Abstract

Context: Integrated care models may improve health care for children and young people (CYP) with ongoing conditions.

Objective: To assess the effects of integrated care on child health, health service use, health care quality, school absenteeism, and costs for CYP with ongoing conditions.


Study selection: Inclusion criteria consisted of (1) randomized controlled trials, (2) evaluating an integrated care intervention, (3) for CYP (0–18 years) with an ongoing health condition, and (4) including at least 1 health-related outcome.

Data extraction: Descriptive data were synthesized. Data for quality of life (QoL) and emergency department (ED) visits allowed meta-analyses to explore the effects of integrated care compared to usual care.

Results: Twenty-three trials were identified, describing 18 interventions. Compared with usual care, integrated care reported greater cost savings (3/4 studies). Meta-analyses found that integrated care improved QoL over usual care (standard mean difference = 0.24; 95% confidence interval = 0.03–0.44; \( P = .02 \)), but no significant difference was found between groups for ED visits (odds ratio = 0.88; 95% confidence interval = 0.57–1.37; \( P = .57 \)).

Limitations: Included studies had variable quality of intervention, trial design, and reporting. Randomized controlled trials only were included, but valuable data from other study designs may exist.

Conclusions: Integrated care for CYP with ongoing conditions may deliver improved QoL and cost savings. The effects of integrated care on outcomes including ED visits is unclear.
Child mortality has decreased markedly over the last 50 years, and the epidemiological transition toward chronic conditions, such as asthma and neurodevelopmental disorders, applies across many countries and regions.\(^1\) These broad population-level changes mean that health systems and health care models must adapt accordingly, providing high-quality care for all childhood conditions.

International data indicate that current models of care are inadequate and unsustainable. In England, there was a 58% rise in children and young people (CYP) attending hospital emergency departments (EDs) from 2007 to 2016,\(^6\) with a further 50% to 60% increase projected by 2030.\(^7\) Eighty-five percent of current ED attendances by CYP are of low severity,\(^8\) manageable in primary care, and the number of children presenting to EDs with minor ailments increases 5% annually.\(^9\) In the United States, only 47% of clinical contacts adhere to clinical guidelines across 11 conditions (2007)\(^10\) and 60% across 17 conditions in Australia (2018).\(^11\)

Authors and policy makers have argued that the current health care model, in the context of the wider health and social system, delivers suboptimal health outcomes for children.\(^12\)–\(^15\) However, new models of integrated care are largely focused on adults, with adults’ needs in mind, including multiple comorbidities and frailty.\(^16\) Definitions of integrated care vary, but consistent themes that apply to any population include person-centered care delivered by professionals and organizations cooperating effectively. Integrated care is thought to increase the patient focus of care and deliver more cost-effective health care.\(^17\) For adults, integrated care has been associated with positive outcomes, with some evidence for reductions in hospital activity and costs.\(^18\)–\(^24\) For children, there are reviews and meta-analyses examining specific conditions or circumstances, for example by Asarnow et al\(^25\); however, we found no systematic reviews examining evidence for comprehensive broad population-based integrated care for CYP. In this systematic review, we assess the impact of integrated care interventions for CYP with ongoing conditions on important outcomes for children including health, health service use, health care quality, education, and costs.

**METHODS**

We conducted a systematic literature search (1990–2018) following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines. The protocol for this review was registered in the International Prospective Register of Systematic Reviews (CRD42016045215).

Two authors completed the search in July 2017, which was updated in September 2018. Five databases were searched: Medline, Embase, PsycINFO, Cumulative Index to Nursing and Allied Health Literature, and the Cochrane Library. Two authors independently screened titles, abstracts, and full texts against the inclusion criteria. A third author informed final decisions when disagreements occurred.

Search criteria comprised 3 categories: (1) population (age, condition), (2) intervention (integrated care), and (3) study design (see Supplemental Information for the full search strategy). Additional articles were identified through reference tracking of included studies. Studies were eligible for review if they (1) reported randomized controlled trials (RCTs) published between 1990 and September 2018, (2) evaluated an integrated health care intervention designed to improve CYP’s health, (3) included CYP (0–18 years) with an ongoing health condition (defined as any health problem requiring clinical follow-up for >12 months in 50% or more cases),\(^26\) (4) included at least 1 health-related outcome indicator, and (5) were a peer-reviewed publication available in English. Eligible articles were screened to identify references reporting additional results from the described intervention, which were included if they assessed the same intervention and sample as the original article.

**Integrated Care Definition**

Many definitions of integrated care have been used in practice and the scientific literature, but no standardized widely accepted definition exists.\(^27\) The World Health Organization defines integrated care as “health services organized and managed so that people get the care they need, when they need it, in ways that are user-friendly, achieve the desired results and provide value for money.”\(^20\) This broad definition describes only the principles of integrated care and is not suitable to assess the presence or extent of integration. For practical application and to reflect the different integration needs of CYP contrasted with adults, we use the Wolfe et al\(^15\) definition, which adapts the World Health Organization principles of integration into domains specifically for the individual- and population-level health care needs of children. The first domain is vertical integration between primary and secondary care, which is important at the population level because minor and acute illness management is a large part of children’s health service use and is crucial for effective gatekeeping. An example is general practitioners or primary care pediatricians working effectively with secondary care pediatricians and mental health professionals. Horizontal integration is both between the health and education sectors, which is important for all CYP, and integration between health and social care, which is important for CYP with complex...
needs. Finally, longitudinal integration is important for age-appropriate care, at key times of developmental transition (eg, between pediatric and adult services). We used these categories of integration to identify if an intervention was integrated and which domain applied. Interventions that included ≥1 of these integration types were eligible for inclusion.

**Data Extraction and Quality Assessment**

Interventions were defined as "integrated care" (yes or no) for each of the domains of integration described above,13 with a detailed rationale documented explaining how the intervention met these criteria. Data were extracted from reports of interventions integrated in at least 1 domain, with outcomes relating to (1) child health outcomes, (2) health service use, (3) health care quality, (4) education (school absenteeism), and (5) cost savings and cost-effectiveness. Population data including sociodemographic details, comparator groups, intervention type and category of integration, study design, and outcome measures were extracted. Bias was assessed by using the Cochrane Collaboration tool.29 Two authors assessed the articles and extracted data, with discrepancies resolved by discussion. When protocol articles were available, these were consulted to assist in data extraction and quality assessment.

**Data Analysis**

Meta-analyses of the effectiveness of interventions were conducted on data from studies in which there was homogeneity of the outcome variable: rates of ED visits and child-related quality of life (QoL) measures. For these 2 outcome measures, when possible, we summarized data at 3 follow-up time points (3, 6, and 12 months) and when reported, data from intention-to-treat analyses were used. If 2 interventions were compared against a control group, data from both intervention arms were included, and the number of participants in the control group was halved for each comparison. Postintervention outcomes, reported as mean and SD, were used for analysis; when postintervention scores were unavailable, mean change scores were used. When mean and SD values were unavailable, missing data were imputed by estimating the SD from the confidence intervals (CIs). Standardized mean differences (SMDs) with 95% CIs were calculated and pooled by using a random effects model for all studies. Dichotomous and continuous outcomes were merged by using Comprehensive Meta-Analysis software (version 2.2; Biostat, Englewood, NJ) to produce SMDs (equivalent to Cohen’s d) for each study. SMD values of at least 0.2, 0.5, and 0.8 indicate small, medium, and large effect sizes, respectively.30 Heterogeneity across studies was assessed by using Cochran Q test and I² test statistics.

Integration was defined as present or absent (as specified above), and because there is no established measure of extent of integration, a description of the integrated service was obtained. Because we included a wide breadth of interventions to capture the diversity of interventions for any and all children’s health care, we used descriptive assessment and clinical judgment of included articles to support the statistical assessment of homogeneity.

**RESULTS**

The search generated 3956 articles; 23 articles, reporting 18 interventions, met the inclusion criteria (Fig 1). Risk of bias overall was moderate; a lack of blinding participants was the most common reason for downgrading quality (n = 8 interventions). An overview of results is provided in Table 1 and Supplemental Table 2.

**Study Characteristics**

Sample sizes ranged between 24 and 1316 CYP, and the mean age was 8.9 years. Asthma was the most commonly studied condition (n = 6). Most (n = 13) studies were conducted in North America.31-44 Comparison groups were treatment as usual (n = 14), waitlist control (n = 2), or enhanced usual care (n = 2); however, a description of usual care was typically not provided.

The 18 interventions included in this review are described in Table 1. Gorelick et al38 and Seid et al44 compared 2 integrated interventions to treatment as usual. When outcomes for the same intervention and patient sample were reported across several articles,34-36,40,41,45-48 the results were combined to reflect the overall impact of the intervention.

**Intervention Characteristics**

All interventions were focused on improving child health; however, the content differed. All articles described the evidence used for intervention development; however, there was limited detail on hypothesized mechanisms of action provided, and no article included a logic model. Most interventions were complex, incorporating multiple elements to achieve integration, summarized in Table 1. Ten interventions included aspects of vertical integration,31,38,40,41,43-50 10 included horizontal integration,32-37,39,40,42,44,51 and no interventions described longitudinal integration.

**Study Outcomes**

Results are described according to the 5 categories defined in the aims of this review: (1) child health outcomes, (2) health service use, (3) health care quality, (4) education (school absenteeism), and (5) cost savings and cost-effectiveness. Meta-analysis was possible only for child health outcomes (QoL) and health...
service use (ED visits) because of data heterogeneity.

Child Health Outcomes (18 Interventions)

Health Status and Functional Symptoms

Studies of 14 interventions assessed changes in 19 functional health indicators, such as improvement in condition-related symptoms. The quality of evidence was moderate (42.9% at low risk of bias). Five of these 19 indicators (26.3%) revealed an improvement among CYP in the intervention group, compared with controls. These indicators included hemoglobin A1c (a measure of diabetes control), pain, disability, symptoms of attention-deficit/hyperactivity disorder, viral load, and symptoms of asthma. Effect sizes for comparisons across intervention and control were not reported. Authors of only 1 article assessed maintenance of effect, finding improved functional status for the intervention compared to the control at 12 months but not maintained at 18 months.49

All interventions in which authors reported a significant health improvement effect for integrated care, compared to the control, included education for caregivers. Regarding improvements in asthma symptoms, Seid et al44 compared 2 integrated interventions to controls (rated at low risk of bias), which revealed contrasting results 3 months postintervention. Both integrated care interventions reported aspects of vertical and horizontal integration: hospital service providers communicated with primary care providers via the electronic health record and referred to community services. CYP receiving integrated care interventions had a one-third odds of having >1 asthma-related nighttime symptom per week compared with controls but only when a psychoeducational workshop for parents was included.

QoL

Studies of 8 interventions assessed changes in QoL between intervention and controls; the quality of evidence was good (62.5% at low risk of bias). Authors of 3 interventions reported greater improvements in QoL for the intervention compared to the control. A meta-analysis to quantify the size of effect for QoL was only possible with data for 6-month follow-up. Meta-analyses (see Fig 2) revealed that interventions had a significant positive effect in improving QoL scores over usual care in 4 studies (5 comparisons) when data were available for pooling (SMD = 0.24; 95% CI = 0.03–0.44; \( P = .02 \)). There was no statistical heterogeneity between studies (\( I^2 = 0\% \); \( P = .48 \); however, studies were heterogeneous regarding clinical populations and control group characteristics. Authors of 1 article compared the intervention to enhanced usual care31; the authors of the remaining articles compared the integrated health care intervention to treatment as usual. The condition studied varied across articles and included diabetes,45–48 asthma,44 and obesity.31,50

Psychological Indicators

Studies of 8 interventions assessed changes in 14 psychological indicators across the intervention and control; the quality of evidence overall was poor (2 at low risk of bias). Indicators included anxiety (\( n = 2 \)), distress and depression (\( n = 4 \)), psychosocial functioning and/or adjustment (\( n = 2 \)), well-being (\( n = 1 \)), body dissatisfaction (\( n = 1 \)), behavioral symptoms (\( n = 1 \)), and self-esteem or self-worth (\( n = 3 \)). Of these 14 indicators, statistically significant improvements were found for 5 of 14 indicators (35.7%) when compared with controls. Psychological outcomes varied within studies. For example, Glisson et al37
found no effect of integrated care interventions on well-being, which improved in both intervention and control groups during the study period, but girls receiving integrated care showed significant improvements in self-efficacy (effect sizes across subscales range between 0.41 and 0.69) compared with girls receiving usual care who showed a significant decline in self-efficacy. No changes in self-efficacy were found for boys across the control and intervention arms.

**TABLE 1** Characteristics of Interventions Included in Review

<table>
<thead>
<tr>
<th>Article</th>
<th>Condition</th>
<th>Child health</th>
<th>Health status</th>
<th>QoL</th>
<th>Psychological</th>
<th>Health service use</th>
<th>1ry, 2ry care use</th>
<th>Health care quality</th>
<th>Education</th>
<th>Cost savings</th>
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**Primary and Secondary Care Use**

**Health Service Use**

*ED Visits*

Studies of 9 interventions reported health service use; the quality of evidence overall was moderate (20% at low risk of bias). Studies of 7 interventions assessed ED visits. In all studies, authors hypothesized that integrated care interventions would reduce ED visits. There was sufficient evidence to examine the effect of interventions on visits to the ED (measured by parent recall) with 2 comparisons (from 2 studies) but only at 6-month follow-up. Meta-analyses (Fig 3) revealed that integrated care did not significantly reduce ED visits between interventions and control groups (odds ratio [OR] = 0.88; 95% CI = 0.57–1.37; P = .57). There was no statistical heterogeneity between studies (I² = 0%; P = .74).

**Primary and Secondary Care Use**

Six studies used a variety of indicators to assess the impact on secondary care use. These included attendance at mental health services (n = 1), urgent doctor appointments (n = 1), and mental health services (n = 1). The hypothesized outcome was contact with the health care service, with some aiming to increase CYP contact with the health care service, and others to reduce contact. Overall, the studies reported mixed results. No consistent patterns were observed, and the quality of evidence varied across studies.
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<th>Article</th>
<th>Kolko et al40, Yu et al41</th>
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<th>McWhirter et al43</th>
<th>O'Shea et al45</th>
<th>Seid et al44</th>
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<th>Simon et al49</th>
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<td>Horizontal; health and social services; United States; intervention and control were as effective as each other&lt;sup&gt;b&lt;/sup&gt;</td>
<td>Horizontal; nurse and school staff; England; positive effects for intervention for child health (viral load)&lt;sup&gt;c&lt;/sup&gt;</td>
<td>Vertical; 1ry care and 2ry care nurse and neonatologist; United States; intervention and control as effective as each other&lt;sup&gt;d&lt;/sup&gt;</td>
<td>Vertical; home health visitors and 1ry care, and 2ry care nurse and neonatologist; United States; intervention and control as effective as each other&lt;sup&gt;e&lt;/sup&gt;</td>
<td>Vertical; home health visitors and 1ry care, and 2ry care nurse and neonatologist; United States; intervention and control as effective as each other&lt;sup&gt;f&lt;/sup&gt;</td>
<td>Vertical; home health visitors and 1ry care, and 2ry care nurse and neonatologist; United States; intervention and control as effective as each other&lt;sup&gt;g&lt;/sup&gt;</td>
<td>Vertical; multidisciplinary team in 2ry care and 1ry care provider; United States; positive effects of intervention for quality of care at 18 mo and service use; intervention more expensive&lt;sup&gt;a&lt;/sup&gt;</td>
<td>Vertical integration between pediatrician health and school staff; Sweden; Australia; intervention and control were as effective as each other&lt;sup&gt;h&lt;/sup&gt;</td>
<td>Vertical integration between pediatrician and dietitian in 2ry care, and general practitioner; 1ry care and 2ry care nurse and school staff; Sweden; intervention and control were as effective as each other&lt;sup&gt;i&lt;/sup&gt;</td>
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<sup>a</sup> For risk of bias, green indicates low risk.
<sup>b</sup> For risk of bias, red indicates high risk.
<sup>c</sup> For risk of bias, amber indicates moderate risk.
4 of 10 indicators met their desired outcome and revealed greater improvements in secondary care use for integrated care over control.

Simon et al\textsuperscript{49} reported that the use of a multidisciplinary team and shared care records between the secondary and primary care provider resulted in a greater reduction in total hospital days and length of stay (\textgreater;7 days) for CYP with medical complexity when compared to treatment as usual over a period of 12 months. Improvements were maintained at 18 months postintervention for length of stay (\textgreater;7 days) but not for total hospital days. However, no change was found between the intervention and control for inpatient admissions, length of stay (per child-year), or ICU visits. No effect sizes were reported, but the authors anticipate that the intervention has increased accessibility to health care services through its focused care coordination. Grupp-Phelan et al\textsuperscript{51} used an integrated intervention aiming to enhance access and increase contact with mental health services. The trial reported no difference among CYP receiving the intervention compared to the control regarding scheduling a mental health appointment (OR = 2.29; 95% CI = 0.41–12.7/1.0) but greater likelihood of attending their first mental health appointment at 60 days postintervention (OR = 9.62; 95% CI = 1.38–67.25/1.0).

**Health Care Quality (n = 6 Interventions)**

Six studies reported indicators for health care quality; 5 of these were related to service satisfaction. The quality of evidence among included studies was moderate. One article\textsuperscript{49} included a combined measure of several indicators of health care quality including support for coordinating multiple providers, getting care needed quickly, prescription medication, getting specialized services, shared decision-making, and getting needed information. Authors of 2 studies reported increased satisfaction with integrated interventions when compared with controls\textsuperscript{40,41,45–48}; however, no further information to quantify these effects was provided. For the combined measure of health care quality, Simon et al\textsuperscript{49} reported greater improvements for the intervention over the control at 12 months postintervention, but...
these effects were not maintained at 18 months. For individual indicators, there was a greater impact. For example, help coordinating multiple providers and getting care quickly were improved for the intervention, over the control, and were sustained at 18 and 12 months, respectively.

**Education (n = 3 interventions)**

Three studies were used to assess school absenteeism as an education-related outcome regarding integrated health care. Authors of 1 study found statistically significant reductions in school absenteeism for the intervention compared to the control,33 reporting a 69% reduction for school absenteeism among CYP with asthma; no estimates of effect size were reported.

**Cost Savings and Cost-effectiveness (n = 5 Interventions)**

Four studies included a cost-savings analysis of integrated health care interventions compared to controls.34–36,39–41,49 Authors of 1 study reported analysis of cost-effectiveness.45–48 The quality of studies overall was moderate (2 at low risk of bias). All studies assessed changes in health care costs from the health system perspective, using health service use data, and 2 also assessed the societal perspective.39,45–48 Overall cost savings were reported for 3 of 4 interventions when compared to the control; no difference was found between the intervention and control for cost-effectiveness.45–48

From the health system perspective, integrated care interventions were more expensive to implement when compared to controls.34–36,39–41,49 but these costs were offset by savings from health care usage.34–36,40,41 For example, Yu et al41 reported a lower cost per integrated care patient associated with mental health services, when compared with controls, during the 6-month trial and at 6- to 12-month follow-up points. However, these differences narrowed at 18 to 24 months as CYP moved further from the intervention period. For Simon et al,49 the high implementation costs were not offset by cost savings over an 18-month period.

From the societal perspective, Hechler et al49 provided CYP and families with self-management support and worked alongside schools. This intervention was associated with significant reductions in parental financial burden compared with controls, and indirect costs to employers decreased as parental work absenteeism decreased significantly. Ellis45–49 found no changes in cost savings from the societal perspective.

**DISCUSSION**

This is the first article to present a systematic assessment of the effectiveness of integrated health care interventions on important outcomes for children with ongoing conditions including health, health service use, health care quality, education, and cost savings. Using a broad systematic search, we identified varied, and often contradictory, outcomes among 18 integrated care interventions. Studies were often limited by poor reporting of statistics or the quality of evidence.

Overall, integrated care for CYP with ongoing conditions appears to offer statistically significant improvements in health-related QoL. However, for other child health outcomes, health service use, health care quality, education (school absenteeism), and cost savings and cost-effectiveness, the evidence is mixed.

Caregiver and education involvement appear to be important in delivering positive changes in CYP health status. At the health system level, the evidence about the influence of integrated care on secondary care usage was mixed and depended on whether the intervention aimed to increase accessibility of services or reduce reliance on secondary care services. Surprisingly, no articles reported on the impact of integrated care on primary care usage.

The meta-analysis indicated limited effectiveness of integrated care in reducing ED visits for CYP with ongoing conditions. However, the broad CIs highlight the heterogeneity of the evidence on the impact of integrated care on ED visits. Of note, the integrated care interventions described in this review did not target minor ailments that cause large numbers of ED visits in CYP with ongoing conditions. Our findings on ED attendance contrast with recent observational studies that have shown up to a 40% decrease in ED presentations for children with medical complexity.53

This review highlights important concerns about the design and evaluation of children’s integrated care. Information regarding the design of integrated interventions in the articles included in this review is limited because no studies presented or described the use of logic models to inform complex intervention development, which have been shown to improve intervention efficiency54,55 or the use of appropriate theoretical frameworks to guide the selection of interventions and outcome indicators. Perhaps as a result of these omissions, the interventions in this review were challenging to describe because of the complex nature of overlapping intervention components.

Evaluation of integrated care interventions among the articles included was limited by variable quality (despite selecting only RCTs), heterogeneity of outcomes and lack of standardized outcome measures, and insufficient follow-up periods. The contrasting results between studies using observational and trial designs and the variable quality of published studies suggests there is much to be gained from further high-quality
recommendations and control groups. Additionally, although heterogeneity prevented the comparison of studies assessing health care costs in this review, authors of 3 of 4 articles reported cost savings associated with integrated care despite higher implementation costs. However, it is not clear whether these cost savings were sustained. Again, standardized measures and appropriate follow-up periods are essential to improve the quality of evidence available.

**Recommendations**

With this review, we present a synthesis of the best available data on integrated care for CYP. We find that integrated care may deliver improvements in QoL and may offer cost savings; however, there is insufficiently strong evidence currently to make firm recommendations regarding the overall effectiveness of integrated services.

Our findings highlight important gaps in the evidence. We recommend that future studies should follow established guidelines on the development and evaluation of complex health system interventions. Using logic models and intervention design methods that include behavior change mechanisms at the level of the individual and the system are also important. Agreed definitions of integration and a validated measure to assess the level of integration in health care interventions would be useful.

To improve the quality and usefulness of research, robust evaluation design, standardization of outcome measures (eg, ED visits), and appropriate follow-up periods are essential, as is informative standardized reporting, for example, of effect sizes between intervention and control groups.

**Strengths and Limitations**

This is the first systematic review of children’s integrated care interventions that includes a comprehensive variety of interventions to capture the diversity needed for all children’s health care. However, the breadth of interventions included also brought challenges. Articles were limited in their descriptions of usual care, making comparison with control groups challenging. In addition, with the available data, we could not discern which children benefited most from integrated health care. Severity of symptoms and socioeconomic background may be important determinants that influence health care outcomes. Furthermore, no conclusions could be made about the extent of integration that may be beneficial for child health or system measures because there is currently no validated measure of extent of integration.

Of the 18 interventions identified, studies on 6 interventions reported on CYP with asthma. Eleven of 18 studies were set in North America; therefore, generalizability of the findings may be limited to these groups, particularly given the differing nature of health care systems.

Despite low statistical heterogeneity, descriptive analysis indicated that studies included in the meta-analyses differed in intervention characteristics and evaluation design; therefore, the meta-analysis needs to be interpreted with a clear understanding of these inherent challenges of health services research. To aid interpretation in the context of these contrasting statistical and descriptive findings, we have presented detailed descriptions of each study included in our study tables and highlighted the impact of each study in turn. This is intended to support the reader to assess the impact of integrated care in each outcome domain (Supplemental Table 2).

**Implications for Clinicians and Policy Makers**

The evidence for integrated care interventions improving child health is limited but encouraging. However, the best available data are currently insufficiently robust to make firm recommendations regarding the overall effectiveness of integrated services to improve children’s health. We recommend strongly that research efforts are directed urgently toward improving the design and evaluation of integrated health care models for children.

**CONCLUSIONS**

Health systems need to adapt to improve quality and meet evolving health needs, so there is an urgent need for high-quality evidence. Integrated models of care may deliver some important improvements in QoL for CYP with ongoing health conditions and potentially save money for the stretched health system. However, the evidence base remains insufficient, and there is an urgent need for high-quality research to develop and evaluate theoretically framed integrated models of care to help CYP living with ongoing health conditions.

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**ABBREVIATIONS**

CI: confidence interval
CYP: children and young people
ED: emergency department
OR: odds ratio
QoL: quality of life
RCT: randomized controlled trial
SMD: standardized mean difference
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