

Alternative community-based models of care for young people with anorexia nervosa: the CostED national surveillance study

Sarah Byford,^{1*} Hristina Petkova,¹ Ruth Stuart,¹
Dasha Nicholls,² Mima Simic,³ Tamsin Ford,⁴
Geraldine Macdonald,⁵ Simon Gowers,⁶
Sarah Roberts,¹ Barbara Barrett,¹ Jonathan Kelly,⁷
Grace Kelly,⁸ Nuala Livingstone,⁸ Kandarp Joshi,^{9,10}
Helen Smith¹¹ and Ivan Eisler^{1,3}

¹Health Service and Population Research Department, Institute of Psychiatry, Psychology & Neuroscience, King's College London, London, UK

²Department of Medicine, Imperial College London, London, UK

³South London and Maudsley NHS Foundation Trust, London, UK

⁴Institute of Health Research, Department of Psychological Sciences, University of Exeter Medical School, University of Exeter, Exeter, UK

⁵School for Policy Studies, University of Bristol, Bristol, UK

⁶University of Liverpool, Liverpool, UK

⁷Beat, Norwich, UK

⁸School of Social Sciences, Education and Social Work, Queen's University Belfast, Belfast, UK

⁹Child and Adolescent Mental Health Service, Royal Cornhill Hospital, NHS Grampian, Aberdeen, UK

¹⁰Mental Health Division, University of Aberdeen, Aberdeen, UK

¹¹NHS Greater Glasgow and Clyde, Glasgow, UK

*Corresponding author s.byford@kcl.ac.uk

Declared competing interests of authors: Tamsin Ford is chairperson of the Child and Adolescent Psychiatry Surveillance Service that was used to run part of the study, which is an unpaid position (other than travel expenses). Kandarp Joshi reports grants from Sunovion Pharmaceuticals, Inc., outside the submitted work. He was principal investigator for the Aberdeen site for a Sunovion-sponsored multisite trial on the efficacy and safety of lurasidone in paediatric schizophrenia. Jonathan Kelly reports that Beat has contracts with some NHS trusts and clinical commissioning groups that provide and commission, respectively, community eating disorders services for children and young people. In these contracts, Beat works with the local services to deliver awareness-raising training for professionals and, in some cases, to offer peer coaching for carers. The funding Beat receives in return for its role in this project is not 'on my behalf'. Beat is a co-applicant on another research project called 'TRIANGLE' (short title), which is also funded by the National Institute for Health Research. The funding Beat receives in return for its role in this project is not 'on my behalf'. Beat is a co-applicant on another research project called 'Mind the Gap' (short title), which is funded by the Welsh Assembly Government and is investigating referral pathways from identification to specialist treatment for people with eating disorders (of all ages) in Wales.

Published October 2019

DOI: 10.3310/hsdr07370

Scientific summary

The CostED national surveillance study

Health Services and Delivery Research 2019; Vol. 7: No. 37

DOI: 10.3310/hsdr07370

NIHR Journals Library www.journalslibrary.nihr.ac.uk

Scientific summary

Background

Evidence suggests that investing in specialist eating disorders services for young people with anorexia nervosa could have important implications for the NHS, with the potential to improve health outcomes through reductions in relapse rates, to reduce costs through reductions in hospital admissions and to improve the quality of life of young people and their families.

Objectives

The primary aims of the Cost-effectiveness of models of care for young people with Eating Disorders (CostED) study were to evaluate the cost and cost-effectiveness of alternative community-based models of service provision for child and adolescent anorexia nervosa and to model the impact of potential changes on the provision of specialist services using decision-analytic techniques. The specific objectives were to (1) identify all new incident cases of anorexia nervosa in young people aged 8 years to 17 years and 11 months in the UK and the Republic of Ireland (RoI) over an 8-month period and provide incidence estimates; (2) classify the model of care provided for each community-based case identified at baseline as either a specialist eating disorders service or a generic child and adolescent mental health service (CAMHS); (3) calculate the relative cost of all notified community-based cases of child and adolescent anorexia nervosa in the UK and RoI and determine the cost-effectiveness of different models of care at 6- and 12-month follow-ups; and (4) model the impact on cost and cost-effectiveness of potential changes to the provision of specialist services.

Methods

An observational surveillance study was undertaken using the Child and Adolescent Psychiatry Surveillance System (CAPSS), a system designed to ascertain cases of rare childhood mental health conditions in the UK and RoI through monthly reporting by clinicians. Clinicians were asked to report cases of young people aged 8–17 years in contact with CAMHS for a first episode of anorexia nervosa in accordance with *Diagnostic and Statistical Manual of Mental Disorders*, Fifth Edition, diagnostic criteria between 1 February 2015 and 30 September 2015.

Clinicians notifying a positive case of anorexia nervosa were sent a baseline questionnaire and, if patients were assessed as eligible for inclusion, follow-up questionnaires after 6 and 12 months. Baseline questionnaires contained sections on the following: characteristics of the notifying service to enable services to be classified as specialist eating disorders services or generic CAMHS, limited patient identifiers to provide basic sociodemographic information and check for duplicate notifications, clinical characteristics to assess case eligibility and outcome data against which to assess clinical improvements over follow-up. The 6- and 12-month follow-up questionnaires contained the same sections, but also included a section on the use of secondary and tertiary mental health services for costing purposes.

The primary outcome measure was the Children's Global Assessment Scale (CGAS). Secondary outcome measures included percentage of median expected body mass index (BMI) for age and sex (%mBMI) and the Health of the Nation Outcome Scales for Children and Adolescents (HoNOSCA). Cases were also assessed for remission and relapse status.

The incidence of anorexia nervosa overall and at each age for all ages of the young people in the study was estimated as the rate per 100,000-person population at risk, with adjustments for missing notifications and missing baseline questionnaires, providing a range within which the true incidence is likely to fall.

To assess which services met criteria for classification as a specialist eating disorders service, a Delphi survey was undertaken that collected opinions from a range of stakeholders interested in eating disorders. The aim was to reach consensus on the key criteria for a community-based mental health service to be classified as a specialist eating disorders service. The Delphi survey was carried out online to maximise the number of respondents from the following groups: service users and their families, child and adolescent psychiatrists, paediatricians, other eating disorders professionals and service commissioners. Respondents were asked to consider the importance of a range of criteria in considering whether or not a service can be classified as specialist. Criteria were identified using existing guidelines for specialist eating disorders services and clinical expertise, and respondents rated them on a five-point Likert scale from 1 = not important to 5 = extremely important. The Delphi survey involved two rounds, with round 1 responses that failed to achieve consensus in accordance with prespecified thresholds being entered into round 2, when participants were given a second opportunity to respond and achieve consensus.

Data on inpatient, outpatient and day-patient mental health service contacts were used to calculate the 6- and 12-month costs of all cases eligible for follow-up. All resource use data related to the period February 2015 to September 2016, and costs, in Great British pounds, were for the 2015/16 financial year. The total costs per participant in each group were compared using standard parametric *t*-tests, with the validity of this approach confirmed using bootstrapping.

The cost-effectiveness of specialist versus generic services was explored in a decision-making context, with a focus on the probability of specialist services being cost-effective compared with generic services given the data available, rather than a focus on statistical significance. Measures of effect analysed in the cost-effectiveness analysis were CGAS score (primary) and %mBMI (secondary). Incremental cost-effectiveness ratios were calculated as the difference in mean cost between specialist services and generic services, divided by the difference in mean effects. To explore uncertainty, cost-effectiveness acceptability curves plotting the probability of one intervention being cost-effective compared with another were generated for a range of possible values of willingness to pay for unit improvements in outcome.

Decision modelling was used to explore the impact of changes to the proportion of specialist versus generic services, using a decision tree structure. The model used data collected in the CostED study and followed the same timeline (12 months), with young people following one of two pathways (specialist or generic) and grouped by remission and relapse status at 6 and 12 months. Cost-effectiveness was explored in terms of total cost per 10-point increase in CGAS score for the full sample. CGAS scores are used to classify patients into one of 10 categories of outcome, with each category covering 10 points on the scale; thus, this approach generates a cost per improvement in functioning from one CGAS category to the next, which is more clinically meaningful than cost per 1-point CGAS gain. Incremental analysis was not appropriate for the economic modelling because there was no comparison between the two groups; instead, the model explored the total cost per gain in outcome for the full population of young people, dependent on the proportion of those young people who are initially assessed in specialist services or generic CAMHS. The proportion of young people in the cohort initially assessed in specialist eating disorders services was varied from 0% to 100% to determine if there is an optimal structure that minimises the costs for the benefits gained. Two-way sensitivity analyses were carried out using different assumptions about baseline CGAS score and the treatment effect, and a probabilistic sensitivity analysis was undertaken to test the overall robustness of the model.

Results

A total of 305 incident cases of anorexia nervosa were eligible for inclusion in the incidence study and 298 cases were assessed as eligible for inclusion in the follow-up study of cost and cost-effectiveness. Clinicians completed and returned a 6-month follow-up questionnaire for 220 of these cases (74%) and a 12-month follow-up questionnaire for 187 cases (63%). The vast majority of the sample were girls (91%), from England (70%) and were coded as white (92%; any white background); their mean age was 14.6 years (± 1.66 years).

The missing data-adjusted estimate of the incidence of anorexia nervosa among young people aged 8–17 years, which we hypothesised to be the most accurate of three estimates presented, was 14 per 100,000. Comparison with previous studies suggests that overall incidence rates have remained steady over the last decade or so, with 13.1 per 100,000 reported in a study of 10- to 14-year-olds carried out between 2000 and 2009, compared with 12.6 per 100,000 estimated in the CostED study for young people of the same age. However, there was evidence to suggest that rates among younger children have increased, with a rate of 3.2 per 100,000 estimated in the CostED study for young people aged between 8 and 12 years, which is substantially higher than 2.1 per 100,000 for young people of the same age estimated from a previous study carried out between 2005 and 2006.

In the Delphi survey, three criteria achieved consensus for inclusion (offering specialist outpatient treatment for eating disorders, providing multidisciplinary specialist outpatient clinics dedicated to eating disorders and holding weekly multidisciplinary meetings dedicated to eating disorders). These criteria suggest that, to be classified as a specialist eating disorders service for young people, a service must provide evidence-based services for the treatment of eating disorders, must be multidisciplinary and must have a clear focus on, and expertise in, eating disorders.

A fourth criterion (the number of cases of eating disorders seen per year) remained uncertain after both rounds of the Delphi survey, with approximately half (52%) of respondents rating it as either very important or extremely important. Previous evidence supports the concentration of treatment for rarer conditions within a small number of services that can develop expertise in that area, particularly in surgery. However, the validity of this argument is less clear for treatments commonly provided to young people with anorexia nervosa, which tend to be psychological and family based, albeit alongside management of physical complications.

The lack of consensus in the CostED study regarding the number of cases of eating disorders seen by a service per year may reflect differences in the configuration of services across geographical regions, with more heavily populated areas being more likely than rural areas to receive a large number of eating disorders referrals, and thus rural areas being less likely to rate this as important. There may be an argument, therefore, for including this criterion only in reference to services likely to receive a minimum number of referrals per year given the size of the population they serve, and not to allow it to penalise services that may be 'specialist' in accordance with all other criteria, but where the size of the population that they serve will make it hard to achieve a specified minimum number of cases per year.

To explore the implications of the uncertainty around this fourth criterion, the criterion was excluded from the main analyses but included in sensitivity analyses, with services being required to see a minimum of 50 cases per year to be classified as specialist (the number of cases selected by the majority of respondents to the Delphi survey).

In terms of service use, the number of hospital admissions (mean 0.54 specialist, 0.60 generic), the length of hospital admissions (mean 32 nights specialist, 31 nights generic) and the number of outpatient contacts [including CAMHS contacts (mean 30 specialist, 27 generic)] were similar in the specialist and generic CAMHS groups. Young people initially assessed in specialist services were more likely to receive care in

an eating disorders facility whereas young people initially assessed in generic CAMHS were more likely to receive care in general psychiatry facilities, which may simply reflect geography and the location of services.

There were no significant differences in total costs over the 12-month follow-up period between the specialist and generic groups in either the unadjusted (mean difference £1230, 95% CI –£14,529 to £16,988; $p = 0.878$) or the adjusted (mean difference –£7106, 95% CI –£23,590 to £9379; $p = 0.396$) analyses. However, adjustment for baseline variables resulted in observed differences favouring participants in specialist services (costs were lower, on average) because of significant baseline differences, with those initially assessed in a specialist service having poorer CGAS scores and %mBMI, both of which were prespecified covariates. This suggests that specialist services were more likely than generic services to assess more severely ill young people or those with more complex needs.

In terms of outcomes, at 6 months, %mBMI was significantly higher in the specialist group than in the generic group (mean difference 2.58%, 95% CI 0.16% to 5.01%; $p = 0.037$), but this difference was no longer significant at 12 months. There were no significant differences in CGAS score or HoNOSCA at either the 6- or the 12-month follow-up. In terms of remission and relapse rates, at 6 months, the proportion of young people exhibiting partial remission was greater in specialist services (25%) than in generic services (22%), but rates were similar by 12 months (27% in both groups). Full remission was evident only for a small proportion of the total sample (approximately 10% at 6 months and 24% at 12 months) and was lower in the specialist group than in the generic group at both 6 months (6% specialist, 14% generic) and 12 months (21% specialist, 27% generic). Relapse rates at 12 months were low and differed little between groups (5% specialist, 6% generic).

Cost-effectiveness analyses suggest that initial assessment in a specialist service has a higher probability (> 50% irrespective of willingness to pay) of being cost-effective than initial assessment in generic CAMHS, for both the CGAS and %mBMI. However, no firm conclusion can be drawn without knowledge of society's willingness to pay for improvements in these outcomes. Decision modelling did not support the hypothesis that changes to the provision of specialist services would generate savings for the NHS, with results and sensitivity analyses suggesting that cost per 10-point improvement in CGAS score varies little as the percentage of participants taking the specialist or generic pathway is varied.

Sensitivity analysis including the fourth criterion (> 50 cases seen per year), which failed to achieve consensus for either inclusion or exclusion, resulted in a decrease in the proportion of cases judged to have been initially assessed in specialist services. Although follow-up outcomes remained similar between the two groups, differences in baseline clinical scores were more pronounced, with those classified as being assessed in a specialist service being more severely ill at the point of assessment, and differences in cost were more pronounced and higher in specialist services than generic services. These Delphi-informed sensitivity analyses suggest that, in common with the results of the main analysis, more severely ill young people are more likely to be referred to specialist services and, despite this baseline disparity, specialist eating disorders services achieve outcomes similar to those of generic CAMHS, although at greater cost. In terms of cost-effectiveness, results were less favourable for specialist services in the sensitivity analysis as a result of the greater cost differences. Specialist services remained more likely to be cost-effective than generic CAMHS when using %mBMI as the outcome measure, but in the case of CGAS score this finding applied to only low levels of willingness to pay. Beyond willingness-to-pay levels of £3000 per unit of improvement in CGAS score, there was a higher probability of generic services being cost-effective. Thus, the results were found to be sensitive to the inclusion of this criterion, but only for the CGAS analysis.

Conclusions

Anorexia nervosa is a relatively rare disorder, making it difficult to recruit adequately powered samples for clinical trials. The CostED surveillance study was able to gather eligible case notifications from almost 80 services across the UK and the RoI and, although follow-up rates were lower than expected at both the 6- and 12-month follow-up points, the sample was still larger than has been achieved to date in randomised clinical trials carried out in this population. The results suggest that, on average, young people with anorexia nervosa initially assessed in specialist eating disorders services or generic CAMHS had similar outcomes and costs at 12 months' follow-up, but those assessed in specialist services were more severely ill when they were first diagnosed, which resulted in specialist eating disorders services having a higher probability of being cost-effective than generic CAMHS. This finding was true for a wide range of values of willingness to pay for outcome gains; however, the lack of a clear willingness-to-pay threshold for the outcomes measured in the study means that no firm conclusions can be reached. The results did not suggest that providing more specialist services would save money for the NHS, given similar costs and outcomes, so decisions about which service type to fund could be made with reference to other factors, such as the preferences of patients and carers.

Future research should focus on the evaluation of eating disorders services as they evolve following the publication of commissioning standards for the provision of community-based eating disorders services for young people in June 2015 and the announcement of £30M of recurrent funding to support the transformation of these services. In addition, given the chronic nature of anorexia nervosa, longitudinal follow-up is needed to provide data to support assessment of the longer-term costs and benefits of CAMHS for young people as they transition into adulthood, and whether or not lifespan services provide a cost-effective alternative to age-specific care. In terms of methods, future research should consider approaches for the collection of outcome data suitable for economic decision-making, such as generic, preference-based measures capable of generating quality-adjusted life-years, currently recommended by the National Institute for Health and Care Excellence and similar health technology assessment bodies in a number of countries around the world.

Trial registration

This trial is registered as ISRCTN12676087.

Funding

Funding for this study was provided by the Health Services and Delivery Research programme of the National Institute for Health Research.

Health Services and Delivery Research

ISSN 2050-4349 (Print)

ISSN 2050-4357 (Online)

This journal is a member of and subscribes to the principles of the Committee on Publication Ethics (COPE) (www.publicationethics.org/).

Editorial contact: journals.library@nihr.ac.uk

The full HS&DR archive is freely available to view online at www.journalslibrary.nihr.ac.uk/hsdr. Print-on-demand copies can be purchased from the report pages of the NIHR Journals Library website: www.journalslibrary.nihr.ac.uk

Criteria for inclusion in the *Health Services and Delivery Research* journal

Reports are published in *Health Services and Delivery Research* (HS&DR) if (1) they have resulted from work for the HS&DR programme or programmes which preceded the HS&DR programme, and (2) they are of a sufficiently high scientific quality as assessed by the reviewers and editors.

HS&DR programme

The HS&DR programme funds research to produce evidence to impact on the quality, accessibility and organisation of health and social care services. This includes evaluations of how the NHS and social care might improve delivery of services.

For more information about the HS&DR programme please visit the website at <https://www.nihr.ac.uk/explore-nihr/funding-programmes/health-services-and-delivery-research.htm>

This report

The research reported in this issue of the journal was funded by the HS&DR programme or one of its preceding programmes as project number 11/1023/17. The contractual start date was in June 2013. The final report began editorial review in February 2018 and was accepted for publication in August 2018. The authors have been wholly responsible for all data collection, analysis and interpretation, and for writing up their work. The HS&DR editors and production house have tried to ensure the accuracy of the authors' report and would like to thank the reviewers for their constructive comments on the final report document. However, they do not accept liability for damages or losses arising from material published in this report.

This report presents independent research funded by the National Institute for Health Research (NIHR). The views and opinions expressed by authors in this publication are those of the authors and do not necessarily reflect those of the NHS, the NIHR, NETSCC, the HS&DR programme or the Department of Health and Social Care. If there are verbatim quotations included in this publication the views and opinions expressed by the interviewees are those of the interviewees and do not necessarily reflect those of the authors, those of the NHS, the NIHR, NETSCC, the HS&DR programme or the Department of Health and Social Care.

© Queen's Printer and Controller of HMSO 2019. This work was produced by Byford *et al.* under the terms of a commissioning contract issued by the Secretary of State for Health and Social Care. This issue may be freely reproduced for the purposes of private research and study and extracts (or indeed, the full report) may be included in professional journals provided that suitable acknowledgement is made and the reproduction is not associated with any form of advertising. Applications for commercial reproduction should be addressed to: NIHR Journals Library, National Institute for Health Research, Evaluation, Trials and Studies Coordinating Centre, Alpha House, University of Southampton Science Park, Southampton SO16 7NS, UK.

Published by the NIHR Journals Library (www.journalslibrary.nihr.ac.uk), produced by Prepress Projects Ltd, Perth, Scotland (www.prepress-projects.co.uk).

NIHR Journals Library Editor-in-Chief

Professor Ken Stein Professor of Public Health, University of Exeter Medical School, UK

NIHR Journals Library Editors

Professor John Powell Chair of HTA and EME Editorial Board and Editor-in-Chief of HTA and EME journals. Consultant Clinical Adviser, National Institute for Health and Care Excellence (NICE), UK, and Senior Clinical Researcher, Nuffield Department of Primary Care Health Sciences, University of Oxford, UK

Professor Andrée Le May Chair of NIHR Journals Library Editorial Group (HS&DR, PGfAR, PHR journals) and Editor-in-Chief of HS&DR, PGfAR, PHR journals

Professor Matthias Beck Professor of Management, Cork University Business School, Department of Management and Marketing, University College Cork, Ireland

Dr Tessa Crilly Director, Crystal Blue Consulting Ltd, UK

Dr Eugenia Cronin Senior Scientific Advisor, Wessex Institute, UK

Dr Peter Davidson Consultant Advisor, Wessex Institute, University of Southampton, UK

Ms Tara Lamont Director, NIHR Dissemination Centre, UK

Dr Catriona McDaid Senior Research Fellow, York Trials Unit, Department of Health Sciences, University of York, UK

Professor William McGuire Professor of Child Health, Hull York Medical School, University of York, UK

Professor Geoffrey Meads Professor of Wellbeing Research, University of Winchester, UK

Professor John Norrie Chair in Medical Statistics, University of Edinburgh, UK

Professor James Raftery Professor of Health Technology Assessment, Wessex Institute, Faculty of Medicine, University of Southampton, UK

Dr Rob Riemsma Reviews Manager, Kleijnen Systematic Reviews Ltd, UK

Professor Helen Roberts Professor of Child Health Research, UCL Great Ormond Street Institute of Child Health, UK

Professor Jonathan Ross Professor of Sexual Health and HIV, University Hospital Birmingham, UK

Professor Helen Snooks Professor of Health Services Research, Institute of Life Science, College of Medicine, Swansea University, UK

Professor Ken Stein Professor of Public Health, University of Exeter Medical School, UK

Professor Jim Thornton Professor of Obstetrics and Gynaecology, Faculty of Medicine and Health Sciences, University of Nottingham, UK

Professor Martin Underwood Warwick Clinical Trials Unit, Warwick Medical School, University of Warwick, UK

Please visit the website for a list of editors: www.journalslibrary.nihr.ac.uk/about/editors

Editorial contact: journals.library@nihr.ac.uk