients were re-assessed. Twelve were not followed as one had left the country and 11 chose not to participate. These 12 were comparable to the participants with regard to sex, age, disease duration, number of active joints, and physician's global assessment (24). In the present study, median (min-max) 18.3 (17.0–28.9) years after symptom onset, 55 (65.5%) patients responded to postal questionnaires on self-rated health. One had died, four were lost to follow-up, and 12 refused to participate. These 17 were comparable to the participants with regard to sex, age, disease duration, number of active joints, and physician's global assessment at first follow-up. However, a significantly higher proportion was in the JSpA disease subtype (chi-square value=8.077, p=0.007). The inclusion criteria for participating in the second follow-up study was belonging to the first follow-up cohort (24). Characteristics of responders at second follow-up are outlined in Table I.

Table I. Characteristics of responders at second follow-up 18 years after symptom-onset.

	Responders at second follow-up
Total	n=55
Age, years, median (min-max)	28 (19-35)
Years from symptom onset, median (min-max)	18.3 (17.0-28.9)
Female, no. (%)	33 (60)
Male, no. (%)	22 (40)
JRA[a] no. (%)	45 (81.8)
Female	27
Male	18
JSpA[a] no.(%)	10 (18.2)
Female	6
Male	4

[a]As assessed at first follow-up (24). JRA: Juvenile Rheumatoid Arthritis including pauciarticular and polyarticular arthritis; JSpA: Juvenile Spondyloarthropathy including syndrome of seronegative enthesopathy and arthropathy (SEA), juvenile ankylosing spondylitis (JAS), arthritis in inflammatory bowel disease (IBD), and juvenile psoriatic arthritis (JPsA).

Assessment of physical functioning and health status at second follow-up

In the second follow-up the postal questionnaires comprised the following instruments (Table II). The Health Assessment Questionnaire (HAQ) was used to measure physical functioning (25). This 20-item questionnaire measures the patient's level of functional ability in eight areas over the past week: dressing and grooming, arising, eating, walking, hygiene, reach, grip, and common daily activities. For each item, there is a 4-level difficulty scale that is scored from 0 to 3: no difficulty (0), some difficulty (1), much difficulty (2), and unable to do (3). Dependence on equipment or physical assistance raises a lower score to level 2 to more accurately represent underlying disability. The eight category scores are averaged into an overall HAQ disability index (HAQ-DI) score on a scale from 0 (no disability) to 3 (completely disabled). Generally, scores of 0 to 1 are considered to represent mild to moderate difficulty, 1 to 2 moderate to severe disability, and 2 to 3 severe to very severe disability (26). However, in the present study the cut-off point for the HAQ-DI was set at 0, dividing the patients into those with no disability (HAQ-DI=0) and those with disability (HAQ-DI>0), making possible a comparison with the first follow-up (24). The validity and reliability of the questionnaire is proved acceptable in numerous translations and cultural adaptations (26-30).

The Visual Analogue Scale (VAS) (31) was used to assess pain, fatigue and illness. The scale consists of a 100 mm doubly-anchored, horizontal line, rated from left ("no pain", "no fatigue" and "no illness" respectively) to right ("the worst pain", "severe fatigue" and "severe illness" respectively). The patient rates his/her assessment of the particular phenomenon over the past week on the scale. The score is the number of millimetres between the patient's rate and the left anchor of the scale. VAS score >10 defines illness, pain and fatigue respectively (32). The VAS has been widely used in clinical everyday and in different research settings (7, 23, 29, 30).

The General Health Questionnaire (GHQ-30) (33) was used to assess psychosocial health. The instrument includes 30 items covering symptoms considered to reflect distress and psychopathology in five factor-analysed dimensions corresponding to anxiety, feelings of incompetence, depression, difficulty in coping, and social dysfunction over the past 2 weeks (34). GHQ-30 includes both positively- and negatively-designed questions. As a screening test the responses to the items in the questionnaire are weighted as a Likert scale (0-1-2-3) with a possible score of 0-90. A high score indicates more psychosocial distress (33). To detect cases of psychiatric distress the responses are weighted binominally as GHQ-30 case scoring with weights (0-0-1-1) with a possible score of 0-30. A cut-off point of 5+ indicates psychiatric distress in the clinical range (33). Reliability and validity are acceptable in a variety of studies (33). The Norwegian translation of GHQ-30 has been used in several studies (20, 35, 36).

Assessment of physical functioning and health status at first follow-up

Measures at first follow-up relevant for comparisons at second follow-up (Table II) comprised HAQ for patients 18 years and older, HAQ adapted for children (CHAQ) (37), with scoring similar to the HAQ-DI, completed by the 16-17-year-old patients and by a parent of patients below 16 years old, VAS pain and VAS fatigue, completed by patient and parent, and physician's global assessment of disease activity (PGA). To measure psychiatric dis-

tress, the following questionnaires were employed; Child Behaviour Checklist (CBCL) (38) completed by parents of patients <18 years old, Youth Self-Report (YSR) (39) completed by patients 12-17 years old, and Hopkins Symptom Checklist (HSCL) 25-item version (40) completed by patients 18 years and older. Additionally, psychiatric diagnosis, a global assessment of functioning (GAS/CGAS) (41, 42) and a global score of chronic family difficulties (CFD) (43) were assessed based on interviews of all the patients as well as the parents of patients <18 years old. The interview provided diagnoses according to diagnostic criteria in the Diagnostic and Statistical Manual of Mental Disorders, third revised edition (DSM-3-R) (44). The variables are described in detail (21) (24).

The questionnaires, CBCL, YSR, HSCL-25, and GHQ-30, are all measuring general emotional state, CBCL and YSR are constructed for patients <18 years old (38, 39), and HSCL-25 and GHQ-30 for adults (33, 40). The GHQ-30 is considered more suitable and valid in samples of patients with physical illness as physical symptoms as indicators of distress are avoided (33). Therefore, GHQ-30 was preferred in the second follow-up. For all these tests there have been estimated case cut-off scores indicating psychiatric distress in the clinical range (33, 38, 39, 45). Thus, to make comparisons of psychiatric distress possible we used these cut-off scores.

Statistical analyses

Kolmogorov-Smirnov Tests of normality were used to test the distribution of data overall and group-wise for diagnose subtype and sex. For non-normal distributed variables, central tendencies are given in medians (min-max). Otherwise descriptive data were calculated as proportions and means (SD). Differences between two unrelated groups were tested by Mann-Whitney Tests for non-normal distributed variables, by the Chi-square Tests for categorical variables, and by an independent sample t-test for the means. Differences within two related groups were tested by Wilcoxon Signed-Rank Tests for non-normal distributed variables, and by McNemar Tests for categorical variables. Correlations of non-parametric variables were expressed as Spearman rank coefficient correlations. In all the analyses, limit values for significance were set at $p<0.05$. The Statistical Package SPSS version 16 was used for all calculations (46).

Ethical considerations

The study was approved by the Norwegian Data Inspectorate and the Regional Committee for Medical Research Ethics in Norway, and performed in accordance with the Helsinki declaration (47). The participants were informed that participation was voluntary, that they could withdraw whenever they wished and have their contribution deleted, and that all material is treated confidentially. Written information was given together with the postal questionnaires. Thus, returned filled-in questionnaires were accepted as informed consent.

Table II. Assessment measures utilised at first (1994/-95) and second (2004) follow-up used for comparisons at second follow-up.

	Measures at first follow-up		Measures at second follow-up
Age/number	<18 years n=31	≥18 years n=41	>18 years n=55
Physical variables			
Disease activity	PGA	PGA	–
Physical disability	CHAQ	HAQ	HAQ
Pain	100 mm VAS	100 mm VAS	100 mm VAS
Fatigue	100 mm VAS	100 mm VAS	100 mm VAS
Illness	–	–	100 mm VAS
Mental and psychosocial variables			
Mental health	CBCL/YSR	HSCL-25	GHQ-30
	DSM-III-R	DSM-III-R	–
	CGAS	GAS	–
Family variables			
Chronic family difficulties	Parent interview	Patient interview	–

PGA: Physician's global assessment of disease activity; CHAQ/HAQ: Childhood or Adult Health Assessment Questionnaire; VAS: Visual Analogue Scale; CBCL: Child Behaviour Checklist; YSR: Youth Self Report; HSCL: Hopkins Symptom Checklist; DSM-III-R: the *Diagnostic and Statistical Manual of Mental Disorders*, 3rd ed., revised; CGAS/GAS: Children's or Adults' Global Assessment Scale; GHQ-30: General Health Questionnaire 30-item version.

Table III. Outcome variables for physical functioning, illness, pain, fatigue and psychosocial health at second follow-up, and comparisons of physical functioning, pain and fatigue from first to second follow-up (n=55).

	First follow-up	Second follow-up	*p*-value
CHAQ/HAQ-DI[a] median (range)	0.00 (0-3)	0.00 (0-2.5)	0.76[e]
CHAQ/HAQ no. (%)			
No disability: Score =0	32 (58.2)	34 (61.8)	0.008[f]
Disability: Score >0	23 (41.8)	21 (38.2)	0.031[f]
VAS 100 mm[b] median (min-max)			
Pain	6,5 (0-85)	6 (0-72)	0.64[e]
Fatigue	13.5 (0-97)	14.5 (0-90)	0.52[e]
Illness	–	4 (0-74)	–
GHQ-30 Likert scale 0-90[c] mean (SD)	–	24.2 (10.8)	–
GHQ-30 Case score 0-30[c] mean (SD)	–	3.1 (5.1)	–
GHQ-30 Case score 5+[d] no. (%)	–	12 (21.8)	–

[a]CHAQ/HAQ Disability Index: 0 = no disability; 3 = completely disabled; [b]VAS: 0 = no pain, no fatigue, no illness respectively, 100 = the worst pain, severe fatigue, severe illness respectively; [c]GHQ-30 Likert/Case score: 0 = the best psychosocial health, 90/30 = severe psychological distress; [d]GHQ-30 Case score 5+ = psychiatric distress; [e]Wilcoxon Signed-Rank Tests; [f]McNemar Tests.

Table IV. Spearman's correlations of psychosocial health, physical functioning, pain, fatigue, and illness at second follow-up, n=55.

	VAS pain score	VAS fatigue score
GHQ-30 Likert score[a] correlation coefficient	0.282 (p=0.037)	0.416 (p=0.002)
HAQ-DI score[b] correlation coefficient	0.537 (p=0.000)	0.291 (p=0.033)
VAS pain score[c] correlation coefficient	–	0.426 (p=0.001)
VAS fatigue score[c] correlation coefficient	0.426 (p=0.001)	–
VAS illness score[c] correlation coefficient	0.761 (p=0.000)	0.367 (p=0.006)

[a]GHQ-30 Likert scale: 0 = the best psychosocial health, 90 = severe psychological distress; [b]HAQ DI: 0 = no disability, 3 = completely disabled; [c]VAS: 0 = no pain, no fatigue, no illness respectively, 100 = the worst pain, severe fatigue, severe illness respectively.

Results

Of the 55 respondents, six (<27 years, 11%) still lived with their parents, 33 (60%) lived with partner. Thirteen females (24%) had children, compared to three males (6%).

Results from the questionnaires at second follow-up

Outcome variables at second follow-up are presented in Table III. Thirty-four patients (62%) scored =0 on the HAQ-DI indicating no physical disability and 21 (38%) scored >0 indicating physical disability from mild/moderate to severe/very severe. Twelve patients (22%) scored within the clinical range of GHQ-30 case score indicating psychiatric distress, and four of these patients also scored >0 on HAQ-DI.

Twenty-six patients (47%) scored 10 or more on VAS illness scale, and on VAS pain and VAS fatigue scales respectively, 27 (49%) and 33 (60%) patients scored 10 or more, indicating experienced ill-health physically and emotionally to a certain extent.

VAS pain correlated significantly with psychiatric distress by GHQ-30 Likert score, with physical disability by HAQ-DI, with VAS fatigue, and VAS illness scores respectively. Further, VAS fatigue correlated significantly with psychiatric distress, physical disability, and illness scores respectively (Table IV). Moreover, physical disability by HAQ-DI correlated significantly with VAS illness score, r_s .438 (p=.000).

Comparison of variables from first to second follow-up

There were no significant differences from first to second follow-up in physical disability score by HAQ-DI, or in VAS pain and VAS fatigue scores, indicating no significant change in physical function, pain or fatigue during the time span (Table III). Individually, eight patients (14.5%, five females) improved from HAQ-DI >0 to =0 from first to second follow-up while six others (11%, four females) deteriorated from HAQ-DI =0 to >0. Twenty-six patients (47%) remained with HAQ-DI =0, while 15 patients (27%) remained with HAQ-DI >0 from first to second follow-up. VAS fatigue score tended to be higher at both times compared to VAS pain score (Table III).

No significant differences were found between disease subtypes or sex from first to second follow-up. However, both the JSpA and the JRA groups showed a tendency of decrease in HAQ-DI score, although they showed a tendency of increased VAS pain and VAS fatigue scores. The JSpA group tended to score higher than the JRA group on VAS pain and VAS fatigue both times. Males showed a tendency of increase in HAQ-DI score, and a stronger tendency of increase in VAS pain and VAS fatigue scores from first to second follow-up, in contrast to the tendency of decrease in the same scores in females (Table V).

Self-rated psychiatric distress by GHQ-30 case cut-off score at second follow-up was compared to self-rated psychiatric distress by CBCL, YSR and HSCL-25 cut-off scores at first follow-up. One child patient had a cut-off score in the clinical range (>63) of CBCL and one youth patient had a cut-off score in the clinical range (>63) of YSR. None of the adult patients at first follow-up had a HSCL-25 cut-

Table V. Outcome variables of physical functioning, pain and fatigue at first (t-1) and second (t-2) follow-up by all responders and by disease subtype and sex[a].

	No. of patients	CHAQ/HAQ-DI[b] Median (min-max)		VAS pain[c] Median (min-max)		VAS fatigue[c] Median (min-max)	
		t-1	t-2	t-1	t-2	t-1	t-2
All responders	55	0.00 (0-3)	0.00 (0-2.5)	6.5 (0-85)	6.0 (0-72)	13.5 (0-97)	14.5 (0-90)
Disease subtype[d]							
JRA	45	0.00 (0–3)	0.00 (0–2.5)	4.5 (0–85)	5.0 (0–72)	9.5 (0–97)	12.0 (0–90)
JSpA	10	0.125 (0–1.125)	0.00 (0–0.875)	19.0 (0–56)	21.0 (0–49)	39.5 (0–71)	41.0 (0–73)
Sex:							
Female	33	0.00 (0–3)	0.00 (0–2.5)	10.0 (0–71)	6.0 (0–67)	17.0 (0–97)	12.0 (0–87)
Male	22	0.00 (0–0.875)	0.00 (0–1.75)	3.0 (0–85)	12.5 (0–72)	11.0 (0–76)	22.5 (0–90)

[a]Wilcoxon Signed-Rank Tests; [b]CHAQ/HAQ-DI: 0 = no disability, 3 = completely disabled; [c]VAS: 0 = no pain, no fatigue respectively, 100 = the worst pain, severe fatigue respectively; [d]As assessed at first follow-up (24). JRA: Juvenile Rheumatoid Arthritis including pauciarticular and polyarticular arthritis; JSpA: Juvenile Spondyloarthropathy including syndrome of seronegative enthesopathy and arthropathy (SEA), juvenile ankylosing spondylitis (JAS), arthritis in inflammatory bowel disease (IBD), and juvenile psoriatic arthritis (JPsA).

off score in the clinical range (>1.75). However, by interviews at first follow-up, 10 other patients were diagnosed with a psychiatric diagnosis according to diagnostic criteria in DSM-3-R (44). Of the 12 patients at second follow-up who had a GHQ-30 case cut-off score in the clinical range, none scored within the clinical range of psychiatric distress at first follow-up. However, of the 10 patients at first follow-up who had a psychiatric diagnosis, four scored within the clinical range of psychiatric distress by GHQ-30 case cut-off score at second follow-up.

Psychiatric distress at second follow-up (total GHQ-30 Likert score) was tested against psychiatric distress (total t-scores of CBCL and YSR, and total HSCL-25 score) at first follow-up. Furthermore, psychiatric distress at second follow-up (GHQ-30 Likert and case scores) was tested against physical and psychosocial functioning at first follow-up (PGA, HAQ, CGAS/GAS) and, finally, against number of chronic family difficulties at baseline and first follow-up respectively. No significant correlations were found.

Discussion

Generally, the favourable improvement in physical and psychosocial functioning described at first follow-up seems to persist at second follow-up of this cohort of 55 patients 18 years after baseline. This seems to be in line with Oen (8) and Fantini (9), who found that the rate of remission reached its peak 5-10 years after disease onset, after which the trend reversed. Also at second follow-up a considerable proportion of the patients reported ill-health to a certain extent. Nearly half the patients reported illness and pain above 10 on the VAS scale, nearly two third reported fatigue above the same limit, and 38% reported physical disability by HAQ-DI to various extent. Furthermore, 22% reported psychiatric distress correlating significantly with pain and fatigue. These findings indicate that arthritis-related ill-health constantly has an impact on every-day life, physically and psychosocially, for a considerable proportion of adult patients. This is in line with recent studies reporting active disease in adulthood in an average of 40% of the patients, with a considerable impact on general health status, although only a few patients were severely disabled (8, 9).

Through the time span investigated there were no significant changes in average level of reported physical disability, pain or fatigue. However, individually there was a significant change in physical disability from first to second follow-up as six patients deteriorated, while eight others improved. This might be associated with disease fluctuations and the impact of residua on physical functioning (48). It reminds us of the unpredictable nature of the rheumatic diseases, implying constant uncertainty about the future. This is supported by results reported from the qualitative interview study on a smaller sample of the current cohort (49) and may influence the decisions facing young adults in this important period of life (50).

Males showed a tendency of deterioration in the physical domain in contrast to the tendency of improvement among females from first to second follow-up. This male tendency may represent a true deterioration in disease status, or may represent a stronger tendency in males to deny disease-related problems during adolescence, while they are not so reluctant to report problems as young adults (51). Although the JRA and JSpA groups showed a tendency of improvement in physical disability from first to second follow-up, both groups tended to report increased pain and fatigue. However, the JSpA group reported less physical disability and more pain and fatigue both times compared to the JRA group. Although the proportion of JSpA among the responders at second follow-up was smaller compared to the non-responders, it may seem as the different rheumatic disease subtypes have a slightly different symptom impact.

Fatigue seemed to be prominent at both first and second follow-up and was a significant correlate of pain, psychiatric distress, illness, and physical disability respectively at second follow-up; hence fatigue seems to be an overarching aspect of the health status. This is in line with fatigue as a common symptom related to physical and psychiatric conditions and longstanding experience of pain, such as fibromyalgia (52). The unpredictability of the disease course and hence uncertainty related to the future may also contribute to the experience of fatigue (49). It may also be that some patients have a tendency to experience and express fatigue rather than more specific feelings of anxiety and depression.

In the psychosocial domain there seems to be a change from first to second follow-up. Although psychiatric distress was not closely related to disease severity at first follow-up (21), it seems to be an association at second follow-up as psychiatric distress was a significant correlate of pain and fatigue. This may reflect the longstanding chronicity of the disease and its impact on the entire adult life course. At first follow-up, the adolescents reported increased closeness to parents and a tendency to being overprotected (53). In childhood and adolescence, increased closeness may have protected the patients from experiencing psychiatric distress. However, overprotection may have put the patients' achievement of independence at risk and thus have a negative impact on emotional well-being in adult life (53). Thus, the disease seems to be one of several distress factors contributing to psychiatric distress in adulthood, which is in line with results from the qualitative interview study mentioned above (49).

The association between psychosocial functioning and chronic family difficulties observed at first follow-up (21) is not evident at second follow-up. Rather, at second follow-up the scores indicate an improvement from psychiatric distress associated with chronic family difficulties. At second follow-up the patients have established independent lives and are not so closely related to their origin family with its strengths and problems.

The major advantages of this study are the longitudinal perspective of a representative cohort and the inclusion of both physical and psychosocial measures at baseline and at first and second follow-up. The respondent rate was satisfactory. Yet, certain limitations must be considered when drawing conclu-

sions. First, the age change through the follow-up period influenced the choice of assessment methods employed on the different occasions; only at second follow-up all the questionnaires were in adult versions and answered without parents' or observers' influence. Second, at second follow-up only postal self-administered questionnaires were used, and disease activity was not confirmed due to no physical examination by the physician. These facts made test comparisons between first admission and second follow-up less valid. Therefore, these results are only discussed in the text.

In summary, the favourable physical and psychosocial outcome reported at first follow-up seems to persist; yet arthritis-related ill-health in adulthood is still evident. The acknowledgement of the uncertainty involved in the unpredictability of the disease course should have implications for clinical practice. Furthermore, individual changes are particularly important to keep in mind.

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