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Title

Economic evaluation of a trial exploring the effects of a web-based support tool for parents of children with Juvenile Idiopathic Arthritis.

Authors

Chris Flood[1 & 2*], Shashivadan P Hirani[2], Kathleen Mulligan[2,3], Jo Taylor[2], Sally Harris[4], Lucy R Wedderburn[5,6,7], Stanton P Newman[2], on behalf of the WebParC Investigator group[8]

[1] Institute of Health and Social Care, London South Bank University

[2] Centre for Health Services Research, School of Health & Psychological Sciences, City, University of London, London, United Kingdom

[3] East London NHS Foundation Trust, London, United Kingdom

[4] University Hospitals Sussex NHS Trust, Brighton, United Kingdom

[5] University College London Great Ormond Street Institute of Child Health, University College London, London, United Kingdom

[6] Centre for Adolescent Rheumatology Versus Arthritis at University College London Great Ormond Street Institute of Child Health, University College London, United Kingdom

[7] National Institute for Health Research Biomedical Research Centre at Great Ormond Street Hospital, London, United Kingdom

[8] See Acknowledgements

Corresponding author: Prof Stanton Newman Centre for Health Services Research, School of Health & Psychological Sciences, City, University of London, London, United Kingdom EC1V 0HB (Stanton.Newman.1@city.ac.uk**) https://orcid.org/0000-0001-6712-6079**

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Abstract

Objective

To explore the cost-effectiveness of a web-based support tool for parents of children with Juvenile Idiopathic Arthritis.

Methods

A multi-centred randomised controlled trial was conducted in paediatric rheumatology centres in England. The WebParC intervention consisted of online information about JIA and its treatment and a toolkit using cognitive-behavioural therapy principles to support parents manage their child's JIA. An economic evaluation was performed alongside the randomised controlled trial involving 220 parents. The primary outcome was the self-report Pediatric Inventory for Parents measure of illness-related parenting stress, with two dimensions; difficulty and frequency. These measures along with costs were assessed post intervention at 4 months and at 12 months follow up. Costs were calculated for healthcare usage using a UK NHS economic perspective. Some data was also collected and analysed on the impact of caring costs on families. Uncertainty around cost effectiveness was explored using bootstrapping and cost-effectiveness acceptability curves.

Results

At 4 months, the intervention arm showed improved Pediatric Inventory for Parents (PIP) scores with a mean difference between trial arms in the dimensions of frequency and difficulty scores of 1.5 and 3.6 respectively. At 12 months, the mean difference between trial arms in frequency and difficulty scores were 0.35 and 0.39, again representing improved PIP scores for the intervention arm.

At both 4 and 12 month follow up the average total cost per case was higher in the control group when compared to the intervention arm with mean differences of £360 (95% CI £29.6 to £691) at 4 months and £203 (95% CI £16 to £390) at 12 months.

At both 4 month and 12 month follow up, the majority of costs associated with health service use were found to belong to secondary care for both arms.

Cost data was further analysed in combination with effectiveness data from the Pediatric Inventory for Parents measure. The probability of being cost effective ranged between 49% and 54%.

Conclusion

The WebParC online intervention led to reductions in primary and secondary healthcare resource use that translated into reduced costs at 4 and 12 months. The intervention demonstrated particular savings for rheumatology services at both follow ups. Future economies of scale could be realised by health providers with increased opportunities for cost effectiveness over time.

Keywords

Juvenile arthritis, cost effectiveness, parent support, telehealth, economic evaluation.

Key Messages

1. This study reports on a relatively low-cost, online digital intervention for reducing parental stress.
2. Cost savings within the National Health Service were achieved particularly within rheumatology services.
3. Further reductions in parental stress and online support operating costs could extend cost effectiveness.

Introduction

Parents with a child diagnosed with Juvenile Idiopathic Arthritis (JIA) have to manage their child's medication, pain, distress, physical difficulties, impact on schooling, hospital visits, time off work and financial issues. Parents of children with chronic illness can experience considerable stress, more than those with healthy children. Early intervention to support parents may facilitate better outcomes for their children with JIA The previously published trial paper (1) associated with the economic evaluation reported in this paper, demonstrated that web-based interventions for parents of children with JIA can result in significant benefits, in terms of reduced illness-related parent stress. Using e-health as a vehicle to support parents may be an effective tool providing easy access to support. WebParC is a website developed for parents of children with recently diagnosed JIA to complement usual clinical care with the aim of helping parents cope with the stress of managing their child's illness. The WebParC web-based tool was specially designed to provide information, support and practical skills in dealing with carer challenges.

The aim of this study was to determine whether the WebParC online tool is cost-effective in comparison to standard management.

Methods

Study design

An economic evaluation was undertaken alongside a two-arm multi-centre RCT in sixteen National Health Service tertiary paediatric rheumatology services in England. Details of the study are available on the ISRCTN registry at http://www.isrctn.com/ISRCTN13159730. Ethical approval was obtained from the Health Research Authority London Bridge Research Ethics Committee, reference 13/LO/0288. This paper reports the economic evaluation of WebParC with the clinical study and outcomes reported elsewhere [1]. Participants were recruited at paediatric rheumatology outpatient clinics. Inclusion criteria for participants included parents that were aged ≥18 years, with a child aged ≤12 years who had been diagnosed with JIA within the previous 6 months. Parents needed to speak and read English. Other exclusion criteria included the presence of a current severe mental illness such as identifiable psychosis in parents, a major problem with literacy making the questionnaire completion impossible, or likely to be distressed by the study as judged by their child's rheumatologist. Lastly those without internet access were also excluded. For contextual information table 1 details participant characteristics at baseline.

For the control arm of the study child participants with JIA and their parents continued to receive standard clinical care, with no alternative educational intervention provided. In addition to standard care, the intervention arm gave access to a specially designed website for parents. To avoid biasing clinical staff, randomisation was concealed from clinical teams. A blocked randomization procedure was performed using computer-generated random allocation sequences. Block sizes were varied, so that study site staff could not guess which group a block had been randomized to.

Website content was written by a multidisciplinary team of healthcare professionals, comprising 13 rheumatologists, four rheumatology nurse specialists, two clinical psychologists, two physiotherapists, a podiatrist, occupational therapist, ophthalmologist and social worker, with research assistant support. The website content included information about JIA, its treatment, a JIA toolkit (based on cognitive-behavioural therapy principles aimed to help promote coping), problem-solving communication with family members and pain management support.

The primary outcome was parental stress at 4 and 12 months post randomisation, measured with the Pediatric Inventory for Parents (PIP) [2], a validated measure to assess difficult events that parents may face.

Perspective taken.

A UK NHS economic perspective was adopted, with some consideration given to costs impacting on carers. Undiscounted costs are reported given time frames did not exceed more than a year. Costs are reported in £s sterling.

Resource use data collected and costing analysis methods.

Resource use was collected using the Client Service Resource Inventory (CSRI) [3] (paper based and online) at 4 and 12 months post intervention to understand the potential impact of the web-based support on different types of health care services used, and any expenditure incurred on families arising from having a child with JIA. Participants provided information

about what services they had used in the prior 3 months at both the 4 and 12 month follow ups. Data was collected for each parent participant for a child with JIA.

Resource use collected included hospital outpatient appointments and/or admissions. Additional data collected included different types of health care services used by participants and where the resource-use would impact, for example GP and practice nurse appointments within primary care. For secondary care resource use, data collected included contact time with professions such as rheumatologists, rheumatology nurse specialists, ophthalmologists, physiotherapists, occupational therapists and psychologists. Also collected were resources associated with specialist joint injections and urgent care in the form of hospital visits to accident and emergency. The number of appointments/visits/or occasions participants had contact with each of the relevant professions was recorded for all resources used and these were then entered into an Excel spreadsheet.

Individual participant admission costs were estimated by collating the number of admissions for an individual along with each recorded length of stay in days. This total number of days was then multiplied against the relevant officially published reference or unit cost for an admission day. The unit costs applied to the totals for resources used by participants were measured in UK pound sterling (£) for the years 2017-2018. Unit costs were derived from the National Schedule of Reference costs (2017/2018) [4], or from 2017 Unit Costs of Health and Social Care [5].

Unit costs were applied to each resource use variable to calculate the individual total participant costs or total cost per case, across both the control and intervention arms. A total average cost per trial arm could thereafter be calculated. Overall costs and averaged costs for

each of the health care resources used were calculated along with standard deviations and the percentage of the total overall cost for each. The mean difference in costs between trial arms were reported with 95% confidence intervals and *p* values showing statistically significant differences in costs where they occurred.

As part of detailing the intervention costs, specialist intra-articular joint injections of Triamcinolone Hexacetonide, a key therapy used to treat JIA, were also reported. Typically the dose of this drug varies from anything from 10 mg for one small joint to 100 mg for up to 4 joints. Unit costs of £120 per ampoule were provided by the Great Ormond Street Hospital finance team and verified on the National Institute for Health and Care Excellence/British National Formulary medicinal forms webpages [6]. According to advice from the finance team in the lead Trust, these administration costs included the cost of; pathology tests; pharmacy staff in the clinical units; pharmacy department staff in the dispensing team; radiology tests; theatre use; specialty overheads and the costs for occupied bed days. The procedure for administering the injections was described as taking on average about 15-20 minutes in total and this was factored into the resources used and the final cost calculation. The average cost for the injections based on these procedures and staff involved was calculated to be £1,060.

Costs associated with the setup of the WebParC website included IT consultant time and advice, annual website support, user testing sessions, continued ongoing hosting of the website and data back-ups costs. These costs were only applied to the intervention arm.

To assess the impact of caring for a child with Juvenile Idiopathic Arthritis on carers, out-of-pocket costs borne by parents and carers across trial arms were also calculated. Using the

CSRI questionnaire parents were asked to report their ability to work and whether they were receiving any benefits or allowances. Respondents were asked to record what typically these amounts might total in a month.

Missing data are very common in cost-effectiveness analyses where health outcome data and costs, involve using multiple variables. Multiple imputation was used to impute any missing data using the multiple imputation function in SPSS (2020) [7] and were based on variable data that was available. Item level variable data (as opposed to aggregated data at the participant level) for both the 4 month and 12month time points were imputed. This entailed creating multiple data sets (10) for each variable [8], [9]. Data values generated from the multiple data sets were pooled and combined to produce overall average values to use as the imputation values for any of the missing values associated with PIP and resource use data variables.

Cost effectiveness analyses

Further cost effectiveness analyses were performed whereby clinical outcome data was combined with the cost analyses. The previously described PIP measure was used for the effectiveness part of the cost effectiveness calculations. The PIP measure with its two dimensions of difficulty and frequency, when combined with the cost data provided two cost effectiveness outputs; namely cost per unit change in difficulty associated with dealing with stressful events and cost per unit change in frequency associated with stressful events.

To calculate the two PIP scores for experienced difficulty and frequency, a change in score from baseline to the first follow-up period and a change in score from the first follow up to the second follow time point were conducted.

Scores from the two sub scale parts of the scale were used in combination with the resource and cost use data to determine Incremental Cost Effectiveness Ratios (ICERs). An ICER essentially measures the difference in average costs between the trial arms divided by the differences in average effects and create a point estimate for helping to understand overall cost-effectiveness. The ICER is therefore the cost of providing an extra unit of health outcome improvement and helps decision makers weigh up if the extra cost at which the new intervention (potentially) buys an extra unit of health outcome is deemed acceptable. In such circumstances a new intervention may be recommended. In contrast, circumstances may also arise where a new model of care could be 'dominated' by existing services if it costs more and provides less 'quality of life', 'gain' or 'improvement.'

The combined distribution of costs and effects once analysed, are plotted on a cost effectiveness plane. This plane is divided into four quadrants with each quadrant having a different implication for economic evaluation decision making. For example, if all the combined data falls into the south east quadrant, this would reflect negative costs and positive effects and therefore represent an intervention which is more effective and less costly than the control group. In these circumstances the intervention would be considered cost effective. In reality the combined data can fall across any or all of the quadrants on the scatter plot, creating uncertainty about whether or not the intervention is cost-effective or not.

Whilst it is correct to refer to cost effectiveness planes and ICERs in any primary analysis, (with their calculation being useful for establishing an initial sense of the data and point estimates), data presented in this way can be easily misinterpreted. For example, an intervention that is less expensive and more effective will generate a negative ICER, but so will an intervention that is more expensive and less effective. Furthermore, there can be additional difficulties due to the ratio statistic nature of an ICER and the need to produce confidence intervals [10]. A well-established statistical solution to the problem of estimating confidence limits for ICERs is the use of nonparametric bootstrapping. This technique produces the cost effectiveness acceptability curve (CEAC), which has become a mainstream part of economic evaluations that support decision-makers facing uncertainty about the efficacy of new healthcare treatments.[11].

In the WebParC study, we used repeat re-sampling from the costs and effectiveness data using non-parametric bootstrapping to generate a distribution of mean costs and effects for the two trial arms. Statistical sensitivity analysis was conducted by estimation of confidence intervals for mean difference in costs and effects per participant, and by plotting cost-effectiveness acceptability curves. Cost-effectiveness acceptability curves allow decision makers to assess the overall probability of an intervention being cost-effectiveness given particular thresholds. Typically, NICE uses a threshold of between £20 to £30 K per QALY with some [12] proposing that such a threshold should increase to £100k for rare conditions.

All costing and cost effectiveness analyses with their produced tables and figures were generated using Excel (2018) [13] with the exception of the multiple imputation calculations which used SPSS software (2020, version 27) [7] to replace missing values.

Results

Resources use and cost outcomes

Baseline data collected and analysed showed no statistically significant differences in costs between the trial arms or for any of the individual service use variables used to establish costs. At 4 months and 12 months the same health services resource variables were again converted into cost data and are presented in tables 2 and 3. These tables show the differences respectively between trial arms in terms of average costs per participant for health service costs. In the 4 month and 12 month follow up tables, costs associated with health service use are further divided into primary care costs and secondary care costs associated with the types of resources used. For example, GP and practice nurse appointments represent the primary care costs and secondary care costs are associated with appointments and sessions related to other staff, including rheumatologist, rheumatology nurse specialist, ophthalmologist, physiotherapy, occupational therapy and psychologist time. Urgent care in the form of Accident and Emergency attendance and the intervention costs are reported separately within the tables.

Rather than rely on a complete case analysis approach, where data gaps were found, or the assuming of zero costs, multiple imputation methods were used [8,9] to achieve a full data set. Prior to multiple imputation methods being used, 64.5% of participants showed some data gaps and 44.5% of data on average was recorded as not having had data returned completed.

Data analysed at 4 months showed statistically significant differences (p= 0.03) between the total mean costs between the intervention and the control group. The total cost per case was higher in the control compared to the intervention arm by £360 (95% CI £29.6 to £691). Secondary care costs represented the majority of the overall costs in both arms (84% for the intervention and 94% for the control allowing for rounding of figures).

This analysis indicated that there were statistically significant cost differences between standard care and the WebParC intervention at 4 months related to the specialist consultations with the rheumatologist consultant, rheumatologist nurse specialist appointments, physiotherapy, occupational therapy and visits to accident and emergency. Overall the other costs borne by primary and secondary care service were all lower (though not statistically significantly so) for the intervention arm compared to control conditions, (with the exception of ophthalmology and psychology where mean differences in costs were marginally higher in the intervention arm by £1 and £4 respectively).

At 12 months the analysis showed statistically significant cost differences between the two trial arms with mean differences in costs of £203, 95% CI £16 to £390, p=0.04). Consultations with the consultant rheumatologist, the nurse specialist appointments and appointments with a psychologist still showed cost differences that were statistically significantly lower for the intervention arm of the study. With the exception of resource use and costs associated with GP visits, (which were more or less equal across trial arms; mean cost per participant £27 as compared to £26 for the control arm of the study), all other primary and secondary care resource variables had lower costs for the WebParC intervention arm. Differences in these variables were not statistically significant. As with the 4 month follow up, the majority of costs

associated with resources used at 12 months were found to belong to secondary care for both arms (76% for the intervention and 92% for the control) of the study.

In terms of any economic impact on carers, data showed a reduction in absence from work due to less Juvenile Idiopathic Arthritis related illness episodes (on average 3 days less and 2.5 days less for the intervention arm at 4 and 12 months respectively), as well as a reduction, although not statistically significant, in the use of state benefits (£273 per month on average and £164 per month on average) associated with the control group and the intervention group respectively at the 12 month follow up.

Cost effectiveness analysis results

The cost-effectiveness planes in Figures 1 and 2 show the distribution of 1000 replicates of cost and effects for the WebParC intervention versus care as usual at 4 and 12 months follow up. The data uses a change in both the PIP sub scales score for stress frequency and associated difficulties alongside a change in costs to determine (cost) utility scores. As can be seen replicates are distributed across all four quadrants on both the cost-effectiveness planes, with the brown squares representing the frequency scores combined with cost data whilst the blue circled data points represent data from the difficulty scores combined with cost data.

At 4 months, the mean difference between trial arms in their frequency and difficulty scores were 1.5 and 3.6 respectively, representing improved PIP scores for the intervention arm. These scores along with mean differences in costs between trial arms of -£360, reflecting lower costs in the intervention arm lead to an ICER point estimate of -£241 for frequency and -£99 for difficulty.

At 12 months, the mean difference between trial arms in their frequency and difficulty scores were 0.35 and 0.39 respectively, again representing improved PIP scores for the intervention arm. These scores along with mean differences in costs between arms of -£203, again reflecting lower costs in the intervention arm provide an ICER point estimate of -£587 for frequency and -£525 for difficulty.

Cost-effectiveness acceptability curves are shown in Figures 3 and 4 based on the analysis at 4 months and 12 months using both the PIP subscale scores for stress and associated difficulties. Again brown (lines) represent the frequency scores combined with cost data whilst the blue lines represent data points from the difficulty scores combined with cost data. These analyses indicate that the probability of the WebParC intervention being cost-effectiveness was between 49% to 54% at different thresholds of expenditure up to £50,000.

Discussion

Previous research has called for economic evaluations of digital approaches and telemedicine in paediatric health care [14]. To the authors' best knowledge this is the first cost-effectiveness analysis of an internet-based support program for parents of children with JIA. This economic evaluation contributes to a relatively limited research literature using online support for parents with children with long term conditions, particularly those with arthritis. The findings reported indicate that a web-based tool, used for supporting parents of children with Juvenile Idiopathic Arthritis led to reductions overall in the use of primary and secondary care services at 4 and 12 months.

Juvenile arthritic disorders are costly to treat. Treatment often involves numerous health care professionals' time and services, medication costs and frequent outpatient visits. There are periods of hospitalization, possibly the need for surgery, medical equipment costs and in particular costs associated with aids and appliances. All of which can contribute to high financial costs for health care systems especially when there are children with particularly complex care requirements.

The results of this study show the potential for relatively low-cost support services to be delivered online. Whilst costs would be associated with maintaining and updating webpage support and online materials to support families, future economies of scale could be achieved, across greater numbers of health care organisations choosing to share costs. Cost benefits may accrue with longer time frames, where reductions in hospital admissions and overall disability especially for younger patients and their families are realised.

Web based health technology interventions have a potential to reduce social and economic burden on parents and carers allowing them to better manage their work and caring responsibilities. Technology developed in this way may also not be so susceptible to the same constraints of availability of staff and these factors overall may increase equity of access.

Strengths and limitations of this study

This economic evaluation was part of a multi-centre trial which increases the external validity for the study findings [15]. With the exception of the inclusion of the EQ-5D, this study adhered to the NICE recommendations [16] for economic evaluations in health technology assessments.

A limitation of this study includes recruitment criteria that excluded certain participants on the basis of; language (non-English speakers), a mental health or learning disability or not having access to an internet connection. Access to the WebParC website was available via smartphones, although often those from lower income households are also less likely to have access to smartphones, whilst having more expensive mobile phone contracts and data plans [17], adding another potential barrier to accessing information online.

Future research

Reduced access to digital technologies and widening inequalities is already of concern [18]. Future study designs exploring the cost effectiveness of web-based support, should aim to maximise the most inclusive study participation possible to improve the generalisability of findings. Thereafter future economic evaluation designs will be well-positioned to explore any

extra cost associated with additional support, including the translation of instruments for data collection and online materials.

Recent literature also points to the limited use of methods used to generate utility values in NICE appraisals from the child and adolescent population [19,20]. Future research could consider measuring quality of life directly from children and adolescents with JIA using the EQ-5D-Y [20]. This would not be without its challenges as younger populations have considerable additional life years available for the most part, increasing estimates of quality-of-life gains in any economic evaluation, possibly limiting comparison for any QALY gains to studies with other younger populations. Future economic evaluations of JIA could also consider using the EQ-5D to measure carer quality of life.

Conclusion

At 4 and 12months cost differences between trial arms were particularly notable for the rheumatology services. These cost savings show the potential to provide supportive online care whilst releasing opportunity costs to develop service provision elsewhere within the specialty. When costs and effects were combined and analysed jointly, they did not show the WebParC intervention to be conclusively cost-effective. Nonetheless it would be wrong to simply interpret these findings as a negative. The probability of the service being cost-effective overall ranged from 49% to 54% with future cost-effectiveness potentially being driven by further reductions in parental stress and operating costs.

Supplementary material

Supplementary information to support the main paper is available at *x (advised by journal editors)* and provides more detailed explanation around the study design, measures and analysis and some background context to economic decision making in health care.

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Author contributions

All authors were involved in drafting or revising this article and all approved the final version. Dr Chris Flood led on the economic evaluation data design and data analysis and first drafting of the paper.

Study conception and design

Stanton Newman, Kathleen Mulligan, Shashivadan Hirani, Lucy Wedderburn, Sally Harris,

Economic evaluation design

Chris Flood.

Acquisition of data

Jo Taylor, Kathleen Mulligan.

Analysis and interpretation of data

Chris Flood, Shashivadan Hirani, Kathleen Mulligan, Stanton Newman.

Conflicts of Interest

None declared

Data Availability Statement

Data underlying this article were provided by City, University of London and the National Health Service under licence / by permission and may not be shared publicly for ethical/privacy reasons. (e.g. for the privacy of individuals that participated in the study and gave consent to use their data only in specified ways). Summary data may be shared on reasonable request to the corresponding author with permission of these third parties.

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