

Co-ordinated care for people affected by rare diseases: the CONCORD mixed-methods study

Stephen Morris,^{1*} Emma Hudson,¹ Lara Bloom,²
Lyn S Chitty,³ Naomi J Fulop,⁴ Amy Hunter,⁵
Jennifer Jones,⁵ Joe Kai,⁶ Larissa Kerecuk,⁷
Maria Kokocinska,⁷ Kerry Leeson-Beevers,⁸
Pei Li Ng,⁴ Sharon Parkes,⁷ Angus IG Ramsay,⁴
Amy Simpson,⁵ Alastair Sutcliffe,⁹ Christine Taylor⁴
and Holly Walton⁴

¹Department of Public Health and Primary Care, University of Cambridge, Cambridge, UK

²The Ehlers-Danlos Society, London, UK

³North Thames Genomic Laboratory Hub, Great Ormond Street NHS Foundation Trust and the University College London Great Ormond Street NHS Foundation Trust, London, UK

⁴Department of Applied Health Research, University College London, London, UK

⁵Genetic Alliance UK, London, UK

⁶Centre for Academic Primary Care, University of Nottingham, Nottingham, UK

⁷Birmingham Women's and Children's NHS Foundation Trust, Birmingham, UK

⁸Alström Syndrome UK, Torquay, UK

⁹Great Ormond Street Hospital for Children NHS Foundation Trust, University College London, London, UK

*Corresponding author sm2428@medsch.cam.ac.uk

Declared competing interests of authors: Stephen Morris has been a member of the following National Institute for Health Research (NIHR) committees: NIHR Heath and Social Care Delivery Research (HSDR) Funding Board (2014–19), NIHR HSDR Commissioning Board (2014–16), NIHR HSDR Evidence Synthesis Sub-board (2016), NIHR Health Technology Assessment (HTA) Clinical Evaluation and Trials Board (associate member 2007–9), NIHR HTA Commissioning Board (2009–13), NIHR Public Health Research Funding Board (2011–17) and NIHR Programme Grants for Applied Research Expert Sub-Panel (2015–19). Naomi J Fulop is a NIHR Senior Investigator and was a member of the NIHR HSDR Funding Board (2013–18). Amy Hunter and Jennifer Jones have both received funding from Alexion Pharmaceuticals (Boston, MA, USA) outside the submitted work. Angus IG Ramsay was an associate NIHR HSDR Funding Board member (2015–18).

Plain English summary

The CONCORD study

Health and Social Care Delivery Research 2022; Vol. 10: No. 5

DOI: 10.3310/LNZZ5321

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

Plain English summary

More than 3.5 million people in the UK are affected by a rare condition. These conditions can be severe and disabling and affect more than one part of the body, meaning that patients often require treatment from different experts. Many people have to visit a variety of health services to receive care, some of which, such as the local hospital or general practitioner, may be close to home, and others, such as hospitals where they can see different specialists, may be further away. This can cause problems because co-ordination between these health-care providers is often not very good and there may be gaps in care due to patients not seeing the right professionals. In addition, when patients do see the right professionals the information to facilitate the right care may not be available. The aim of this study was to find out how well the care of people with rare conditions is co-ordinated in the UK and how people affected by rare conditions would like their care to be co-ordinated. We undertook a review to develop a definition of co-ordinated care and identify what co-ordinated care might involve. We undertook a survey of patients, parents/carers and health-care professionals to find out about current experiences of care co-ordination. The survey found that for the majority of people affected by rare diseases care was not well co-ordinated, with limited access to care co-ordinators, specialist centres and care plans. We interviewed people affected by rare conditions and found that poorly co-ordinated care can have negative impacts on their mental and physical health, as well as on their finances. We found that patients and families would like their care to be better co-ordinated and, hypothetically, are willing to pay for this. We developed a way of classifying how care of rare diseases can be co-ordinated, which could help to improve the experiences of people affected by rare conditions.

Health and Social Care Delivery Research

ISSN 2755-0060 (Print)

ISSN 2755-0079 (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

This journal was previously published as *Health Services and Delivery Research* (Volumes 1–9); ISSN 2050-4349 (print), ISSN 2050-4357 (online)

The full HSDR 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 and Social Care Delivery Research* journal

Reports are published in *Health and Social Care Delivery Research* (HSDR) if (1) they have resulted from work for the HSDR programme, and (2) they are of a sufficiently high scientific quality as assessed by the reviewers and editors.

HSDR programme

The HSDR 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 HSDR programme please visit the website at <https://www.nihr.ac.uk/explore-nihr/funding-programmes/health-and-social-care-delivery-research.htm>

This report

The research reported in this issue of the journal was funded by the HSDR programme or one of its preceding programmes as project number 16/116/82. The contractual start date was in June 2018. The final report began editorial review in February 2020 and was accepted for publication in September 2021. The authors have been wholly responsible for all data collection, analysis and interpretation, and for writing up their work. The HSDR 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 HSDR 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 HSDR programme or the Department of Health and Social Care.

Copyright © 2022 Morris et al. This work was produced by Morris et al. under the terms of a commissioning contract issued by the Secretary of State for Health and Social Care. This is an Open Access publication distributed under the terms of the Creative Commons Attribution CC BY 4.0 licence, which permits unrestricted use, distribution, reproduction and adaption in any medium and for any purpose provided that it is properly attributed. See: <https://creativecommons.org/licenses/by/4.0/>. For attribution the title, original author(s), the publication source – NIHR Journals Library, and the DOI of the publication must be cited.

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 Professor of Digital Health Care, Nuffield Department of Primary Care Health Sciences, University of Oxford, UK

Professor Andrée Le May Chair of NIHR Journals Library Editorial Group (HSDR, PGfAR, PHR journals) and Editor-in-Chief of HSDR, 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 Consultant in Public Health, Delta Public Health Consulting Ltd, UK

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

Ms Tara Lamont Senior Adviser, Wessex Institute, University of Southampton, UK

Dr Catriona McDaid Reader in Trials, 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 Emeritus Professor of Wellbeing Research, University of Winchester, 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, Child and Adolescent Mental Health, Palliative Care and Paediatrics Unit, Population Policy and Practice Programme, UCL Great Ormond Street Institute of Child Health, London, 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

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

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