The clinical effectiveness, cost-effectiveness and acceptability of community-based interventions aimed at improving or maintaining quality of life in children of parents with serious mental illness: a systematic review

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Scientific summary

Interventions for improving or maintaining QoL in children of parents with SMI

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Scientific summary

Background

Improving the lives of children born to parents with serious mental illness (SMI) is an urgent political and public health concern. The best estimates suggest that 50–66% of people with SMI may be living with one or more children under the age of 18 years. The burden placed on these young people is substantial. Research shows that serious parental mental illness is associated with increased risk of adverse outcomes in children. Short-term outcomes include poorer mental and physical health as well as increased risk of a range of behavioural, social and educational difficulties. Longer-term outcomes may extend into adulthood and include social or occupational dysfunction, lower self-esteem, increased psychiatric morbidity and alcohol or substance misuse. This evidence synthesis sought to assess the clinical effectiveness, cost-effectiveness and acceptability of community-based interventions aimed at increasing or maintaining quality of life (QoL) in children of parents with SMI.

Objectives

The objectives of the evidence synthesis were:

- to provide a systematic and descriptive overview of all the evidence for community-based interventions for improving QoL in children and adolescents of parents with SMI, with specific reference to intervention format and content, participant characteristics, study validity and QoL outcomes measured
- to examine the clinical effectiveness of community-based interventions in terms of their impact on a range of predetermined outcomes, particularly those likely to be associated with QoL for children and adolescents of parents with SMI
- to examine, when possible, potential associations between intervention effect and delivery including intervention format and content, prioritisation of child outcomes, child age group, parental mental health condition, family structure and residency
- to explore all available data relating to the acceptability of community-based interventions intended to improve QoL for children and adolescents of parents with SMI, with specific reference to intervention uptake, adherence and patient satisfaction
- to assess key factors influencing the acceptability of and barriers to the delivery and implementation of community-based interventions for improving QoL in children and adolescents of parents with SMI
- to provide a systematic and descriptive overview of all the economic evidence for community-based interventions for improving QoL in children and adolescents of parents with SMI, with specific reference to intervention resources, cost burden, study validity, method of economic evaluation and economic outcomes measured
- to examine the cost-effectiveness of community-based interventions in improving QoL for children and adolescents of parents with SMI using a decision-analytic model
- to identify, from the perspective of the UK NHS and personal social services, research priorities and the potential value of future research into interventions for improved QoL in this population.

Methods

Data sources

Comprehensive, systematic searches were undertaken using 19 health, allied health and education databases, searched from inception until January 2011, with an update search being performed in May 2012. Nine psychiatry, psychology and child health journals were hand searched. In addition, grey

literature (e.g. conference proceedings and voluntary organisation publications), dissertations, ongoing research registers and bibliographies from the texts of relevant trials and reviews were searched. Forward citation tracking of all included trials was undertaken. Key authors and specialists in the field were contacted.

Study selection

Study participants were children or adolescents aged ≤ 18 years of age and/or the parents of these children. To be eligible for inclusion, ≥ 50% of the sample had to have a SMI as defined by a current or lifetime clinical diagnosis or comparable symptom profile. SMI was defined to include schizophrenia and schizoaffective disorder, puerperal and non-puerperal psychosis, borderline personality disorder and and personality disorder, with or without substance misuse and other mental health co-morbidities. Severe unipolar depression and severe postnatal depression were also included.

Eligible interventions comprised any community-based (i.e. non-residential) psychological or psychosocial intervention that involved professionals or paraprofessionals and parents or children, for the purposes of changing knowledge, attitudes, beliefs, emotions, skills or behaviours related to health and well-being.

Comparisons of two or more active interventions or of an active treatment with a 'no treatment' comparator were included. The 'no treatment' category extended to include waiting list controls, delayed treatment and usual care management.

Primary outcomes comprised validated measures of children's QoL and/or children's emotional well-being. Secondary outcomes were derived from UK policy and stakeholder consultation. These comprised measures of children's physical health, safety, social function, self-esteem, mental health literacy, coping skills, family function and parental mental health symptoms. Acceptability was defined in terms of intervention uptake, adherence and participant satisfaction or views.

Data extraction and synthesis

Quantitative and qualitative data relating to study design, quality, sample characteristics, interventions and comparators, and clinical, economic and acceptability outcomes were extracted using a standard proforma. Study quality was assessed according to the Cochrane collaboration risk of bias assessment tool for randomised controlled trials (RCTs), the Cochrane guidance for non-randomised designs and the Critical Appraisal Skills Programme (CASP) tool for qualitative research. Economic studies were assessed for quality using a standard critical appraisal checklist for economic evaluations.

Studies targeting the parents or children of parents with SMI were synthesised separately to those for severe depression. Our primary analysis focused on data from RCTs or quasi-RCTs. Lower levels of evidence were retained and summarised for the purposes of future research priority setting. Continuous data from RCTs were translated to standardised mean difference effect sizes (ESs). Dichotomous data were translated into standardised ESs using logit transformation. ESs were pooled using random-effects modelling. Clinical and methodological heterogeneity were explored whenever possible via subgroup and sensitivity analyses. Alternative sources of bias were investigated using funnel plots when data allowed. In instances for which data pooling was inappropriate, narrative synthesis was employed. Cost-effectiveness data were summarised and acceptability data were synthesised using a textual narrative approach.

Results

Our searches generated 34,659 hits and identified 57 eligible studies, of which 29 were RCTs or quasi-RCTs. Only three of these trials targeted the parents or children of parents with SMI. Twenty-six trials targeted the parents or the children of parents with severe depression.

Evidence of clinical effectiveness

All three trials pertaining to SMI recruited mothers (or the children of mothers) with psychosis or psychotic symptoms and all compared one or more active interventions to a treatment as usual control. Children were aged ≤ 12 years. Overall, the three trials reported on five different interventions. Two were cognitive–behavioural interventions delivered directly to children, two sought to indirectly influence children's QoL through an enhancement of mothers' parenting behaviours and one integrated a home-based parenting intervention alongside an intervention for mothers' mental health symptoms. None of the trials reported primary outcomes relevant to children's overall QoL.

Analysis of secondary outcomes was limited by clinical and methodological heterogeneity between the trials and poor reporting of outcome data. Data pooling was not possible. All three trials were judged to be at a high or unclear risk of bias and all were published in the USA between 1982 and 1984. The generalisability of this evidence to the contemporary UK health context is questionable.

Twenty-six trials (90%) focused on severe parental depression, 18 of which evaluated community-based interventions for the parents of children in the first 2.5 years of life. Only two trials evaluated interventions aimed at preschool and/or primary school-aged children (aged 2.5–12 years) and six studies evaluated interventions relevant to children of primary school age or beyond (6–18 years).

Overall, the 26 trials reported on 38 interventions. Thirty-one (82%) were solely, or predominantly, parent-based interventions, 21 of which were psychotherapies aimed solely at improving parents' mental health. Six interventions (16%) targeted the parent–child dyad and one (3%) was delivered to children alone. In total, 14 interventions (37%) sought to enhance some aspect of parenting behaviour or family function.

Pooling of data was feasible for only four short-term outcomes. Five depression trials contributed data to a meta-analysis comparing the effect of any variant of community-based intervention to a waiting list/treatment as usual control on children's short-term emotional health. Pooled ESs suggested no significant short-term effect of intervention; however, the width of the accompanying confidence intervals (CIs) for the effect did include effects of potential clinical significance (standardised ES 0.06, 95% CI –0.20 to 0.33). The small number of trials contributing to this analysis prevented any meaningful subgroup analyses and these results should be treated with caution.

Eight depression trials contributed data to a meta-analysis comparing the effect of any variant of community-based intervention to a waiting list/treatment as usual control on children's social functioning and behaviour. Pooled ESs suggested no significant short-term effect of intervention, although, once again, the width of the CIs reported included effects of potential clinical importance (standardised ES 0.23, 95% CI 0.00 to 0.46). The small number of trials contributing to this analysis prevented any meaningful subgroup analyses. Results should be treated with caution.

Seventeen depression trials contributed data to a meta-analysis comparing the effect of any variant of community-based intervention to a waiting list/treatment as usual control on parents' short-term mental health. Pooled ESs suggested a significant medium to large effect of intervention (standardised ES 0.73, 95% CI 0.51 to 0.94). Clinical and methodological heterogeneity was evident and marked statistical heterogeneity was observed ($I^2 = 67.8\%$, $p = 0.000$). Preliminary evidence from a smaller number of trials reporting longer-term outcomes suggested that these clinical effects may diminish over time.

Dividing the trials according to intervention type resulted in a smaller short-term effect for psychoeducational and psychosocial models than for psychotherapeutic interventions, whereas dividing the trials according to child age ranges revealed medium to large effects for both children aged 0–4 years and those aged 6–18 years. No trials were identified for children between 4 and 6 years. Grouping the trials by intervention target resulted in a medium to large effect for parent-based interventions and a large effect for dyadic interventions. Finally, pooling trials by intervention objectives revealed a medium to large effect

for interventions targeting parental well-being and a small to medium, non-significant, effect for the small number of interventions targeting the parent–child relationship. The limited number of comparisons in some groups limits the utility of these findings.

Six trials contributed data to a meta-analysis comparing the effect of any variant of community-based psychosocial intervention to a waiting list/treatment as usual control on parents' responsiveness to their children. Pooling these data produced a medium to large effect of intervention on short-term parenting behaviours (standardised ES 0.67, 95% CI 0.32 to 1.02). The small number of trials contributing to this analysis prevented any meaningful subgroup analyses.

All but one of the trials pertaining to severe parental depression were judged to be at a high or unclear risk of bias, indicating a relatively poor level of trial quality overall. Particular methodological problems were noted in relation to randomisation and allocation procedures, sample size, potential attrition biases and selective outcome reporting. Interpretation of the findings was further limited by a lack of existing data and marked heterogeneity in the populations, interventions and outcomes assessed. Children's self-reported outcomes were rare, and only four trials were conducted in UK settings. The majority of the evidence base remains biased towards parent-based interventions targeting severely depressed mothers of infant children. The generalisability of these findings to other diagnoses, to older children and to the children of fathers with SMI is unclear.

Evidence of cost-effectiveness

No economic evaluations or cost or resource-use studies were identified that focused on the children of parents with SMI. Only one economic evaluation focusing on severe parental depression was found. This study was at high risk of bias and reported a narrow assessment of costs and effects. Costs and benefits were presented from the perspective of the mother and could not be meaningfully used to support resource allocation decisions aimed at improving children's subjective QoL.

Planned economic synthesis included decision-analytic modelling alongside the narrative synthesis of any economic evaluations found. The absence of any rigorous evidence to support the clinical effectiveness of specific interventions, combined with an absence of economic evidence, rendered this economic modelling impossible. Value of information analysis (VOI) was also infeasible.

Evidence of acceptability

The acceptability review synthesised data relating to intervention uptake, adherence, patient satisfaction and patient views. Rates of intervention uptake and adherence were inconsistently reported across the studies included in our syntheses and a lack of data from high-quality RCTs made meta-analysis inappropriate. No rigorous high-quality qualitative data were found.

Limited quantitative evidence suggested that child custody losses and conflicting life circumstances may act as potential barriers to intervention access. The available qualitative data highlighted the importance of developing intervention models and delivery mechanisms capable of transcending the high levels of social isolation and stigma faced by families living with SMI. Children's views of community-based interventions were lacking. Preliminary data suggests that children may value peer interactions and normalising activities, although further research is needed to confirm these findings.

Limitations

Current evidence of the clinical effectiveness of community-based interventions for children of parents with SMI remains heavily focused on interventions for depressed mothers with infant children. The generalisability of these findings to families living with other diagnoses, to older children and adolescents, and to families in which fathers have SMI is not clear. Too few studies are available to ascertain medium- and long-term follow-up effects or to fully consider the associations between different

intervention characteristics and intervention effect. Potential biases arise from selective outcome reporting in the primary studies and the inclusion of quasi-randomised studies in the review.

UK studies that have reported child-specific QoL outcomes within the last decade have typically targeted high- or multirisk families for whom risk is defined in terms of social deprivation. The needs of children within such families may be qualitatively very different from those in our syntheses. Further consideration should be given to the optimal method of identifying families and children affected by serious parental mental illness and to the possibility that functional outcomes, rather than diagnostic indicators, may be more appropriate markers of illness severity.

Conclusions

Implications for practice

Evidence for community-based interventions to enhance QoL in children of parents with SMI is underdeveloped and, in its current state, does not provide any rigorous rationale to underpin UK service development and delivery.

Implications for research

Future research must include designs with properly framed a priori research questions and adequate power to deliver answers. Trials must follow appropriate randomisation and allocation procedures, with formal monitoring of intervention uptake and adherence rates. Validated, child-centred and age-appropriate primary outcome measures for QoL should routinely be employed and trials should ensure full reporting of this outcome data. The need to measure longer- as well as shorter-term QoL outcomes and to nest in-depth acceptability studies within these trials cannot be overemphasised. High-quality cost data must be collected.

Manualised parenting interventions with proven efficacy in multirisk families and group-based psychoeducational programmes that target similar outcomes to those prioritised in our stakeholder group exist as two potential candidates for modification and piloting via an exploratory RCT.

Consistent with the philosophy of the Medical Research Council framework for RCT development, a substantial programme of pilot work is first advocated. Greater evidence is needed to underpin the development of feasible and acceptable interventions for this population. This work may usefully include a scoping review of current provision across statutory and non-statutory service provision, a series of professional stakeholder consultation events designed to ascertain the likely facilitators and constraints in the host health-care systems and a programme of qualitative work undertaken with children and families with experience of parental SMI. New, age-appropriate instruments that better reflect the life priorities and unique challenges faced by the children of parents with SMI may also need to be developed.

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