

Adolescents with juvenile idiopathic arthritis: who cares after the age of 16?

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Abstract

Objectives

Medical follow-up in the most appropriate treatment setting is important for patients with juvenile idiopathic arthritis (JIA). The aims of this study were 1) to identify the settings in which JIA patients are followed up after leaving paediatric rheumatology, and 2) to compare the clinical profile of patients in different settings.

Methods

The Short Form-36, Health Assessment Questionnaire, and linear analogue scale for quality of life were sent to JIA patients older than 16 years, who had been followed in one academic paediatric rheumatology centre from 1994 to 2007 and who did not participate in a structured transitional care program. Forty-four patients participated in this cross-sectional, comparative study.

Results

Thirteen patients were no longer in medical follow-up, 6 patients were followed by their general practitioner, and 25 patients were followed by a rheumatologist. All patients treated with glucocorticosteroids, DMARDs and anti-TNF were followed by a rheumatologist. Patients under the care of a rheumatologist had worse physical functioning ($U=33.5$, $p<0.001$); greater disability ($U=49.0$, $p=0.001$); more pain ($U=59.0$, $p=0.002$); and lower quality of life ($U=69$, $p=0.02$) than patients not in follow-up. Of the patients no longer in follow-up, 2 (16.7%) had disabilities and 5 (41.7%) reported persistent pain.

Conclusion

The present data indicated that JIA patients with persistent disease and associated functional disabilities tend to remain in the rheumatology circuit. However, the disease of patients leaving specialised rheumatology care is not necessarily controlled. These data may be helpful for organising the proper transfer of patients from paediatric to adult-focused care.

Key words

Arthritis, juvenile rheumatoid, adults, patient care management, nursing.

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Introduction

Juvenile idiopathic arthritis (JIA) is the most common chronic rheumatic disease in children and is an important cause of short-term and long-term disability. Different studies in the past decade have shown that only 40–60% of patients experience inactive disease or clinical remission at follow-up (1, 2).

The clinical subtypes of JIA are defined by clinical characteristics within the first six months of disease onset. The three major subgroups are oligoarticular JIA (oligo-JIA), polyarticular JIA (poly-JIA), and systemic JIA (sys-JIA). Ten years after onset, 22–41% of patients with oligo-JIA, 45–50% of patients with poly-JIA, and 27–48% of patients with sys-JIA have persistent arthritis (3). Overall, 30–60% of JIA patients are in remission as they enter adulthood.

When patients with JIA reach adulthood, follow-up at paediatric rheumatology care is no longer possible. Nonetheless, this important group of patients requires further specialist follow-up to guarantee optimal treatment; adjustment of disease-modifying antirheumatic drugs (DMARDs) and anti-TNF therapy, if necessary; regular assessment of disease activity, pain, and function; and care modification, if needed (4). Also specific aspects, such as dealing with functional limitations in their professional life and social relationships may need to be addressed. The most appropriate follow-up (general practitioner vs. rheumatologist) depends also on the JIA subtype and the severity and duration of the disease. Patients can be discharged from paediatric rheumatology without needing any further medical follow-up.

Hence, medical follow-up in the most appropriate treatment setting for JIA is of relevance. Therefore, the aims of this study were 1) to identify the settings in which patients with JIA are followed up after leaving paediatric rheumatology, and 2) to compare the clinical profile of patients in each setting.

Methods

Study population

We conducted a cross-sectional, comparative study. Patients were recruited

from the database of paediatric rheumatology (1994–2007) of the University Hospitals Leuven, Belgium. Inclusion criteria were as follows: a) born in 1992 or before, b) consulted the paediatric rheumatologist at the University Hospitals Leuven at least two times, and c) no longer in follow-up with a paediatric rheumatologist. Patients followed up by the paediatric rheumatologist for disorders other than JIA were excluded. Overall, 98 patients met the inclusion criteria and were therefore eligible.

Of the 98 patients invited to participate in the study, 44 patients completed the questionnaires, yielding a response rate of 44%. Fifty-four questionnaires (56%) were either not returned or returned but filled out incompletely. The non-responders and responders did not differ in age ($U=1050$, $p=0.70$); gender ($\chi^2=3.05$, $p=0.08$); or subtype of JIA ($\chi^2=4.6$, $p=0.10$).

Variables and measures

– *Demographic and clinical variables* included gender; age; disorder subtype (oligo-JIA, poly-JIA, and sys-JIA); and drug therapy. These data were collected from the medical records. By means of a self-report questionnaire, patients were asked what type of doctor followed them for their rheumatic disorder, the frequency of follow-up, whether they visited a physiotherapist, and their educational and employment status.

– *General health status* was measured using the Short Form-36 (SF-36) (5). The SF-36 is a generic instrument constructed to measure eight health attributes: physical functioning, role-physical functioning, bodily pain, general health, vitality, social functioning, role-emotional functioning, and mental health. The raw scores for each subscale were translated onto a scale ranging from 0 to 100. A lower score indicates worse health status. The SF-36 has been shown to have good internal consistency, validity, and reliability (6–8). The SF-36 has been used extensively in various patient populations, allowing comparison of the SF scores of patients with a rheumatic disorder with those of healthy individuals. Gender and age-

Competing interests: none declared.

adjusted data on the SF-36 from the general population in the Netherlands are available (9), enabling comparison of our patients with a normative population.

– *Functional status* was measured using the Health Assessment Questionnaire (HAQ), which consists of the HAQ disability index (DI) and the HAQ pain scale (10). The HAQ DI assesses eight categories (dressing, arising, eating, walking, hygiene, reaching, gripping, and outside activity), comprising a total of 20 items, and is measured on a 4-point ordinal scale, from 0 (without any difficulty) to 3 (unable to do). The highest score in each of the 8 categories is averaged to obtain a disability index, which is set on a scale of 0 (no disability) to 3 (complete disability). The HAQ pain scale evaluates the presence or absence of arthritis-related pain and severity using a single-item, 10 cm double-anchored visual analogue scale (VAS), which is scored from 0 (no pain) to 3 (severe pain). By convention, the disability index is expressed on a scale of 0–3 units, representing the mean of the eight domain scores. A HAQ DI of 0 indicates no functional disability, while a disability index of 3 indicates severe functional disability. A healthy individual is expected to have a HAQ DI of 0. While there is no official consensus as to what constitutes mild, moderate, or severe disability, a score of 0 is considered to be no disability, a score of 0 to 1 is mild, a score of >1 to 2 is moderate, and a score of >2 is severe. For the HAQ pain scale, we used the same categories. The HAQ has been shown to have good validity (11, 12) and very good reliability (11).

– *Quality of life* was measured with a linear analogue scale (LAS). The LAS that we used consisted of a vertically oriented, 10 cm line, graded with indicators ranging from 0 (worst imaginable quality of life) to 100 (best imaginable quality of life). Patients were asked to rate their overall quality of life by marking the point on the scale that indicates how good or bad their quality of life is in their opinion. The use of this LAS in different patient populations has shown that it is valid and reliable for assessing quality of life (13).

Procedure

A researcher (F. C.) contacted the patients by mail. The package included an introductory letter detailing the aims of the project, an informed consent form, and the questionnaires. The patients were asked to return the completed instruments, including the signed informed consent form, in a pre-addressed and stamped envelope.

The study protocol was evaluated and approved by the Institutional Review Board of the University Hospitals Leuven and was performed in accordance with ethical standards, as described in the latest Declaration of Helsinki. Patients were only included in our study if they provided written informed consent. If patients were younger than 18 years, informed consent from their parents was also requested.

Statistical analysis

Data were analysed with SPSS statistical software version 15.0 (SPSS Inc., Chicago, IL, USA). Nominal data were expressed in frequencies and percentages. Medians and quartiles (Q1–Q3) were calculated for continuous, non-normally distributed variables. For group comparisons, according to the follow-up setting we used a Bonferroni-adjusted Kruskal-Wallis test for 3-group comparisons. An adjusted Mann-Whitney U-test was used for post hoc analyses between two groups. All the tests were two-sided.

Differences in the SF-36 scores of JIA patients and those of the general population were expressed as mean standardised differences from the norm data. For each patient, the norm score of individuals with the corresponding age and gender was subtracted from the patient's score and divided by the standard deviation from the norm data, generating a standardised difference for that patient. The averaging of this difference over all patients resulted in a mean standardised difference value. Values less than zero indicated that the perceived health of JIA patients is lower than that of the norm group, adjusted for age and gender. To measure the size of the observed differences in the SF-36 scores, we used Cohen's *d*. To appraise the magnitude of the standardised differences in terms of

effect size, we considered a difference of 0.2 to 0.5 to be a small effect, that of 0.5 to 0.8 to be a medium effect, and that of 0.8 or higher to be a large effect (14).

Results

Patient characteristics

In this study, 44 patients with JIA participated; 15 were male and 29 were female. Nineteen patients had oligo-JIA, 16 patients had poly-JIA, and 9 patients had sys-JIA. The age of these patients ranged from 16 to 30 years, with a median age of 20 years. Patient characteristics are presented in Table I.

Current medical follow-up

Of the 44 patients participating in this study, 13 patients (29.6%) were not in regular medical follow-up, 6 patients (13.6%) were in follow-up with their general practitioner, and 25 patients (56.8%) were in follow-up with a rheumatologist (Table II). Of the latter group, 20 patients (80%) were treated in a tertiary care centre and 5 patients (20%) were treated by a local rheumatologist. Of the patients no longer

Table I. Patient characteristics.

| Variables | (n=44) |
|---------------------------------|---|
| Gender: | |
| Male | 15 (34.1%) |
| Female | 29 (65.9%) |
| Median age (years) | 20 (Q1=19.0, Q3=22.6) range: 16 - 30 |
| Subtype | |
| Oligoarticular JIA | 19 (43.2%) |
| Polyarticular JIA | 16 (36.4%) |
| Systemic JIA | 9 (20.1%) |
| Marital status (n = 43) | |
| Unmarried | 37 (84.1%) |
| (living with parents) | |
| Living alone | 0 (0%) |
| Married or cohabiting | 6 (15.9%) |
| Employment status | |
| Student | 23 (52.3%) |
| Employed | 16 (36.4%) |
| Unemployed/ looking for work | 4 (9.0%) |
| Not able to work/ disability | 1 (2.3%) |
| Education level: (n=43) | |
| Primary education | 8 (18.5%) |
| Secondary education | 23 (53.4%) |
| Higher education | 7 (16.2%) |
| University | 5 (11.1%) |

Table II. Subtypes and medical follow-up in 44 patients with juvenile idiopathic arthritis (JIA).

| Subtype | n (%) | Medical follow-up | | |
|------------------|-------|-----------------------------|----------------------------|-----------------------|
| | | No medical follow-up (n=13) | General practitioner (n=6) | Rheumatologist (n=25) |
| oligo-JIA (n=19) | | 9 (20.5%) | 3 (6.8%) | 8 (18.2%) |
| poly-JIA (n=16) | | 2 (4.5%) | 2 (4.5%) | 11 (25.0%) |
| sys-JIA (n=9) | | 2 (4.5%) | 1 (2.3%) | 6 (13.7%) |

Oligo-JIA: oligoarticular JIA; poly-JIA: polyarticular JIA; sys-JIA: systemic JIA.

Table III. Medication history, current medication and medical follow-up in 44 patients with juvenile idiopathic arthritis (JIA).

| Medication History | n (%) | Medical follow-up | | |
|----------------------|-------|-----------------------------|----------------------------|-----------------------------|
| | | No medical follow-up (n=13) | General practitioner (n=6) | General practitioner (n=25) |
| no medication | | 1 | 0 | 0 |
| analgesics/NSAID's | | 11 | 5 | 25 |
| glucocorticosteroids | | 6 | 1 | 19 |
| DMARDs | | 3 | 1 | 16 |
| growth hormones | | 1 | 0 | 2 |
| Anti-TNF | | 0 | 0 | 10 |
| Current Medication | | | | |
| no medication | | 9 | 4 | 3 |
| analgesics/NSAID's | | 4 | 2 | 16 |
| glucocorticosteroids | | 0 | 0 | 9 |
| DMARDs | | 0 | 0 | 12 |
| growth hormones | | 0 | 0 | 1 |
| Anti-TNF | | 0 | 0 | 8 |

DMARDs: disease-modifying antirheumatic drugs; Anti-TNF: anti-tumour-necrosis-factor.

in medical follow-up, 9 patients had oligo-JIA, 2 patients had poly-JIA, and 2 patients had sys-JIA. Of the patients being treated by a general practitioner, 3 patients had oligo-JIA, 2 patients had poly-JIA, and 1 patient had sys-JIA. Finally, of the patients being treated by a rheumatologist, 11 patients had poly-JIA, 8 patients had oligo-JIA, and 6 patients had sys-JIA.

Medication intake

Table III gives an overview of the medication history of included patients according to the follow-up setting: 41 (93.2%) patients took analgetics or NSAID's, 26 (59.1%) took glucocorticosteroids, (45.5%) took DMARDs and three (6.8%) took growth hormones. Ten (22.7%) patients were treated with anti-TNF-antagonists. Twenty-eight patients (63.6%) were still on medication, of which 22 (78.6%) took analgesics or NSAIDs, 12 (42.9%) took DMARDs, 9 (32.1%) took glu-

cocorticosteroids, and one (3.6%) received growth hormones therapy. Eight (18.2%) patients are still under anti-TNF therapy. Only patients under the care of a rheumatologist took glucocorticosteroids and/or DMARDs and TNF-antagonists.

General health status

Scores on the eight dimensions of the SF-36 ranged from 57.0 for general health in patients followed by a rheumatologist to 97.9 for role-physical functioning in patients no longer in medical follow-up. As shown in Figure 1, the Kruskal-Wallis test revealed a statistically significant difference only for 'physical functioning' (corrected for multiple testing; 8 pairs, $\alpha=0.00625$) between patients in different follow-up settings. Moreover, post hoc analysis (3 pairs, $\alpha=0.02$) showed that this difference was significant between patients treated by a rheumatologist and patients not in medical follow-up (Fig.

1). No other statistically significant differences in SF-36 dimensions were found between patients in the different follow-up settings.

Figure 1 also shows the mean standardised differences per type of follow-up. Data from patients treated by a rheumatologist showed the largest differences when compared to the norm data. In these patients, large standardised differences were found in the dimensions 'physical functioning' (-3.12), 'general health' (-1.52), 'bodily pain' (-1.31), and 'role-physical functioning' (-0.92). Patients under the care of their general practitioner displayed large differences in physical functioning (-1.21) and bodily pain (-0.79), when compared to normative individuals.

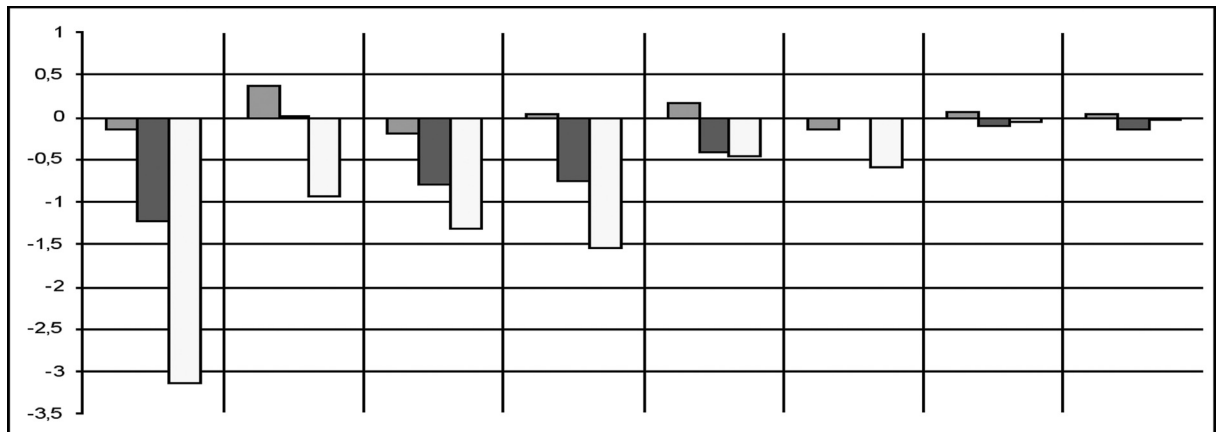
Functional status

HAQ DI scores ranged from 0 to 2 for individual patients, with a median score of 0.125 (Q1=0.0, Q3=0.75). HAQ pain scores (VAS) ranged from 0 to 1.6, with a median score of 0.31 (Q1=0.01, Q3=0.85) on a scale of 0 to 3. Significant differences across the groups were observed both on the HAS DI and the HAQ pain scale. Post hoc analyses revealed that patients in follow-up with a rheumatologist had significantly worse functional status (U=49.0, $p=0.001$) and significantly more pain (U=59.0, $p=0.003$) than patients not in follow-up (Table IV).

Sixteen patients (37.2 %) had a mild disability, while 7 patients (16.3%) had a moderate disability. Of the 23 patients with disabilities, 6 patients were in follow-up with a rheumatologist. No patients had a severe disability (Table V). Eleven (25.6 %) of the 44 patients were pain free. Five patients (11.6 %) not in follow-up perceived mild pain; these patients had oligo-JIA. Six patients (13.9 %) in follow-up with a rheumatologist suffered from moderate pain. No patients experienced severe pain (Table V).

Quality of life

LAS scores for quality of life ranged from 25.0 to 100.0, with a median of 80.0. The median quality of life of patients no longer in follow-up (86.0; Q1= 80.0, Q3= 92.3) was significantly



| | PF $\bar{x} \pm SD$ | RP $\bar{x} \pm SD$ | BP $\bar{x} \pm SD$ | GH $\bar{x} \pm SD$ | VT $\bar{x} \pm SD$ | SF $\bar{x} \pm SD$ | RE $\bar{x} \pm SD$ | MH $\bar{x} \pm SD$ |
|------------------------------------|---|-----------------------------|-----------------------------|-----------------------------|-----------------------------|-----------------------------|-----------------------------|-----------------------------|
| Age and gender adjusted norm value | 93.9 ± 2.4 | 87.8 ± 2.1 | 81.9 ± 4.2 | 77.8 ± 2.0 | 70.2 ± 3.9 | 87.6 ± 1.5 | 83.07 ± 5.3 | 77.6 ± 2.5 |
| No medical follow-up | 93.8 ± 8.0 | 97.9 ± 7.2 | 81.5 ± 20.9 | 80.4 ± 19.0 | 74.6 ± 21.0 | 87.5 ± 31.1 | 88.8 ± 29.6 | 79.3 ± 27.5 |
| General practitioner | 80.0 ± 24.9 | 87.5 ± 30.6 | 65.2 ± 34.5 | 65.0 ± 31.8 | 62.5 ± 27.5 | 87.5 ± 25.0 | 77.8 ± 40.4 | 74.6 ± 17.3 |
| Rheumatologist | 62.6 ± 23.1 | 66.0 ± 42.6 | 57.2 ± 22.6 | 57.0 ± 22.6 | 63.8 ± 0.11 | 79.5 ± 19.7 | 85.3 ± 33.4 | 78.2 ± 14.2 |
| Kruskal-Wallis | $\chi^2 = 15.1$ $p=0.001$ | $\chi^2 = 6.0$ $p=0.048$ | $\chi^2 = 8.2$ $p=0.017$ | $\chi^2 = 9.3$ $p=0.009$ | $\chi^2 = 3.4$ $p=0.184$ | $\chi^2 = 5.1$ $p=0.077$ | $\chi^2 = 0.7$ $p=0.722$ | $\chi^2 = 1.9$ $p=0.391$ |
| Post-hoc analyses | No medical follow-up versus rheumatologist U = 33.5; $p<0.02$ | | | | | | | |

Fig. 1. Differences in health status (SF-36) of the study population: comparison with norm data and data obtained from patients in different follow-up settings.

PF: Physical function; RP: role-physical; BP: bodily pain; GH: general health; VT: vitality; SF: social function; RE: role-emotional; MH: mental health Cohen’s *d* cut-offs: 0.2 to 0.5: small effect; 0.5 to 0.8: medium effect; 0.8 or higher: large effect. χ^2 : Kruskal-Wallis test; U: Mann-Whitney U-test.

Table IV. Disability index and pain score of 44 patients with JIA.

| | No medical follow-up (n=13)* | General practitioner (n=6)* | Rheumatologist (n=25)* | Test value <i>p</i> -value | Post hoc analyses |
|----------------|---------------------------------|--------------------------------|---------------------------|-------------------------------|---|
| HAQ DI | 0.00 (0.00, 0.00) | 0.06 (0.00, 0.63) | 0.50 (0.63, 1.06) | $\chi^2=12.17$ $p=0.002$ | No medical follow-up vs. rheumatologist U = 49.01, $p=0.001$ |
| HAQ Pain Scale | 0.00 (0.00, 0.48) | 0.02 (0.00, 0.5) | 0.6 (0.25, 1.03) | $\chi^2=11.34$ $p=0.003$ | No medical follow-up vs. rheumatologist U = 59.0, $p=0.002$ |

*Values are medians (first quartile, third quartile). χ^2 : Kruskal-Wallis test; U: Mann-Whitney U-test.

higher than that of patients in follow-up with a rheumatologist (80.0; Q1=70, Q3=85.0) (U=69.0, $p=0.02$) or a general practitioner (75.0; Q1=58.8, Q3=81.8) (U=12.0, $p=0.02$).

Discussion

In the present study, we aimed to identify the settings in which patients with

JIA are followed up after the age of 16 and to compare the clinical profile – medical treatment, functional, general health status, and quality of life – of patients in different settings.

Patients not in medical follow-up

We found that one-third of JIA patients older than 16 were no longer in follow-

up. In general, these patients perceived no pain, did not take medication such as DMARDs or glucocorticosteroids, and experienced no disabilities. Most of these patients were most likely to be in remission; thus, follow-up may have been judged as unnecessary. However, in the non-follow-up group of patients, 2 patients (16.7%) had a mild disabil-

Table V. Proportion of patients with no, mild, or moderate disabilities and no, mild, moderate, or severe pain*.

| HAQ DI | No medical follow-up (n=12) [†] | | | | General practitioner (n=6) | | | | Rheumatologist (n=25) | | | |
|-----------|---|-----------------|---------------------|-------------------|-------------------------------|-----------------|---------------------|-------------------|--------------------------|-----------------|---------------------|-------------------|
| | no disability | mild disability | moderate disability | severe disability | no disability | mild disability | moderate disability | severe disability | no disability | mild disability | moderate disability | severe disability |
| oligo-JIA | 7 | 1 | 0 | 0 | 1 | 1 | 1 | 0 | 3 | 4 | 1 | 0 |
| poly-JIA | 1 | 1 | 0 | 0 | 2 | 0 | 0 | 0 | 2 | 4 | 5 | 0 |
| sys-JIA | 2 | 0 | 0 | 0 | 1 | 0 | 0 | 0 | 1 | 5 | 0 | 0 |

| HAQ pain scale | No medical follow-up (n=12) [†] | | | | General practitioner (n=6) | | | | Rheumatologist (n=25) | | | |
|----------------|---|-----------|---------------|-------------|-------------------------------|-----------|---------------|-------------|--------------------------|-----------|---------------|-------------|
| | no pain | mild pain | moderate pain | severe pain | no pain | mild pain | moderate pain | severe pain | no pain | mild pain | moderate pain | severe pain |
| oligo-JIA | 3 | 5 | 0 | 0 | 0 | 2 | 1 | 0 | 1 | 4 | 3 | 0 |
| poly-JIA | 2 | 0 | 0 | 0 | 2 | 0 | 0 | 0 | 0 | 8 | 3 | 0 |
| sys-JIA | 2 | 0 | 0 | 0 | 1 | 0 | 0 | 0 | 0 | 6 | 0 | 0 |

*Disability scores: no disability (0), mild (0 to ≤1), moderate (>1 to ≤2); pain scores: no pain (0), mild (0 to ≤1), moderate (>1 to ≤2), severe pain (>2).

[†]One missing value.

ity and 5 patients (41.7%) perceived mild pain. These patients would arguably benefit from further medical surveillance, at least either by a rheumatologist or by a general practitioner. In these patients, the absence of medical follow-up is probably inappropriate.

Drug use and medical treatment

Our findings of this study that patients treated with glucocorticosteroids DMARDs and anti-TNF – the use of which indicates a more severe disease – are typically followed up by a rheumatologist and are consistent with previous findings from patients with rheumatoid arthritis that DMARDs and anti-TNF therapy are rarely prescribed by primary care physicians (15). Good communication between general practitioners and rheumatologists should be encouraged, as the coordination of care would greatly benefit this patient population. Indeed, general practitioners need to identify candidate patients who may benefit from DMARDs and timely refer these to a rheumatologist; they also need to be aware of and monitor for potential treatment-related adverse effects (16, 17).

Health profile of patients in specialised care

Overall, patients in follow-up with a rheumatologist had higher degree of disability and higher pain scores than patients no longer in follow-up. Nonetheless, their HAQ scores were lower than those of young adults with JIA

examined in previous studies. Two studies in this patient population were published, reporting mean HAQ scores of 0.9 ± 0.5 (18) for patients comparably aged to our patients, and 0.8 ± 0.6 for patients older than our patients (19). Contrary to the subjects of Packham *et al.*'s study, no patients in our study had severe pain complaints. The continuous advances in therapy, like anti-TNF medication, for patients with rheumatic disorders might explain the successive improvements in functional outcome and pain that we observed in the present study.

In the literature, we could find only one comparable study about the general health status of adults with JIA (18). Foster *et al.* showed that JIA has a major impact on generic health status: 18.2% of their patients scored 0 (*i.e.* worst possible functioning) in at least one domain of the SF-36 (18). In our patient sample, 31.8% of the patients scored 0 in at least one SF-36 domain. When comparing the general health status of patients according to the type of follow-up, we found a significant difference only for the domain physical functioning between patients not in follow-up and patients in follow-up with a rheumatologist.

In our study, we assessed functional status using the HAQ. A recent study concludes, however, that a functional measure focused to assess the function of individual joint groups may detect with greater precision the functional impact of arthritis in specific body ar-

eas. Hence, future research on the functional status of patients with JIA is encouraged to use this approach (20).

Despite a comparably worse health and functional status in patients followed by a rheumatologist, these patients reported a relatively high median quality of life score. This confirms that health status, functional status, and quality of life are distinct constructs that, although related, cannot be interchanged (21, 22). This illustrates that, in addition to other patient-reported outcome measures, the measurement of quality of life in this patient population is important. Since the health-related quality of life improves in those patients who showed better treatment-responses, special attention should be paid to those patients with poor treatment responses to prevent the negative impact of arthritis on patients' functioning and well-being (23).

Importance of transition and transfer

There are many possible reasons why patients with JIA stop ongoing follow-up: i) Patients are in remission and no longer experience a need for medical surveillance (24); ii) patients are not informed about their rheumatic disease and associated complications (25); and iii) health professionals are not educated and trained in transitional care for adolescents with JIA (26). Although our findings indicate that ceasing further medical follow-up is inappropriate for some of our patients, we cannot precisely estimate the magnitude of

the problem. Furthermore, we do not know the consequences of lapse of care in these patients. We make a plea for this issue to be put on future research agendas.

Two fundamental principles are important for preventing patients from inappropriately interrupting their care. First, interdisciplinary care programs are needed throughout the follow-up of children with JIA. In such programs, patients and parents should be educated about the disease and complications. A study demonstrates that an interdisciplinary approach – including monitoring drug treatment and assessment of socio-professional functioning and psychological well-being guided by a rheumatology nurse specialist – leads to significantly lower disease activity, higher rates of remission, and better functional status than usual care approaches (27). A recent randomised controlled assessor blind trial showed that patient education also leads to a significant improvement in disease-specific knowledge and general health perception (28).

Second, a proper transfer from paediatric to adult-focused care is essential. Guidelines on standards of care for persons with rheumatic arthritis state that all young persons with rheumatoid arthritis should undergo smooth transfer from the care of a paediatric rheumatology team to that of an adult rheumatology team (29). A multicenter study in the UK showed substantial improvements in the level of knowledge of JIA patients after participation in a transitional care program (30). In an interdisciplinary transition program, a nurse specialist can provide information about the best follow-up setting. Although the importance of transition programs is beyond dispute, the evidence of the effectiveness of these programs is scarce (31). Moreover, it is unknown to what extent such transition programs are implemented. Thus, we advocate an international survey investigating the attitude toward and the current practice of transfer and transition of adolescents with JIA. Such surveys have been conducted previously in cystic fibrosis (32, 33), congenital heart disease (34), endocrine pathology (35), and dialysis (36) patients.

Methodological issues

This is the first study to scrutinise the setting of follow-up care after JIA patients leave paediatric rheumatology care and to examine the profile of patients in each setting. The response rate was 44%. However, there were no significant differences in age, gender, and type of JIA among the responders and non-responders. Selection bias in this study is therefore unlikely. Nonetheless, there were some methodological limitations that should be considered in interpreting the study findings. First, this was a single centre study with a small sample size (n=44). Due to this limitation we may assume that our study sample is not representative for the whole population. Therefore, our study findings cannot be generalised.

Second, this study is conducted in a large tertiary care centre in Belgium, having both paediatric and adult rheumatology programs. Belgium is a small country with a healthcare system that allows easy access to tertiary care. Hence, continuous follow-up with a rheumatologist is facilitated. A study like ours in other healthcare systems may yield different results.

Third, we limited our study to the assessment of general health, functional status, and quality of life. Other aspects of JIA such as fatigue, coping, and comorbidity might also be considered in decisions regarding referral to and continuity of rheumatology care. These outcomes, along with those dealing with functional disability, health status, and pain, should be considered in future studies of patients with JIA.

Conclusion

In this study, we identified the settings in which patients with JIA are followed up after leaving paediatric rheumatology care. We also compared the clinical profile of patients in each setting. We found that one-third of our patients were no longer in follow-up. In addition, we determined that patients who received glucocorticosteroid, DMARD or anti-TNF treatment were followed by a rheumatologist. Although patients in follow-up with a rheumatologist had good quality of life, they had worse physical functioning, higher degree of

disability, and higher pain scores than patients not in follow-up. However, two of the patients not in follow-up experienced disabilities; five of these patients reported pain. The absence of medical follow-up in these patients is probably inappropriate. A well-prepared transfer from paediatric to adult-focused care may optimise appropriate follow-up for all patients with JIA.

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