Clinical effectiveness and patient perspectives of different treatment strategies for tics in children and adolescents with Tourette syndrome: a systematic review and qualitative analysis

Chris Hollis, 1* Mary Pennant, 2 José Cuenca, 1 Cris Glazebrook, 1 Tim Kendall, 2 Craig Whittington, 2 Sarah Stockton, 2 Linnéa Larsson, 2 Penny Bunton, 3 Suzanne Dobson, 4 Madeleine Groom, 1 Tammy Hedderly, 5 Isobel Heyman, 6 Georgina M Jackson, 1 Stephen Jackson, 7 Tara Murphy, 8 Hugh Rickards, 9 Mary Robertson 10 and Jeremy Stern 4

Declared competing interests of authors: Madeleine Groom reports grants from Shire Pharmaceuticals and personal fees from Janssen Pharmaceuticals outside the submitted work.

¹Division of Psychiatry and Applied Psychology, Institute of Mental Health, University of Nottingham Innovation Park, University of Nottingham, Nottingham, UK

²National Collaborating Centre for Mental Health, Royal College of Psychiatrists, London, UK

³School of Psychological Sciences, University of Manchester, Manchester, UK

⁴Tourettes Action, The Meads Business Centre, Farnborough, Hampshire, UK

⁵Paediatric Neurology Department, Kings College Hospital NHS Foundation Trust, London, UK

⁶Department of Child and Adolescent Mental Health, Great Ormond Street Hospital for Children, London, UK

⁷School of Psychology, University of Nottingham, Nottingham, UK

⁸Institute of Neurology, University College London, London, UK

⁹National Centre for Mental Health, Birmingham, UK

¹⁰Department of Neurology, St George's University Hospitals NHS Foundation Trust, London, UK

^{*}Corresponding author

Plain English summary

Strategies for tics in children and adolescents with Tourette syndrome

Health Technology Assessment 2016; Vol. 20: No. 4 DOI: 10.3310/hta20040

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

Plain English summary

rourette syndrome (TS) is a common condition affecting up to 1% of children and young people and is characterised by motor and vocal tics. TS frequently occurs together with attention deficit hyperactivity disorder, autism spectrum disorder and obsessive-compulsive disorder. The main treatments for tics are medication and behavioural interventions. The study has two main components: part 1 – to identify from a systematic review of the published research literature of controlled trials which treatments for tics are most effective and part 2 – to understand from the perspective of young people with TS and their parents how they experienced treatment and care for TS, including which interventions they found helpful or harmful and what outcomes they valued most. Part 2 involved a national online survey of 358 parents/carers (295 contributing usable data) and in-depth interviews with 40 young people with TS. In the part 1, study a ntipsychotics and noradrenergic agents [clonidine (Dixarit®, Boehringer Ingelheim)] were found to be effective for treating tics in children and young people with TS. However, antipsychotics can cause troublesome adverse effects (sedation, weight gain and neurological symptoms) and aripiprazole (Abilify®, Otsuka) may be better tolerated than other antipsychotics. Habit reversal therapy was also found to be effective for treating tics. Overall, the number and quality of clinical trials is low and this downgrades the strength of the evidence and conclusions. Larger and better-conducted trials addressing important clinical uncertainties are required. The key themes identified in part 2 were difficulties in access to specialist care, delay in diagnosis, limited access to behavioural interventions (only 25% of young people had a behavioural intervention), the importance of anxiety and emotional symptoms in TS, lack of provision of information to schools and inadequate information regarding medication and adverse effects.

HTA/HTA TAR

Health Technology Assessment

ISSN 1366-5278 (Print)

ISSN 2046-4924 (Online)

Impact factor: 5.116

Health Technology Assessment is indexed in MEDLINE, CINAHL, EMBASE, The Cochrane Library and the ISI Science Citation Index.

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

Editorial contact: nihredit@southampton.ac.uk

The full HTA archive is freely available to view online at www.journalslibrary.nihr.ac.uk/hta. 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 Technology Assessment journal

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

Reviews in *Health Technology Assessment* are termed 'systematic' when the account of the search appraisal and synthesis methods (to minimise biases and random errors) would, in theory, permit the replication of the review by others.

HTA programme

The HTA programme, part of the National Institute for Health Research (NIHR), was set up in 1993. It produces high-quality research information on the effectiveness, costs and broader impact of health technologies for those who use, manage and provide care in the NHS. 'Health technologies' are broadly defined as all interventions used to promote health, prevent and treat disease, and improve rehabilitation and long-term care.

The journal is indexed in NHS Evidence via its abstracts included in MEDLINE and its Technology Assessment Reports inform National Institute for Health and Care Excellence (NICE) guidance. HTA research is also an important source of evidence for National Screening Committee (NSC) policy decisions.

For more information about the HTA programme please visit the website: http://www.nets.nihr.ac.uk/programmes/hta

This report

The research reported in this issue of the journal was funded by the HTA programme as project number 10/142/01. The contractual start date was in June 2012. The draft report began editorial review in August 2013 and was accepted for publication in August 2014. The authors have been wholly responsible for all data collection, analysis and interpretation, and for writing up their work. The HTA editors and publisher have tried to ensure the accuracy of the authors' report and would like to thank the reviewers for their constructive comments on the draft 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 HTA programme or the Department of Health. 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 HTA programme or the Department of Health.

© Queen's Printer and Controller of HMSO 2016. This work was produced by Hollis *et al.* under the terms of a commissioning contract issued by the Secretary of State for Health. 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).

Editor-in-Chief of Health Technology Assessment and NIHR Journals Library

Professor Tom Walley Director, NIHR Evaluation, Trials and Studies and Director of the HTA Programme, UK

NIHR Journals Library Editors

Professor Ken Stein Chair of HTA Editorial Board and Professor of Public Health, University of Exeter Medical School, UK

Professor Andree Le May Chair of NIHR Journals Library Editorial Group (EME, HS&DR, PGfAR, PHR journals)

Dr Martin Ashton-Key Consultant in Public Health Medicine/Consultant Advisor, NETSCC, UK

Professor Matthias Beck Chair in Public Sector Management and Subject Leader (Management Group), Queen's University Management School, Queen's University Belfast, UK

Professor Aileen Clarke Professor of Public Health and Health Services Research, Warwick Medical School, University of Warwick, UK

Dr Tessa Crilly Director, Crystal Blue Consulting Ltd, UK

Dr Peter Davidson Director of NETSCC, HTA, UK

Ms Tara Lamont Scientific Advisor, NETSCC, UK

Professor Elaine McColl Director, Newcastle Clinical Trials Unit, Institute of Health and Society, Newcastle University, UK

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

Professor Geoffrey Meads Professor of Health Sciences Research, Health and Wellbeing Research and Development Group, University of Winchester, UK

Professor John Norrie Health Services Research Unit, University of Aberdeen, UK

Professor John Powell Consultant Clinical Adviser, National Institute for Health and Care Excellence (NICE), 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 Institute of Child Health, UK

Professor Helen Snooks Professor of Health Services Research, Institute of Life Science, College of Medicine, Swansea University, 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 members of the NIHR Journals Library Board: www.journalslibrary.nihr.ac.uk/about/editors

Editorial contact: nihredit@southampton.ac.uk