

Institution: The Open University

Unit of Assessment: A3 Allied Health Professions, Dentistry, Nursing and Pharmacy

Title of case study: Autism – Impact on clinical practice and raising awareness

1. Summary of the impact

Dr Rosa Hoekstra engages in research on the biological, cultural and cognitive aspects of autism, and in communicating the findings of these studies to a wider audience. Her involvement in developing a rapid quantitative instrument of autistic traits has aided diagnostic practice. Through The Open University's OpenLearn website and open educational resources associated with the Health Education And Training (HEAT) programme, Hoekstra contributes to raising autism awareness both in the UK and abroad.

2. Underpinning research

Dr Hoekstra's autism research is multidisciplinary, with topics ranging from assessing the genetic and environmental influences on autism and autistic traits to exploring cognitive phenotypes of autism to studying autism in Ethiopia. Her twin, family and questionnaire development studies link the variability in genetic and environmental processes with cognitive and behavioural outcomes in autism, leading to a better understanding of the heterogeneity inherent within the autism spectrum. In her more recent work in Ethiopia, she examined how to increase autism awareness using a low cost and scalable method that reaches the most under-served communities in the world. Her research efforts include:

1. Study of the aetiology of autism using twin and family designs. She conducted the first twin family study of autistic traits in a non-child sample, providing evidence that autistic traits are under substantial genetic influence in late adolescence [3.3]. Her studies in twins and families provide evidence for the strong genetic influences on autism and autistic traits (see [3.5] for a review) and show that sub-clinical autistic traits are more frequently observed in first degree relatives of people with autism [3.6]. During part of this work, Hoekstra was supported by a fellowship awarded by the Netherlands Organisation for Scientific Research (€87,480).
2. Dr Hoekstra's research has contributed to the development and translation of the Autism-spectrum Quotient (AQ), a quantitative measure to assess individual differences in autistic traits. The original measure was developed by Baron-Cohen and colleagues. Hoekstra was involved in developing the adolescent version of this questionnaire [3.1]. She also led the validation of the Dutch translation [3.2] and the development of an abridged version of the questionnaire – the AQ-Short [3.4].
3. Hoekstra received \$199,750 funding from the USA-based charity Autism Speaks to conduct a study of the effectiveness of mental health training for rural health extension workers. The project aims to raise autism awareness and decrease stigma related to mental health problems in rural Ethiopia. Rural health extension workers in Ethiopia provide essential basic health care services to a population of over 80 million people. The Health Education And Training (HEAT) upgrading programme was developed by The Open University in collaboration with the Ethiopian Federal Ministries of Health and Education, UNICEF and the African Medical and Research Foundation (AMREF). Hoekstra and colleagues evaluated the effectiveness of the mental health part of the HEAT curriculum by interviewing health extension workers who had just completed the training and comparing their knowledge, attitudes and practices with health workers who were not yet trained. The first findings indicated that trained health workers had fewer stigmatising attitudes and incorrect beliefs regarding the causes and treatment of autism compared to untrained health workers. However, there were several remaining gaps in the knowledge of HEAT-trained health workers [5.5]. In addition, Hoekstra and colleagues surveyed parents with a child with autism in Ethiopia (Nigussie et al., IMFAR autism conference 2013) and interviewed stakeholders involved in autism service provision in Ethiopia (Tekola et al., Autism Europe conference, 2013); both studies indicated the lack of diagnostic and educational service provision and high levels of stigma experienced by families affected by autism.

3. References to the research

1. Baron-Cohen, S., Hoekstra, R.A., Knickmeyer, R. and Wheelwright, S. (2006) 'The Autism-Spectrum Quotient (AQ) – adolescent version', *Journal of Autism and Developmental Disorders*, vol. 36, pp. 343–350.
2. Hoekstra, R.A., Bartels, M., Cath, D.C. and Boomsma, D.I. (2008) 'Factor structure, reliability and criterion validity of the Autism-spectrum Quotient (AQ): a study in Dutch population and patient groups', *Journal of Autism and Developmental Disorders*, vol. 38, pp. 1555–1566.
3. Hoekstra, R.A., Bartels, M., Verweij, C.J.H. and Boomsma, D.I. (2007) 'Heritability of autistic traits in the general population', *Archives of Paediatrics & Adolescent Medicine*, vol. 161, pp. 372–337.
4. Hoekstra, R.A., Vinkhuyzen, A.A., Wheelwright, S., Bartels, M., Boomsma, D.I., Baron-Cohen, S., Posthuma, D. and van der Sluis, S. (2011) 'The construction and validation of an abridged version of the autism-spectrum quotient (AQ-Short)', *Journal of Autism and Developmental Disorders*, vol. 41, pp. 589-596.
5. Ronald, A. and Hoekstra, R.A. (2011) 'Autism spectrum disorders and autistic traits: a decade of new twin studies', *American Journal of Medical Genetics Part B*, vol. 156B, pp. 255–274.
6. Sucksmith, E., Allison, C., Baron-Cohen, S., Chakrabarti, B. and Hoekstra, R.A. (2013) 'Empathy and emotion recognition in people with autism, first-degree relatives, and controls', *Neuropsychologia*, vol. 51, pp. 98–105.

Grants

2012-2014. Autism Speaks \$199,750. Title: Increasing autism awareness in Ethiopia: The HEAT+ project. PI: Hoekstra, R.A and Long, L-A., Davey, G.C.B., Hanlon, C., Fekadu, A.

2007-2009. Netherlands Organisation for Scientific Research €87,480. Title: Refining the autism phenotype: parent and twin studies of the Autism-Spectrum Quotient and cognitive endophenotypes. PI: Hoekstra, R.A.

4. Details of the impact

Hoekstra's autism research is making an impact on several levels:

- clinical practice (through the development of a rapid quantitative instrument of autistic traits)
- by raising awareness, both in the UK and in the Netherlands (through a book, conference talks, articles and interviews in the popular media such as the *New Scientist* and the BBC)
- by raising mental health and autism awareness in Ethiopia (Hoekstra's mental health research project).

Clinical impact and impact on families affected by autism

The Autism-spectrum Quotient (AQ) is an instrument to assess autistic traits, both in the research context and in the clinic. According to the Dutch national psychiatry consortium for autism spectrum disorders in adults [5.1], the Dutch translation of the AQ (validated by Hoekstra et al., [3.2]) is the most commonly used instrument in clinical practice to aid autism spectrum diagnoses in adults in the Netherlands.

The British National Collaborative Centre for Mental Health (NCCMH) developed the UK NICE guidelines for autism spectrum disorders in adults simultaneously and in collaboration with the team devising the multidisciplinary guidelines for the Netherlands. The Dutch guidelines recommend using Hoekstra's Dutch translation of the AQ as a case identification instrument [5.2]. Apart from the paper and pencil version, the Dutch AQ is now also included as a digital test application and even though the digital application only started recently, it has been used 34 times by 22 different institutions over one month (July 2013) [5.3].

The international impact of Hoekstra's research is further evidenced through her development, in collaboration with Ethiopian psychiatrists and mental health experts, of mental health education materials for the HEAT programme in Ethiopia [5.4]. A first cohort of 1,367 health extension

workers started studying these materials in 2010/2011, a further 20,000 students are expected to enrol in the programme. The first 204 students graduated in 2012; Hoekstra and colleagues evaluated the mental health study sessions of HEAT in this first cohort of graduated students, and compared their knowledge, attitudes and skills with health workers who had not yet received training. The first findings of the study with HEAT-trained health workers showed less stigmatising attitudes and incorrect beliefs regarding the causes and treatment of autism. Based on the findings from this study, the research team produced additional training materials comprising five videos showing how to conduct a clinical interview with parents of a child with intellectual disability or autism, and a mental health pocket guide with dedicated sections on child mental health including autism and intellectual disability. The Ethiopian Federal Ministry of Health has formally approved these study materials and intends to distribute the pocket guide to all 39,000 health extension workers in the country. All study sessions and training materials are free and openly available online, with the potential for re-use in other under-resourced countries. Hoekstra is included in the World Health Organisation's global autism network. In 2013 she presented the first project findings at an autism consultation organised by the World Health Organisation and Autism Speaks [5.5].

Public engagement

Research advances made by Hoekstra informed her book, *The Autism Spectrum in the 21st Century: Exploring Psychology, Biology and Practice*, co-authored with Drs Ilona Roth, Chris Barson, Greg Pasco and Terry Watson [5.6]. The book is seen as 'a must-have manual' of 'extraordinarily good value' [5.7]. Parents of children with autism have commented on how helpful they have found the book: (i) 'every parent should be given this book upon receiving such a devastating diagnosis, as although my son was diagnosed more than a year ago, and despite reading much on the internet, I have learnt so much more from the book. ...' (ii) 'I find it [a] very good summary ... and [the book] actually helped me to adjust the course of actions in ... helping my son.'

Hoekstra's research also features on The Open University's OpenLearn website, for which she and colleagues Ilona Roth and Mark Hirst produced the following materials:

- Autism quiz: http://www.open2.net/autism/autism_quiz.html
- What is autism?: <http://www.open.edu/openlearn/body-mind/health/health-sciences/what-autism>
- The genetics of autism: <http://www.open.edu/openlearn/body-mind/the-genetics-autism>
- Autism, mind and brain: <http://www.open.edu/openlearn/body-mind/autism-mind-and-brain>
- Autism and talent: http://www.open2.net/autism/autism_and_talent.html
- Unzip your genes animated quiz: <http://www.open.ac.uk/openlearn/science-maths-technology/science/biology/unzip-your-genes>.

Altogether, these autism-related educational pages have attracted over 62,000 unique visitors in the period between April 2011 and July 2013. The 'Unzip your genes' page has attracted more than 37,800 unique visitors since April 2011; and more than 20,000 people have completed the Unzip your genes quiz. The 'Unzip your genes' page includes not only a quiz, but also two animated videos explaining 'what is meant by genetic difference' and 'what are twin studies'. The two animations have been viewed around 26,000 and 35,500 times between April 2011 and July 2013. The videos can be found on the 'unzip your genes' page, direct links to the YouTube videos are: <http://www.youtube.com/watch?v=a5yzRRvROpE> and <http://www.youtube.com/watch?v=BTYCv1ObZrI>. Hoekstra frequently gives talks to a wider audience [5.8] and her research and her comments on autism research findings are covered in the popular media [5.9].

5. Sources to corroborate the impact

1. Organisation that can corroborate claim of impact: CASS18+ (Dutch Psychiatry Consortium): <http://www.cass18plus.nl/>. The Psychiatrist and CLASS 18+ board member can be contacted regarding the claim that the Dutch translation of the AQ, validated by Hoekstra et al., 2008, is the most commonly used instrument in clinical practice to aid autism spectrum diagnoses in adults.
2. Dutch multidisciplinary guidelines autism spectrum disorders in adults [Multidisciplinaire

Impact case study (REF3b)

- richtlijn Autismespectrumstoornissen bij volwassenen. Richtlijn voor de diagnostiek en behandeling van autismespectrumstoornissen bij volwassenen.] Final concept guidelines for authorisation, October 2012: www.ggzrichtlijnen.nl.
3. Organisation that can corroborate claim of impact: TelePsy: www.telepsy.nl. The Director, Telepsy can be contacted regarding the digital application of the AQ and can confirm how frequently it is used by clinical care institutions in the Netherlands.
 4. The Open University's Health Education and Training (HEAT) Programme: www.open.ac.uk/heat
 5. Consultation on ASD and other developmental disorders: *From Awareness Raising to Capacity Building*, 16–18 September 2013, World Health Organisation Headquarters, Geneva.
 6. Roth, I. with Barson, C., Hoekstra, R., Pasco, G. and Watson, T. (2010) *The Autism Spectrum in the 21st Century: Exploring Psychology, Biology and Practice*, London, Jessica Kingsley Publishers.
 7. Ratcliffe, book review of 'The Autism Spectrum in the 21st Century: Exploring Psychology, Biology and Practice', *Learning Disability Today*, June/July 2012. p. 30.
 8. Selected talks by Dr Hoekstra:
Dutch Autism Society Conference, 7–8 October 2011 (<http://www.autisme.nl>; audience around 1200 people); BPS London Lectures, December 2010 (<http://www.bps.org.uk/news/london-lectures>; audience 800 people); National Autism Conference, Rotterdam, 16 March 2012 (<http://www.nationaalautismecongres.nl/>; audience 600 clinical and academic professionals) and its sister conference in Frankfurt, 17 March 2012 (<http://www.autismus-kongress.de/>; audience 200 clinical and academic professionals).
 9. Selected media coverage:
New Scientist (online and print edition) 20 August 2011: Commentary on new autism family research [<http://www.newscientist.com/article/mg21128264.000-autism-risk-rises-for-siblings-of-autistic-children#.UpMOR9JSh8E>]; *New Scientist*, 25 June 2011: Coverage of research findings [<http://www.newscientist.com/article/dn20589-childhood-autism-spikes-in-geek-heartlands>]; *Financial Times*, 15 July 2011 [<http://www.ft.com/cms/s/2/03cb43a0-acff-11e0-9623-00144feabdc0.html#axzz2ju6OKHRS>]; Dutch newspapers, 20–21 June 2011: 'NRC Handelsblad', 'Trouw', 'AD', 'de Telegraaf', 'Metro', 'de Pers', 'Eindhovens Dagblad'; 23 October 2010.