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Mikola, K.

2022-11-02

Mikola , K , Rebane , K , Glerup , M , Kautiainen , H , Relas , H , Peltoniemi , S & Aalto , K
2022 , ' Transition to adult care in Finnish adolescents with juvenile idiopathic arthritis ' ,
Scandinavian Journal of Rheumatology , vol. 51 , no. 6 , pp. 490-494 . <https://doi.org/10.1080/03009742.2022.2040117>

<http://hdl.handle.net/10138/356093>

<https://doi.org/10.1080/03009742.2022.2040117>

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Title page

Brief Communication

Transition to adult care in Finnish adolescents with juvenile idiopathic arthritis

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Short running title: Transition in JIA

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Abstract

Objective: The symptoms of juvenile idiopathic arthritis (JIA) and the necessity for continuous treatment may persist in adulthood. Therefore, patients with JIA need to be appropriately transferred to adult care. We aimed to analyze the timing of the patients' transition to adult care, patients' self-management skills with the process and the quality of the transition.

Methods: This study included 161 Finnish participants of the population-based Nordic JIA cohort who attended a 17-year follow-up appointment. Special attention was paid to the three groups: those referred by the pediatric rheumatology outpatient clinic to primary health care (PHC), those who were directly transferred to adult rheumatology care, and those who were later referred.

Results: A total of 136 (84%) patients were eligible to participate in the study, and 40% of them were directly transferred to an adult rheumatology clinic. 72% of the patients eventually ended up being referred to an adult rheumatology outpatient clinic. However, 16% of the patients in the PHC group had an active disease during the study appointment and were referred to adult services after the study visit.

Conclusion: This study revealed the need to improve the transition process from pediatric care to adult care and to find the variables that can indicate the need for immediate transition. Albeit challenging, it is important to avoid treatment delay in adult patients with JIA who may have an active disease but who do not have an appropriate access to an adult rheumatologic outpatient clinic.

Introduction

Juvenile idiopathic arthritis (JIA) is a heterogeneous disease that begins before the age of 16 years (1). It is divided into seven categories, based on the disease course and outcome (2,3).

Although many children with JIA achieve remission, half of them still have an active disease at adulthood. At the follow-up studies of the Nordic cohort, it was found that approximately half the patients with JIA were not in remission indicating the chronicity of JIA and the need of adult health care follow-up (4,5).

The process of transition to adult rheumatology care can be unsuccessful in up to 50% of the patients (6). Some transition protocols have been validated (7) and there is a recent European League Against Rheumatism recommendation on transition (8), but there is no gold standard for successful transition (9).

In Finland, the transition to adult care takes place at the age of 16 years and characteristically patients have ongoing antirheumatic medication and a stable disease status. In 2011, a distinct rheumatology outpatient transition clinic was founded in the Hospital District of Helsinki and Uusimaa (HUS) area (10). Patients in remission who have been off medication for at least 2 years are not transferred to the adult rheumatology clinic but are directed to primary health care (PHC) (10). Patients are advised to contact their general practitioner if they develop joint symptoms. The contact to PHC is free of charge. If these patients have a manifest disease flare, they are referred to the adult rheumatology clinic.

We aimed to investigate the transition rate of young adults with JIA. We analyzed the possible differences in clinical characteristics among the three patient groups. Finally, we aimed to evaluate the patients' experiences and self-management skills with the transition process.

Methods

This Finnish transition study is part of the previously described Nordic JIA cohort study (4,5).

All the patients with JIA onset within the period from January 1997 to June 2000 with at least two previous follow-up visits were eligible to participate. Accordingly, the cohort of 161 patients was invited to attend a follow-up appointment (mean 17 years after disease onset). A

standardized telephone interview was conducted for the patients who were unable to personally attend a follow-up appointment (n = 57). During the appointment, a joint examination was performed, clinical data and the medical history was updated. The Juvenile Arthritis Damage Index (JADI-A and JADI-E) was used to assess articular and extra-articular damage (11).

Remission was defined according to the preliminary Wallace criteria (12). The patients filled out the Health Assessment Questionnaire (HAQ) (13) and accomplished a global assessment of their overall well-being.

Based on the information gathered the patients were divided into three groups: those who were referred to PHC, those who were directly transferred to adult rheumatology care, and those who were later referred to the latter after initially being discharged from pediatric care. The PHC group consisted of patients who were not followed up by a rheumatologist after being discharged from the pediatric department. Patients still attending Children's clinic were not included.

A modified questionnaire about transition, Care Transitions Measure® (3-CTM in Finnish) was used to evaluate the quality of care in transition (14).

The approval of the study was acquired from the local Medical Ethics Committee.

Statistical analyses

Statistical comparisons between the transition groups were made using analysis of variance for the continuous variables and chi square test or the Fisher–Freeman–Halton test for the categorical variables. The Kaplan–Meier method was used with age as the underlying time scale to estimate the crude cumulative transition. Bootstrap or permutation methods were used when the theoretical distribution of the test statistics was unknown or when there was a violation of the assumption (e.g., non-normality). Hommel’s adjustment was applied to correct the levels of significance for multiple testing (post hoc), if appropriate. The Stata 17.0 statistical package (StataCorp LP, College Station, TX, USA) was used for the analyses.

Results

Of the 161 eligible study participants, 84% completed the follow-up visit and the background demographics are presented in Table 1. 67% of the directly transferred patients had polyarticular rheumatoid factor negative or extended oligoarticular JIA. 59% of the participants in the PHC group had a persistent oligoarticular disease course. There was no statistically significant difference in sex or age between the three transition groups. However, there was a significant difference in the distribution of JIA subtypes ($p < 0.001$), in particular for persistent oligoarthritis ($p = 0.007$), which was more frequent in the PHC group.

At 16 years of age, 54 patients with JIA (40%; 95% CI: 32–48) were directly transferred from the pediatric care department to an adult rheumatology care clinic. A total of 101 patients were referred at some point (estimate of cumulative transition rate 72%; 95% CI: 59–84) during the follow-up period.

The patients' disease characteristics are provided in Table 2. Differences in disease activity were found among the transition groups at the follow-up visit. Of the patients who were not in remission, 30% belonged to the later referred group, 51% belonged to the directly transferred group, and remarkably, 19% belonged to the PHC group ($p < 0.001$). 89% of the directly transferred patients were on medication at the time of transition (76% at the study visit), and 93% of them had no active joints. The directly transferred patients had a significant uveitis burden: 22% of them had JADI-E > 0 . Moreover, 26% of the directly transferred patients had JADI-A > 0 mainly because of temporomandibular joint involvement.

In the PHC group, 75% of the patients had been off medication during the previous 9 years before the follow-up visit compared to 2% in the directly transferred group and 8% in the later referred group ($p < 0.001$). None of the patients in the PHC group with previous uveitis had active joints at the 17-year follow-up visit.

The CTM questionnaire was accomplished by 66% of the patients. The survey showed significant differences among the three patient groups (Figure 1). Most of those who were directly referred felt that their own opinions were considered at their last visit at the pediatric care. They also had the best awareness of health issues and knowledge about medicines.

Discussion

Our main finding was that over time, nearly three-quarters of the patients were eventually transferred to an adult rheumatology clinic. Originally 40% were directly transferred. The most striking finding was that around one fifth of the patients in the PHC group and half of the patients in the later referred group had disease activity at the follow-up visit. On the other hand, these patients had low disease activity, and low JADI and pain scores. However, although their

disease activity was mild, they were lost to follow-up even in a healthcare system that is free of charge. In a Canadian study, a link between mild disease at the last pediatric visit and the risk of dropping out from the adult follow-up was detected, supporting our findings (6). This group should be given special attention during the transition and low-threshold access to adult rheumatology clinics.

In the beginning of the present follow-up study the patients were referred to an adult clinic without any special procedures. From the year 2011 there has been a designated adult rheumatologist and a team consisting specialized nurse and other healthcare workers (10). The monitoring of the patients is more stringent, e.g., new appointments are reserved and more actively offered if a patient seems to be at risk of dropping out. However, these changes did not solve some problems detected in our study.

Directly transferred patients were found to be most properly prepared for the transition when asked about their medications and health maintenance. There were fewer positive answers to the questions regarding the transition process in the PHC group than in the directly transferred group.

The proportion of patients with active disease but no rheumatological contact was surprisingly high at our study. We acknowledge that transferring adolescents with a history of a chronic disease to PHC, which is a healthcare system requiring self-initiative and independent skills to operate in, may deserve re-evaluation.

As a conclusion, a successful transition of young patients with JIA to adult care is challenging even in a public healthcare system. The need for standardized measures of outcome and effectiveness of transition is obvious and more research is required to understand the

perspectives and needs of young patients regarding transition (15). Our study showed that the patients need clear guidance regarding whom to contact in the healthcare system if they develop symptoms after several years of remission. The challenge is to identify the patients who do not need adult care at the time of transition but who experience symptoms or flare later. A possible option is to change our current practice and transfer all patients, except maybe the persistent oligoarthritis group, to an adult rheumatological clinic.

The strength of this study is the long follow-up period in this initially prospective research. To our knowledge, this is the first population-based JIA follow-up study that reported the transition of patients over time. We did not study the predictors of the transition process. Focusing only to the Finnish patients could be also considered as a limitation and the results may not apply to other countries.

Acknowledgements

We thank all the patients and the pediatric rheumatologist involved in this study.

Conflict of interest

The authors declare no potential conflict of interest.

Funding

The study received funding from The Finnish Society of Rheumatology and the Pediatric Research Center.

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Table 1. Background demographics of the transition study groups of JIA

	PHC group (n = 56)	Directly transferred (n = 54)	Later referred (n = 26)
Female, n (%)	32 (57)	30 (56)	20 (77)
Age at disease onset, mean (SD)	5.8 (4.0)	6.1 (4.1)	8.0 (4.1)
JIA subtype, n (%):			
Systemic onset	1 (2)	0 (0)	0 (0)
RF-positive polyarthritis	0 (0)	1 (2)	0 (0)
RF-negative polyarthritis	6 (11)	20 (37)	5 (19)
Psoriatic	5 (9)	4 (7)	3 (12)
Enthesitis related	3 (5)	7 (13)	3 (12)
Undifferentiated	2 (4)	1 (2)	3 (12)
Persistent oligoarthritis	33 (59)	5 (9)	7 (27)
Extended oligoarthritis	6 (11)	16 (30)	5 (19)

PHC = primary health care; SD = standard deviation; JIA = juvenile idiopathic arthritis; RF = rheumatoid factor

Table 2. Disease characteristics at the 17-year follow-up visit

	PHC group (n = 56)	Directly transferred (n = 54)	Later referred (n = 26)	P-value [multiple comparison] *
Disease duration, years, mean (SD)	17.9 (1.0)	17.1 (1.2)	17.0 (1.2)	..
Age at follow-up, years, mean (SD)	23.7 (4.2)	23.2 (4.2)	25.0 (4.3)	0.21
Body mass index, kg/m ² , mean (SD)	23.7 (4.6)	23.2 (3.8)	23.3 (3.5)	0.64
Remission on/off medication, n (%)	47 (84)	30 (56)	12 (46)	< 0.001 [P/D, P/L]
Off medication	47 (84)	13 (24)	7 (27)	
On medication,	0 (0)	17 (31)	5 (19)	
Inactive disease** 8–17 years n (%)	42 (75)	1 (2)	2 (8)	< 0.001 [P/D, P/L]
Cumulative number of involved joints, median (IQR)	2 (1, 5)	11 (6, 16)	6 (4, 9)	< 0.001 [P/D, P/L, D/L]
Disease Activity Scale (DAS28), mean (SD)	1.93 (0.56)	1.81 (0.84)	1.83 (0.76)	0.76
Health Assessment Questionnaire (HAQ), mean (SD)	0.07 (0.44)	0.14 (0.32)	0.09 (0.23)	0.64
Visual Analogue Scale (VAS), mm, mean (SD)				
Pain	5.1 (10.5)	15.9 (22.6)	21.8 (26.5)	0.002 [P/D, P/L]

Physicians' global assessment of overall disease activity	2.4 (6.3)	15.7 (23.7)	19.0 (28.5)	0.005 [P/D, P/L]
Patients' global assessment of overall disease activity	3.8 (7.4)	16.2 (22.6)	16.2 (22.3)	0.002 [P/D, P/L]
Fatigue	40.3 (22.8)	35.3 (25.1)	42.4 (23.6)	0.49
JADI-A, n (%)				< 0.001 [P/D, P/L]
0	56 (100)	40 (74)	23 (88)	
1	0 (0)	8 (15)	2 (8)	
≥2	0 (0)	6 (11)	1 (4)	
JADI-E > 0, n (%)	3 (5)	12 (22)	2 (8)	0.021 [P/D]

*Hommel's multiple comparison was used to correct significance levels for post hoc testing ($p < 0.05$).

**Inactive disease from 8 years (mean) to 17 years (mean) of follow-up.

PHC = primary health care; JADI-A, the Juvenile Arthritis Damage Index assessment of articular damage; JADI-E, the Juvenile Arthritis Damage Index assessment of extra-articular damage.

P = PHC group; D = directly transferred; L = later referred

Figure 1. Patients' experiences and self-management skills according the CTM-3 transition questionnaire

Caption:

The percentage of positive responses ("agree" or "strongly agree") to the questionnaire accomplished by the patients in the three groups regarding their feelings during their final visit to the children's clinic, regardless of whether they were transferred at that time or were not. The error bars show 95% confidence intervals.

Opinions: The staff in the children's clinic took my opinions and those of my family into account when deciding what is required to treat my disease when I left the children's clinic.

Health: When I left the children's clinic, I knew the things I must do in order to maintain my health.

Medicine: When I left the children's clinic, I was aware of all my medicines (if any) and how to use them.