

# The impact of a coordinated transitional care programme on adolescents with juvenile idiopathic arthritis

J. E. McDonagh, T. R. Southwood and K. L. Shaw on behalf of the British Society of Paediatric and Adolescent Rheumatology

**Objective.** There is an extensive evidence base for the need of transitional care, but a paucity of robust outcome data. The aim of the study was to determine whether the quality of life of adolescents with juvenile idiopathic arthritis (JIA) could be improved by a co-ordinated, evidence-based programme of transitional care.

**Methods.** Adolescents with JIA aged 11, 14 and 17 yrs and their parents were recruited from 10 rheumatology centres in the UK. Data were collected at baseline, 6 and 12 months including core outcome variables. The primary outcome measure was health-related quality of life (HRQL): Juvenile Arthritis Quality of Life Questionnaire (JAQQ). Secondary outcome measures included: knowledge, satisfaction, independent health behaviours and pre-vocational experience.

**Results.** Of the 359 families invited to participate, 308 (86%) adolescents and 303 (84%) parents accepted. A fifth of them had persistent oligoarthritis. Median disease duration was 5.7 (0–16) yrs. Compared with baseline values, significant improvements in JAQQ scores were reported for adolescent and parent ratings at 6 and 12 months and for most secondary outcome measures with no significant deteriorations between 6 and 12 months. Continuous improvement was observed for both adolescent and parent knowledge with significantly greater improvement in the younger age groups at 12 months ( $P = 0.002$ ).

**Conclusions.** This study represents the first objective evaluation of an evidence-based transitional care programme and demonstrates that such care can potentially improve adolescents' HRQL.

KEY WORDS: Adolescent, Juvenile idiopathic arthritis, Transitional care, Quality of life.

Transitional care is a holistic, multidimensional, multidisciplinary and active process that addresses the medical, psychosocial and educational/vocational needs of adolescents as they move from child- to adult-centred care. Generic development in this area is supported by major policy documents in the UK and US [1–6] and the non-categorical nature of issues facing adolescents with chronic illnesses and/or disability [7].

To date, there have only been limited evaluations of transitional care programmes that support beneficial outcomes in a number of chronic diseases (e.g. diabetes [8, 9]; cystic fibrosis [10, 11]). However, a recent commissioned review of transitional care practice reported a 'paucity of robust evidence' [12]. The need for transitional care in rheumatology is now well-established [13–17], but only one study has addressed the outcome of transitional care in terms of improved follow-up in adult care [18].

The aim of the study was to determine whether the quality of life of adolescents with JIA could be improved by a co-ordinated, structured multidisciplinary, evidence-based, transitional care programme.

## Methods

### Participants

The cohort has been described elsewhere [17]. In summary, patients and parents were recruited from 10 paediatric rheumatology centres represented in the British Society of Paediatric and

Adolescent Rheumatology (BSPAR). Eligibility criteria for patients included that they: (i) had a diagnosis of juvenile idiopathic arthritis (JIA) as defined by the revised International League Against Rheumatism criteria [19]; (ii) were expected to remain in the paediatric care of a consultant member of the BSPAR for at least 6 months; and (iii) were aged 11, 14 or 17 yrs (i.e. they were in their 11, 14 or 17th yr  $\pm$  1 month). These age-specific entry criteria were selected because they (i) reflect the stages of adolescent development, (ii) facilitate exploration of the importance of timing in transitional care, and (iii) avoid discrimination between patients attending the same centre since all children in each centre would become eligible for the study when they reached 11, 14 or 17 yrs of age. Only one parent of a given patient was recruited. The participation of a parent was not contingent on their child's participation.

Due to the complexity, heterogeneity, individual-centred and evolving nature of transitional care provision in the UK at the time of the study, a randomized control trial either within a single centre or between centres was not considered possible or ethical. Study design was further complicated by the multidisciplinary nature and the heterogeneity of adolescent rheumatology provision in the UK [13], the underdeveloped nature of adolescent healthcare in the UK in general [13, 20] and the unmet training needs amongst professionals [21]. Due to the study being conducted on behalf of BSPAR, changes in practice in many member centres were observed during the development phase of the study, simply by virtue of the study conception.

Division of Reproductive and Child Health, University of Birmingham, Institute of Child Health, Birmingham, UK.

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Correspondence to: Dr Janet E. McDonagh, MD, FRCP, ARC Senior Lecturer in Paediatric and Adolescent Rheumatology, Division of Reproductive and Child Health (University of Birmingham), Institute of Child Health, Steelhouse Lane, Birmingham B4 6NH, UK. E-mail: j.e.mcdonagh@bham.ac.uk

Randomization by site would potentially mean postponing such developments in control sites and this was not considered ethical. Although the study did not have a true control group, the 17-yr-olds' scores were used as comparative data or 'pseudo-control' group for the 11- and 14-yr-olds'. The '17-yr-old' baseline data reflected the status of patients who would soon enter adult-care in UK healthcare, without any experience of a formal transitional care programme [17]. The specific unmet needs of the latter cohort have been previously reported by the authors [17].

Patients and parents were all sent introductory and explanatory letters detailing the aims of the project. All participants gave written, informed consent. Ethical approval was obtained from the West Midlands Multicentre Research Ethics Committee and the Local Research Ethics Committees by each of the participating centres.

### Intervention

A programme of transitional care (PTC) was co-ordinated within each centre by a local programme co-ordinator (LPC), funded for 1 day per week for the duration of the study, with assistance from the local BSPAR consultant rheumatologist. The details of the PTC have been reported elsewhere [22]. In brief, the programme centred on templates for individualized transition plans (ITP). The ITP templates were created for the young person and the parent to reflect both adolescent development as well as the components of the transition programme in terms of health, home and school. They were designed to reflect the developmental stages of adolescence (early, mid and late) and were reviewed at each clinic visit and/or every 6 months. Once each template was complete, the young person would be invited to move on to the next plan. The other key components of the programme included a local project coordinator, age and developmentally appropriate informational resources for adolescents, informational resources for parents and local programme coordinators addressing the needs identified in the first phase of the project [14–16, 21], and a departmental transition policy template for completion by each participating centre. Further details of the programme can be obtained from the authors and revised format of resources via their team website [www.dreamteam-uk.org](http://www.dreamteam-uk.org).

### Assessment

The study employed a repeated measures design with multiple baselines. Data were collected at three time points: prior to entering the PTC (baseline) and again, at 6 and 12 month clinic follow-up visits. Study duration was limited by available funding. Clinical data were provided by the senior clinician. Patient and parent data were collected using individual questionnaires designed for self-completion, with support from LPCs where necessary. Questionnaires were sent to 'no shows' by mail when possible.

**Primary outcome measure.** Health-related quality of life (HRQL) was assessed using the Juvenile Arthritis Quality of Life Questionnaire (JAQQ) [23]. Detailed discussion of the baseline data and use of this measure in a UK population has been published elsewhere [24]. The JAQQ consists of 74 items grouped into four dimensions: gross motor function (17 items), fine motor function (16 items), psychosocial function (22 items) and systemic symptoms (19 items). The response format for each item uses a 7-point Likert-type scale of frequency/difficulty scale anchored by 'None of the time—never' at 1 and 'All of the time—always' at 7. Each item also has a 'Does not apply to me' option at 0. The mean score for the five highest scoring items in each of the four dimensions is computed as the dimension score; the total JAQQ score is computed as the mean across the four dimensions still with a score range from 1 to 7. Higher scores indicate lower HRQL.

To determine the sample size necessary for the study, data regarding the primary outcome measure i.e. the JAQQ was used in collaboration with the authors of the questionnaire [23]. With a sample size of 500 patients, at least 90% power was predicted to detect an effect size of 0.15 based on the difference from baseline in enrolled patients. For comparison between the three recruitment age-groups, at least 80% power is needed to detect an effect size of 0.25 [23, 25].

**Secondary outcome measures.** Due to the lack of adolescent-specific measures currently available in this area, the following outcome tools and question sets were designed by the authors and based on the prior needs [14–16, 21]. Data collection regarding the validation of these measures is still on-going and has not been presented in detail further here.

**Arthritis-related knowledge.** Adolescent and parent knowledge were assessed using a 16-item disease-specific multidimensional measure with multiple-choice response format. Final knowledge scores range from 0–16 where higher scores indicate greater knowledge.

**Satisfaction with rheumatology care.** Patient and parent satisfaction were measured using 22- and 27-item measures, respectively. Items include statements about issues specifically identified in the earlier needs assessment [14–16, 21] including the physical environment, clinic procedures, relationships with healthcare personnel, information and support. Respondents are asked to rate the extent to which each item represents (i) 'best' and (ii) 'current' care using a 7-point Likert scale anchored by 'strongly disagree' at 1 and 'strongly agree' at 7. Satisfaction with each item is conceptualized as the 'gap' between their 'best' and 'current' score (i.e. their 'gap' score). Thus, a score of 0 shows that there is no difference ('gap') between 'best' and 'current' care. In contrast, a positive score indicates that 'best' practice is rated higher than 'current' care; the greater the 'gap' score, the lower the level of satisfaction. Conversely, a negative score indicates that the rating of 'current' exceeds 'best' practice; thus, the more negative a score, the greater the level of satisfaction. Overall satisfaction is computed as the mean of all the 'gap' scores and ranges between –7 (most satisfied) and 7 (most unsatisfied).

**Independent health behaviours.** These included self-medication and independent consultations and were assessed using closed questions.

**Pre-vocational experience.** Pre-vocational experience was explored in terms of household chores, work experience, career advice and career aspirations. It was measured using a series of closed and open questions.

**Other data.** Demographic data were collected at baseline including age, gender, ethnicity, socio-economic status based on the reduced 5-class version of National Statistics-Social Economic Classification (NS-SEC) [26], family structure, parental marital status, educational qualifications and employment, and the adolescent's educational status.

**Clinical data** included JIA onset subtype, age at onset, age at diagnosis and disease duration.

**Core outcome variables** as described by Giannini *et al.* [27] were collected. Adolescents and parents also completed the Childhood Health Assessment Questionnaire (CHAQ), as modified for use in the UK [28]. To enhance completion by young people, an adolescent version of the CHAQ was developed. This version was written in the first person and omitted developmentally inappropriate words (e.g. potty) and the 'not applicable' response, the latter being absent in the adult HAQ [29].

**Analyses.** The Statistical Package for the Social Sciences (SPSS 11.0) was used to perform all the analyses on the quantitative data (Norusis/SPSS, 1993) [30]. The Kolmogorov–Smirnov test was used to check for skewness in the distribution of values of each study variable. There was evidence of skewness for some

variables, which led to the choice of non-parametric inferential statistics.

Group differences were analysed using chi-square, Mann–Whitney, Kruskal–Wallis or Jonckheere–Terpstra tests, as appropriate. Associations between the study variables were analysed using Spearman Rho correlations. The strength of statistically significant correlations were defined as very weak ( $r_s < 0.20$ ), weak ( $r_s = 0.20–0.39$ ), moderate ( $r_s = 0.4–0.69$ ), strong ( $r_s = 0.70–0.89$ ) and very strong ( $r_s = 0.90–1$ ) [31].

Comparisons for all study variables were made between (i) baseline and 6 months and (ii) baseline and 12 months using the Wilcoxon matched pairs signed-ranks test for ordinal data and McNemar's Test for categorical data. Sustainability of the outcome changes between 6 and 12 months were also evaluated.

Stepwise linear regression analyses were performed to explore the relationship between possible predictor variables and the change in primary outcome measure (JAQQ) at 6 and 12 months. Variables entered included those that displayed significant univariate relationships with the JAQQ at 6 and 12 months ( $P < 0.05$ ). This was to limit the potential explanatory variables to a moderate number. Appropriate normality tests on residuals were implemented. The change in  $R^2$  for the regression upon entry of a variable was taken as the proportion of the variation in final JAQQ change scores explained by that variable.

In view of the relatively small numbers of certain JIA subtypes in each age group [17], patients were divided into two main groups; 'oligoarthritis persistent' and 'other' that were, by definition, primarily polyarticular and/or systemic. Ethnicity categories were similarly collapsed into 'White/European' and 'other'.

Due to the use of multiple baselines and for the sake of brevity, the term 'withdrawals' encompasses participants, who chose to withdraw from the study or were lost to follow-up or were not able to be seen by the clinical research team within the study period, due to the local constraints of service provision.

Due to the exploratory nature of the study, the level of significance was set at  $P < 0.01$  (unless stated otherwise) to adjust for multiple testing. All analyses were undertaken using the SPSS 11.0 (Chicago, IL, USA).

## Results

### Participant characteristics

The cohort has been described in detail elsewhere [17]. In summary, of the 359 families invited to participate, 308 (85.79%) adolescents and 303 (84.4%) parents accepted, the majority of whom were mothers 252 (83.2%). Although less than the original target sample size, it was calculated that a sample size of 308 would give 74% power to detect an effect size of 0.150 but, more usefully, would have 90% power to detect the slightly larger effect size of 0.185 [23, 24]

Analyses of available data showed no significant differences between the participants and non-participants ( $n = 51$ ). There was

no consistent reason for non-participation. Completion rates are shown in Table 1.

Key demographic and disease-related data at baseline are presented in Tables 2 and 3. Analyses between the three age-groups, indicated no significant differences in the demographic data. However, older age was significantly associated with increased duration of care ( $P = 0.002$ ), age at onset ( $P < 0.001$ ), age at diagnosis ( $P < 0.001$ ), disease duration ( $P < 0.001$ ), older parents ( $P < 0.001$ ), increased frequency of non-oligoarticular JIA and decreased frequency of persistent oligoarthritis. ( $P < 0.001$ ). There were no differences in the core outcome variables. Older groups were also more likely to be self-medicating (40.7, 76.9 and 80% of 11, 14 and 17-yr-olds, respectively,  $P < 0.001$ ) and making independent visits (23.3, 42.2 and 63.2% of 11, 14 and 17-yr-olds, respectively,  $P < 0.001$ ).

There were no significant differences at baseline between male and female caregivers for parent proxy scores of their son/daughter's CHAQ ( $P = 0.739$ ); pain ( $P = 0.302$ ); overall well-being ( $P = 0.467$ ); satisfaction ( $P = 0.252$ ) and JAQQ ( $P = 0.987$ ). Female caregivers had significantly better baseline knowledge than male caregivers ( $P = 0.001$ ) with respective median scores of 11 (1–16) and 10 (1–14).

### Primary outcome data: HRQL

Compared with baseline values, significant improvements were reported for adolescent and parent-proxy ratings of JAQQ at 6 and 12 months with no significant deterioration between follow-up assessments (Table 4). Age-group did not significantly influence the scores at any time-point, nor the change in scores at either follow-up (data not shown).

With respect to familial comparisons, adolescent scores showed both agreement and consistency (i.e. there were no significant differences between adolescent and parent-proxy scores at any time point and strong positive correlations between them at baseline ( $r_s = 0.754$ ,  $P < 0.001$ ), 6 months ( $r_s = 0.746$ ,  $P < 0.001$ ) and 12 months ( $r_s = 0.816$ ,  $P < 0.001$ )). Greater change in adolescent JAQQ scores was also related to greater change in parent-proxy scores at both 6 months ( $r_s = 0.551$ ,  $P < 0.001$ ) and 12 months ( $r_s = 0.490$ ,  $P < 0.001$ ).

Variables exhibiting significant relationships with JAQQ change scores (data not shown) were entered into stepwise linear multiple regression analyses. With respect to adolescent-rated HRQL, the final regression models explained 38.1% of the variation in the JAQQ change scores at 6 months (Table 4). Change in adolescent-rated pain explained 28.7% of variation, to which change in adolescent-rated disability added a further 5.2%, independent visits 2.2% and physician-rated disease activity 2.0%. At 12 months, the final model explained 27.6% of the variation in scores. Change in adolescent-rated disability explained 18.4% and self-medication at baseline added a further 9.2%.

Likewise, with respect to parent-proxy-rated JAQQ the final regression models explained 68.9% of the variation in the JAQQ change scores at 6 months (Table 4). Change in parent-proxy-rated disability explained 63.2% of the variation in JAQQ change

TABLE 1. Availability of complete data within the study period

	Adolescents			Parents		
	Baseline <i>n</i> (%)	6 mth <i>n</i> (%)	12 mth <sup>a</sup> <i>n</i> (%)	Baseline <i>n</i> (%)	6 mth <i>n</i> (%)	12 mth <sup>a</sup> <i>n</i> (%)
All	308 (100.0)	228 (74.0)	122 (52.6)	303 (100.0)	209 (69.0)	106 (46.3)
11 yrs	103 (100.0)	78 (75.7)	37 (50.7)	103 (100.0)	76 (73.8)	35 (47.9)
14 yrs	128 (100.0)	93 (72.7)	50 (49.0)	128 (100.0)	88 (68.7)	45 (44.1)
17 yrs	77 (100.0)	57 (74.0)	35 (61.4)	72 (100.0)	45 (62.5)	26 (48.1)

<sup>a</sup>Because of multiple baselines, the completion rates are based on the 232 adolescents and 229 parents who were eligible for 12 month assessments. Incomplete data include the participants, who chose to withdraw from the study or were lost to follow-up or were not able to be seen by the clinical research team within the study period, due to the local constraints of service provision.

TABLE 2. Adolescent characteristics at baseline [17]

Median (min, max) unless stated otherwise	All <i>n</i> = 308	11 yrs <i>n</i> = 103	14 yrs <i>n</i> = 128	17 yrs <i>n</i> = 77
Age, yrs	14.2 (10.9,18.0)	11.5 (10.9,12.1)	14.3 (13.9,15.2)	17.3 (16.8,18.0)
Gender, M:F	1:1.5	1:1.8	1:1.3	1:1.7
Ethnicity: white/European, <i>n</i> (%)	274 (91.0)	97 (94.2)	112 (88.2)	64 (91.4)
JIA subtype, <i>n</i> (%)				
Oligoarthritis -persistent	60 (19.5)**	31 (30.1)	23 (18.0)	6 (7.8)
Others	248 (80.5)**	72 (69.9)	105 (82.0)	71 (92.2)
Duration of care at current hospital, yrs	3.5 (0.0,15.8)**	2.9 (0.0,10.6)	3.0 (0.0,14.3)	4.1 (0.3,15.8)
Age at diagnosis, yrs	9.0 (1.0,17.0)***	7.8 (1.0,11.8)	9.5 (1.1,14.3)	11.4 (1.3,17.0)
Disease duration, median (minimum, maximum) yrs	5.7 (0.0,16.3)***	3.9 (0.1,10.9)	5.8 (0.3,14.3)	6.9 (0.0,16.3)
Core outcome Variables [27]				
Physician Global Assessment (PGA) of disease activity	10 (0,99)	11 (0,97)	8 (0,99)	10 (0,72)
Active joint count	0(0,42)	0 (0,42)	0 (0,31)	1 (0,24)
Limited joint count	2 (0,71)	2 (0,71)	2 (0, 52)	2 (0,52)
Adolescent-rated pain score (VAS-pain)	16 (0,100)*	14 (0,99)	15 (0,100)	30 (0,93)
Adolescent-rated CHAQ	0.5 (0.0, 3.0)	0.5 (0.0, 3.0)	0.4 (0.0, 2.9)	0.7 (0.0, 2.6)
Adolescent-rated VAS-global	17 (0,94)	14 (0,86)	19 (0,94)	23 (0,89)

Significant differences between age-groups: \**P* < 0.05; \*\**P* < 0.01; \*\*\**P* < 0.001.

TABLE 3. Comparison of study variables at all the three time points and between 6 and 12 months

Study variables (min, max)	Baseline		Change scores (6 months-baseline)		Change scores (12 months-baseline)		Change scores (12 –6 months)	
	Median (min, max)	<i>n</i>	Median (min, max)	<i>n</i>	Median (min, max)	<i>n</i>	Median (min, max)	<i>n</i>
JAQQ (0, 7)								
Adolescent rating	2.7 (1.0, 6.8)	308	−0.3 (−3.8, 5.6)***	224	−0.3 (−4.8, 4.1)**	122	0.0 (−2.7, 5.0)	107
Parent-proxy rating	2.8 (1.0, 7.0)	300	−0.2 (−6.0, 4.9)**	210	−0.4 (−4.2, 3.0)***	110	−0.1 (−2.3, 3.7)	96
Knowledge (0, 16)								
Adolescent rating	8 (0, 15)	307	2 (−6, 13)***	221	2 (−7, 12)***	121	0 (−6, 9)**	106
Parent rating	11 (1, 16)	300	1 (−4, 11)***	209	1 (−3, 9)***	108	1 (−4, 5)**	97
Satisfaction (−7, 7)								
Adolescent rating	0.3 (−5.2, 4.8)	301	−0.2 (−4.7, 2.8)**	211	−0.3 (−3.5, 6.0)**	114	0.2 (−2.4, 4.4)	102
Parent rating	0.7 (−1.5, 5.2)	286	−0.4 (−5.0, 6.1)***	200	−0.6 (−3.8, 3.0)***	106	0.0 (−7.6, 2.3)	94
Independent health behaviours								
Self-medicating: %	64.9	151	−0.9	108	4.2	48	−5.9	68
Independent visits: %	41.0	307	4.9	223	5.0	121	4.7	106
Pre-vocational readiness markers								
Work experience	0 (0, 4)	218	−0 (0, 3)***	130	0 (0, 2)***	73	0 (0, 6)***	67
Career advice	0 (0, 6)	219	−0 (0, 5)***	118	0 (0, 4)***	61	0 (0, 2)***	57
Household activities: %	77.4	292	1.6	188	8.2	110	8.5	82
Vocational plans: %	67.7	251	6.3	142	6.8	84	7.2	66

Significant improvements compared with baseline: \**P* < 0.05; \*\**P* < 0.01; \*\*\**P* < 0.001.

Negative scores indicate improvement in HRQL and Satisfaction.

scores to which change in parent-proxy-rated pain added a further 5.7%. At 12 months, the model explained 46.0% of the variation. Change in parent-proxy-rated disability explained 39.2% of the variation and adolescent-rated disability contributed a further 6.8%.

### Secondary outcomes

**Knowledge.** With respect to change, continuous improvement was observed for both adolescent and parent knowledge (Table 4). Better knowledge was associated with increasingly older age-group at baseline (respective median scores of 6, 8 and 9, *P* < 0.001), 6 months (9, 10 and 11, *P* < 0.001) but this did not achieve significance (*P* > 0.01) at 12 months (11, 11 and 12, *P* = 0.026). Age-group did not influence the change in knowledge at 6 months but at 12 months, greater improvement was significantly associated with younger age-group (*P* = 0.002).

Parental knowledge was not influenced by the adolescent's age-group.

With respect to familial relationships, parents had significantly greater knowledge than adolescents at baseline (respective median scores of 11 and 8, *P* < 0.001), 6 months (12 and 10, *P* < 0.001) and 12 months (12 and 11, *P* < 0.001 throughout). These differences remained significant, irrespective of age-group (*P* < 0.001). However, higher adolescent knowledge showed weak-to-moderate significant relationships with higher parental knowledge (*r*<sub>s</sub> = 0.321 *P* < 0.001; *r*<sub>s</sub> = 0.461, *P* < 0.001; *r*<sub>s</sub> = 0.300, *P* = 0.002 at baseline, 6 and 12 months, respectively). With respect to change, adolescents had significantly greater improvement in their scores compared with parents at the 6 month assessment (respective median scores of 2 and 1, *P* < 0.001) and at the 12 month-assessment (respective median scores of 2 and 1, *P* = 0.007).

**Satisfaction.** Compared with baseline values, significant improvements were reported for adolescent and parent's

TABLE 4. Multivariate relationships with HRQL

	B	P	Increments in R <sup>2</sup>
<b>Adolescent-rated JAQQ</b>			
Change in scores at 6 months			
Change in VAS-pain (adolescent)	0.020	<0.001	28.7
Change in CHAQ (adolescent)	0.485	0.010	5.2
Making independent visits (at 6 months)	0.375	0.026	2.2
Change in PGA	0.008	0.024	2.0.
Constant	-0.896	0.001	
Total			38.1
Change in scores at 12 months			
Change in CHAQ (adolescent)	1.003	0.006	18.4
Change in self-medication (adolescent)	0.724	0.048	9.2
Constant	-1.192	0.023	
Total			27.6
<b>Parent-proxy rated JAQQ</b>			
Change in scores at 6 months			
Change in CHAQ (parent-proxy)	1.260	<0.001	63.2
Change in VAS-pain (parent-proxy)	0.013	0.032	5.7
Constant	-0.036	0.812	
Total			68.9
Change in scores at 12 months			
Change in CHAQ (parent-proxy)	0.938	<0.001	39.2
Change in CHAQ (adolescent)	0.558	0.003	6.8
Constant	-0.244	0.018	
Total			46.0

Where B = regression co-efficient for each variable; R<sup>2</sup> = percentage of the variation explained.

satisfaction scores at 6 and 12 months with no significant deterioration between follow-up assessments (Table 4). Age-group had no significant influence upon adolescent or parent satisfaction (at all time-points) or upon the changes in satisfaction at the follow-up assessments.

With respect to familial data, parents had significantly less satisfaction than their adolescents at baseline (respective median scores of 0.3, 0.7,  $P=0.001$ ), and no significant differences in scores were observed at the follow-up assessments. Correlational analyses between adolescent and parent scores showed only one significant result: 6 month scores displayed a weak positive relationship ( $r_s=0.211$ ,  $P=0.003$ ), but adolescent satisfaction appeared to be unrelated at baseline and 12 months. Parents showed more change than adolescents at both the 6 month assessment (respective median scores of -0.4 and -0.2,  $P=0.041$ ) and the 12 month assessment (respective median scores of -0.3 and -0.6,  $P=0.029$ ), although this did not achieve significance ( $P>0.01$ ).

**Independent health behaviours.** No significant changes in self-medication or independent consultations were observed at either the 6 or the 12 month assessments (Table 4). With respect to age-group, independent visits increased with older age at each time point ( $P<0.001$  in all cases). At baseline and 6 months, the 11-yr-old groups were less likely to be self-medicating than the 14- and 17-yr-olds groups that had similar proportions of adolescents who were self-medicating. However, at 12 months, there were no significant differences between any age-groups and while the 17-yr-olds continued to show the highest level of self-medication (80.0%), the 11-yr-olds now displayed the same level of self-medication as the 14-yr-olds (66.7%).

**Vocational markers.** Compared with baseline values, significant improvements were reported for work experience and career advice at all time points. (Table 4). However, the amount of improvement was small (median change=0 episodes in all cases, ranging between 2 and 6). As would be expected, the number of work experiences and episodes of career advice significantly

increased with older age-group at each assessment ( $P<0.001$  in all cases). However, the difference in the median number of episodes between the youngest and oldest groups at each time-point was small (a difference of 1 for improvements of work experience, a difference of 1 for career advice at the 6-month assessment and 2 at the 12-month assessment). Changes in the number of work-experience and career-advice episodes were not influenced by the agegroup.

Whilst the proportions of adolescents with vocational plans and performing household activities increased over time, these changes were not statistically significant. The proportions of adolescents who had vocational plans increased with older age-group at baseline (54.4, 65.8 and 88.5% for the 11-, 14- and 17-yr-olds, respectively,  $P<0.001$ ), and 6 months (54.0, 79.7 and 84.2%,  $P=0.001$ ), but not at 12 months (64.5, 75.0 and 92.3%,  $P=0.047$ ). Household chores, however, did not vary significantly by age-group.

#### '17 yr-old pseudo-control group' comparative data

**Primary Outcome data (HRQL).** In terms of the comparison group, the 17-yr-olds had worse HRQL than the other age-groups at every time point, although these differences did not reach significance. At the end of the study, all groups had significantly improved and the gap between the median JAQQ scores of the 17-yr-olds and the younger groups had decreased (from 0.8 to 0.3).

**Secondary outcomes.** At baseline the 17-yr-olds had a median knowledge score of 9, which was significantly higher than the scores of younger groups. However, by the end of the study, 14-yr-olds had a median knowledge score of 11, which was significantly better than the 17-yr-old baseline score ( $P=0.005$ ).

Whilst satisfaction had improved, there were no significant differences between the age-groups and whilst the median satisfaction scores of the 11- and 14-yr-olds at follow-up were better than what the 17-yr-olds had at baseline, this difference was not statistically significant.

With respect to independent behaviours, the overall levels of self-medication and independent consultations did not significantly improve. However, at the end of the study, the proportion of 11-yr-olds who were self-medicating had increased and was no longer significantly lower than the older groups. Moreover, there were no statistical differences between the proportion of 11-yr-olds who were self-medicating (66.7%) and the 17-yr-old control group (80.0%). This was not the case for independent consultations, where after 12 months, the 11- and 14-yr-olds remained less likely to be seen alone compared with the 17-yr-old control group.

With respect to vocational markers, the 17-yr-old baseline scores for number of work experience and career-advice were significantly better than the 11- and 14-yr-olds' scores at 12 months, despite their improvement from baseline.

#### Who stayed in the programme?

There were no significant differences in baseline characteristics between the adolescents who participated at 6 months and those who withdrew following recruitment and/or had incomplete data. There were similarly no differences between parents, with the exception that parents who completed assessments at 6 months had greater baseline knowledge than those who had incomplete data (respective median scores of 11 and 10,  $P=0.005$ ).

Due to time constraints and the use of multiple baselines, only 232 (75.6%) adolescents were eligible for a 12 month assessment. Of these, complete data were available for 121 adolescents and 108 parents. Reasons for incomplete data included no clinical indication for review within the designated follow-up period, limited capacity of service for follow-up, non-adherence to appointments and/or true 'withdrawals'. When compared with

those with incomplete data ( $n = 111$ ), adolescents who completed the study were more likely to have a higher socio-economic classification ( $P = 0.002$ ). However, there were no significant differences with respect to any other demographic characteristics or core outcome variables.

In terms of the primary outcome variable, there were no statistically significant differences in baseline HRQL between those adolescents who completed the study and those who did not (respective median adolescent-rated JAQQ scores of 3.2 and 2.6,  $P = 0.039$ , and median parent-rated JAQQ scores of 3.3 and 2.9,  $P = 0.111$ ). Nor was there any significant difference in the amount of change in HRQL at 6 months for those who completed the study compared with those who did not. This was true for both adolescent JAQQ scores ( $-0.4$  and  $0.0$ ,  $P = 0.023$ ) and parent-proxy scores ( $-0.4$  and  $-0.1$ ,  $P = 0.017$ ).

With respect to the secondary outcome variables, adolescents with complete data had significantly better baseline knowledge than those who had incomplete data (respective median scores of 8.0 and 7.0,  $P = 0.003$ ). However, there were no differences in parental knowledge scores (respective median scores of 11.0 and 11.0,  $P = 0.156$ ). Comparison between those who completed the study and those who did not, also showed no significant differences in the 6 month improvement for both adolescents (2.0 and 2.0,  $P = 0.964$ ) and parents (1.0 and 1.0,  $P = 0.216$ ). In terms of satisfaction, there were no differences between those who completed the study and those who did not for either adolescent baseline scores (0.4 and 0.3,  $P = 0.665$ ) or parental baseline scores (0.6 and 0.8,  $P = 0.234$ ). Nor were there any significant differences in improvement in satisfaction at 6 months for adolescents ( $-0.3$  and  $-0.1$ ,  $P = 0.431$ ) or parents ( $-0.4$  and  $-0.5$ ,  $P = 0.451$ ).

There were no significant differences between those who stayed in the PTC and those who left, for baseline work experience or career advice, nor the changes in these at the 6 month assessments.

## Discussion

This study is the first to provide objective data of the immediate potential impact of a PTC [22] with respect to adolescents with a chronic illness and their parents. The beneficial impact is reflected by the improvement in adolescent HRQL, adolescent and parental knowledge and satisfaction, and pre-vocational readiness markers (Table 3).

### *Predictors of improvement in HRQL*

The key predictors of improvement in HRQL in adolescents with JIA, irrespective of age, identified in this study were functional ability (CHAQ) and pain (Table 4). These are the internationally accepted core outcome variables in JIA [27] and such data support the routine documentation at clinic visits of these variables and highlights the importance of addressing their management within a PTC.

The contribution of self-medication and independent clinic visits with improvement in adolescent HRQL is interesting and supports the philosophy of transitional care, which aims to 'promote skills in communication, decision-making, assertiveness, self-care and self-advocacy' [32]. The duration of this study may have been too short to observe significant changes in self-advocacy, which potentially varies significantly with the individual and family. Independent visits have been identified as an 'important and highly feasible component of best practice', in an earlier Delphi study of users and providers of transitional care in JIA, during the needs assessment phase of this project [16], and were specifically recognized as important by the adolescents themselves [14]. Reid *et al.* [33] also reported that independent visits were a predictor of successful transfer in a cohort of adolescents with congenital heart disease. Independent healthcare behaviours are particularly pertinent during adolescent

development as this is the time when young people become new users of health services. Furthermore, expectations of self-management within adult healthcare are higher than in paediatrics and therefore an important aspect of preparation for transfer. In adolescent diabetes, coping skills training has been reported to be associated with improved disease control, better self-efficacy and less impact of HRQL [34]. Finally, independent visits by the adolescents, however, may also have influenced the reduction in parental completion rates at follow-up.

### *Who benefited most from the PTC?*

The lack of major differences between the age groups would suggest that all ages benefit from a PTC, even the 17 yr-olds who are about to enter adult care. The comparison between the 11- and 14-yr-olds at follow-up compared with the baseline 17-yr-old data in knowledge would support the philosophy of starting a PTC early in adolescence [32]. The greater change in JAQQ scores in older adolescents and those who had developed JIA later supports the need for further developments specifically in adolescent rheumatology.

### *Limitations*

It could be argued that the observed changes in HRQL cannot be definitively attributed to participation in the PTC and may simply reflect improvement in disease activity. There was however no change in markers of disease activity (e.g. number of active joints, ESR/CRP, physician's global assessment of overall disease activity) over time and neither did such markers come out in the regression as explanatory variables of the variance in HRQL. Transitional care, by definition is not exclusive of medical aspects of care, and therefore includes control of disease activity. Furthermore, delivery of transitional care is limited during active flares of disease [32] and is more appropriately delivered during periods of remission when young people are likely to be more receptive to such interventions.

The observed improvements were also seen across all the domains which might simply reflect greater maturation. Conversely, lack of observed improvement could also represent delayed maturation as a result of a chronic illness. The spectrum of improvement observed may also reflect the comprehensive nature of the PTC, which was designed to address all domains and based on an extensive needs assessment [14–16, 21].

The lack of a true control group and the challenges of performing a randomized controlled trial in transitional care have already been discussed in the 'Methods' section. Normative data regarding certain independent behaviours and vocational readiness are also not readily available for comparison and definitive attribution of the PTC in these areas cannot be specified. For example, the provision of work experience within the national curriculum in the UK may simply reflect the observed increases in vocational readiness at follow-up. However, one could argue that young people with chronic illnesses need more career counselling compared with their healthy peers acknowledging the reported unemployment rates in young adults with JIA, and 14% of the 17-yr-olds who had had no such input at baseline remain a cause for concern [17]. Although the changes in vocational readiness markers were not large, they were statistically significant and encouraging given the short-time framework of the study and we would hope to see further improvement over time. Moreover, the data reveals that some improvement is possible even for those 17-yr-olds who are just about to exit the transitional care programme.

The lack of a true comparison group also made it impossible to control for non-specific effects of potential confounding variables such as the staff-patient relationships, clinic settings, etc. thereby determining which components of the PTC influenced the HRQL. The impact of such effects are illustrated by the reported improved documentation of transitional care issues in a case-note audit

performed in the participating centres and including patients *Not* recruited to the programme in the participating centres [35].

In the final regression model for adolescent JAQQ change score, only 27.6% of the variation was explained (Table 4). Other non-disease-related factors which were not objectively measured e.g. specific PTC components, time spent with the young person, etc. are other potential influences worthy of further consideration and study. A further example is the potential impact of the 'Hawthorne effect' i.e. the generic professional contact with the LPC, a nurse or allied health professional, funded for 1 day per week during the study, was as effective as the specific intervention. In support of this, adolescents reported greater acceptability of the LPC role compared with the paper-based resources ( $P < 0.001$ ) [22]. Provider behaviour has been reported to be important in adolescents' satisfaction with health care provision [36]. The communication skills of the adolescents and the health professionals in the educational 'encounters' were also not assessed. Additionally, no data were collected on the amount of time allocated for the PTC, a resource that may be determined by the professional and the health care system rather than the patient. Both of these factors have been shown to be important in adolescent clinics [21, 37] and are potential limitations of this study.

Another important limitation of this study is the limited completion rates at follow-up. However, the sample size of 308 still had 74% power to detect an effect size of 0.15 and 90% power to detect an effect size of 0.185. The limited completion reflects the use of multiple baselines, the short timescale due to limited available funding, limited flexibility in service to ensure reviews within study period in addition to the generic challenges of clinical research with this age-group [38] including non-adherence to appointments [39], particularly when their disease is quiescent, and/or adolescents choosing to attend the clinic independently of their parents. Ideally, analyses would have been performed when all the enrolled patients had completed a 12 month follow-up in order to maximize the results, but unfortunately this was not possible within the funding constraints of this study.

As well as being a relatively new area of research, transitional care development in the clinical setting (as the rest of adolescent healthcare) is only in its infancy in the UK [1–4, 13, 20]. The multidimensional and multidisciplinary nature of transition in addition to the time requirement to detect change presents further challenges in developing an evidence base within a time and financially limited research project. Furthermore limited clinic time was highlighted as one of the three main barriers to the provision of developmentally appropriate transitional care prior to commencement of this study [21] which was conducted within the National Health Service rather than research-based clinics. As mentioned earlier, many variables studied are likely to take longer than 12 months to both change and/or result in measurable outcomes e.g. prevocational markers, actual employment, etc. Furthermore, whereas there may be a significant improvement following entry to the PTC, subsequent changes may be more gradual thereafter, reflecting the process rather than event nature of transition. Notably, in author's centre, of the 53 adolescents recruited, preliminary data reveals at 24 months since the completion of the project, that 28 have been successfully transferred to adult care, five have been discharged from hospital care, 19 have individualized transition plans in progress and one is currently lost to follow-up [40].

The differences in the sample at 12 months compared with baseline raise important questions requiring further consideration. Did those who failed to complete the study perceive that the programme was less relevant to them, or was it a lack of professional skill of engagement of such young people from the outset in that they tended to be from a lower socio-economic class or was it simply a marker of limited capacity of the service to review participants within the time available? This group also had lower baseline knowledge and if follow-up data had been available

on those who were still participating in the programme, but had just not been reviewed within the study period, the improvements may have been greater.

Finally, there remains a need to address resourcing issues and the cost-effectiveness of PTCs. Acknowledging the financial climate of the health service, the PTC in this study utilized (where available) the currently available resources in the UK [22], existing members of the rheumatology team, other statutory services (e.g. [www.connexions.gov.uk](http://www.connexions.gov.uk)) and the voluntary sector (e.g. [www.ccaa.org.uk](http://www.ccaa.org.uk), [www.arthritiscare.org.uk](http://www.arthritiscare.org.uk)) with an emphasis on further signposting of additional resources for both professionals and young people.

## Conclusions

This study reports a positive impact across adolescence of an evidence-based, structured PTC [22] developed in response to a prior needs assessment in the target patient group [14–16, 21]. It is the first such study in any chronic illness and compared with baseline, improvements were noted in most outcome measures (HRQL, knowledge, satisfaction, vocational readiness markers), irrespective of age with baseline functional disability, self-medication and pain predicting improvement. In view of the generic nature of transitional care issues [7], this study has important relevance to other childhood onset chronic illnesses beyond those within rheumatology. The challenges of transitional care research and data collection with adolescents have been highlighted by the limitations of the study results. The initiation and maintenance of a structured-PTC for adolescents with JIA is therefore likely to be effective in the short-term but cost implications and medium and long-term outcome remain unresolved.

**Contributors:** Eileen Baildam, Jeremy Camelleri, Diane Coulson, Joyce Davidson, Sue Ferguson, Helen Foster, Paul Galea, Janet Gardner-Medwin, Janine Hackett, Ann Hall, Gill Jackson, Jane Kelly, Sue Kemp, Nicki Kennedy, Ruth McGowan, Steph Phillips, Clarissa Pilkington, Alison Swift, Helen Venning, Tracey Whitely, Jane Wilby, Sue Wyatt, Helena Wythe.

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