



# THE AUTISM DIVIDEND

Reaping the rewards of better investment

By Valentina Lemmi, Martin Knapp and Ian Ragan  
on behalf of the National Autism Project



NATIONAL  
AUTISM  
PROJECT



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From Professor Sir Michael Rutter

The National Autism Project is to be warmly congratulated in producing this highly informative and broad-ranging, but easily accessible, report. Bravely, it highlights the fact that most policy and practice is not supported by evaluation of effectiveness (let alone cost-effectiveness) by studies of good quality. Rightly, this leads to a call for much greater research funding, but further discussion is needed on both the level of research expertise that is available and the types of research that are most needed.

Whereas far too many reviews shy away from recommending that some treatments should be dropped because of the evidence indicating that they do not work, this report points out that drug trials have mainly failed to show any effects of medication on core symptoms. The report notes some of the ineffective treatments (such as secretin) and those where there are harms as well as benefits – such as the antipsychotics (for example, risperidone) that carry a substantial risk of metabolic ill-effects.

Early screening is advocated but it is unfortunate that there was no discussion of the fact that few of the measures covered the broader phenotype.

Quite rightly, the report recognises that it has to be based on currently available findings, whilst noting that basic science advances in the years ahead are very likely to lead to changes in treatment recommendations based on a better understanding of the mechanisms underlying the biology.

Most of all, the Project has admirably succeeded in achieving a balance between hope and caution on what we really know. That was no easy matter given that some of the findings and conclusions are bound to be somewhat controversial. Overall, it is clear that this is much the best review of the field that has been undertaken to date. Researchers, clinicians and policy makers will all find much to think about in this excellent report.

**Professor Sir Michael Rutter**  
**Professor of Developmental**  
**Psychopathology, Social, Genetic,**  
**and Developmental Psychiatry Centre,**  
**Institute of Psychiatry, Psychology and**  
**Neuroscience, King's College London**

# FOREWORD

From Dr Elizabeth Vallance JP PhD  
Chair, National Autism Project Strategy Board

Some 700,000 people in the UK are autistic – more than the entire population of Sheffield. And although the social cost of autism is reckoned to be a staggering £32 billion a year, annual research funding is a mere £4 million. This clear asymmetry was the starting point of the National Autism Project (NAP) and our report is its response.

We are hugely grateful to the Shirley Foundation for financing the project in its entirety, which has meant we have not had to seek funding out of the area's limited existing financial cake. Indeed, as I said at NAP's launch in April 2015, we hope substantially to increase the size of that cake!

The original aim of the Project was to map research in the area, identify effective, evidence-based and cost-effective practice and then to be the authoritative voice for the dissemination of such interventions and practice while advocating increased research funding in areas where further evidence was required. It became clear from the research undertaken by the LSE, however, that the evidence base for much existing practice was not robust, so that policy and interventions are rarely supported by clear evidence of effectiveness. There is the opportunity, therefore, for what we call the

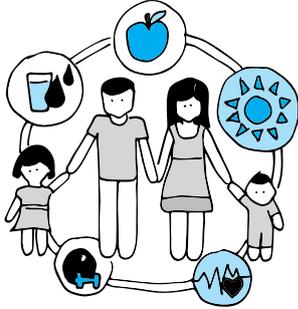
Autism Dividend, which would be the result of properly evidenced practice not only improving the lives of autistic people but making sure that society's resources are allocated in the most cost-effective ways.

NAP has benefitted from the help and support of many people. We have been able to draw on the experience of many experts, from diverse backgrounds, who have worked together always putting the interests of the project and of the autistic community first. Our report has been immeasurably strengthened, too, by the participation at every stage in the project of autistic people who in contributing their time and expertise have helped us to avoid mistakes of emphasis and direction we might have made without their perceptive input. Thanks, too, to the Strategy Board for being the guiding light in this long and complex piece of work.

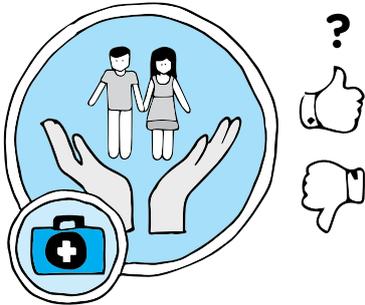
We hope that our report's findings will usher in a new era in research and practice, which will unequivocally improve the lives of all autistic people.

# EASY READ SUMMARY

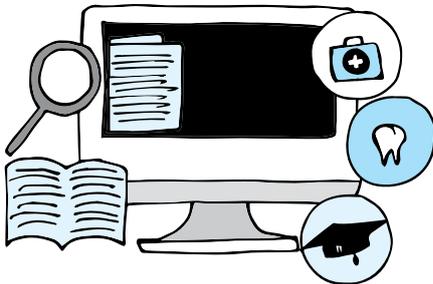
## The National Autism Project



We want autistic people and their families to get what they need to live happy and healthy lives.



Most autistic people need extra support in their lives. But we don't know very much about how good this support is.



This report looks at lots of information about the services and activities that can be used by autistic people and their families.



It has been written by autism experts and researchers. Our Autistic Advisory Panel made sure that the views of autistic people are included.

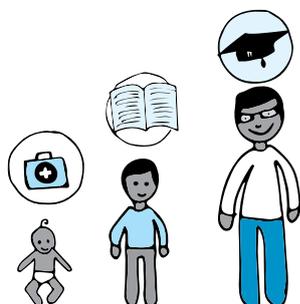


It covers England, Northern Ireland, Scotland and Wales.

## This is what we did



We looked at lots of services and activities used by autistic people.



These services are used by people at different times in their lives, like at school, going to the doctor or getting a job.



We tried to understand whether these services and activities are helpful. We have also tried to work out if they are good value for money.



We found that often there is not enough evidence about services or activities.



We want to see more evidence to help autistic people and local services choose the best option.



## We have 10 ideas to help make the situation better



- 1.** People's autism should be identified as early as possible. This will help them throughout their lives. They should then get access to good support.



- 2.** There should be better evidence that services and activities are good and helpful. Things that we know are bad or dangerous should not be available.



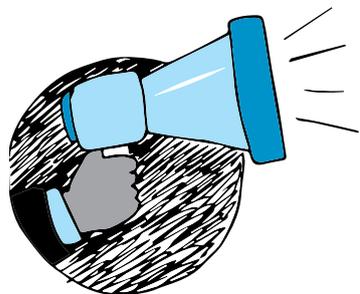
- 3.** People in charge of autism services should think about how much a service or activity that is helpful really costs. Sometimes services or activities for autistic people may cost a lot of money in the beginning. But they might also help to save money in the future.



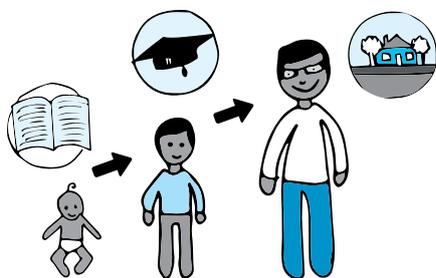
- 4.** More should be done to remove the things that stop autistic people from getting the services or activities that they want or would be good for them. It should be easier for autistic people to get a job and have regular health checks.



- 5.** Lots of autistic people get very stressed or anxious. There should be support for them and their families in the community. Everyone should understand more about what can cause anxiety or stress for autistic people and the simple things they can do to help.
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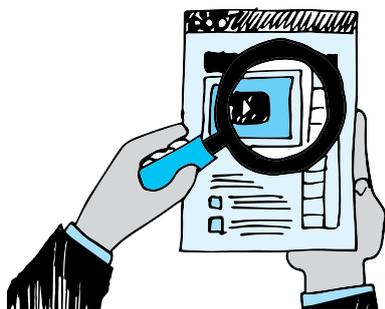
- 6.** There should be a campaign to help everyone understand more about autism. A national campaign is helping to change people's views about mental health. We want one for autism.



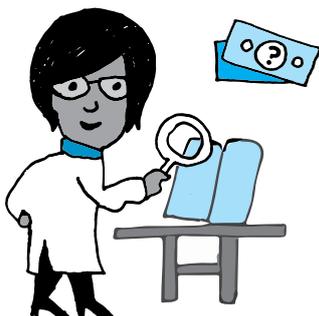
- 7.** There should be better planning for when autistic people have to make big changes in their lives, like leaving school or moving house. Autistic people and parents and carers need good information to help them.



- 8.** Services working with autistic people should work together better. They should think about how money to pay for autism services is best used.



- 9.** We need to collect more information about autistic people. This would help services make better plans.



- 10.** There should be much more money spent on autism research so we can understand better what makes different services and activities good or bad. For example, we need to know more about how to help autistic people get and keep jobs and how to prevent bullying.

# EXECUTIVE SUMMARY

## Background

In 2007, research led by Professor Martin Knapp from the London School of Economics and Political Science demonstrated that the economic consequences of autism in the UK totalled £28 billion per annum. The study, financed by The Shirley Foundation, stimulated other work such as the National Audit Office report in 2009, *Supporting People with Autism through Adulthood*, which showed that effective support could substantially reduce costs. The Autism Act (2009) aimed to improve diagnosis and support for autistic adults in England, while in Wales the first ever national strategy for autism was published in 2008, and other political initiatives followed in Scotland (the Scottish Strategy for Autism launched in 2011) and Northern Ireland (the wide-ranging Autism Act (NI) 2011). These initiatives in all four nations led to further development of action plans, revised strategies and new guidance. It looked as though a new era of more effective recognition and support for autistic people was beginning. Yet nearly a decade on, the needs of autistic people are still unmet and the expected economic dividend never materialised. When the LSE revisited the figures for 2014 using more accurate information the total came to £32 billion. Something is clearly not working.

Concerned that this situation could not continue, The Shirley Foundation established the National Autism Project in early 2015 and asked Professor Knapp and his team to map the existing evidence base and identify clear research gaps. The discovery that the evidence base for the effectiveness of many support practices and interventions in current use was limited or of poor quality led to a shift in focus to whether these offerings were effective and, if effective, whether they made economic sense.

The Shirley Foundation believes that a focus on actions that the evidence shows are both effective and cost-effective will benefit the autistic community and contribute to a reduction in the huge impact of autism on the UK economy. This is the Autism Dividend.

The analysis of evidence was carried out over a period of 21 months. It was based on an extensive literature search of priority areas and topics which were selected with the advice of a team of external experts recruited to the project. These included an Autistic Advisory Panel composed of autistic adults. The experts were also important in helping understanding of the practical and conceptual challenges to developing the economic evaluations and models that underpin the analyses.

The work identified many gaps in the evidence base, leading us to conclude that most policy and practice is not supported by evaluation of effectiveness and cost-effectiveness in studies of good quality. This is hardly surprising in view of the very small sums of money spent on autism research (only £4m per year in the UK) compared with other societal issues of comparable size and importance.

From these gaps, we have selected some of the areas of particular relevance to the lives of autistic people as topics for further research to support good practice. The recommendations of this report therefore fall into two categories: those in policy and practice for which we have made a case based on the existing evidence of effectiveness and cost-effectiveness; and those where research is needed to build the evidence base.

## The Four Principles

In considering the wide range of possible interventions in areas such as education, health, social care and employment, we came across consistent themes or principles that underlie both our recommendations and the challenges most commonly faced by autistic people.

### Principle 1: Personalised actions

Recognising people's individual characteristics and circumstances, different needs and individual preferences.

### Principle 2: Choice and control

Balancing the wish of many autistic people for greater autonomy with effective support structures to meet their needs.

### Principle 3: Addressing inequalities

Providing support and building better understanding and tolerance of difference to address inequalities in access to healthcare, education and employment.

### Principle 4: A life-long perspective

Ensuring early identification and evidence-based interventions during childhood or adolescence, and at all stages of life.

## The Policy Recommendations

Each of the recommendations listed below encompasses very broad areas of policy and practice. To illustrate the evidence base on which they are founded, we have included case studies in the report that contain detailed evidence on reported effectiveness and, where available, cost-effectiveness.

The recommendations are:

1. Ensure timely identification and diagnosis
2. Provide evidence-supported interventions
3. Make the economic case for intervention
4. Remove barriers to access
5. Tackle environmental and other stressors
6. Fight stigma and discrimination
7. Ensure better transitions (for example between child and adult services)
8. Coordinate action across sectors
9. Build better information systems

We are not the first to call on policy makers to implement recommendations of this type. However, our approach has been a rigorous evaluation of evidence backed by expert opinion which we hope will strengthen the case and encourage and support responsible authorities to take action.

## The National Workshops

We recognised that any implementation of the policy recommendations will take different pathways in England, Northern Ireland, Scotland and Wales because of the continuing process of devolution of powers from central government. In order to understand what policies were already in place in the four nations, how present policy is addressing our recommendations and how changing policy might affect their future implementation, workshops were held in London, Belfast, Alloa and Cardiff with charities, professionals, policy makers and autistic people and parents. The outputs of these workshops together with descriptions of national policy and policy initiatives are included in the Appendices of this report.

## The Research Recommendations

A number of research areas emerged from the analysis of the evidence-base where knowledge is currently weak. With stronger supporting evidence though, these areas could have significant impact on the lives of autistic people. Other ideas came from proposals made in the national workshops.

Many of these proposals have been identified by others as important topics for research and some are already under investigation. We hope, however, that the approach we have taken to identify these topics will spur further effort and encourage funders to look at some different areas for research. Most importantly we hope that there will be recognition of the current inadequacy of autism research funding, and understanding that such research is a good investment, one that is vital for the wellbeing of autistic people.

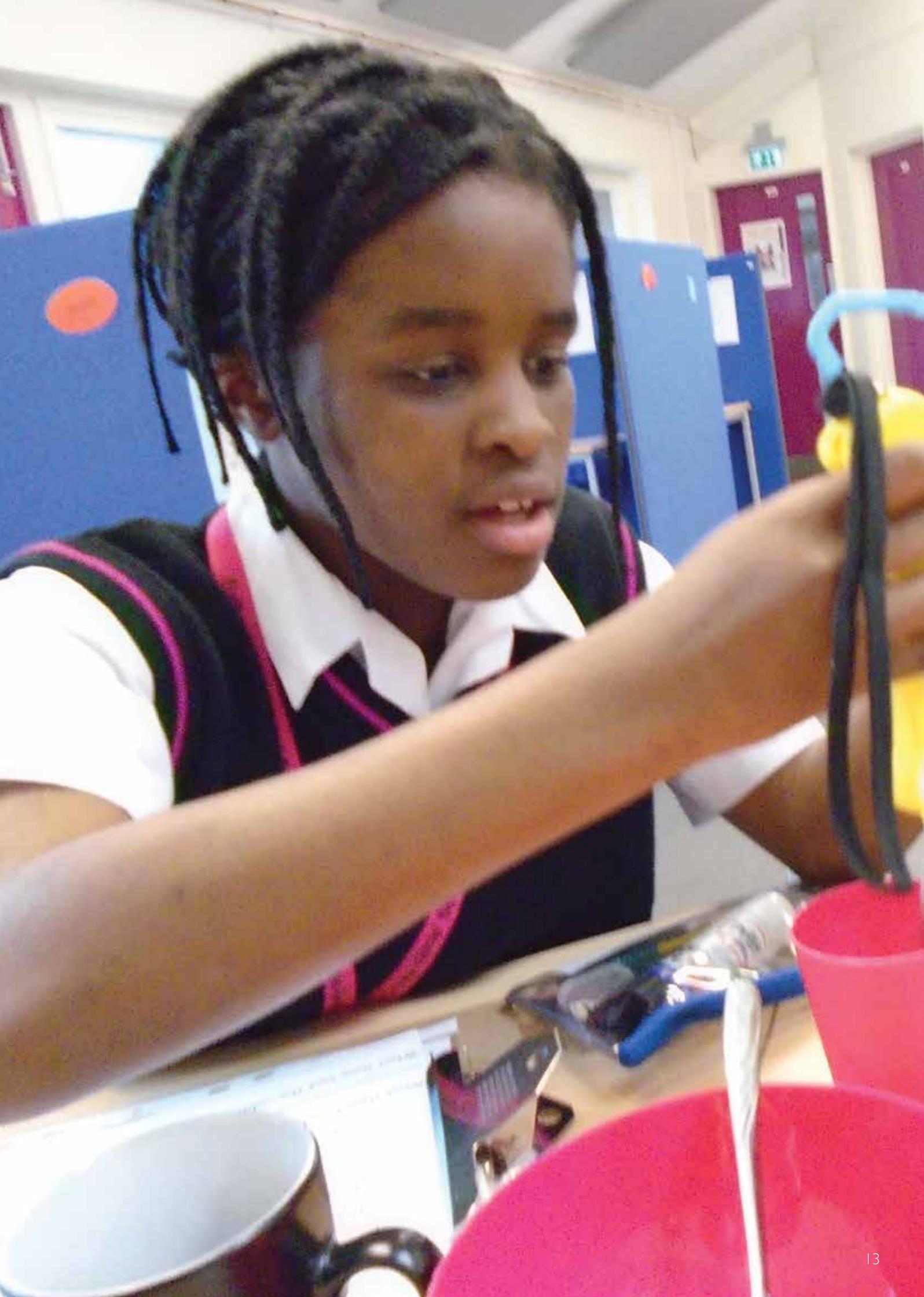
## Conclusions

Our work has found that a great deal more could and should be done to generate evidence to shape policy and improve practice in autism. Too often, the evidence base was found to be weak on benefit and even weaker on cost-effectiveness. Efforts to collect data to substantiate the effectiveness of interventions are inadequate and research to underpin practice is woefully underfunded. The result is a UK economic impact of £32 billion per annum while the needs of autistic people remain unmet. This is an unsustainable state of affairs and must be addressed.

We call on national governments to acknowledge our findings and recommendations on evidence-based practice, to empower and resource service providers in all areas of autism provision to implement the recommendations, and to take our findings into account in their own future planning.

We also call on national governments and research funders to increase investment in autism research from its current low base in order that future decision-making is properly knowledge-based.

Over the next year we will be campaigning to promote the Autism Dividend described in this report so that not only autistic people but society as a whole can reap the rewards of better investment. We will be inviting people associated with the project and others, to write about the “future they would like to see” on our website.



# THE NATIONAL AUTISM PROJECT

Autistic people live in a world that was not designed by or for them to meet their needs. Even the most gifted may require support in order to achieve an acceptable quality of life, and the approximately one third who have learning disabilities<sup>1</sup> are likely to be heavily dependent on service provision.

In 2007, research led by Professor Martin Knapp from the London School of Economics and Political Science demonstrated that the economic consequences of autism in the UK totalled £28 billion per annum<sup>2,3</sup>. The study, financed by The Shirley Foundation, stimulated other work such as the National Audit Office report in 2009, *Supporting People with Autism through Adulthood*, which showed that effective support could substantially reduce costs. The Autism Act (2009) aimed to improve diagnosis and support in England while in Wales the first ever national strategy for autism was published in 2008 and other political initiatives followed in Scotland (the Scottish Strategy for Autism launched in 2011) and Northern Ireland (the wide-ranging Autism Act (NI) 2011). These initiatives in all four nations led to further development of action plans, revised strategies and new guidance. It looked as though a new era of more effective recognition and support for autistic people was beginning. Yet nearly a decade on, the needs of autistic people are still unmet

and the expected economic dividend has never materialised. When the LSE revisited the figures for 2014 using more accurate information the total came to £32 billion, more than that of heart disease, cancer and stroke combined<sup>4</sup>. Something is clearly not working.

“ Nearly a decade on, the needs of autistic people are still unmet. ”

We attribute much of this failure to the fact that research funding for autism lags far behind the amount spent on these or other conditions that, like autism, have a major impact on the wellbeing of so many people. Only about £4 million is spent each year on autism research<sup>5</sup>; equivalent to just £6 for each of the estimated 700,000 autistic people

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<sup>1</sup> We use the term 'learning disability' throughout this report to refer to 'a significantly reduced ability to understand new or complex information, to learn new skills ... with a reduced ability to cope independently, which started before adulthood, with a lasting effect on development' (from the Valuing People White Paper 2001). Another term used to describe the same characteristics is 'intellectual disability'. Learning disability is not the same as 'learning difficulty': the latter refers to one or more specific problems processing certain types of information, such as dyslexia or dyspraxia, and does not affect general intelligence.

<sup>2</sup> Knapp, M., Romeo, R. and Beecham, J. (2007). The economic costs of autism. Foundation for People with Learning Disabilities. [http://eprints.lse.ac.uk/3462/1/Economic\\_consequences\\_of\\_autism.pdf](http://eprints.lse.ac.uk/3462/1/Economic_consequences_of_autism.pdf)

<sup>3</sup> Knapp, M., Romeo, R. and Beecham, J. (2009). Economic cost of autism in the UK. *Autism*, 317-336.

<sup>4</sup> Buescher, A., Cidav, Z., Knapp, M. and Mandell D (2014). Costs of autism spectrum disorders in the United Kingdom and United States of America. *JAMA Pediatrics*, 168, 721-728.

<sup>5</sup> Pellicano, L., Dinsmore, A. and Charman, T. (2013). *A Future Made Together: Shaping Autism Research in the UK*. London: Institute of Education.

in the UK<sup>6</sup>. As a result, there are significant gaps in our understanding of autism and the evidence about the most effective approaches to care, support and service design.

This makes it all the more important that what research money there is should be spent on studies that produce the most benefit for autistic people and their families. There is no overarching strategy for autism research in the UK, and a survey conducted by the Centre for Research in Autism and Education in 2013 revealed that the priorities of researchers are quite different from those favoured by autistic people themselves, their families or other carers<sup>7</sup>. However, a survey from Autistica in 2014 showed that autistic people are enthusiastic about research and would like their views to be considered when deciding on research questions<sup>8</sup>.

With all this in mind, the National Autism Project was created in January 2015 as a three-year project funded by The Shirley Foundation. Its original purpose was to map the existing research base and provide

“ Central to this was the need to understand what was truly important and beneficial across the whole spectrum of activities that affect the lives of autistic people. ”

authoritative recommendations on what further research was needed, with the ultimate aim of substantially increasing the amount of funding that this would then attract.

Central to this was the need to understand what was truly important and beneficial across the whole spectrum of activities that affect the lives of autistic people – both children and adults, as well as their families and carers. In doing so we found several activities and practices that are widely accepted as beneficial, and for some, there was evidence that they were also cost-effective, that is, their use produced benefits that were significant enough to justify the resources needed to achieve them. The evidence base for the effectiveness of many practices which claim to support autistic people was, however, often of limited or poor quality, thereby pointing to the need for further research.

The project is, therefore, now focused on identifying the most effective interventions (i.e. those for which there is robust supporting evidence) and, wherever possible, using economic evidence to examine their cost-effectiveness. By this means we aim to promote the wider use of effective and cost-effective interventions by service providers. We also want to encourage increased research intensity in areas where the evidence base needs to be strengthened.

The selection of the interventions was based on literature reviews and analyses undertaken by a small team at the London School of Economics and Political Science (LSE) in collaboration with a group of experts recruited to the project.

We have also considered the means by which our recommendations might be put into practice. Those which require further research can be directed to the appropriate funders, but those which involve changes to practice need to be mapped onto the policies and service delivery systems currently in place. As these differ in the four devolved nations of the UK, workshops were conducted with charities,

<sup>6</sup> <http://www.autism.org.uk/about/what-is/myths-facts-stats.aspx>.

<sup>7</sup> Pellicano et al. (2013) op.cit.

<sup>8</sup> Wallace, S., Parr, J., Hardy, A. (2014). One in a Hundred: putting families at the heart of autism research. London: Autistica.

professionals, policy makers and autistic people in England, Northern Ireland, Scotland and Wales to consider the recommendations, their relevance to each nation and which policies, current and future, might support and be supported by their implementation. For this, invaluable assistance was offered by the autism charities operating in the four nations.

Over the coming year, we will be taking the findings and recommendations to key decision makers and funders.

## The people involved

The National Autism Project Strategy Board is chaired by Dr Elizabeth Vallance. Members of the Board come from widely different backgrounds and include people on the autism spectrum. Each member is a highly respected figure in his or her own field.

The Board has profited from the advice of two additional groups:

- The Autistic Advisory Panel composed of people with a diagnosis of autism and whose chair, Dr Dinah Murray, is also a member of the project's Strategy Board. The Panel has been influential in helping the neurotypical<sup>9</sup> members of the project to understand better what autistic people really need.
- The Expert Group comprising 25 subject experts who are UK leaders in their respective fields and who have helped to refine the conclusions and recommendations.

Membership of these three bodies is given in Appendix D.

“Over the coming year, we will be taking the findings and recommendations to key decision makers and funders.”

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<sup>9</sup>We use the term 'neurotypical' for all those without a diagnosis of autism.

# WHY THE PROJECT IS NEEDED

## Basic facts about autism

Throughout this report we use the term autism to include everyone on the autism spectrum, regardless of their autistic traits, their intellectual capacity or their dependence on support and services. The term therefore includes those with a diagnosis of Asperger syndrome, those labelled as 'high-functioning', those with severe and complex autism and those with co-occurring conditions. It is generally accepted that these labels are quite inadequate to describe the variety of ways in which autism presents itself. For example, someone of great intellectual ability may be regarded as high-functioning yet be incapable of what would be considered routine and simple activities for the neurotypical majority. Nevertheless, these labels are useful generalisations to illustrate the range of different needs of members of the autistic community, here meaning not only the autistic person but their family and carers as well.

It is often said that 'When you have met one

autistic person you have met one autistic person', meaning that no two autistic people are the same. But why should we expect them to be? Perhaps this is a relic of a medical model which regards autism as an illness with a characteristic set of symptoms. We have moved away from talking about 'people with

“Autism is no more likely to manifest itself identically amongst autistic people than musicality or skill at football.”

autism' to referring to 'autistic people' in order to emphasise that, in this country at least, autism is not regarded as something additional to the individual, like a broken bone or a mental illness, but something intrinsic and no more likely to manifest itself identically amongst autistic people than musicality or skill at football.

## Some facts about autism

- There are at least 700,000 autistic people currently living in the UK, which is around 1% of the population.
- Autism is apparently much more prevalent in males than females but part of this difference may be due to under-reporting of females.
- Autism is a life-long condition for which there is no 'cure'. Indeed the idea of a cure is deeply resented by many autistic people.
- Autism causes difficulties with social communication and interactions, as well as a restricted and repetitive range of behaviours, activities or interests.
- Many autistic people experience sensory-processing difficulties e.g. becoming overwhelmed by their environment, avoiding or not noticing certain sensations, or seeking out certain sensations. These difficulties may heighten their anxiety and stress, impact on their participation and at times cause pain.
- 80% of autistic people, at some point in their lives, have mental health problems such as anxiety and depression, and the majority rate their anxiety as the number one problem in their lives.
- About one in 10 autistic adults has epilepsy, but among those with intellectual disability the prevalence is much higher - up to 30%.
- Approximately one third of autistic people have learning or intellectual disabilities.
- Autistic people are socially disadvantaged: they are more likely to be excluded from schools, to suffer poor healthcare, to be underemployed and to be badly served by the criminal justice system.

Even so, there are features in common among autistic people and these are listed in Box 1. Of course, in a neurotypical world, autistic people are measured against neurotypical norms and not the other way round. One of the most important challenges for society is to reconfigure the neurotypical world to be more accepting of difference.

## The four core principles

From our extensive reading of the research literature, our analyses and our interactions with researchers, our engagement with practice and policy communities, and our many and often deep discussions with autistic people, we have concluded that both the issues facing the autistic community and the recommendations we propose can be rooted in four core principles. While none of these principles is exclusive to autism and indeed may apply to other conditions and interventions, they relate in particular and important ways, outlined below, to interventions in this area.

These principles reflect the challenges that autistic people may face. They underpin how society - neurotypical and autistic - should act. Not surprisingly, these principles apply, on the whole, to how the neurotypical majority should make efforts to accommodate the autistic minority and improve the quality of their lives. But the acceptance of neurodiversity also means that responsibility lies on both sides, and everyone must accept that there are practical limits to such accommodations.

### Principle 1: Personalised actions

Autistic people are as diverse a group as any other, characterised by atypical

and very uneven interests and skills. It is therefore essential for them to have support tailored to their individual needs, strengths and preferences. Policy frameworks and resource commitments should – where this is appropriate and expected to be beneficial – be targeted according to those individual characteristics and circumstances.

This necessity for personalised actions arises from the diverse nature of autism. We do not fully understand its biological basis although we suspect that it is to a great extent genetic. However, the genetic foundation is typically complex and requires the combined effect of variants of many genes in many environments. It is not surprising, therefore, that autism is described as a spectrum condition, but its variability in presentation also leads to variability in needs and strengths, and variability in the actions or interventions to meet those needs.

Personalised actions need to be differentiated not only by how autism itself presents, but also by age, gender, degree of learning disability, sensory sensitivities and co-occurrence of other health problems – and, of course, by recognising individual preferences. The lack of a true biological understanding of autism results in the absence of diagnostic tests that are timely, precise and predictive of what actions and interventions are going to be most beneficial. Parents, carers and practitioners are left with observation and experience to guide them, and autistic children and adults are therefore reliant on the individual skills of those supporting them, skills that in practice are often limited.

The vast range of autistic individuals' capacities and responses indicates that 'one size' clearly cannot fit all. Poorly adjusted

“Of particular concern are interventions that may train autistic people to be unquestioningly compliant, increasing their vulnerability.”

intervention, and lack of timely, tailored social support, may have huge repercussions in terms of life expectancy, risk of depression, anxiety and suicide<sup>10,11,12</sup>. Proxy decision-makers who are not usually autistic themselves tend to make decisions in terms of what autistic people lack. They are seldom told by autistic people what a good autistic life is like. An autistic person may not place a high value on a trait that non-autistic people consider essential, and may have, and value, abilities or affinities that have never occurred to non-autistic people. Intervention may amount to trading one unconventional set of traits with a more conventional, but not objectively superior, set. Assumptions that everyone wants, needs and values the same things must be confronted and reframed to determine their applicability to any individual. Infliction of conventional values may infringe individuals' rights to self-determination.

Of particular concern are interventions that may train autistic people to be unquestioningly compliant, increasing their vulnerability. There is also the potential loss of connection to people who are 'like them' and the risk of ending up rejecting all that is autistic while not being able to pass well enough to succeed in non-autistic terms. And then, there are the adjusted expectations that the autistic person is now doing 'so well' that they can manage

without support and have no excuse for continuing to have difficulty.

Particularly for intensive childhood interventions, there is also considerable risk of turning every activity into therapy with 'learning goals' and 'observations', jeopardising the right of children to have free time for play and recreation (UN Convention on the Rights of the Child). Disabled children are not an exception, and turning every activity into therapy does not constitute giving them 'free time'.

## Principle 2: Choice and control

Autistic people often express the wish for greater autonomy - more choice in the opportunities available to them and greater control over decisions that affect their lives - but empowerment may be pointless unless there are effective support structures in place for some people. For example, personal budgets for social care are a form of empowerment but can be hard to get in practice despite the requirements of the Care Act (2014)<sup>13</sup>. Co-production<sup>14</sup> requires active collaboration to achieve a mutually desirable result.

The diverse nature of autism can lead to disagreement between autistic people, family members and practitioners over what constitutes benefit to the autistic individual (particularly young children and individuals lacking capacity) and what kinds of intervention are acceptable and effective. Ideally, a range of possibilities should be made available to suit people across the autism spectrum, and that reflect the wide variety of co-occurring conditions such as learning disability, epilepsy, anxiety or depression. It is also possible that an autistic individual may choose not to seek assessment or to reveal the results of such an assessment if undertaken, perhaps because they consider a formal label or diagnosis to be a hindrance.

<sup>10</sup> Autistica (2016). Personal Tragedies, Public Crisis. [www.autistica.org.uk/wp-content/uploads/2016/03/Personal-tragedies-public-crisis.pdf](http://www.autistica.org.uk/wp-content/uploads/2016/03/Personal-tragedies-public-crisis.pdf).

<sup>11</sup> Croen, L.A., Zerbo, O., Qian, Y. et al. (2015). The health status of adults on the autism spectrum, *Autism*, 19, 814-823.

<sup>12</sup> Hirvikoski, T., Mittendorfer-Rutz, E., Boman, M. et al. (2016). Premature mortality in autism spectrum disorder. *British Journal of Psychiatry* 208, 232-238.

<sup>13</sup> [www.legislation.gov.uk/ukpga/2014/23/contents/enacted](http://www.legislation.gov.uk/ukpga/2014/23/contents/enacted).

<sup>14</sup> Co-production refers to active involvement of people who use social care, health or other services in decisions about their lives and treatment. Among other things, co-production emphasises that individuals have experience and skills, as well as needs.

This right should be respected, although for children it is necessary for the assessment or diagnosis to be shared in order for support arrangements to be set up. In this as in other respects, diversity should be respected.

Society in general has a right to control the fair and reasonable distribution of resources to individuals. This may at times conflict with the rights of autistic adults to autonomy, which includes the right to make decisions that others may consider unwise. In most cases this conflict can be successfully resolved though co-production as provided for in England by the Care Act (2014).

### Principle 3: Addressing inequalities

Some kinds of inequalities are unavoidable and are not necessarily all unfair. That Usain Bolt can run faster than anyone else is an example of inequality, but unless his ability stems from illicit drug use, for example, then it is not unfair. The term 'inequity' is sometimes used to refer to unfair and avoidable differences arising from neglect, discrimination, poor and corrupt practice, while 'inequality' may simply be a description of the difference between two things.

The analysis of the additional cost of autism to the UK economy<sup>15</sup> revealed many examples of inequalities that are unfair yet could be addressed in principle. For autistic children, particularly those with a learning disability, major contributors to this additional social cost are special education, service use and disrupted parental employment (resulting in lower incomes for them and lost productivity for the national economy). For adults, the major contributors to cost were special accommodation, medical and nonmedical services and productivity losses of the autistic individuals themselves, all of these magnified for those with co-occurring learning disabilities.

Clearly some of these costs are unavoidable, necessary and – because of the care and support services they make possible – also potentially beneficial to the individual and family. But other costs are not; they arise from inequities in access to healthcare or to educational and employment opportunities, and from variations in understanding and tolerance of difference by those coming into contact with autistic people.

### Principle 4: A life-long perspective

Every autistic child becomes an autistic adult. It is not an unreasonable assumption, therefore, that actions taken in infancy and childhood can have lasting benefits in adulthood – personal, societal and economic benefits – and so considerable importance is rightly attached to early identification and appropriate evidence-supported interventions during childhood or adolescence. But actions

“ Every autistic child becomes an autistic adult.”

whose consequences play out over many years are not always immediately attractive to governments who often seek, not entirely unreasonably, a return on investment in the shorter term. With public sector resources so very stretched, actions need to be affordable.

There is also much that can and should be done with immediate benefit to the quality of life of adults as well as children. Ongoing support during adulthood is associated with higher costs because of its duration, but evidence for the effectiveness or cost-effectiveness of such support is weak, and research on autism in adults is particularly underrepresented. Nevertheless, there must be a question about how much of

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<sup>15</sup> Buescher et al. (2014) op.cit.

what is needed now in terms of support for adults (to address mental health needs, for example) could have been avoided by better care, support and education at a younger age. This is an experiment in progress; most autistic adults in the UK today will not have had the level of support as children that nowadays would be regarded as normal. Consequently, their current circumstances are not necessarily a good indication of the life-chances of the next generation. Effective interventions and support are needed and will continue to be needed at all stages of life, whether these are remediating the effects of earlier poor care or not.

Again, from an economic perspective, there may be little incentive for one government department or sector to invest in interventions (such as education) that benefit the budgets

“The idea of spending now to save later or save elsewhere is exactly the strategic approach needed.”

of other departments at some future date (such as health or work and pensions). But ever-tightening budgets and ever-present performance targets have a tendency to encourage short-termism and narrow horizons. The idea of spending now to save later or to save elsewhere - or indeed both (what is sometimes called 'diagonal accounting') – may be challenging for policy makers but is exactly the strategic approach needed to improve the life-chances and wellbeing of autistic people over the life-course.

## Gaps in knowledge, gaps in funding

What emerges from our work is that the legal frameworks designed to support autistic people throughout their lives are fundamentally well-meaning and thoughtfully designed, but often fail to be implemented as intended. Even when entitlement to diagnosis and assessment of need is enshrined in law there are systemic barriers and long delays. Lack of resources, lack of knowledge, inadequate specialist training and poor understanding of autism by key professionals - whether teachers, healthcare practitioners, social care workers or those in the criminal justice system – and by employers, leads to inappropriate responses that blight the lives of autistic people and prevent them from achieving the fulfilment to which they are entitled. Lack of evidence about the best ways to support autistic people results in the wrong choices being made, in turn leading to poor outcomes and a waste of effort and resources. Lack of funding for research, training and provision of services leads to a failure of care and to greater costs in the long run.

The National Autism Project seeks to address the gaps in – and misinterpretations of - knowledge and the gaps in funding through recommendations based on the best available evidence, supported where possible by economic arguments. While the aim is ultimately to improve the quality of life of the autistic community we hope that the effectiveness and economic evidence will encourage policy makers and research funders to reprioritise their efforts to take advantage of 'spending now to save later'.

# METHOD OF WORKING

The research that underpins this report was conducted in the Personal Social Services Research Unit (PSSRU) at the London School of Economics and Political Science (LSE), led by Professor Martin Knapp and Valentina Iemmi. One particular primary aim of the study was to examine the economic case for a range of interventions in the autism area.

By *intervention* we mean any action (including diagnosis) with the potential to improve the lives of autistic individuals and their families, across any relevant dimension of life, at any stage in the life-course, in any 'system' (including education, health, social care, housing, employment and community development) and delivered in any 'sector' (public, third sector, private for-profit, or 'informal').

By *economic case* we mean one or more of a number of things: what is the impact on public or private expenditure? What is the impact on societal resources more generally (including 'hidden' costs such as unpaid support from family or friends)? Does spending on an intervention generate savings in the short term or over a longer period? If it does, who benefits and when? If the net effect of an intervention is to increase costs, is it nevertheless likely to be seen as cost-effective because the outcomes are considered to be worth spending the resources needed to achieve them?

The main research activities were:

- to review evidence on the effectiveness of interventions in the autism area;
- to review and summarise evidence on the cost-effectiveness of interventions;

- to carry out some new empirical analyses to examine the economic case (as the need and opportunity arose); and
- to consult with experts from the research, policy and practice communities, as well as autistic people, about research currently underway, in order to understand the relevance and significance of evidence, to explore the relevance of emerging interventions, and to identify data that could be used in our own analyses.

The work at LSE started in March 2015 with identification of the most promising, high-impact interventions on the basis of completed research, so long as that research was relevant to the UK context. A fuller account of the research methods employed in the project is provided in Appendix A and analysis of some interventions that were looked at in detail is described in the case studies of Appendix B.

This has been the most comprehensive review of a wide range of forms of evidence on autism intervention ever conducted and it has been uniquely informed by the extensive input of the Autistic Advisory Panel and the Expert Group. Nonetheless, the scope of the case studies has been determined by the existing range of relevant research, and as a result much has been left out. There may well be potentially constructive, very low-cost interventions, such as getting carers or teaching staff to tune in to the focused interests that are a diagnostic feature of autism, that have rarely been the focus of formal research.

# THE MEANING OF EVIDENCE

In addition to the four underlying principles that emerged in the course of the National Autism Project, two themes, each of paramount importance, recurred during our examination and discussion of the evidence.

Firstly, better agreement is needed on **what constitutes valid evidence**. Robust evaluations are necessary to generate an evidence base, although we should not only be interested in randomised controlled trials. Positive impacts need to be validated outside the somewhat unreal contexts in which some such trials are conducted before scaling up to wider implementation. Data from observational study designs may help to identify the impacts of risk factors and interventions over comprehensive periods of the life-course. Experiential and other qualitative evidence is essential to complement quantitative data, although it can be hard to generalise from small-sample anecdotal reports. A related issue is whether evidence from research outside the UK can be transferred to the UK practice context, and we discuss this further in some of the case studies in Appendix B.

“ Involving autistic people in evidence-generation greatly improves the chances of finding viable solutions to real problems.”

Available evidence on autism interventions is limited in many respects, including in duration of follow-up, breadth of measured impact, and setting. Much of the research we reviewed during the project was weak in design and execution, and some researchers appeared to over-claim the relevance of their findings for real-world implementation. It was suggested to us that standards of acceptable evidence

in autism are lower than in other areas of healthcare, leading to a plethora of poorly tested approaches. Better overall standards are needed in intervention research: autistic people and their families deserve no less.

It is fundamentally important that multiple perspectives are considered: what might be considered effective to the service provider might be undesirable or completely unacceptable to the service user. A parent’s view on effectiveness might diverge from their autistic adolescent’s view. This does not mean that any of these views are wrong, just that the potential for differences must be factored into the design and interpretation of research evidence, and subsequently into any plans for implementation.

Over and above these potential differences in viewpoint between the autistic person, family member and practitioner, there is the complication that an intervention in one sector (for example, education) could have its main impacts in another sector (health or employment, perhaps) and perhaps only some years later. This may create perverse incentives (the ‘diagonal accounting’ challenge referred to before) when decisions need to be taken about public investment.

Evidence that is intended to be about autistic people should be produced *with* autistic people. Co-production can take many forms, but should permeate all stages of research, from issue identification and question framing, to study design, data analysis and interpretation, and to knowledge exchange and impact. Involving autistic people in evidence-generation greatly improves the chances of finding viable solutions to real problems.

A second recurrent theme throughout the project was the need for more and better informed discussion as to **what are appropriate criteria of success**. There is clearly a need to move beyond 'simply' improving what might be considered 'clinical' outcomes, such as social interaction, communication skills and behaviours, although these are undoubtedly important for many autistic people. Other dimensions to include might be autonomy by reference to individual circumstances and preferences (for example, with the help of assistive technology); having a purposeful life again as defined by the individual (for example, inclusion in school with the appropriate educational support); and improved general wellbeing (for example, being able to participate in individually tailored, meaningful activities with the help of person-centred support).

Assessing success should take into account the perspectives of different stakeholders, seeking to achieve whatever is considered to be the right balance between the competing interests and commitments of autistic people, family members and other carers, public sector commissioners and wider society.

None of this is uncontroversial.



# POLICY RECOMMENDATIONS

## Recommendation 1. Ensure timely identification and diagnosis

Timely identification and subsequent formal diagnosis of autism are surely the most basic of building blocks for an effective and cost-effective autism strategy. In an ideal world, identification of needs should be sufficient for action to be taken, but in practice a diagnosis is often required to access support and services. Identification and diagnosis open the door to assessment of an individual's needs for support, and of how their personal strengths and preferences might affect what should be done. This is also the way to understand the family and community context in which the autistic individual is living, studying, working, participating and engaging.

An issue with autism is the lack of biological criteria on which to make a diagnosis. The *Diagnostic and Statistical Manual of Mental Disorders* (DSM-5, used primarily in the US<sup>16</sup>) makes the point that symptoms must be present in the early developmental period 'but may not become manifest until social demands exceed limited capacities'. This is a common occurrence - children and adults may manage adequately under one set of circumstances, but as a result of increasing demands as they get older or their environment changes, they may reach a point when they are unable to cope. This can result in behavioural problems or mental health issues. The need for tools to diagnose autism before this stage is reached would be hugely beneficial and might emerge from research on the genetic and neurological basis of autism. No such diagnostic tools exist today.

There are screening interventions that aim to identify autistic individuals at an early stage, as well as assessment interventions that aim to diagnose the type of autism and the associated needs. GPs are typically the gateway to a diagnostic assessment which is then carried out by a multidisciplinary team of specialists. It must be competently conducted and accurate, and the findings must be sensitively communicated. The National Institute of Health and Care Excellence (NICE) has produced materials to support diagnosis and multi-agency assessment, as well as recommendations for commissioning support<sup>17</sup>.

“ Identification and assessment must not only be timely but also a springboard to action.”

Typically, autism is diagnosed in the UK at around age five years; this situation has not changed for a decade. There are, however, many adults whose autism has never been identified, in part because services a few decades ago were not geared up to do so. Those adults may therefore not have received the support that could help them and their families. In addition, we do not know whether current diagnostic processes serve women and men equally well; girls are diagnosed later than boys, on average, and there is a relative lack of research on the female presentation of autism.

<sup>16</sup> *Diagnostic and Statistical Manual of Mental Disorders Fifth Edition: DSM-5 (2013)* American Psychiatric Association.

<sup>17</sup> National Institute for Health and Care Excellence (<https://www.nice.org.uk/guidance>).

Identification and assessment must not only be timely but also a springboard to action. Timely diagnosis can open doors to specialist support (in school, for example) and rights (perhaps to welfare benefits). It can be the route for families to get the support they need. It can help to remove some of the uncertainty about why certain traits or behaviours occur ('a relief' to many families); indeed, it may remove guilt from parents who blame themselves for 'bad parenting'.

It can improve the chances of identifying commonly co-occurring conditions such as anxiety and epilepsy, and should therefore contribute to better treatment and health. It

can help someone gain a sense of community. Timely identification and diagnosis might also help to avoid crises which result in unwanted, distressing and costly admissions into hospital, psychiatric intensive care units or residential care settings. However, these potential benefits might not materialise if information systems are inadequate or if key data are not shared appropriately.

Although it was not possible to find evidence to examine the economic case for timely identification, diagnosis and assessment, in Appendix B we summarise the main effectiveness evidence.



## Recommendation 2. Provide evidence-supported interventions

Identification and diagnosis of autism may be very important to the individual but will have little consequence if appropriate services and supports are not subsequently made available. Those services and supports – whether for the individual or their family – should be ‘evidence-supported’. We discussed earlier the sometimes contested meaning of ‘evidence’ in the autism area; differences in viewpoint should be celebrated rather than quashed, but still need to be managed.

A service that is seen as ‘good’ or ‘desirable’ to at least some people could be one that responds promptly to needs, or one that emphasises the individuality of the person and as far as possible takes account of their preferences. It might be seen as ‘good’ because it is effective in meeting those needs and improving wellbeing, or because decisions are shared and those outcomes are co-produced. It might ensure that individuals are supported with dignity, or that family strengths or community solidarities are not undermined. It might promote social inclusion<sup>18</sup>. It might support autonomy, but without leaving any individual exposed to unreasonable risk of harm or excessive stress. It might respond to differences in gender, age, ethnicity, language, religion, culture, sexual preference or socio-economic group in ways that are not discriminatory or unfair. It might make efficient use of scarce resources.

In reviewing the research literature we sought evidence on interventions by reference to all or any of the above criteria. Although our focus was slightly different, we built on the excellent wide-ranging work of Research Autism<sup>19</sup>, and the rigorously developed clinical guidelines

developed by NICE<sup>20</sup>. What is abundantly clear from our work and that of others is that the evidence base is both limited and very uneven in quality and coverage.

The balance between what is known from good research to work for children and what works for adults may also warrant attention. Previous research at LSE showed that 10% of the overall cost of autism arose in childhood and 90% in adulthood<sup>21</sup>. Effective actions in childhood – such as the early interventions that we describe in more detail in Appendix B – might well head off some of those later adulthood costs, as well as being cost-effective in childhood. But more research attention certainly needs to be given to the needs of autistic adults. Evidence on cost, cost-effectiveness and affordability is especially sparse across the life-course and there is almost no research on autism in old age.

“What is abundantly clear is that the evidence base is both limited and very uneven in quality and coverage.”

A marked policy trend in the UK over the past two decades – and one that is observable across many policy areas – is the gradual but inexorable shift in the balance of risk and responsibility from the state to the *individual*. This can be seen, for example, in relation to choice of school, funding of university education, choice of GP and hospital, personal

<sup>18</sup> The term ‘social inclusion’ implies voluntary participation by autistic people.

<sup>19</sup> <http://researchautism.net/>.

<sup>20</sup> For example, feeding into NICE’s autism quality standards: <https://www.nice.org.uk/guidance/qs51>.

<sup>21</sup> Buescher et al. (2014) op.cit.



budgets in social care, pensions and publicly supported housing. This trend goes hand in hand with recognition that many health, social care, education and other interventions will be better at meeting needs or satisfying wants if they recognise and respond to the characteristics, circumstances and preferences of individuals. This is clear from the evidence on personal budgets in social care that we summarise in Appendix B. In the broad medical context (i.e. not specifically in relation to autistic people) there is rapidly growing interest in 'precision medicine' - using diagnostic testing to deliver the right medicine at the right dose to patients with particular genetic or symptomatic profiles.

Research has not kept up, and cannot yet provide the evidence that would help to 'personalise' support for autistic individuals. Nevertheless, access to evidence-supported

interventions in the 21st-century must surely mean access to interventions that are known to be both suitable and acceptable to the individual and their family, whilst ensuring that the already wide inequalities in access are not widened further.

It is also surely time to avoid wasting public money on interventions that are harmful or for which there is negative evidence, such as certain diets, chelation therapy, vaccine avoidance, homeopathic preparations and facilitated communication techniques.

In situations where the evidence base is under-developed or uncertain - such as in relation to mentoring programmes, for which there is encouraging but preliminary evidence - interventions could be supported on the understanding that they will be rigorously evaluated at the earliest opportunity.

## Recommendation 3. Make the economic case

When deciding whether to deliver or fund an intervention, the core question must surely ask if it is going to be effective: does it meet needs, improve functioning or improve wellbeing? Does it do what autistic people or parents believe will be of help and not harmful to them? If the intervention is intended to be preventative, does it actually reduce the risk of illness or other unwanted outcomes? But because resources are scarce – and *always* scarce – decision-makers will also want to know the economic consequences.

Interventions must therefore make economic sense. First, they must be feasible, in the sense that they use resources that can be made available in the UK, such as suitably trained professionals to deliver an early intervention service or clinical psychologists to deliver a psychosocial therapy. Second, they must be affordable within current and expected future budgets, especially given the enormous pressure on public resources today. Third, the intervention must be cost-effective in the sense that the outcomes achieved are sufficiently important to justify the resources that must be spent to generate them. This should not be taken to imply that we believe that the resources presently allocated to autism in the UK are sufficient: they are most clearly not, by reference to both efficiency and equity criteria.

The cost-effectiveness argument is important. For an intervention to make ‘economic sense’ it might not need to be cost-saving. It might actually cost *more* than what is currently done, but the better outcomes are considered by the decision-maker to be worth paying for because the resources that have to be committed cannot be put to better use

elsewhere. It is rarely easy to decide whether the costs are justified by the outcomes: this is a value judgement rather than a scientific ‘result’ and is the reason why NICE decisions and recommendations can sometimes prove controversial.

“ For an intervention to make ‘economic sense’ it might not need to be cost-saving.”

There is another consideration to bring in here. The overall economic impact of autism – estimated to be £32 billion in the UK in 2011 – is an aggregate of all identifiable costs across the life-span. Some of those costs are, in the short term at least, undeniably desirable: they represent the appropriate ways that health, education and other systems identify, assess and respond to the needs of autistic individuals and families. But some costs might result from avoidable crises or because interventions are delivered for which there is either no evidence, or perhaps even evidence of harm.

Reducing the latter (the ‘bad costs’) whilst increasing the former (the ‘good costs’) could represent a better and more equitable way to use society’s scarce human, capital and other resources. Bringing down the overall costs in the long run - by perhaps preventing unnecessary needs from emerging or tackling those that do emerge early enough to avoid distressing difficulties later on - would obviously be more efficient in the grander scheme of things.



2014/12/0

## Recommendation 4. Remove barriers to access

Even when there is evidence that an intervention is effective, and perhaps also cost-effective, it does not immediately follow that policy or practice will change. There might simply not be enough resources available: no spare capacity in the budget, or too few staff with the right skills to deliver the intervention. Some of the approaches to early intervention for young children evaluated in the US, for instance, appear to require large commitments of staff time. This does not mean that those interventions should not be explored, but delivery at scale in the UK looks unlikely at the present time without a substantial shift in how resources are allocated.

Another barrier to access could be that services are only available in some parts of the country but not others, perhaps in the big cities and not in rural areas, or only where there are enthusiastic ‘autism champions’. It may be that the services that are available do not match the needs or preferences of autistic people. Another barrier could be the seemingly perennial difficulty of cross-agency coordination (see Recommendation 8).

Everyone in the UK will be aware of and will face barriers stopping them from accessing all of the healthcare or other services they might want, or taking up desired opportunities to enhance their skills or progress in their careers. But the autistic minority face many more barriers than the neurotypical majority, and that is fundamentally unfair. Two examples can be given to illustrate this: health checks and employment support.

Autistic people generally have more health

problems than other people, and a higher risk of premature death, including a nine-times higher risk of suicide<sup>22</sup>. (We stress though that the latter figure is from a Swedish study and may not translate directly to the UK.) There are many reasons, summarised very well in reports from Autistica (*Personal Tragedies, Public Crisis*<sup>23</sup>) and the Westminster

“Autistic people generally have more health problems than other people, and a higher risk of premature death.”

Commission on Autism (*A Spectrum of Obstacles*<sup>24</sup>). They include above-average rates of co-occurring conditions such as epilepsy and anxiety; lack of understanding and awareness about autism among health and other professionals; difficulties faced by some autistic people in recognising symptoms and expressing their needs; poor information systems; and the fact that many autistic people are economically and socially disadvantaged. Regular health checks would be both effective and cost-effective and they are now being strongly advocated by NHS England. Training healthcare staff so that they have a better understanding of autism would also help considerably. Where co-occurring conditions do arise, such as anxiety, there are interventions that work for some people and that are cost-effective, such as cognitive behavioural therapy (CBT). We discuss these interventions in more detail in Appendix B.

<sup>22</sup> Hirvikoski, T. et al. (2016) op. cit.

<sup>23</sup> [www.autistica.org.uk/wp-content/uploads/2016/03/Personal-tragedies-public-crisis.pdf](http://www.autistica.org.uk/wp-content/uploads/2016/03/Personal-tragedies-public-crisis.pdf).

<sup>24</sup> The Westminster Commission on Autism (2016) *A Spectrum of Obstacles*. Huddersfield: National Children's Group.

We also need to be aware of the risk of double jeopardy: the cumulative disadvantages experienced by many autistic people over their lives could push them into lower socioeconomic groups, and it is these groups in the UK population who on average get a worse deal from the NHS than the middle classes<sup>25</sup>.

There are also many barriers to opportunity. Education support for autistic children has improved immensely over recent years, but not every autistic child gets the opportunity to achieve their full potential. Even if they do well at school or in higher education, they will very often face major barriers entering or remaining in the world of work. According to the National Autistic Society, only 16% of diagnosed autistic adults in England are in full-time employment, and two-thirds are not working at all<sup>26</sup>. Most of those who are not employed do not believe or know whether they will ever be employed.

Employment is the main source of income for most people in this country, but can also affect someone's social status and roles, their social participation, and their self-esteem. Long periods of unemployment or a succession of short term, insecure jobs can be not only economically damaging (perhaps pushing someone into poverty or unmanageable debt) but also utterly demoralising and a major source of stress and poor health. From the perspective of the national economy, it is a huge waste of potential.

It does not have to be like that. Of course, gainful use of time is not just about paid

work, and opportunities must also not be closed off to other rewarding activities such as volunteering and studying.

There are interventions that can make a difference, such as structured employment support, which can be delivered so as to achieve a number of desirable outcomes in a way that is economically attractive (see Appendix B). Interventions that help to build resilience, or address mental health symptoms are also potentially very important. In addition, of course, changing the mind-set of employers could make a huge difference to the chances of autistic people getting and keeping meaningful employment.

<sup>25</sup> Cookson, R., Propper, C., Asaria, M., Raine, R. (2016) Socio-economic inequalities in healthcare in England. *Fiscal Studies*, forthcoming.

<sup>26</sup> National Autistic Society (2016). *The autism employment gap. Too Much Information in the workplace*. London: National Autistic Society

## Recommendation 5. Tackle environmental and other stressors

The very nature of autism leaves people exposed to much higher risk of stress from external demands, and a more frequent and severe experience of it. Launching their campaign for more research into stress in autism, Research Autism noted on their website<sup>27</sup> the very high proportions of autistic adults reporting stress as 'a significant issue' (98%), having 'a high or very high effect on their sleep' (74%) and 'a high or very high effect on their mental health' (86%). Parents and carers reported how stress had 'a high or very high effect on their autistic child's education or work' (95%) and relationships (84%), and how almost all (98%) found it 'difficult or impossible to find effective support for their children'.

Stresses of that intensity greatly increase the risk of anxiety and behaviour that could challenge autistic individuals themselves, their families or others. Interventions that aim to address those behaviours have had some favourable reports - positive behavioural support in the form of 'active support' may work for some children and families - but this approach is not suitable for everyone, and indeed is not without controversy.

What is clear, however, is that far too many children and adults with a learning disability and/or autism who display behaviours that challenge either themselves or others ('concerning behaviours') find themselves accommodated for long periods in unsuitable inpatient settings.

The Transforming Care programme in England – prompted by the abuses in Winterbourne

View hospital - aims to improve the lives of these people, particularly by supporting them in community settings, but NHS England's focus up until now has been to get people out of residential care rather than meeting their needs more broadly. More than a third of the 2600 people with learning disabilities in inpatient settings are autistic, but progress in moving them out of inpatient settings has been slow. There are many more people currently in the community thought to be at risk of being admitted to an inpatient setting. Supporting people in community settings has the potential to save money, and opens up the possibility of reinvesting resources in more cost-effective ways<sup>28</sup>, but may well require a different way of understanding and managing the concept of risk.

Tackling misconceptions about autism, improving awareness of how environmental factors and aspects of the built environment can easily become major stressors and providing appropriate structures and support are all necessary responses. Discrimination and inequality are also both likely to contribute to heightened levels of anxiety, with the danger that someone might get trapped in a never-ending cycle of uncertainty, anxiety, and concerning behaviour. But doing more to avoid those environmental stressors in the first place would obviously be desirable. The Kingwood Trust has, for example, undertaken work in partnership with the Royal College of Art on adapting physical environments to reflect sensory challenges and preferences in autistic people with learning disabilities, suggesting that even low-cost adaptations may be able to reduce stress.

<sup>27</sup> <http://researchautism.net/about-us-research-autism/beating-stress-in-autism>.

<sup>28</sup> Lemmi, V., Knapp, M., Gore, N. et al. (2016). What is standard care for people with learning disabilities and behaviour that challenges and what does it cost? *British Journal of Learning Disabilities*, 44, 309-319.

## Recommendation 6. Fight stigma and discrimination

Access to services and opportunities – and consequently also to the benefits they can generate – are affected by many things, including societal attitudes. Few autistic people or families feel that the UK general public has a good understanding of autism or what it means to be autistic.

Worryingly, professionals in key positions in health, education, housing, criminal justice and other systems often appear to be untrained and under-prepared for effective engagement with autistic people. This ignorance can manifest itself in many ways: annoyance from the public when children ‘misbehave’, peer-victimisation and bullying at school, hate-crime, social isolation, difficulties in getting a job or pursuing vocational options, poor access to appropriate health and social care, psychological distress and even suicide. Also, family members will often experience negative attitudes and social isolation.

Stigma and discrimination seem to be endemic: some of the associated problems stem from lack of awareness, and some perhaps from more insidious suspicion and hostility. At a societal level, anti-stigma interventions are needed to raise awareness, improve knowledge, reduce prejudice and directly tackle discrimination. At the level of individual experience, anti-bullying and similar interventions are needed to offer protection from what could be a lifetime of distress and damage.

In fact, there has been very little research focused on whether these initiatives work in relation to autism (see Appendix B). In

other fields – especially in the mental health area – there are now a number of studies pointing to the success of some national anti-stigma campaigns (such as *Time to Change* in England, and *See Me* in Scotland). Social attitudes can be changed, albeit slowly. Those campaigns also appear to be cost-effective because they help people living with mental illness to access treatment and to gain paid employment<sup>29</sup>. It is not yet known whether similar efforts being made in the autism field will produce similar results. Nor do we really know whether policy-led ‘reasonable adjustments’ to mainstream services (premises, processes, communications, planning and preparation<sup>30</sup>) make a difference to the lives of autistic people.

There is a similar story when it comes to targeted efforts such as anti-bullying initiatives: there is no evaluative evidence *specifically* relating to autism, but there are broadly encouraging findings from studies in other and wider settings. For example, some school-based anti-bullying programmes and some efforts to tackle cyberbullying have been found to be both effective and cost-effective. But whether those same results apply to the risk of bullying of autistic children or adults is not known.

<sup>29</sup> I Evans-Lacko, S., Courtin, E., Fiorillo, A., et al. (2014) The state of the art in European research on reducing social exclusion and stigma related to mental health: a systematic mapping of the literature. *European Psychiatry*, 29, 381-389.

<sup>30</sup> Department of Health (2014) *Think Autism: Fulfilling and Rewarding Lives*. London: HM Government.

## Recommendation 7. Ensure better transitions

Over the course of our lives, each of us will make many transitions, some of which go smoothly and some of which do not. They could include starting school for the first time or moving between schools, starting and leaving university, beginning our first job, losing a job unexpectedly, moving house, becoming a parent, coping with the death of someone close to us, or suddenly discovering that we have a serious illness.

For anyone with long-term health problems or care needs or requiring special support in their education, there may also be the difficult transition from child and adolescent services to adult services. Transitions of this kind can be stressful for anyone, but particularly so for an autistic person who seeks sameness. Instability can cause enormous anxiety, but this may not be recognised by others or responded to adequately. Recognition and response may be even less likely if the professionals seen by an autistic person in the health, social care, education or housing sectors are themselves changing all the time.

Some transitions are certain to happen and their timing can be predicted – such as leaving school - and so can be prepared for well in advance. Others are common but their timing is unknown, which requires a different kind of preparation. The needs of autistic women undergoing the transitions of pregnancy, labour and early motherhood are poorly recognised and rarely addressed, creating unnecessary risk to both mother and child.

There are also transitions that are always likely to be crises or emergencies such as the unexpected illness or death of the primary carer or supporter. It should now be possible

to pre-empt such crises by ensuring that a full assessment of needs is in place but there is a widespread lack of implementation.

There needs to be better awareness of the stress that any such transitions can cause autistic people: unpredictability is especially dangerous. Parents and other carers need information and support to help them to prepare, particularly when a young person with moderate or severe learning disabilities is facing the very difficult transition into the adult care system. Interventions that can build resilience can also help. The 'reasonable adjustments' recommended for a range of different settings have the potential to help, including housing and designing environments that work throughout the lifespan, but they do actually need to be implemented.

## Recommendation 8. Coordinate action across sectors

Autistic people may have needs that require support from many different systems - health, social care, education, housing, employment, welfare benefits and so on. If those needs are to be appropriately and efficiently met then coordination of support between systems, organisations, budgets and professionals is crucial. This will help to avoid wasteful duplication of effort or (more commonly) avoid gaps developing between systems through which vulnerable people might fall, with distressing and costly consequences.

A more coordinated, informed approach as required by law could also help to 'personalise' assessments and support. The four national workshops (see Appendix C) showed that even within national legislative and policy frameworks there is great variety in how local areas choose to structure their autism services. Local autism leads may be positioned within education, healthcare or social services and the extent to which they are able to coordinate services also varies greatly.

This does not require vertical or horizontal organisational integration: the prevalence of autism is too low to make such macro-level changes sensible in local contexts, and anyway many of the grand attempts at integration within and between health and social care systems have failed. Legislative change is unlikely to be sufficient: implementation requires commitment, ownership, leadership and funding.

One thing that must be done is to overcome the 'silo-budgeting'<sup>31</sup> that is not only common but becoming harder to break down in these fiscally straitened times. (Ironically, it is of course precisely at a time of such profound

resource scarcity that inter-budget and inter-professional coordination is needed.) This requires *proportionate coordination* - creating the means by which different organisations and the professionals within them can work together to find pragmatic solutions for individual autistic people. No two autistic people are the same and so coordination should be as flexible as resources and local contexts allow.

As we have already mentioned, there is a related need to address the 'diagonal accounting' challenge: finding a way to overcome the common situation where an intervention that is known to be effective requires investment in one sector but seems to generate most of its economic pay-offs not only some years later but also in another sector. Such a pattern creates complicated disincentives that require long-term strategic planning.

“ Legislative change is unlikely to be sufficient: implementation requires commitment, ownership, leadership and funding. ”

<sup>31</sup> This occurs when decision-makers are focused intently on protecting their own resources and carefully managing their own spending, but in doing so they shift costs and dump problems onto other decision-makers and budgets.

## Recommendation 9. Build better information systems

Running through many of the above recommendations is a clear need to develop and fully utilise better *information systems* and data on autistic people. For example, coordination of action across different services and systems will inevitably require some sharing of information on needs, previous patterns of support, family and other contexts, and so on. Reliable prevalence data are needed for any strategic planning. Information is also required to monitor progress in relation to, say, assessments, reasonable environmental adjustments or health checks.

The challenges in this regard are all too familiar. Few information systems join up and speak to each other, even within the same system (a situation that has plagued the NHS for a long time), never mind between systems. No single coding is used consistently in primary care to record autism. There is also the very necessary challenge of ensuring proper data protection.

The recent reports from Autistica<sup>32</sup> and the Westminster Commission on Autism<sup>33</sup> recommended an 'autism mortality review' to gather data on health risks associated with autism, as well as better data generally. The Government's Think Autism statement proposed to establish a working group to look at data and information availability and needs<sup>34</sup>. We welcome those suggestions and efforts, and urge that action is taken quickly to support the other recommendations made here through better information gathering.

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<sup>32</sup> [www.autistica.org.uk/wp-content/uploads/2016/03/Personal-tragedies-public-crisis.pdf](http://www.autistica.org.uk/wp-content/uploads/2016/03/Personal-tragedies-public-crisis.pdf).

<sup>33</sup> The Westminster Commission on Autism (2016) *A Spectrum of Obstacles*. Huddersfield: National Children's Group.

<sup>34</sup> Department of Health (2014) *Think Autism: Fulfilling and Rewarding Lives*. London: HM Government.



# RESEARCH RECOMMENDATIONS

The many gaps that we encountered in the evidence base suggest themselves as potential topics for research, and increasing investment in research is our final recommendation which we expand on in this section of the report.

We have already pointed out the very small amount of money spent on autism research in the UK. Approximately £4m per annum works out as £6 per autistic person per annum compared with £220 for each cancer patient and £110 for each person living with dementia. Moreover, since most current UK research on autism is skewed towards children, research spend per autistic adult is even lower, perhaps no more than £1.50 per annum.

The case studies in the Appendices make frequent reference to research conducted outside the UK, including many studies carried out in the USA and Australia. Both countries invest much more heavily in autism research than the UK (£75<sup>35</sup> and £31<sup>36</sup> per autistic person per annum respectively). However, we cannot rely on other countries to do the research that we should be undertaking ourselves. This is particularly the case where service models have to be designed or adjusted to fit into UK-specific funding, access and provision arrangements, and have to take into account the local availability of skilled professionals to deliver them. We have treated findings from non-UK research with some caution in the knowledge that their conclusions and recommendations may not translate directly to the UK context. It is essential that UK-based studies are carried out to test whether encouraging evidence from abroad has relevance here.

Given the enormous societal impact and economic cost of autism, the low spend in the UK seems impossible to justify. Certainly our work and that of others shows no shortage of questions that need answering. The comprehensive review of autism research in the UK undertaken by the Centre for Research in Autism and Education pointed to many gaps as well as achievements<sup>37</sup>.

Recently, Autistica - working in collaboration with the James Lind Alliance, the National Autistic Society, the Autism Alliance and the Autism Research Trust - carried out a nationwide survey among autistic people and their families, charities and clinicians to prioritise the unanswered questions about autism that they agreed were most important, in order to direct future research. This Priority Setting Partnership (PSP)<sup>38</sup> showed that it is still the most fundamental questions that remain unanswered about the effectiveness of interventions in autism. What is striking is the many similarities between these priorities, reached through a wide-ranging survey of opinion, and our own conclusions about the evidence gaps, reached by an entirely different approach. Their top ten questions are shown in Box 2.

Basic biomedical research aimed at furthering understanding of the neurological basis of autism cannot be subject to the same kind

<sup>35</sup> Pellicano et al. (2013) op.cit.

<sup>36</sup> Herbert, D.L., Barnett, A.G., White, R. et al. (2016) Funding for cerebral palsy research in Australia, 2000–2015: an observational study. *BMJ Open*, 6, e012924.

<sup>37</sup> Pellicano et al. (2013) op. cit.

<sup>38</sup> [www.autistica.org.uk/research/top10/](http://www.autistica.org.uk/research/top10/).

## Top ten questions for autism research from the James Lind Alliance Priority Setting Partnership

1	Which interventions improve mental health or reduce mental health problems in autistic people? How should mental health interventions be adapted for the needs of autistic people?
2	Which interventions are effective in the development of communication/language skills in autism?
3	What are the most effective ways to support/provide social care for autistic adults?
4	Which interventions reduce anxiety in autistic people?
5	Which environments/supports are most appropriate in terms of achieving the best education/life/ social skills outcomes in autistic people?
6	How can parents and family members be supported/educated to care for and better understand an autistic relative?
7	How can autism diagnostic criteria be made more relevant for the adult population? And how do we ensure that autistic people maximise their potential and performance in the workplace?
8	How can we encourage employers to apply person-centered interventions and support to help autistic people maximise their potential and performance in the workplace?
9	How can sensory processing in autism be better understood?
10	How should service delivery for autistic people be improved and adapted in order to meet their needs?

### Box 2

of approach as we have used to look at the effectiveness and cost-effectiveness of interventions. The impact of basic research is likely to be some years in the future and its magnitude and importance are difficult to predict. We understand however, that the PSP is exploring the role of both basic and applied research in taking forward its work, and we too believe that answers can best be provided by a combination of the two, basic research acting as a driver to progress in all areas of applied research and practice.

Basic science is essential to give us the understanding of the biology of autism needed to underpin future evidence-supported

practice. For example, better understanding of the genetics and neurobiology of autism could lead to more precise diagnostic tools, rooted in real biological understanding. These tools might enable the identification of subtypes of autism and the better prediction of interventions that are effective and beneficial, so moving towards personalised actions with less recourse to trial and error. Basic research may also be needed to understand the vulnerability of autistic people to co-occurring conditions (e.g. epilepsy, and the mental health problems and anxiety identified in the PSP priorities) and whether their responses to drug treatments for these conditions are different to those of the neurotypical majority.

Recent changes in national and international views on research into mental health and neurodevelopmental disorders are likely to be relevant. The National Institute of Mental Health in the US is moving towards research that is symptom-led and formulated around well-understood neurobiology rather than the classical psychiatric classification models<sup>39</sup>.

For these reasons, it would be a mistake to shift resources away from basic to applied research in the belief that this would in the long term provide greater benefit to the autistic community and indeed, basic research is strongly supported by the autistic community even while they recognise that its current scope does not always match their immediate priorities.

The way to reconcile the disparity between the current scope of research spending and the needs expressed by the autistic community is to increase investment in both basic and applied research with the expectation that, in the long run, this investment will pay a return in improved quality of life as well as economic benefits. The UK has strengths in mental health research and in basic and clinical studies in autism which should be supported. There has also been the important initiative to establish a database of more than 5000 autistic children, adults and families who are potentially willing to participate in research studies. Led by Dr Jeremy Parr at Newcastle University, funded by Autistica, and supported by the NAS and Autism Alliance, the ASD-UK database is already gathering data, and could be used to gather much more<sup>40</sup>.

In Box 3 we list the most important areas for increased research funding that have been identified from our work and which reflect the need for both basic and applied research. We also include ideas that emerged from the four national workshops. This list is not meant to imply in any way that work is not being undertaken in these areas. Quite the contrary, we are aware of current projects addressing several of these topics. We hope though that the list and the supporting evidence will encourage research funders to increase their investment in research of the highest quality in these priority areas.

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<sup>39</sup> [www.nimh.nih.gov/research-priorities/rdoc](http://www.nimh.nih.gov/research-priorities/rdoc).

<sup>40</sup> [www.asd-uk.com/aboutasd-uk/](http://www.asd-uk.com/aboutasd-uk/).

## Research Recommendations

1. Development of new gender- and age-appropriate diagnostic tools and interventions.
2. Basic and applied research to support personalised actions.
3. Basic and applied research on the conditions that co-occur with autism and strategies to prevent or ameliorate their frequency, severity and consequences.
4. High-quality, ideally long-term, studies of interventions of all kinds and across all relevant systems - education, health, social care, housing, employment, criminal justice and beyond - to establish their acceptability, effectiveness and contribution to wellbeing, especially in adults.
5. High-quality studies on the real-world implementation of emerging interventions to establish their feasibility, generalisability, affordability and cost-effectiveness; and development of tools to conduct such studies (such as routine measurement of service utilisation, economic effects on parents, and autism-sensitive measures for health-related quality of life).
6. Research into ways to change social attitudes, prevent bullying and stop discrimination.
7. Studies to find the best ways to coordinate actions and interventions across different systems and different professional groups to stop autistic people falling through the gaps, especially during periods of transition in their lives.
8. Concerted efforts to ensure that research on autism is co-produced with autistic people.
9. Investment in the collection of data on autistic people so as to build information systems that underpin the other recommendations of this report.

### **Research topics from the workshops:**

- a. Research to explore why the average age of diagnosis has not declined over the last ten years despite major research into the early identification of autism.
- b. Collecting evidence on the value of peer support and mentoring.
- c. Exploring through the experience of both autistic children and adults whether a formal label or diagnosis is a help or a hindrance in combating discrimination.
- d. Research into the role of the family in supporting interventions.
- e. Exploring what the outcomes have been for autistic individuals who are signed off from supported services.

# CONCLUSION

Our work has found that a great deal more could and should be done to generate evidence to shape policy and improve practice in autism. Too often, the evidence base was found to be weak on benefit and even weaker on cost-effectiveness.

Efforts to collect data to substantiate the effectiveness of interventions are inadequate and research to underpin practice is woefully underfunded. The result is that autism costs the UK economy £32 billion per annum while the needs of autistic people remain unmet. This is an unsustainable state of affairs and must be addressed.

This report brings together evidence on what works well for autistic people and what makes economic sense, and provides convincing arguments about how it is possible to improve the quality of life of autistic people and their families while at the same time reducing this huge economic cost. This is the Autism Dividend that can be won through better informed decision making and wiser allocation of resources.

The weak evidence base on which decisions are made is not helped by the shamefully low level of funding of autism research. Even a ten-fold increase in current research investment would only bring the UK to a level comparable to the USA and Australia. A sum of £40 million per annum is still a mere fraction of that spent in the UK on cancer research (£500 million per annum) or dementia (currently £90 million per annum, and with substantial increases already budgeted). The Autism Dividend will not be realised fully until major deficiencies in our knowledge of autism and of the effectiveness of interventions are remedied.

We call on national governments to acknowledge our findings and recommendations on evidence-based practice, to empower and resource service providers in all areas of autism provision to implement the recommendations and to take our findings into account in their own future planning.

We also call on national governments and research funders to increase investment in autism research from its current low base in order that future decision-making is properly knowledge-based.

Over the next year we will be campaigning to promote the Autism Dividend described in this report so that not only autistic people but society as a whole can reap the rewards of better investment. We will be inviting people associated with the project and others, to write about the “future they would like to see” on our website.



# APPENDICES

# APPENDIX A:

## Research methods

We started by identifying a *preliminary list of possible interventions* using NICE guidelines relating to autistic children and adults, the list of interventions on the Research Autism website, and expert opinion. We defined the term ‘interventions’ in a broad sense, including any action with the potential to improve the lives of autistic people and/or their families, across any relevant dimension of life, at any stage in the life-course, in any ‘system’ (education, health, social care, housing, employment, community development, or other) and delivered in any ‘sector’ (public, third (charitable) sector, private (for-profit), or ‘informal’). Risk-reduction or prevention strategies could also be included.

Our initial scope was wide-ranging, and included: early interventions; multidisciplinary services; employment support; cognitive-behavioural therapies for individuals with anxiety; person-centred skilled support (e.g. positive behavioural support); screening and diagnostic assessment; parent training and support programmes; health checks; awareness, anti-stigma campaigns and anti-bullying programmes; social skills interventions; assistive and adaptive technology (including augmentative communication); sensory-motor interventions; criminal justice services; creative and expressive therapies; self-management; peer-mentoring and advocacy; animal-assisted activities and therapies; standard services; standard social care services; standard educational services; medications for co-occurring conditions; special diets and dietary supplements; alternative medical procedures, complementary and alternative medicine.

We looked for recent systematic or other literature reviews in these areas. We conducted our own rapid literature reviews for each of the identified

interventions. Searches were conducted between February 2015 and August 2016 in electronic databases specialised in medical and social sciences (PubMed, PsycARTICLES, PsychINFO, CINAHL, SOCIndex, IBSS, Econlit, Cochrane Library, Campbell Library), in websites specialised in autism or health technology appraisal (GOV.UK, GOV.SCOT, GOV.WALES, Northern Ireland Executive, Legislation.GOV.UK, World Health Organization, NICE, Social Care Institute for Excellence, National Audit Office, Scottish Intercollegiate Guidelines Network, Public Health England - Improving Health and Lives Learning Disabilities Observatory, Research Autism, National Autistic Society, Autistica, American Speech and Hearing Association), in Google Scholar, and Google Search. We ran additional searches, tracking citations of included studies using Google Scholar and contacting experts (particularly the NAP Expert Group, the NAP Autistic Advisory Panel, government officials, practitioners and researchers working in the autism area).

In our electronic rapid reviews we used search terms for autism and autistic spectrum disorder (ASD) combined with search terms for each intervention and cost or service use. We included studies assessing the effectiveness or cost-effectiveness of the identified interventions for autistic people. We were particularly interested in interventions for which economic data were available. We included different types of studies: health technology assessments, systematic reviews and meta-analyses, randomised controlled trials, other controlled studies, and non-controlled studies. We were interested in both quantitative and qualitative evidence. We included studies published in either scientific journals or grey literature, and written in English or any other language available within the research team

(French, Spanish and Italian). We did not set limitations on the publication date.

We then identified the *final list of interventions*. We organised the evidence into tables to summarise key information: the name of the intervention/ approach, countries where evaluation was conducted, who the intervention was for (e.g. type of autism diagnosis, level of learning disability, age group), description of the intervention itself, setting (e.g. school, health clinic, at home), components of the intervention (e.g. different therapy types), evidence on effectiveness, evidence on cost-effectiveness, and information on costs of delivery. We discussed our emerging findings with some of the experts noted above, who helped us select the final list of interventions.

We examined the economic case for as many of this final list of interventions as possible. By ‘economic case’ we meant one or more of a number of things: what is the impact on public or private expenditure? What is the impact on societal resources more generally (including ‘hidden’ costs such as unpaid support from family or friends)? Does spending on an intervention generate savings in the short- or long-term, and if so to whom and when? If the net effect of an intervention is to increase costs, is it nevertheless likely to be seen as cost-effective because the outcomes are considered to be worth spending the resources needed to achieve them?

In examining the economic case we considered three possibilities, in each case focused on interventions for which there is evidence on effectiveness:

- (a) If there was already robust economic evidence from the UK then we would provide a summary of the economic case without further analysis.

- (b) If there was robust economic evidence from outside the UK, or partial UK evidence, then we would aim to use some form of mathematical modelling to explore the potential economic case in a UK context; model-based estimates are usually not as strong as estimates from well-designed primary-data studies.
- (c) If there was no previous economic evidence but we could access data or summary parameters from previous evaluations (e.g. effectiveness trials or observational studies) then we would again use modelling or possibly new analyses of the primary dataset to examine economic questions.

Parameters for any simulation modelling were drawn from what we found in the rapid literature reviews, from extant datasets (e.g. trials, cohort studies), and from expert opinion where other evidence was not available.

We set out to look at the widest set of economic impacts (and we separated public, private and societal costs) over different time periods (up to 1 year, from 2-5 years, and over 5 years). However, we could only describe, analyse or comment if suitably robust data were available. It will be seen from the case studies that it was not possible to examine the economic case for a number of the interventions for which there was some effectiveness evidence.

A parallel task was to explore the practical and conceptual challenges in conducting economic evaluation in the autism area that informed the design of the economic models. We conducted over 30 interviews with various stakeholders in the autism area (researchers, clinicians, NGO representatives, policy makers, autistic people and

their carers) to better understand issues related to the opportunities and challenges in performing and using economic evaluation in the area (e.g. societal values on which the economic case is built, capturing the attention of decision makers). Details are available in a working paper<sup>41</sup>.

Provided that studies have been well-conducted, we generally give more emphasis to recent UK evidence than to evidence from other countries or from some years ago. Evidence from outside the UK is very often relevant here, but caution should be exercised when transferring it to a

UK context because of systemic differences in healthcare, education, social care, housing and other arrangements. There might also be differences between countries in the availability of the professionals needed to deliver particular interventions. Generally speaking, economic evidence travels less well between countries than effectiveness evidence, because differences in the organisation of healthcare, education and other systems can generate differences in relative costs. As will be seen from the case studies, there is unfortunately a shortage of good UK evidence in relation to some interventions.

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<sup>41</sup> Rupert, A., Lemmi, V., Knapp, M. (2016). Economic evaluation in the field of autism: practical and conceptual challenges. PSSRU, London School of Economics and Political Science.

# APPENDIX B:

## Case studies

We present eleven case studies here. These are the interventions for which we could find evidence of sufficient quality to draw some conclusions about reported effectiveness, even if our conclusions end up sometimes being tentative. For most case studies we have been able to include some discussion of the economic consequences, but a general finding is that there is very little robust economics evidence. Each of the case studies should be read in the light of the discussion in ***The Meaning of Evidence*** section earlier in this report.

As we have just noted, we were particularly interested in evidence from the UK, but there is generally much more evidence from elsewhere, especially from the United States. Research evidence can often be transferred from one country to another, but we have tried to be alert to differences in education, healthcare, social care and other systems – and to differences in labour markets, welfare benefit entitlements and so on – that might then influence the effectiveness or cost-effectiveness of an intervention in the UK setting.

As noted above, we were interested in evidence using a wide range of research designs. Randomised controlled trials (RCT) are often the most robust way to test the effectiveness and cost-effectiveness of an intervention, but they are not necessarily the best design to answer every question. Moreover, it would often be helpful to supplement an RCT with other evidence, particularly the kind of experiential insights that can come from well-conducted qualitative research. We have noted study design in summarising relevant studies, and occasionally added reservations about robustness or generalisability. Very few studies are conducted over long periods, which is a general limitation of evidence in this field.

In some case studies we have summarised evidence for a wider group than autistic people. For example, some evidence on health checks for people with learning disabilities is, we believe, generalisable to the subgroup of people with learning disabilities who are autistic. Another example is that we believe that some of the evidence on anti-bullying programmes in a general population is relevant to autistic people. Wherever possible, we sought and summarised studies that are autism-specific, but we think that some of this more general evidence can be helpful in informing policy discussion.

### **The eleven case studies are:**

1. Screening and diagnostic assessment
2. Early interventions
3. Social skills interventions
4. Parent training and support
5. CBT for anxiety
6. Employment support
7. Health checks
8. Personalised care and support
9. Assistive and adaptive technologies
10. Anti-stigma and anti-bullying interventions
11. Pharmacological interventions

Many of the studies we reviewed are written in academic language or use medical terms and whilst we have endeavoured to use natural language in our introductions, assessments and conclusions, where we report the findings of individual studies, we do so in the language they use. This includes the terms 'high-functioning autism' and 'low-functioning autism' as defined in each paper.

# SCREENING AND DIAGNOSTIC ASSESSMENT<sup>42</sup>

## Target population

Autistic children and adults with or without a learning disability.

## Context

Autism spectrum disorder (ASD) is associated with impairments in social interaction and social communication, and restricted and repetitive interests and behaviours<sup>43</sup>. While core features are usually present during early childhood, they may not become manifest until later. One in a hundred individuals is autistic, and about one in three autistic individuals has a learning disability<sup>44</sup>. The diagnosis has been found to be relatively stable over the life-course. In a review of 23 different studies of children, anywhere from 53% to 100% of them maintained the diagnosis of autistic disorder while between 14% and 100% were diagnosed with another form of autism at follow-up<sup>45</sup>. However, difficulties have been reported in distinguishing preschool children with autistic-like behaviours who will maintain the diagnosis<sup>46</sup>. The underlying prevalence of autism has been stable over recent years, but diagnosis rates have increased<sup>47</sup>. Median age at diagnosis of autism in the United Kingdom is 55 months, with no change found between 2004 and 2014<sup>48</sup>.

Screening and diagnostic interventions aim to identify autistic children as early as possible to enable appropriate support for the child and family to be put in place. NICE recommended early recognition and referral to a multidisciplinary diagnostic assessment team with a pathway in place for onward referral to specialists<sup>49</sup>. Every child or young person diagnosed with autism should have an allocated case manager or key worker. For diagnosis in adults, NICE recommends use of one of a range of formal assessment tools<sup>50</sup>.

## Intervention

There are two main approaches to assessment and diagnosis:

- Screening interventions such as short questionnaires administered face-to-face by teachers and healthcare professionals or remotely, by telephone.
- Assessment interventions, administered by professionals – such as paediatricians, child and adolescent psychiatrists, speech and language therapists, psychologists - which are longer and more complex.

<sup>42</sup> Case study prepared by Valentina Lemmi, Margaret Perkins and Martin Knapp.

<sup>43</sup> NICE (2011). Autism diagnosis in children and young people. Recognition, referral and diagnosis of children and young people on the autism spectrum. CG128. London: NICE.

<sup>44</sup> Mackay, T. et al. personal communication (report forthcoming).

<sup>45</sup> Woolfenden, S., Sarkozy, V., Ridley, G. et al. (2012). A systematic review of the diagnostic stability of autism spectrum disorder. *Research in Autism Spectrum Disorders*, 6, 345-354.

<sup>46</sup> Russell, G., Golding, J., Norwich, B. et al. (2012). Social and behavioural outcomes in children diagnosed with autism spectrum disorders: a longitudinal cohort study. *Journal of Child Psychology and Psychiatry*, 53, 735-744.

<sup>47</sup> Lundström, S., Reichenberg, A., Anckarsäter, H. et al. (2015). Autism phenotype versus registered diagnosis in Swedish children: prevalence trends over 10 years in general population samples. *BMJ*, 350, h1961.

<sup>48</sup> Brett, D., Warnell, F., McConachie, H. et al. (2016). Factors affecting age at ASD diagnosis in UK: no evidence that diagnosis age has decreased between 2004 and 2014. *Journal of Autism and Developmental Disorders*, 46, 1974-1984.

<sup>49</sup> NICE (2011). op.cit.

<sup>50</sup> NICE (2012). Autism in adults: diagnosis and management: Clinical guideline CG142. London: National Institute for Health and Care Excellence.

## Effectiveness

Evaluations of screening and assessment programmes have been conducted in a range of settings, looking for ways to speed up processes and improve consistency between assessors.

### Screening in schools

A study in New Jersey showed that children can be successfully screened for autism by their usual child care providers or by preschool teachers using two standard screening tools, the Modified Checklist for Autism in Toddlers (M-CHAT) and the Social Communication Questionnaire (SCQ)<sup>51</sup>. The screening process also identified some children with other clinical diagnoses, most typically speech-language disorder.

### Screening in healthcare centres

Two further US studies evaluated screening programmes in healthcare centres. The Each Child Study used a systematic autism screening process administered by physicians for toddlers (aged 14-30 months) in a large, community-based paediatric practice<sup>52</sup>. More autistic children were identified in this way than through clinical judgment or caregiver concerns, although no single approach identified all children with early signs of autism. The

second study examined whether organisational changes in a private primary care practice could help overcome screening barriers and implement the use of the M-CHAT. A retrospective chart review for 99 children achieved overall screening fidelity of 91% over a 7-month period at low cost<sup>53</sup>.

The DIANE project reported a large controlled cross-sectional study of children and adolescents (0-11 years) in the Netherlands using a two-stage screening approach that led to the earlier detection of autism, mainly in children with a low IQ<sup>54</sup>. Components included training for professionals to recognise early signs of autism using the Early Screening of Autistic Traits Questionnaire (ESAT), a specific referral protocol and a specialist, experienced multidisciplinary diagnostic team.

A study of 4000 children looked at a screening programme to predict a clinical diagnosis of autism in a general population of children attending the standard 2.5-year-old check-up in Sweden<sup>55</sup>. The M-CHAT was used to screen, together with observations made by trained nurses of the child's joint attention abilities (JA-OBS). Children thought to be autistic were referred to specialist clinics for further assessment. Of the 1.6% of the study group identified through screening as having a possible autism diagnosis, three-quarters subsequently had that diagnosis confirmed.

<sup>51</sup> Janvier, Y. M., Harris, J. F., Coffield, C. N. et al. (2016). Screening for autism spectrum disorder in underserved communities: early childcare providers as reporters. *Autism*, 20, 364-373.

<sup>52</sup> Miller, J. S., Gabrielsen, T., Villalobos, M. et al. (2011). The each child study: systematic screening for autism spectrum disorders in a pediatric setting. *Pediatrics* 127, 866-871.

<sup>53</sup> Gura, G. F., Champagne, M. T., & Blood-Siegfried, J. E. (2011). Autism spectrum disorder screening in primary care. *Journal of Developmental & Behavioral Pediatrics*, 32, 48-51.

<sup>54</sup> Oosterling, I. J., Wensing, M., Swinkels, S. H. et al. (2010). Advancing early detection of autism spectrum disorder by applying an integrated two-stage screening approach. *Journal of Child Psychology and Psychiatry*, 51, 250-258.

<sup>55</sup> Nygren, G., Sandberg, E., Gillstedt, F. et al. (2012). A new screening programme for autism in a general population of Swedish toddlers. *Research in Developmental Disabilities*, 33, 1200-1210.

### Screening by phone/telehealth

Telephone or telehealth screening has been explored in three US studies. One evaluated telephone-based early developmental and autism screening for vulnerable families who called the standard 2-1-1 service (a human services information line). A sample of parent callers of children aged 0–5 years who expressed concern about their child’s development were invited to complete a questionnaire, administered by a trained care coordinator (the Parents’ Evaluation of Developmental Status (PEDS Online) for children aged 5 or the M-CHAT for children aged 16-48 months). A proof-of-concept study found the approach could potentially improve access to and uptake of screening in a population where access to primary care might be problematic<sup>56</sup>.

An integrated telehealth system for remote administration of an adult autism assessment, designed to be as similar as possible to face-to-face assessment, achieved high-quality interaction that was easy to use<sup>57</sup>. The third study described the 7-item Mobile Autism Risk Assessment (MARA), electronically administered to triage children and adolescents considered at highest risk of being autistic. Comparing MARA scores with diagnosis from the child’s first visit to a developmental behavioural paediatric clinic suggested that the screen offered promise as a way to distinguish autism from other developmental or behavioural disorders<sup>58</sup>.

### Assessment in healthcare centres

A small Scottish study compared diagnoses by local assessment teams (each with at least three professionals from relevant disciplines - educational psychology, paediatrics, speech and language therapy, psychiatric nursing and social work) trained by the specialist autism assessment team with diagnoses by the specialist team. The training included developmental and clinical history-taking specific to autism, use of the Autism Diagnostic Observation Schedule–Generic (ADOS-G) and how to reach a diagnosis using ICD-10<sup>59</sup> criteria. There was good correspondence between the diagnoses of the local teams and specialist team, indicating that the approach could shorten waiting times for diagnosis, as well as improve autism-related clinical skills more generally<sup>60</sup>.

State-wide guidelines were introduced in Western Australian to manage assessments and diagnoses for autistic children, with an open forum for clinicians (paediatricians, psychiatrists, psychologists and speech pathologists) to discuss issues and a state-wide register of newly diagnosed cases. To aid consistency, diagnoses used standardised tools together with current diagnostic criteria. The programme improved consistency across assessments and more cohesive working between assessors, and made it easier to analyse diagnoses over time<sup>61</sup>.

<sup>56</sup> Roux, A. M., Herrera, P., Wold, C. M. et al. (2012). Developmental and autism screening through 2-1-1: reaching underserved families. *American Journal of Preventive Medicine*, 43, S457-S463.

<sup>57</sup> Parmanto, B., Pulantara, I.W., Schutte, J. L. et al. (2013). An integrated telehealth system for remote administration of an adult autism assessment. *Telemedicine and e-Health*, 19, 88-94.

<sup>58</sup> Duda, M., Daniels, J., & Wall, D. P. (2016). Clinical Evaluation of a Novel and Mobile Autism Risk Assessment. *Journal of Autism and Developmental Disorders*, 46, 1-9.

<sup>59</sup> ICD-10 is the 10th revision of the International Statistical Classification of Diseases and Related Health Problems (ICD), a medical classification list by the World Health Organization (WHO).

<sup>60</sup> McClure I., MacKay T., Mamdani H., et al. (2010). A comparison of a specialist autism spectrum disorder assessment team with local assessment teams. *Autism* 14, 1-15.

<sup>61</sup> Glasson, E. J., MacDermott, S., Dixon, G. et al. (2008). Management of assessments and diagnoses for children with autism spectrum disorders: the Western Australian Model. *Medical Journal of Australia*, 188, 288-291.

## Economic evidence

The US study of organisational changes in a private primary care practice described above calculated that the cost of screening was offset by revenue generated by identified cases<sup>62</sup>. Although not a comprehensive economic evaluation, the study at least suggested a financial incentive for private practitioners to be more alert to autism.

The only other economic evidence we could find was simulation modelling carried out by the National Audit Office (NAO), looking at the consequences of introducing specialist multi-disciplinary teams to help identify and assess adults with high-functioning autism, and then provide support in the areas of health and social care, housing and employment. Drawing on data from the best local models found by NAO, their analyses suggested that quite substantial savings to the public purse could be achieved even with modest increases in identification rate<sup>63</sup>. More generally, the NAO argued that specialist multi-disciplinary support and joint working across all areas – clinical, social and employment – could improve transition from childhood to adult services, and support high-functioning adults to achieve better quality of life in a way that offered value for money from a public expenditure perspective.

## Conclusions

Identification, diagnosis and assessment are the most important routes into appropriate support and treatment for autistic children and adults. Under-recognition of needs and missed diagnoses can lead to considerable, long-term difficulties for the individual and the family. Delayed recognition is a bigger issue for girls, indicating at least one source of inequality that should be addressed<sup>64</sup>. Under-recognition and inadequate assessment will frequently lead to high and unnecessary costs for both the public purse and for families.

There have been relatively few studies of identification, diagnosis or assessment, especially in the UK. Some recent studies have explored ways to improve identification and diagnosis rates and processes, with some indications of success. Training a wider group of professionals to identify and diagnose autism, and perhaps using telephone or other remote approaches, might need to be the way forward when health and other budgets are so stretched. Encouraging the use of standardised screening tools is clearly sensible – to help practice learn from and contribute to research, to support consistency in service responses and to facilitate data sharing between services and systems.

There is almost no economic evidence to guide decision-makers.

<sup>62</sup> Gura et al. (2011) op.cit.

<sup>63</sup> National Audit Office (2009). *Supporting People with Autism through Adulthood*. London: NAO.

<sup>64</sup> Rutherford, M., McKenzie, K., Johnson, T. et al. (2016). Gender ratio in a clinical population sample, age of diagnosis and duration of assessment in children and adults with autism spectrum disorder. *Autism*, 20, 628-634.

# EARLY INTERVENTIONS<sup>65</sup>

## Target population

Autistic children with or without learning disability.

## Context

Autistic children can experience significant deterioration in their physical and mental health and educational outcomes during childhood compared to their neurotypical peers<sup>66</sup>. In addition, their mothers are more likely to experience poor mental health and families face higher risk of unemployment and low income. These negative consequences can continue into adolescence and adulthood<sup>67</sup>.

Developmental trajectories are associated with a range of factors: individual characteristics (learning disabilities and early language ability), family context, access to services, and other factors such as being bullied<sup>68,69,70</sup>. The average costs of supporting autistic individuals over their life-course are high - estimated in the UK to be £1.5 million for someone with learning disabilities and £0.92 million for someone without (at 2011 price levels)<sup>71</sup>. Almost two-thirds of parents consider that a lack of timely support was responsible for their child having higher long-term support needs<sup>72</sup>.

## Intervention

Early interventions aim to address the core features of autism in young children. There are many different approaches, but they tend to share some common characteristics:

- use of practices based on theoretical principles, such as developmental/relational principles and applied behaviour analysis (ABA);
- specially trained educational or healthcare professionals to deliver the intervention (e.g. teachers, clinical psychologists or speech and language therapists);
- participation of parents;
- delivery in community settings (schools and healthcare centres) and/or at home.

## Effectiveness

*Preschool Autism Communication Therapy (PACT)* is a parent-child early intervention that aims to improve social interaction and communication in young autistic children (2–5 years old)<sup>73</sup>. Based on developmental principles, the intervention includes

<sup>65</sup> Case study prepared by Valentina Lemmi and Martin Knapp.

<sup>66</sup> Dillenburger, K., Jordan, J.A., McKerr, L., Keenan, M. (2015). The Millennium child with autism: Early childhood trajectories for health, education and economic wellbeing. *Developmental Neurorehabilitation*, 18, 37-46.

<sup>67</sup> Steinhausen, H.C., Mohr Jensen, C., Lauritsen, M.B. (2016). A systematic review and meta-analysis of the long-term overall outcome of autism spectrum disorders in adolescence and adulthood. *Acta Psychiatrica Scandinavica*, 133, 445-452.

<sup>68</sup> Magiati, I., Tay, X.W., Howlin, P. (2014). Cognitive, language, social and behavioural outcomes in adults with autism spectrum disorders: a systematic review of longitudinal follow-up studies in adulthood. *Clinical Psychology Review*, 34, 73-86.

<sup>69</sup> Kirby, A.V., Baranek, G.T., Fox, L. (2016). Longitudinal predictors of outcomes for adults with autism spectrum disorder: systematic review. *Occupation, Participation and Health*, 36, 55-64.

<sup>70</sup> Fountain, C., Winter, A.S., Bearman, P.S. (2012). Six developmental trajectories characterize children with autism. *Pediatrics*, 129, e1112-e1120.

<sup>71</sup> Buescher, A.V., Cidav, Z., Knapp, M., & Mandell, D.S. (2014). Costs of autism spectrum disorders in the United Kingdom and the United States. *JAMA Pediatrics*, 168, 721-728.

<sup>72</sup> Rosenblatt, M. (2008). *I Exist: The message from adults with autism in England*. London: National Autistic Society.

<sup>73</sup> <http://research.bmh.manchester.ac.uk/pact>.

one individual session of 2.5 hours on alternate weeks over a 6-month period, followed by monthly maintenance sessions for 6 months, with parents undertaking daily home-practice. PACT has been administered by speech and language therapists in the UK, but other health professionals (including non-specialist health workers in South Asia) have been successfully trained to deliver it<sup>74</sup>. A UK trial that compared PACT with treatment as usual found improvements in relation to autism symptoms (both social-communication and restricted repetitive behaviours) along with parent-child social interaction and communication after 13 months<sup>75</sup>. The improvement in autism symptoms and child communication were then found to be sustained until ages 7-11 years, over six years following the end of intervention<sup>76</sup>. Another UK trial, currently underway, is evaluating an expanded version of PACT simultaneously in both home and education settings (PACT-Generalised)<sup>77</sup>.

Adapted from the Video Interaction for Promoting Positive Parenting (VIPP) programme<sup>78</sup>, *Video Interaction to Promote Positive Parenting (iBASIS-VIPP)* is a parent-child very early intervention that aims to improve social interaction in infants at high risk of autism (9-14 months old). Based on developmental principles, iBASIS-VIPP comprises

six sessions followed by up to six booster sessions over 5 months, with parents undertaking daily home-practice<sup>79</sup>. The intervention is administered by speech and language therapists or psychologists at home. A UK trial compared iBASIS-VIPP with no treatment in infants at familial high risk of autism, finding moderate improvements in parent-child social interaction after 5 months of treatment<sup>80</sup>. While promising, results need to be interpreted with caution due to the small sample size.

*Early Start Denver Model (ESDM)* aims to improve cognitive and adaptive behaviours in autistic children aged 18–30 months. Based on developmental and applied behavioural analysis principles, ESDM comprises two 2-hour individual sessions per day, 5 days per week, over a 2-year period, and separate parent training twice a month. ESDM is administered by both community-based therapists and parents at home. A meta-analysis of five evaluations found a small positive effect of ESDM (compared to treatment as usual) on cognitive and adaptive behaviours<sup>81</sup>. Some individual trials have found other significant effects, and one showed how these improvements in cognitive and adaptive skills were maintained two years after the end of the intervention<sup>82</sup>. Similar results were found for a less intensive version

<sup>74</sup> Rahman, A., Divan, G., Hamdani, S., et al. (2015). The effectiveness of the Parent-mediated intervention for Autism Spectrum disorders in South Asia (PASS): a randomised controlled trial in India and Pakistan. *Lancet Psychiatry*, 3, 128-136.

<sup>75</sup> Green, J., Charman, T., McConachie, H. et al. (2010). Parent-mediated communication-focused treatment in children with autism (PACT): a randomised controlled trial. *Lancet*, 375, 2152-2160.

<sup>76</sup> Pickles, A., Le Couteur, A., Leadbitter, K. et al. (2016). Parent-mediated social communication therapy for young children with autism (PACT): long-term follow-up of a randomised controlled trial. *Lancet*, 388, 2501-2509.

<sup>77</sup> Improving autistic children's social communication with parents in everyday settings. *BioMed Central DOI 10.1186/ISRCTN25378536*.

<sup>78</sup> Juffer, F., Bakerman-Kranenburg, M.J., Van Ijzendoorn, H. (2008). *Promoting positive parenting: an attachment-based intervention*. New York: Taylor Francis.

<sup>79</sup> Green, J., Wan, M.W., Guiraud, J. et al. (2013). Intervention for infants at risk of developing autism: a case series. *Journal of Autism and Developmental Disorders*, 43, 2502-2514.

<sup>80</sup> Green, J., Charman, T., Pickles, A. et al. (2015). Parent-mediated intervention versus no intervention for infants at high risk of autism: a parallel, single-blind, randomised trial. *Lancet Psychiatry*, 2, 133-140.

<sup>81</sup> Canoy, J.P., Boholano, H.B. (2015). Early Start Denver Model: a meta-analysis. *Journal of Education and Learning*, 9, 314-327.

<sup>82</sup> Estes, A., Munson, J., Rogers, S.J. et al. (2015). Long-term outcomes of early intervention in 6-year-old children with autism spectrum disorder.

of ESDM for children aged 18–60 months: there were 15–20 hours of group sessions and 1-hour individual sessions each week, plus six 2-hour parent sessions over ten months<sup>83</sup>. Results from a three-site evaluation of ESDM presented at the International Meeting for Autism Research in 2014 showed no effect of the intervention on the outcome variables reported, but these findings appear not to have been subsequently published and so cannot be verified. An Australian study showed significant improvements in developmental rate and receptive language development at the end of an ESDM programme compared to a 'generic' intervention programme for autism<sup>84</sup>. The study also found better improvement in verbal development in younger children, but not in non-verbal development or autism severity<sup>85</sup>.

*Early Intensive Behavioural Intervention (EIBI)* is a widely applied intervention to alter autistic behaviours in children aged 2–5 years, based on applied behaviour analysis principles and linked to the Lovaas approach<sup>86</sup>. EIBI involves 20–40 hours per week of individual sessions over 1 to 4 years, and is administered at home or school by an ABA-trained therapist. A recent Cochrane review<sup>87</sup>

included five studies (two from the UK<sup>88,89</sup>) and found limited evidence of effectiveness in improving intelligence, communication and language skills. A meta-analysis of eleven EIBI studies found improvements in IQ, non-verbal IQ, expressive and receptive language and adaptive behaviour<sup>90</sup>. However, a more recent UK controlled before-after study showed dilution of the positive effects two years after the end of the intervention<sup>91</sup>.

*Joint Attention Symbolic Play Engagement Regulation (JASPER)* is a carer-mediated early intervention to improve social communication and joint attention in children aged 1–8 years. Based on developmental and applied behavioural analysis principles, JASPER is structured into two 30-minute individual sessions per week over 10 weeks, including active coaching of carers. It is administered by a therapist in a variety of settings (home, school, community centre). Different versions of JASPER have been extensively evaluated through trials in the US over almost two decades, generally showing improvement in joint attention, social communication and social interactions in different settings, with effects

<sup>83</sup> Eapen, V., Crncec, R., Walter, A. (2013). Clinical outcomes of an early intervention program for preschool children with autism spectrum disorder in a community group setting. *BMC Pediatrics*, 13, 1.

<sup>84</sup> Vivanti, G., Prior, M., Williams, K., Dissanayake, C. (2014). Predictors of outcomes in autism early intervention: why don't we know more? *Frontiers of Pediatrics*, 2:58. doi:10.3389/fped.2014.00058.

<sup>85</sup> Vivanti, G., Dissanayake, C., Victorian ASELCC Team. (2016). Outcome for children receiving the Early Start Denver Model before and after 48 months. *Journal of Autism and Developmental Disorders*, 46, 2441-2449.

<sup>86</sup> <http://www.lovaas.com/about.php>

<sup>87</sup> Reichow, B., Barton, E.E., Boyd, B.A. et al. (2012). Early intensive behavioral intervention (EIBI) for young children with autism spectrum disorders (ASD). *Cochrane Database of Systematic Reviews* 10, CD009260.

<sup>88</sup> Magiati, I., Charman, T., Howlin, P. (2007). A two-year prospective follow-up study of community-based early intensive behavioural intervention and specialist nursery provision for children with autism spectrum disorders. *Journal of Child Psychology and Psychiatry*, 48, 803-812.

<sup>89</sup> Remington, B., Hastings, R.P., Kovshoff, H. et al. (2007). Early intensive behavioral intervention: outcomes for children with autism and their parents after two years. *American Journal on Mental Retardation*, 112, 418-438.

<sup>90</sup> Peters-Scheffer, N., Didden, R., Korzilius, H., Sturmey, P. (2011). A meta-analytic study on the effectiveness of comprehensive ABA-based early intervention programs for children with autism spectrum disorders. *Research in Autism Spectrum Disorders*, 5, 60-69.

<sup>91</sup> Kovshoff, H., Hastings, R.P., Remington, B. (2011). Two-year outcomes for children with autism after the cessation of early intensive behavioral intervention. *Behavior Modification*, 35, 427-450.

maintained after one year<sup>92,93,94</sup>.

The *Milton & Ethel Harris Research Initiative (MEHRI)* is another programme that aims to improve social communication and social interaction skills in young children (aged 2–5 years). Based on developmental principles, MEHRI involves a weekly 2-hour session with the child and parent, and meetings with parents every eight weeks over a 1-year period. MEHRI is administered by speech and language or occupational therapists. A Canadian trial compared MEHRI to usual community care, showing greater improvement in social interactions at the end of the intervention, although not in language skills<sup>95</sup>. Further analyses showed greater improvement in functional language skills<sup>96</sup>.

The *AEIOU Programme* aims to improve learning and development in children aged 2.5–6 years, based on educational and behavioural principles<sup>97</sup>. The programme comprises 25–40 hours per week of small group sessions, over a 2-year period. Sessions offer a range of interventions (e.g. behavioural treatment, joint attention intervention, self-management, story-based interventions) and two augmentative communication systems (Picture Exchange Communication System, and a modified sign language), together with regular parent training and home-visits. AEIOU is

administered in classrooms by a multidisciplinary therapy team (psychologists, speech and language therapists, occupational therapists, teachers, and childcare professionals) with a staff-to-child ratio of between 1:2 and 1:4. An Australian non-controlled before-after study found improvements in autism symptoms and educational skills after one year<sup>98</sup>.

## Economic case

Some of these early intervention approaches have also been the subject of economic evaluation. It should be emphasised that, with the exception of the within-trial evaluation of PACT (see next paragraph), all other economic studies have been models, and some have been criticised on methodological grounds.

An economic study conducted as part of the PACT trial in the UK did not support the cost-effectiveness of PACT when added to usual care compared to usual care alone<sup>99</sup>. The cost of PACT was estimated at £4,105 per child (2006-2007 price levels). PACT was associated with significantly higher service costs (mean difference per child of £4,489) and non-significantly lower societal costs that included effects on parental work and productivity (mean difference £1,385) over a 13-month period.

<sup>92</sup> Lawton, K., Kasari, C. (2012). Teacher-implemented joint attention intervention: Pilot randomized controlled study for preschoolers with autism. *Journal of Consulting and Clinical Psychology*, 80, 687-693.

<sup>93</sup> Goods, K.S., Ishijima, E., Chang, Y.C., Kasari, C. (2013). Preschool based JASPER intervention in minimally verbal children with autism: Pilot RCT. *Journal of Autism and Developmental Disorders*, 43, 1050-1056.

<sup>94</sup> Kasari, C., Gulsrud, A.C., Wong, C. et al. (2010). Randomized controlled caregiver mediated joint engagement intervention for toddlers with autism. *Journal of Autism and Developmental Disorders*, 40, 1045-1056.

<sup>95</sup> Casenhiser, D.M., Shanker, S.G., Stieben, J. (2013). Learning through interaction in children with autism: preliminary data from a social-communication-based intervention. *Autism*, 17, 220-241.

<sup>96</sup> Casenhiser, D.M., Binns, A., McGill, F. et al. (2015). Measuring and supporting language function for children with autism: evidence from a randomized control trial of a social-interaction-based therapy. *Journal of Autism and Developmental Disorders*, 45, 846-857.

<sup>97</sup> Synergies Economic Consulting. (2013). *Cost-Benefit Analysis of Providing Early Intervention to Children with Autism: Estimation of the net economic benefit of early intervention for a cohort of children with autism*. Synergies Economic Consulting Pty Ltd.

<sup>98</sup> Paynter, J., Scott, J., Beamish, W. et al. (2012). A pilot study of the effects of an Australian centre-based early intervention program for children with autism. *The Open Pediatric Medicine Journal*, 6, 7-14.

<sup>99</sup> Byford, S., Cary, M., Barrett, B. et al. (2015). Cost-effectiveness analysis of a communication-focused therapy for pre-school children with autism: results from a randomised controlled trial. *BMC Psychiatry*, 15(1), 1.

A UK economic model used data from the first PACT trial<sup>100</sup> as part of a wider look at speech and language therapies to assess cost-savings for autistic children<sup>101</sup>. We agree with the methodological reservations expressed by NICE in their technology appraisal<sup>102</sup>, and we do not discuss this study further.

A Canadian modelling study estimated the value of ESDM compared to parent-delivered ESDM (1-hour therapist sessions each week over 12 weeks and intervention at home by parents) and usual care (EIBI)<sup>103</sup>. Intervention costs were high: CAN\$100,994 for ESDM, CAN\$692 for parent-delivered ESDM and CAN\$112,000 for EIBI (2013 price levels). From the public service perspective – i.e. looking at all public expenditures – the model suggested that ESDM would cost an additional CAN\$23,000 per person per dependency-free life-year to age 65 when compared to usual care; and an additional CAN\$58,000 per dependency-free life-year when compared to parent-delivered ESDM. From a societal perspective – now also including caring activities and productivity losses for parents - ESDM was found to be more effective and less expensive than both usual care and parent-delivered ESDM.

There have been a few economic modelling studies that have looked at EIBI, but they all appear to have methodological limitations. When NICE reviewed these studies in 2013 it concluded that the studies often had serious methodological problems linked to the selective use of clinical effectiveness data, further modifications made for the economic analyses, or the selection of other model parameters<sup>104</sup>. All of these modelling studies were conducted outside the UK, and each reported cost savings with EIBI, but we agree with the NICE appraisal that the findings may not be reliable<sup>105,106,107,108</sup>. A recent Canadian modelling suggested that there could be high lifetime savings if waiting times for intensive behavioural intervention (IBI) could be shortened<sup>109</sup>.

The other economic evaluation in this early intervention area is an Australian modelling study of good practice Autism Specific Early Learning and Care (ASELC), such as the AEIOU programme (averaging 25 hours per week over 2 years)<sup>110</sup>. Cost was estimated to be AUS\$100,000 per child; lifetime savings per autistic individual depended on the level of learning disability: AUS\$0.75 million (without learning disability), AUS\$1.2 million (mild to moderate learning disability), and AUS\$1.3 million (severe learning disability).

<sup>100</sup> Green et al. (2010) op.cit.

<sup>101</sup> Marsh, K., Bertranou, E., Suominen, H., Venkatachalam, M. (2010) An economic evaluation of speech and language therapy. London: Matrix Evidence.

<sup>102</sup> NICE (2013) op.cit. p.287.

<sup>103</sup> Penner, M., Rayar, M., Bashir, N. et al. (2015). Cost-effectiveness analysis comparing pre-diagnosis autism spectrum disorder (ASD)-targeted intervention with Ontario's Autism Intervention Program. *Journal of Autism and Developmental Disorders*, 45, 2833-2847.

<sup>104</sup> NICE (2013) op.cit. p.537-540.

<sup>105</sup> Peters-Scheffer, N., Didden, R., Korzilius, H., Matson, J. (2012). Cost comparison of early intensive behavioral intervention and treatment as usual for children with autism spectrum disorder in the Netherlands. *Research in Developmental Disabilities*, 33, 1763-1772.

<sup>106</sup> Chasson, G.S., Harris, G.E., Neely, W.J. (2007). Cost comparison of early intensive behavioral intervention and special education for children with autism. *Journal of Child and Family Studies*, 16, 401-413.

<sup>107</sup> Jacobson, J.W., Mulick, J.A., Green, G. (1998). Cost-benefit estimates for early intensive behavioral intervention for young children with autism—general model and single state case. *Behavioral Interventions* 13, 201-226.

<sup>108</sup> Motiwala, S.S., Gupta, S., Lilly, M.B. et al. (2006). The cost-effectiveness of expanding intensive behavioural intervention to all autistic children in Ontario. *Healthcare Policy*, 1, 135-151.

<sup>109</sup> Piccininni, C., Bisnaire, L., Penner, M. (2016). Cost-effectiveness of wait time reduction for intensive behavioral intervention services in Ontario, Canada. *JAMA Pediatrics*, 14. doi:10.1001/jamapediatrics.2016.2695

<sup>110</sup> Synergies Economic Consulting (2013). Cost-Benefit Analysis of Providing Early Intervention to Children with Autism, Estimation of the net economic benefit of early intervention for a cohort of children with autism. Synergies Economic Consulting Pty Ltd.

## Conclusions

Many different early intervention approaches have been evaluated, and quite a few of them have been examined from an economics point of view, albeit through the use of modelling methods. Compared to other types of interventions (as covered in other case studies), this area is quite well populated with effectiveness evidence. There are, however, still very few studies conducted in the UK. The evidence from a number of these studies is positive in terms of changes in some aspects of a child's life, at least in the short term. The recently published follow-up findings from the PACT study – which is a low-intensity approach – suggest that some longer-term gains might also be achieved.

One immediate observation from our review is the remarkable range of *intensities* across the different programmes, varying widely in terms of the number of professionals per child (or family), the number and duration of individual sessions, and the period over which a complete programme

is delivered. Some early intervention programmes require many hours of therapist involvement per week, over a period of years. The PACT study, incidentally, would be considered low-intensity within this set of early intervention approaches.

There have been a number of economic modelling studies of intensive behavioural interventions, but most appear to suffer from methodological problems that considerably limit their reliability, and all have been conducted outside the UK, which probably limits their relevance. We cannot be sure that the generally high costs associated with these more intensive programmes – that can have repercussions for all concerned in terms of the opportunity costs of the time commitments needed (by professionals and families) - will be outweighed by savings or other economic benefits over the longer term. Parents may worry about the 'high costs' of EIBI<sup>111</sup>. Some autistic people and families have also expressed reservations that some behavioural approaches are not ethical.

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<sup>111</sup> Tzanakaki, P., Grindle, C., Hastings, R.P. et al. (2012). How and why do parents choose Early Intensive Behavioral Intervention for their young child with Autism? *Education and Training in Autism and Developmental*

# SOCIAL SKILLS INTERVENTIONS<sup>112</sup>

## Target population

Autistic children and adults with or without learning disability.

## Context

There are wide variations in individual characteristics and circumstances, but many autistic people experience difficulties in social participation and social isolation: one study suggested that 22% of young autistic people and 24% of autistic adults in England have no friends<sup>113</sup>. Social isolation in young autistic adults is associated with lower communication and functional skills<sup>114</sup>. It also increases the risk of depression and anxiety, and damages life satisfaction and self-esteem<sup>115</sup>.

## Intervention

Social skills interventions aim to improve social interaction and social communication of autistic people. They are based on social learning principles, usually including a combination of teaching and practising techniques such as instructions, discussion and role play<sup>116</sup>. Content may vary, from recognition of emotions and non-verbal communication, to social rules of communication and interaction. These are usually group interventions facilitated by a

person who may or may not be autistic. More recently, computer-based interventions have been developed.

## Effectiveness

There is mixed evidence on the effectiveness of social skills interventions for autistic children and young adults. A Cochrane review of the effects of social skills groups for children and young people identified five randomised trials of sufficient quality to include, finding some evidence that social skills groups can improve social competence for some autistic children and adolescents, although the authors were cautious about the generalisability of findings<sup>117</sup>.

A review of social skills interventions for autistic adolescents and adults with severe/profound learning disability found a number that demonstrated some degree of effectiveness: video-modelling for promoting social behaviour, intensive interaction on a variety of social behaviours, and peer-mediated intervention to support a number of social and communication behaviours. Structured teaching – and the TEACCH programme in particular (see later) – may have positive effects on a number of positive social behaviours<sup>118</sup>.

The programmes that have been evaluated most frequently are described below.

<sup>112</sup> Case study prepared by Margaret Perkins, Martin Knapp and Valentina Lemmi.

<sup>113</sup> Bancroft, K., Batten, A., Lambert, S. et al. (2012). *The Way We Are: Autism in 2012*. London: National Autistic Society.

<sup>114</sup> Orsmond, G. I., Shattuck, P. T., Cooper, B. P. et al. (2013). Social participation among young adults with an autism spectrum disorder. *Journal of Autism and Developmental Disorders*, 43, 2710-2719.

<sup>115</sup> Mazurek, M. O. (2013). Loneliness, friendship, and well-being in adults with autism spectrum disorders. *Autism*, 18, 223-232.

<sup>116</sup> National Collaborating Centre for Mental Health (2012). *Autism: recognition, referral, diagnosis and management of adults on the autism spectrum*. London: British Psychological Society.

<sup>117</sup> Reichow, B., Steiner, A. M., Volkmar, F. (2012). Social skills groups for people aged 6 to 21 years with autism spectrum disorders. *Cochrane Database of Systematic Reviews*. 11;(7):CD008511.

<sup>118</sup> Walton, K. M., & Ingersoll, B. R. (2013). Improving social skills in adolescents and adults with autism and severe to profound intellectual disability: A review of the literature. *Journal of Autism and Developmental Disorders*, 43, 594-615.

### Learning Experiences and Alternative Program for Preschoolers and their Parents (LEAP)

The LEAP programme offers autistic children systematic, individually designed education plans within integrated settings. A cluster randomised controlled trial (RCT) in the US compared preschools that received 2 years training and coaching in the LEAP approach with preschools that received the intervention manuals only. The study looked at the effects on autistic 4-year olds, and found that the group in the more intensively supported preschools showed significantly greater improvement in a number of areas: cognition, language, social behaviour, behaviour described as ‘problematic’ and autism symptoms<sup>119</sup>. The more closely that teachers followed the recommended programme, the better were the outcomes.

### Treatment and Education of Autistic and related Communication Handicapped Children (TEACCH)

TEACCH, also developed in the US, refers to a range of programmes using a psychoeducational model for individualised social skills training for children, young people and their parents. This ‘structured teaching’ approach comprises four main elements: physical structure, visual schedules, work systems and task organisation. The approach is incorporated in the SPELL approach employed in National Autistic Society services in the UK.

Evidence on its effectiveness is mixed. An early

US study of young autistic adults with learning disabilities focused on farming and landscaping skills, and found increases in structure and ‘individualised programming’ in communication, independence, socialisation, developmental planning, and positive behaviour management compared to participants in a control group. There was no difference in acquisition of skills, but family satisfaction was greater<sup>120</sup>. A second small-sample US study looked at a home-based teaching programme (HTP) for 2-3-year old autistic children and their parents. The intervention group received specialist tuition on how to develop the child’s cognitive, motor and language skills, and how to prompt engagement in activities. Parents learned about autism and did homework to support the training. The small sample size might explain why no significant differences were found in child or parent behaviour<sup>121</sup>.

Barnardo’s Forward Steps Early Intervention Programme (EIP) for preschool children is mainly based on TEACCH principles. The programme comprises one-to-one individual sessions over 10 weeks, including a 2-3 hour session and a home visit of up to 2 hours each week. Trained TEACCH facilitators support each child and encourage them to learn and develop appropriate skills across a range of developmental domains, while parents learned about their child’s autism and how to implement key TEACCH methods. A non-controlled before-after study involving 31 parents of 18 autistic preschool children in Northern Ireland found statistically significant decrease in parental

<sup>119</sup> Strain, P.S., Bovey, E.H. (2011). Randomized, controlled trial of the LEAP model of early intervention for young children with autism spectrum disorders. *Topics in Early Childhood Special Education* 31, 133-154.

<sup>120</sup> Van Bourgondien, M. E., Reichle, N. C., & Schopler, E. (2003). Effects of a model treatment approach on adults with autism. *Journal of Autism and Developmental Disorders*, 33, 131-140.

<sup>121</sup> Welterlin, A., Turner-Brown, L. M., Harris, S. et al. (2012). The home TEACCHing program for toddlers with autism. *Journal of Autism and Developmental Disorders*, 42, 1827-1835.

stress, a high level of parental satisfaction with the programme, and improvements in children's expressive and receptive language skills<sup>122</sup>.

A meta-analysis of 13 studies concluded 'that TEACCH effects on perceptual, motor, verbal and cognitive skills were of small magnitude', effects over adaptive behaviour (communication, activities of daily living, and motor functioning) were negligible or small range, but there were 'moderate to large gains' in social and maladaptive behaviour. The duration, intensity or setting for TEACCH did not make a difference<sup>123</sup>.

### **UCLA Program for the Education and Enrichment of Relational Skills (PEERS)**

The PEERS intervention is a manualised, parent-assisted, social skills programme for autistic adolescents and young adults. It focuses on making and keeping friends and managing peer rejection and conflict. PEERS is based on the principles of an adapted Children's Friendship Training (CFT) developed for a mixed group including high-functioning autistic children. Programmes are delivered weekly, over 12-14 weeks, to adolescents in small groups, with the approach appropriate to the age group. Parents meet regularly and are a key part of the intervention.

An early study of 13-17 year olds found a significant improvement in knowledge of and actual social skills (as reported by parents) compared to a control group<sup>124</sup>. A second study by the same researchers focused on a school-based CFT programme for 6-12 year olds. Improvements were seen in parent measures of their child's social skills and behaviour in play groups, and child measures of popularity and loneliness<sup>125</sup>. A third study evaluated a programme for 12-18 year olds, reporting longer-term outcomes. Between 1 and 5 years after the intervention, the PEERS group maintained gains in terms of social functioning, frequency of peer interactions and social skills knowledge<sup>126</sup>. Another study with this age group (11-16 year olds and their parents) reported beneficial effects in relation to family chaos and parenting self-efficacy<sup>127</sup>.

Two studies have looked at young high-functioning adults (18-24 years). PEERS-based social skills training led to significantly less loneliness and improved social skills knowledge, and caregiver-reported improvements in young adults' overall social skills, social responsiveness, empathy and socialisation<sup>128</sup>. Social skills training sessions focused on making and keeping friends, improved overall social skills, frequency of social engagement and social skills knowledge; it also significantly reduced autism symptoms related

<sup>122</sup> Braiden, H. J., McDaniel, B., McCrudden, E. et al. (2012). A practice-based evaluation of Barnardo's Forward Steps Early Intervention Programme for children diagnosed with autism. *Child Care in Practice*, 18, 227-242.

<sup>123</sup> Virues-Ortega, J., Julio, F. M., Pastor-Barruso, R. (2013). The TEACCH program for children and adults with autism: A meta-analysis of intervention studies. *Clinical Psychology Review*, 33, 940-953.

<sup>124</sup> Laugeson, E. A., Frankel, F., Mogil, C. et al. (2009). Parent-assisted social skills training to improve friendships in teens with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, 39, 596-606.

<sup>125</sup> Frankel, F., Myatt, R., Sugar, C. et al. (2010). A randomized controlled study of parent-assisted Children's Friendship Training with children having autism spectrum disorders. *Journal of Autism and Developmental Disorders*, 40, 827-842.

<sup>126</sup> Mandelberg, J., Laugeson, E. A., Cunningham, T. D. et al. (2014). Long-term treatment outcomes for parent-assisted social skills training for adolescents with autism spectrum disorders: the UCLA PEERS program. *Journal of Mental Health Research in Intellectual Disabilities*, 7, 45-73.

<sup>127</sup> Karst, J. S., Van Hecke, A. V., Carson, A. M. et al. (2015). Parent and family outcomes of PEERS: a social skills intervention for adolescents with autism spectrum disorder. *Journal of Autism and Developmental Disorders*, 45, 752-765.

<sup>128</sup> Gantman, A., Kapp, S. K., Orenski, K. et al. (2012). Social skills training for young adults with high-functioning autism spectrum disorders: a randomized controlled pilot study. *Journal of Autism and Developmental Disorders*, 42, 1094-1103.

to social responsiveness<sup>129</sup>. Caregivers in these studies included parents, other family members, job coaches, life coaches, peer mentors and behavioural coaches.

A group-based, highly structured, manualised psychosocial intervention was offered to high-functioning 7-12 year olds in a 5-week summer programme (*Summer Max*). A recent RCT compared two versions: high-intensity (child-staff ratio of 2:1) and low-intensity (ratio of 4:1). There were no differences between the high- and low-intensity groups, but overall there were significant improvements in non-literal language and emotion recognition, and parent ratings of social skills, autism-related symptoms, withdrawal and behavioural symptoms<sup>130</sup>.

## Economic evidence

There is no cost-effectiveness or other economic evaluative evidence on any of these social skills interventions.

## Conclusions

There is evidence that there are some interventions that can help autistic adolescents and young adults to develop friendships and generally build social skills, with some benefits too for parents or other caregivers. Involving parents in such programmes is important for their success. However, most studies in this area are relatively small, and only one has been conducted in the UK. Whilst the findings show promise, there is also clearly a need for further study, particularly in UK contexts.

Although a few studies report the narrow cost of an intervention, there appears to be no economic evaluative evidence in this area.

NICE Clinical Guidelines recommended, in respect of psychosocial interventions, that it may be helpful to consider play-based strategies with parents, carers and teachers to increase joint attention, engagement and reciprocal communication<sup>131</sup>. For older children, peer mediation may be useful. But NICE also pointed out the limited availability of such programmes, and the additional costs of professionals needed to support such interventions.

<sup>129</sup> Laugeson, E.A., Gantman, A., Kapp, S. K. et al. (2015). A randomized controlled trial to improve social skills in young adults with autism spectrum disorder: the UCLA PEERS® program. *Journal of Autism and Developmental Disorders*, 45, 3978-3989.

<sup>130</sup> Lopata, C., Thomeer, M. L., Lipinski, A. M. et al. (2015). RCT examining the effect of treatment intensity for a psychosocial treatment for high-functioning children with ASD. *Research in Autism Spectrum Disorders*, 17, 52-63.

<sup>131</sup> NICE (2013). *Autism: The Management and Support of Children and Young People on the Autism Spectrum Clinical Guideline 170*. London: National Institute for Health and Care Excellence.

# PARENT TRAINING AND SUPPORT PROGRAMMES<sup>132</sup>

## Target population

Parents of autistic children with or without learning disabilities.

## Context

Parents may encounter many challenges in bringing up autistic children and therefore seek support<sup>133</sup>.

They might experience stress or mental health problems, affecting their ability to function and care for their children<sup>134</sup>, particularly if they have more than one child with special needs<sup>135</sup>. Studies have demonstrated that parents of autistic children have lower quality of life compared to parents of neurotypical children<sup>136,137</sup>. Predictors of poor parental quality of life include being the mother, being unemployed, lacking social support, and children's behavioural difficulties. While parents may adapt to their circumstances over time - with improvements

in psychological wellbeing, social relationships and support - adaptation is more difficult when autistic children exhibit aggressive and/or violent behaviours<sup>138</sup>. There is evidence of earlier mortality of parents of autistic children or children with learning disabilities compared to the general population<sup>139</sup>. No evidence is currently available on the impact of being a mother and also autistic.

Parents may not be able to take paid employment, or may find that their work is disrupted by their care responsibilities, and these effects are greater than for parents of neurotypical children<sup>140</sup>. This can lead to quite high costs for both the family and economy<sup>141</sup>. Additional financial burden may arise because the family feels a need to pay for treatments<sup>142</sup>. The cost borne by families is higher when an autistic child has learning disabilities, and will be higher still if a child has anxiety, epilepsy or irritability<sup>143</sup>.

<sup>132</sup> Case study prepared by Valentina Lemmi, Margaret Perkins and Martin Knapp.

<sup>133</sup> Altieri, M.J., von Kluge, S. (2009). Searching for acceptance: challenges encountered while raising a child with autism. *Journal of Intellectual and Developmental Disability*, 34, 142-152.

<sup>134</sup> Bonis, S. (2016). Stress and parents of children with autism: a review of literature. *Issues in Mental Health Nursing*, 37, 153-163.

<sup>135</sup> Kuhlthau, K., Payakachat, N., Delahaye, J. et al. (2014). Quality of life for parents of children with autism spectrum disorders. *Research in Autism Spectrum Disorders*, 8, 1339-1350.

<sup>136</sup> Vasilopoulou, E., Nisbet, J. (2016). The quality of life of parents of children with autism spectrum disorder: A systematic review. *Research in Autism Spectrum Disorders*, 23, 36-49.

<sup>137</sup> Hsiao, Y.J. (2016). Pathways to mental health-related quality of life for parents of children with autism spectrum disorder: roles of parental stress, children's performance, medical support, and neighbor support. *Research in Autism Spectrum Disorders*, 23, 122-130.

<sup>138</sup> Gray, D.E. (2002). Ten years on: A longitudinal study of families of children with autism. *Journal of Intellectual and Developmental Disability*, 27, 215-22.

<sup>139</sup> Fairthorne, J., Hammond, G., Bourke, J. et al. (2014). Early mortality and primary causes of death in mothers of children with intellectual disability or autism spectrum disorder: a retrospective cohort study. *PloS One*, 9(12), e113430.

<sup>140</sup> Cidav, Z., Marcus, S.C., Mandell, D.S. (2012). Implications of childhood autism for parental employment and earnings. *Pediatrics*, 129, 617-623.

<sup>141</sup> Buescher, A.V., Cidav, Z., Knapp, M., Mandell, D.S. (2014). Costs of autism spectrum disorders in the United Kingdom and the United States. *JAMA Pediatrics*, 168(8), 721-728.

<sup>142</sup> Bonis (2016) op.cit.

<sup>143</sup> Ouyang, L., Grosse, S.D., Riley, C. et al. (2014). A comparison of family financial and employment impacts of fragile X syndrome, autism spectrum disorders, and intellectual disability. *Research in Developmental Disabilities*, 35, 1518-1527.

## Intervention

Parent training and support programmes aim to reduce parental stress and improve the quality of life of both parents and autistic people. They focus on providing parents with the knowledge and skills needed for understanding and supporting autistic people, in particular with social, communication, and behavioural issues, and with co-occurring physical and mental health problems.

## Effectiveness

*Cygnets*<sup>144</sup> is a group intervention for parents of autistic children (5–18 years), structured into six weekly sessions of up to 3 hours and a follow-up session after 6 weeks. It is based on the Family Partnership Model. Sessions include formal teaching, group exercises, discussions and 'homework'. *Cygnets* is administered by a lead trainer and co-trainers to groups of up to 12 parents. A controlled before-after UK study, comparing *Cygnets* to a waiting-list control, showed significant improvements in parent-reported behaviour problems and parenting satisfaction after the intervention and also at 3-month follow-up<sup>145</sup>.

*ASCEND (Autism Spectrum Conditions - Enhancing Nurture and Development)* is a group intervention for parents of autistic children (preschool–19 years), structured into 11 weekly sessions of 2.5 hours.

Sessions include formal teaching, group exercises and discussions. *ASCEND* is administered by a lead trainer and co-trainers to groups of up to 20 parents. A controlled before-after UK study compared *ASCEND* with usual support (waiting-list). It found improvements in parent-reported behaviour problems and parenting competences after the intervention and at 3-month follow-up, although only the former was statistically significant<sup>146</sup>. Parents of autistic children without learning disability showed greater improvements than parents of autistic children who also had learning disability.

*NAS (National Autistic Society) EarlyBird and NAS EarlyBird Plus*<sup>147</sup> are group interventions for parents of children (aged under 5 and 4–8 years, respectively) newly diagnosed as autistic. The intervention comprises 8 sessions, each of 3.5 hours, and home visits over 3 months. Sessions include formal teaching and discussions. *NAS EarlyBird* is administered to groups of up to 12 parents. A controlled before-after UK study showed improvements in parent stress and communication, in parent-reported autistic traits and child development after receiving *NAS EarlyBird* and at 6-month follow-up<sup>148</sup>. Results were similar when the intervention was delivered at the *NAS EarlyBird* centre and at centres run by licensed users. A small New Zealand study also reported positive outcomes<sup>149</sup>.

<sup>144</sup> [www.barnardos.org.uk/cygnets/yk\\_cygnets-parents\\_carers\\_support\\_programme.htm](http://www.barnardos.org.uk/cygnets/yk_cygnets-parents_carers_support_programme.htm).

<sup>145</sup> Stuttard, L., Beresford, B., Clarke, S. et al. (2016). An evaluation of the *Cygnets* parenting support programme for parents of children with autism spectrum conditions. *Research in Autism Spectrum Disorders*, 23, 166-178.

<sup>146</sup> Beresford, B., Stuttard, L., Clarke, S. et al. (2012). *Managing behaviour and sleep problems in disabled children: An investigation into the effectiveness and costs of parent-training interventions*. London: Department for Education.

<sup>147</sup> [www.autism.org.uk/earlybird](http://www.autism.org.uk/earlybird).

<sup>148</sup> Shields, J., Simpson, A. (2004). The *NAS EarlyBird* Programme: preschool support for parents of children with autistic spectrum disorder. *Good Autism Practice*, 5, 49-60.

<sup>149</sup> Anderson, A., Birkin, C., Seymour, F. et al. (2006). *EarlyBird Evaluation*. Wellington: The Ministry of Education.

In Northern Ireland, *KEYHOLE*® offers 10 hours of home-based support and a resource kit for parents. A small non-controlled study reported some improvements in problems relating to play, relationships with others and difficulties with imitation. Mothers reported being less stressed<sup>150</sup>. Adapted from the Incredible Years® programme for children at risk of conduct disorders,<sup>151</sup> *Incredible Years® Autism Spectrum and Language Delays Programme* is a group intervention for parents of young children (2–5 years) diagnosed as autistic and/or with language delay. There are 12 weekly sessions of 2 hours each, including formal teaching, group exercises with role play and home activities. The programme is administered by two clinical psychologists. A non-controlled pilot study in the UK found an increase in pro-social behaviours and a decrease in behaviour problems in children<sup>152</sup>. Similarly, *Incredible Years® Preschool Basic Parent Program* is a group intervention for parents of preschool autistic children, organised into 15 weekly sessions of 2 hours, with an informal dinner 30 minutes before each session. Sessions include formal teaching, group exercises with role play and home activities. In addition, after four sessions, a 'buddy scheme' is introduced to support participants between sessions. The intervention is administered by two facilitators to groups of up to nine parents. A non-controlled before-after study in the US suggested reduction in parental stress, while highlighting some

implementation challenges and opportunities for improvement<sup>153,154</sup>.

*Riding the Rapids* is a group intervention for parents of children (3–11 years) with disabilities (autism, learning disabilities or complex disabilities). There are ten weekly sessions of 2 hours and a follow-up session after 3 months. The programme is based on the STAR approach (Settings Triggers Actions Results). Sessions include problem-solving exercises, positive reinforcement, communication tools and cognitive-behavioural strategies for stress. It is administered by a clinical psychologist and two facilitators to groups of up to 12 parents. A controlled before-after UK study compared *Riding the Rapids* for parents of children with disabilities (autism included) with usual support; it found significant improvements in parent-reported behaviour problems and efficacy<sup>155</sup>.

*Confident Parenting* is a group intervention for parents of children (3–11 years) with disabilities (autism, learning disabilities or complex disabilities). Six weekly sessions of 2 hours – for groups of up to 12 parents - include formal teaching, modelling and discussions. A controlled before-after UK study reported improvements in parent-set goals for their child's behaviours and parent-reported behaviour problems, both immediately after the intervention and 3 months later, though only the former was statistically significant<sup>156</sup>.

<sup>150</sup> McConkey, R., MacLeod, S., Cassidy, A. (2011). The Keyhole® Rainbow Resource Kit: meeting the needs of parents of newly diagnosed preschoolers with ASD. *Early Child Development and Care*, 18, 321-334.

<sup>151</sup> [incredibleyears.com/](http://incredibleyears.com/)

<sup>152</sup> Hutchings, J., Pearson-Blunt, R., Pasteur, M.A. et al. (2016). A pilot trial of the Incredible Years® Autism Spectrum and Language Delays Programme. *Good Autism Practice*, 17, 15-22.

<sup>153</sup> Dababnah, S., Parish, S. L. (2016). Incredible Years Program tailored to parents of preschoolers With autism: pilot results. *Research on Social Work Practice*, 26, 372-385.

<sup>154</sup> Dababnah, S., & Parish, S. L. (2016). Feasibility of an empirically based program for parents of preschoolers with autism spectrum disorder. *Autism*, 20, 85-95.

<sup>155</sup> Stuttard, L., Beresford, B., Clarke, S. et al. (2014). *Riding the Rapids: Living with autism or disability - an evaluation of a parenting support intervention for parents of disabled children*. *Research in Developmental Disabilities*, 35, 2371-2383.

<sup>156</sup> Beresford et al. (2012) op.cit.

Adapted from Triple P Positive Parenting Program<sup>157</sup>, *Stepping Stones Triple P (SSTP)* is a behavioural family intervention for families of children with disabilities (including those who are autistic). It aims to provide parents with positive skills to manage their child's behaviours and to improve parental wellbeing. The programme comprises ten sessions of 1 hour, administered one-to-one by a practitioner at home, and includes formal teaching, observation, practice and feedback. A non-controlled before-after Canadian study reported improvements in parental self-efficacy and wellbeing<sup>158</sup>. A randomised controlled trial in Australia found that a different version of SSTP for families of autistic children (compared to families on the waiting-list) achieved improvements in parent-reported child behaviours, parenting skills and parenting satisfaction after the intervention and at six-month follow-up<sup>159</sup>. This different version of SSTP included group sessions when focusing on teaching and individual sessions when focusing on observation, practice, and feedback.

*More Than Words*® is a group intervention for parents of children at risk of, or diagnosed as autistic (up to age 5)<sup>160</sup>. The intervention includes eight group sessions with parents, and three home-based individual sessions with both parent

and child, delivered over 3 months. The intervention is part of the Hanen Program® administered by speech and language therapists. Sessions aim to teach parents strategies to increase their child's communication. A controlled before-after UK study compared *More Than Words*® for parents of children aged 2-4 years with waiting-list control; it found significant improvements in children's vocabulary, but not in social communication, parental stress or adaptation at 7-month follow-up<sup>161</sup>. A US trial comparing *More Than Words*® for parents of children 15-25 months with treatment as usual found no improvement in parental responsiveness at 5-month or 9-month follow-ups, but improvement in communication in children with lower levels of object interest<sup>162</sup>.

Some parent training and support programmes focus on specific behaviours of autistic children, such as sleeping<sup>163</sup> and eating<sup>164</sup>. Recently, the *NAS EarlyBird Healthy Minds programme*<sup>165</sup> has been developed, adapted from the *NAS EarlyBird programme* to help parents promote the mental health of their children, while in Northern Ireland, *KEYHOLE*® is available.

<sup>157</sup> [triplep.net/glo-en/home/](http://triplep.net/glo-en/home/).

<sup>158</sup> Hodgetts, S., Savage, A., McConnell, D. (2013). Experience and outcomes of stepping stones triple P for families of children with autism. *Research in Developmental Disabilities*, 34, 2572-2585.

<sup>159</sup> Whittingham, K., Sofronoff, K., Sheffield, J. et al. (2009). Stepping Stones Triple P: an RCT of a parenting program with parents of a child diagnosed with an autism spectrum disorder. *Journal of Abnormal Child Psychology*, 37, 469-480.

<sup>160</sup> Sussman, F. (1999). *More Than Words: helping parents promote communication and social skills in children with autism spectrum disorder*. Toronto: Hanen Centre.

<sup>161</sup> McConachie, H., Randle, V., Hammal, D. et al. (2005). A controlled trial of a training course for parents of children with suspected autism spectrum disorder. *Journal of Pediatrics*, 147, 335-340.

<sup>162</sup> Carter, A. S., Messinger, D. S., Stone, W. L. et al. (2011). A randomized controlled trial of Hanen's 'More Than Words' in toddlers with early autism symptoms. *Journal of Child Psychology and Psychiatry*, 52, 741-752.

<sup>163</sup> Stuttard, L., Beresford, B., Clarke, S. et al. (2015). A preliminary investigation into the effectiveness of a group-delivered sleep management intervention for parents of children with intellectual disabilities. *Journal of Intellectual Disabilities*, 19, 342-55.

<sup>164</sup> Sharp, W. G., Burrell, T. L., Jaquess, D. L. (2013). The Autism MEAL Plan: A parent-training curriculum to manage eating aversions and low intake among children with autism. *Autism*, 18, 712-722.

<sup>165</sup> [autism.org.uk/earlybird](http://autism.org.uk/earlybird).

## Economic case

There have been economic evaluations of some of the above parent training and support interventions.

A UK study of Cygnet estimated the mean cost per person for the intervention itself was £351, ranging from £141 to £663 (2009/10 price levels), depending on how many parents there were in each group, patterns of attendance and salary levels for the group facilitators<sup>166,167</sup>. Looking at all services used by children and families, no cost differences were found between the Cygnet or usual support groups, although the small sample size limited the analysis. In the usual support group, costs of services used by children increased over a 3-month period, while in the Cygnet group community health and social care costs increased but hospital and primary care costs decreased.

The mean cost of the *ASCEND* intervention was found to be £615 per person, ranging widely between £201 and £2,543 (2009/10 prices) because of differences in the number of participants and staffing<sup>168</sup>. Total costs of all services used by children and parents were higher in the intervention group, but only significantly higher for hospital services and for community health services after 3 months. Costs were at 2009-2010 price levels.

As regards *Riding the Rapids*, the same UK study calculated the mean cost per person for the programme to be £407, ranging between £80 and £685 (2009/10 prices)<sup>169</sup>. The most commonly used other service was primary care. Mean cost per person for *Confident Parenting* was £391 (range £56 to and £940, 2009/10 prices)<sup>170</sup>. Service costs were not estimated in either the *Riding the Rapids* or *Confident Parenting* studies.

A decision-tree economic model<sup>171</sup> built by NICE allowed examination of the cost-effectiveness of group parent training to help manage behaviour in children and young people with intellectual disabilities (including autism) compared to waiting-list (usual support) over a 9-week intervention period and 52-week follow-up. The cost of achieving one additional quality-adjusted life year (QALY) from group parent training was £27,148 (health and social care perspective), which is between the lower and upper cost-effectiveness thresholds used by NICE. The probability that group parent training would be seen as cost-effective was relatively low: only 29% at the lower threshold, and 52% at the upper threshold.

<sup>166</sup> Stuttard et al. (2016) op.cit.

<sup>167</sup> Beresford et al. (2012) op.cit.

<sup>168</sup> *ibid.*, <sup>169</sup> *ibid.*, <sup>170</sup> *ibid.*

<sup>171</sup> National Institute for Health and Care Excellence (2015). *Challenging behaviour and learning disabilities: prevention and interventions for people with learning disabilities whose behaviour challenges*. NICE Guideline NG11. London: NICE.

## Conclusions

There have been a number of UK studies of parent training and support interventions, although generally they are small-scale, with non-randomised designs. Some have included an economic evaluation component. Despite the limitations, these studies offer encouraging evidence that parent training interventions can reduce parental stress, improve parental wellbeing and competences, and reduce parent-reported behaviour problems shown by their autistic children.

Some of the interventions are not solely focused on parents with autistic children, but may include families where there is a child with learning disabilities or complex disabilities.

The programmes are not expensive, because most are delivered in groups. The cost per family (funded from public budgets in the studies we found) is sensitive to the size of group and the salary levels for the group leaders.

Where there have been economic evaluations, they have focused on health and social care service costs, but not looked at the unpaid care and support that is obviously provided by parents, nor at any employment consequences (and hence potential impact on household income and national productivity).

There is obviously a need for further evaluation (including economic evaluation) in this area, ideally with larger samples, longer follow-ups, and – for the economic component - measuring a wider range of potential economic impacts.

# COGNITIVE BEHAVIOURAL THERAPY FOR ANXIETY<sup>172</sup>

## Target population

Autistic children and adults without learning disability.

## Context

Anxiety affects 42% of autistic children<sup>173</sup>. Two-thirds of autistic adults say they have experienced anxiety because of lack of support and a third have experienced serious mental health problems for this reason<sup>174</sup>. Having mental health problems alongside autism can contribute to premature mortality<sup>175</sup>. Compared to the general population, autistic children are more likely to have anxiety related to specific phobias but they appear to have similar levels of severity, numbers, and types of anxiety disorders<sup>176</sup>. This same Dutch study found that quality of life worsens with increased levels of autism-like behaviour and higher anxiety severity.

There are many causes. Anxiety is more likely in those individuals who are more sensory-sensitive<sup>177</sup>. Anxiety is associated with peer-victimisation<sup>178</sup>, which in turn is strongly associated with social vulnerability<sup>179</sup>. We discuss victimisation and bullying in more detail in another case study<sup>180</sup>.

Children with anxiety problems (whether autistic or not) can generate quite high public sector costs, ranging across the education, health and social care sectors<sup>181</sup>. There is no UK evidence on the incremental cost of anxiety among autistic people, but again, a Dutch study offers some insights, finding no differences in anxiety-related costs when comparing children with and without a diagnosis of autism<sup>182</sup>. There may often be impacts on parents and other carers. General evidence on carers suggests that providing 10 or more hours of unpaid care per week has significant effects on carer mental health<sup>183</sup>, and on employment<sup>184</sup>.

<sup>172</sup> Case study prepared by Valentina Lemmi, Marija Trachtenberg, Margaret Perkins and Martin Knapp.

<sup>173</sup> Simonoff, E., Pickles, A., Charman, T. et al. (2008). Psychiatric disorders in children with autism spectrum disorders: prevalence, comorbidity, and associated factors in a population-derived sample. *Journal of the American Academy of Child & Adolescent Psychiatry*, 47, 921-929.

<sup>174</sup> Rosenblatt, M. (2008). *I Exist: The message from adults with autism in England*. London: National Autistic Society.

<sup>175</sup> Hirvikoski, T., Mittendorfer-Rutz, E., Boman, M. et al. (2016). Premature mortality in autism spectrum disorder. *British Journal of Psychiatry*, 208, 232-238.

<sup>176</sup> van Steensel F., Bögels S.M., Dirksen C.D. (2012). Anxiety and quality of life: clinically anxious children with and without autism spectrum disorders compared. *Journal of Clinical Child & Adolescent Psychology*, 41, 731-738.

<sup>177</sup> Uljarević, M., Lane, A., Kelly, A., Leekam, S. (2016). Sensory subtypes and anxiety in older children and adolescents with autism spectrum disorder. *Autism Research*, 9, 1073-1078.

<sup>178</sup> Storch, E.A., Larson, M.J., Ehrenreich-May, J. et al. (2012). Peer victimization in youth with autism spectrum disorders and co-occurring anxiety: relations with psychopathology and loneliness. *Journal of Developmental and Physical Disabilities*, 24, 575-590.

<sup>179</sup> Sofronoff, K., Dark, E., Stone, V. (2011). Social vulnerability and bullying in children with Asperger syndrome. *Autism*, 15, 355-372.

<sup>180</sup> See the case study on Anti-stigma campaigns and anti-bullying interventions.

<sup>181</sup> Snell, T., Knapp, M., Healey, A. et al. (2013). Economic impact of childhood psychiatric disorder on public sector services in Britain: estimates from national survey data. *Journal of Child Psychology and Psychiatry*, 54, 977-985.

<sup>182</sup> van Steensel F.J.A., Dirksen, C.D., Bögels, S.M. (2013). A cost of illness study of children with high-functioning autism spectrum disorders and comorbid anxiety disorders as compared to clinically anxious and typically developing children. *Journal of Autism and Developmental Disorders*, 43, 2878-2890.

<sup>183</sup> Smith, L., Onwumere, J., Craig, T. et al. (2014). Mental and physical illness in caregivers: results from an English national survey sample. *British Journal of Psychiatry* 205, 197-203.

<sup>184</sup> King, D., Pickard, L. (2013). When is a carer's employment at risk? Longitudinal analysis of unpaid care and employment in midlife in England. *Health and Social Care in the Community*, 21, 303-314.

Being a carer of someone with mental health problems is associated with increased burden in relation to caring time, financial problems, distress and embarrassment, and a higher risk of one's own mental health being affected<sup>185</sup>.

Parents of children with emotional disorders (and not just those who are autistic) have higher levels of psychological stress, which in turn leads them to seek additional services for their children, including health, education and welfare services<sup>186</sup>. Parents with autistic children (and not just those with anxiety) have greater levels of stress than parents of typically developing children or children with other disabilities<sup>187</sup>. The combination of autism and anxiety is therefore likely to be especially demanding.

Looking again at a general population, children with anxiety will have significantly lower earnings at age 30<sup>188</sup> and a significantly lower family income at age 50<sup>189</sup>. Anxiety in adulthood affects service use, employment status, absenteeism from work and days out of role<sup>190</sup>.

## Intervention

Cognitive behavioural therapies (CBT) are widely used to treat mental health problems (including anxiety). They are time-limited, structured

psychological therapies. They aim to '(a) identify the types and effects of thoughts, beliefs and interpretations on current symptoms; (b) develop skills to identify, monitor and then counteract problematic thoughts, beliefs and interpretations related to the target symptoms or problems; and (c) learn a repertoire of coping skills appropriate to the target thoughts, beliefs and/or problem areas'<sup>191</sup>. CBT is usually provided by CBT therapists and/or clinical or counselling psychologists face-to-face, either one-to-one (i.e. individual), in groups or online. NICE guideline 1.6.3 recommends 'adaptations to the method of delivery of cognitive and behavioural interventions for autistic adults and coexisting common mental disorders'. It is not known how often those adaptations are adhered to or if they increase cost-effectiveness.

## Effectiveness

There have been a number of studies of individual and group CBT for autistic children and adolescents with anxiety problems.

A three-arm randomised trial in Australia compared a brief CBT intervention (2-hour, 6-week group-based) delivered either as child-only therapy or as child-parent combined (where parents are trained as co-therapists) with waiting-list control.

<sup>185</sup> Viana MC, Gruber MJ, Shahly V et al. (2013). Family burden related to mental and physical disorders in the world: results from the WHO World Mental Health (WMH) surveys. *Revista Brasileira de Psiquiatria*, 35, 115-125.

<sup>186</sup> Meltzer, H., Ford, T., Goodman, R. et al. (2011). The burden of caring for children with emotional or conduct disorders. *International Journal of Family Medicine* doi: 10.1155/2011/801203.

<sup>187</sup> Hayes, S.A., Watson, S.L. (2013). The impact of parenting stress: A meta-analysis of studies comparing the experience of parenting stress in parents of children with and without autism spectrum disorder. *Journal of Autism and Developmental Disorders*, 43, 629-642.

<sup>188</sup> Knapp, M., King, D., Healey, A., Thomas, C. (2011). Economic outcomes in adulthood and their associations with antisocial conduct, attention deficit and anxiety problems in childhood. *Journal of Mental Health Policy and Economics*, 14, 137-147.

<sup>189</sup> Goodman, A., Joyce, R., Smith, J.P. (2011). The long shadow cast by childhood physical and mental problems on adult life. *Proceedings of the National Academy of Sciences*, 108, 6032-6037.

<sup>190</sup> Alonso, J., Petukhova, M., Vilagut, G. et al. (2011). Days out of role due to common physical and mental conditions: results from the WHO World Mental Health surveys. *Molecular Psychiatry*, 16, 1234-1246.

<sup>191</sup> National Collaborating Centre for Mental Health. (2012). *Autism: recognition, referral, diagnosis and management of adults on the autism spectrum*. London: British Psychological Society.

Study participants were aged 10-12 diagnosed with Asperger's and anxiety<sup>192</sup>. The aim of the study was to teach children how to manage their feelings and encourage a wider range of emotions and behaviour. Therapists were postgraduate psychology students. Children receiving either child-only or child + parent CBT had significantly greater reductions in anxiety and level of social worry, and were better able to identify different strategies for dealing with anxiety in different situations at the 6-week follow-up when compared to the waiting-list control. For each of these three outcomes, the child + parent intervention had better results than the child-only programme.

*Building Confidence* is a family-focused individual-CBT programme combining standard CBT with components to address children's social and adaptive skill deficits, which are hypothesized to be barriers to anxiety reduction<sup>193</sup>. A small trial of Building Confidence compared to a waiting-list control, conducted in the US, enrolled high-functioning autistic children with anxiety aged 7-11 years. Families received 16 weekly 90-minute sessions, with the child receiving 30 minutes and the parents/family 60 minutes. Educational psychologists (or doctoral students) delivered the intervention. Components focused on social skills, adaptive skill deficits, school-based problems and other areas. Social coaching was provided on-site immediately before attempting to join a social activity, whether at home or school. Another adaptation was to set up a peer-buddy and mentoring programme for the child at school

to enhance social acceptance and address social avoidance. The evaluation found that children receiving the intervention had significantly better treatment response, and were more likely to be diagnosis-free (anxiety) both immediately after treatment and at the 3-month follow-up.

Two other US trials have evaluated variants of the Building Confidence approach. One small study extended CBT to a total of 32 weekly sessions, half focused on anxiety symptoms and half on the child's relationships in school and community<sup>194</sup>. Compared to the control group who received treatment as usual, children in the CBT group had greater reductions in anxiety symptom severity and more were diagnosis-free at post-treatment. The other trial added a new component to Building Confidence with the aim of increasing the child's daily living skills. Children receiving the intervention had significantly greater reductions in anxiety and improvements in total and personal daily living skills. Parents could reduce their involvement in their child's private daily routines such as bathing and dressing<sup>195</sup>.

*Exploring Feelings* is a family-focused CBT programme provided to high-functioning autistic children aged 9-13 with anxiety, and their parents. There are weekly 2-hour parallel group sessions (one child-only, one parent-only) for seven weeks. Group leaders were trainee psychologists with experience in group work but not with prior CBT qualifications. Sessions focused on identifying feelings and building a toolbox of physical, social and

<sup>192</sup> Sofronoff, K., Attwoord, T., Hinton, S. (2005). A randomized controlled trial of a CBT intervention for anxiety in children with Asperger syndrome. *Journal of Child Psychology and Psychiatry*, 46, 1152–1160.

<sup>193</sup> Wood, J.J., Drahota, A., Sze, K. et al. (2009). Cognitive behavioral therapy for anxiety in children with autism spectrum disorders: a randomized, controlled trial. *Journal of Child Psychology and Psychiatry*, 50, 224–234.

<sup>194</sup> Fujii, C., Renno, P., McLeod, B.D. et al. (2013). Intensive cognitive behavioral therapy for anxiety disorders in school-aged children with autism: A preliminary comparison with treatment-as-usual. *School Mental Health*, 5, 25-37.

<sup>195</sup> Drahota, A., Wood, J.J., Sze, K.M., Van Dyke, M. (2011). Effects of cognitive behavioral therapy on daily living skills in children with high-functioning autism and concurrent anxiety disorders. *Journal of Autism and Developmental Disorders*, 41, 257-265.

thinking skills appropriate to each child. Homework was provided between sessions. A pilot randomised trial in the UK comparing the 'Exploring Feelings' programme to a waiting-list control group, found that children in the CBT group showed greater reductions in anxiety symptoms and severity 3 months after the start of the programme<sup>196</sup>.

*Discussing + Doing = Daring* is an individual, family-focused CBT programme of 15 sessions in which both parent and child participate. It is for high-functioning autistic children with anxiety aged 8-18. A quasi-randomised study in the Netherlands found no difference between the CBT and a control group (treatment as usual) in reducing anxiety disorder or in health-related quality of life. There were also no cost differences from a societal perspective (health, social care, education, and parents' private costs)<sup>197</sup>.

*Coping Cat* is individualised CBT provided to high-functioning autistic children with anxiety aged 8-14 years. It is a 16-week programme with sessions lasting 60-90 minutes. Eight sessions focus on skills training and eight on exposure tasks. Parents attend two of the sessions, where they receive psycho-education about their child's treatment goals. Homework tasks aim to reinforce skills. A small US trial compared 'Coping Cat' to a waiting-list control group: the CBT intervention led to greater reductions in anxiety symptoms both immediately after treatment ended and 2 months later<sup>198</sup>.

*Multimodal Anxiety and Social Skills Intervention (MASSI)* is aimed at high-functioning autistic adolescents with anxiety aged 12-17. The approach was developed as a manual-supported programme based on the principles of CBT and ABA. It seeks to address adolescents' developmental needs by targeting both anxiety and social disability. There are up to 13 individual sessions of 60-70 minutes and seven group sessions of 75 minutes (group size of 3), plus parent education coaching provided after each individual therapy session. It is delivered over 14 weeks by a clinical psychologist. It has been evaluated in a pilot trial in the US in comparison to a waiting-list control<sup>199</sup>. The MASSI CBT intervention was feasible, acceptable to participants, and led to improvements in parent-reported social responsiveness (measuring changes in communication, social behaviour, interests and unusual behaviour) and developmental global functioning, but did not affect adolescents' anxiety symptoms.

*The Behavioural Interventions for Anxiety in Children with Autism (BIACA)* is family-focused CBT, provided on an individual basis to high-functioning autistic children or adolescents with anxiety. It aims to address anxiety and develop social skills. A small trial in the US compared BIACA to a waiting-list control for adolescents aged 11-15<sup>200</sup>. The programme emphasised exposure, challenging irrational beliefs, and behavioural supports provided by caregivers, as well as numerous autism-specific treatment elements.

<sup>196</sup> McConachie, H., McLaughlin, E., Grahame, V. et al. (2013). Group therapy for anxiety in children with autism spectrum disorder. *Autism* 18, 723-32.

<sup>197</sup> Van Steensel, F.J.A., Dirksen, C.D., Bögels, S.M. (2014). Cost-effectiveness of cognitive-behavioral therapy versus treatment as usual for anxiety disorders in children with autism spectrum disorder. *Research in Autism Spectrum Disorders*, 8, 127-137.

<sup>198</sup> McNally Keehn, R.H., Lincoln, A.J., Brown, M.Z., Chavira, D.A. (2013). The Coping Cat Program for children with anxiety and autism spectrum disorder: a pilot randomized controlled trial. *Journal of Autism and Developmental Disorders*, 43, 57-67.

<sup>199</sup> White, S., Ollendick, T., Albano, A.M. et al. (2013). Randomized controlled trial: multimodal anxiety and social skill intervention for adolescents with autism spectrum disorder. *Journal of Autism and Developmental Disorders*, 43, 382-394.

<sup>200</sup> Wood, J.J., Ehrenreich-May, J., Alessandri, M. et al. (2015). Cognitive behavioral therapy for early adolescents with autism spectrum disorders and clinical anxiety: a randomized controlled trial. *Behavior Therapy*, 46, 7-19.

Eleven 90-minute sessions were provided on a weekly basis (30 minutes each provided separately with the child and parent, and 30 minutes with child and parent). The BIACA intervention led to greater numbers of children having reduced anxiety severity and a greater proportion with a positive treatment response.

A second US trial evaluated a modified version of BIACA for children aged 7-11<sup>201</sup>. At the immediate post-treatment point, children receiving BIACA had greater reductions in anxiety symptoms and severity; these treatment gains were maintained 3 months after treatment ended. Children in the CBT group also had greater improvements post-treatment in overall social responsiveness and in the specific domains of mannerisms and communication. A third US trial involving adolescents aged 11-16, modified the approach further, with at least three sessions devoted to developing coping skills (e.g. behavioural activation, cognitive restructuring) and additional modules offered to address issues such as social and adaptive skill deficits/problems, poor motivation, social/school issues, and co-occurring conditions<sup>202</sup>. At post-treatment and 1 month later, children receiving the intervention had significantly greater reductions in anxiety symptoms and severity, a greater proportion no longer had their primary anxiety diagnosis, and there were reductions in functional impairment and externalising child behaviour.

*Cool Kids* is a family-focused CBT programme for high-functioning autistic young people aged

8-13 with anxiety. It is provided in weekly 2-hour sessions for 12 weeks in groups of 6-8 participants, led by two clinical psychologists. A concurrent group-based parent discussion session is led by another psychologist. The aim is to treat the main components of anxiety rather than a specific anxiety disorder. One Australian trial compared *Cool Kids* to a waiting-list control, finding that *Cool Kids* CBT led to reduced numbers of children with anxiety and greater reductions in symptoms<sup>203</sup>.

*Facing Your Fears* is family-based CBT for high-functioning autistic children aged 7-14 years with anxiety. There are 12 multi-family group sessions of 90 minutes each, with group size between 3-6 children and parents, and led by one clinical psychologist, supported by two co-therapists (clinical psychology trainees). A trial in the US compared *Facing Your Fears* to treatment as usual. The CBT intervention led to reductions in anxiety severity as rated by clinicians (including separation anxiety, social anxiety, specific-phobia anxiety, and generalised anxiety), fewer children with generalised anxiety diagnosis at post-treatment and more children with a positive treatment response<sup>204</sup>. A pilot study has looked at a telehealth version of *Facing Your Fears* to test the feasibility of this mode of delivery in order to increase treatment access, particularly in rural or remote communities<sup>205</sup>. Participants were families with high-functioning autistic young people aged 7-19 with anxiety. Videoconferencing clinic-to-home groups (with two to four families in each) met

<sup>201</sup> Storch, E.A., Arnold, E.B., Lewin, A.B. et al. (2013). The effect of cognitive-behavioral therapy versus treatment as usual for anxiety in children with autism spectrum disorders: a randomized, controlled trial. *Journal of the American Academy of Child and Adolescent Psychiatry*, 52, 132-142.

<sup>202</sup> Storch, E.A., Lewin, A.B., Collier, A.B. et al. (2015) A randomized controlled trial of cognitive behavioral therapy versus treatment as usual for adolescents with autism spectrum disorders and comorbid anxiety. *Depression and Anxiety*, 32, 174-181.

<sup>203</sup> Chalfant, A.M., Rapee, R., Carroll, L. (2007). Treating anxiety disorders in children with high-functioning autism spectrum disorders: a controlled trial. *Journal of Autism and Developmental Disorders*, 37, 1842-1857.

<sup>204</sup> Reaven, J., Blakeley-Smith, A., Culhane-Shelburne, K., Hepburn, S. (2012) Group cognitive behavior therapy for children with high-functioning autism spectrum disorders and anxiety: a randomized trial. *Journal of Child Psychology and Psychiatry*, 53, 410-419. Hepburn, S.L., Blakeley-Smith, A., Wolff, B., Reaven, J.A. (2016). Telehealth delivery of cognitive-behavioral intervention to youth with autism spectrum disorder and anxiety: a pilot study. *Autism*, 20, 207-218.

for 11 sessions of 1 hour. The expectation of this study was to engage more with parents relative to their children; in contrast, standard (i.e. non-telehealth) CBT expects parents and their children to participate to the same degree. This evaluation found that the telehealth version was acceptable to families (high levels of attendance and satisfaction) but there were some (minor) technical problems around connectivity. The telehealth version of Facing Your Fears resulted in greater reductions in parent-reported child anxiety compared to the waiting-list control.

## Economic case

Interestingly, the only within-trial economic study found that the particular CBT approach evaluated (Discussing + Doing = Daring in the Netherlands) was not cost-effective – no reduction in anxiety, no improvement in health-related quality of life, and no cost differences<sup>206</sup>. There appear not to have been within-trial economic evaluations in any of the other studies described above, all of which found some effectiveness gains from CBT.

NICE conducted a meta-analysis of 21 randomised controlled trials that were rated as having moderate to high quality<sup>207</sup>. It then recommended the use of individual and group-based CBT for high-functioning autistic children with anxiety on the basis that both delivery modes were effective compared to waiting-list control. But while both delivery modes are effective, group-based CBT is more cost-effective than individual CBT. The cost-effectiveness ratio calculated by NICE

for group-based CBT was £13,910 per quality-adjusted life-year (QALY), which is well below the lower threshold that NICE uses to inform its recommendations. This cost-effectiveness analysis by NICE only included the cost of the intervention and benefits only to the child, since at the time of model development there was no information on how CBT might alter children's and parents' use of other services, nor how reductions in a child's anxiety might produce benefits for parents.

We therefore extended the NICE model. We were able to bring in additional information to consider more potential benefits and cost savings, particularly impacts on the child's caregivers. Evidence on these potential additional impacts is still limited, but we were able to draw on the findings from one CBT study that showed that parents have increased leisure time and reduced expenditure on household damages due to improvements in behaviour and an increase in the child's independence in personal daily living skills<sup>208</sup>, and on one parent-focused parenting study that reported improved parental mental health and corresponding quality of life<sup>209</sup>. Including these additional impacts, the short term cost-effectiveness of CBT becomes more attractive, even under quite conservative assumptions. The conclusion from our analysis, which takes a societal perspective, is that we are 95% certain that the group-based CBT is cost-effective at £2,550 per QALY. For individual CBT, we are 95% certain that the intervention has a cost-effectiveness of £31,050 per QALY under our most conservative assumptions<sup>210</sup>.

<sup>206</sup> Van Steensel et al. (2014) op.cit.

<sup>207</sup> NICE Clinical Guideline 170.

<sup>208</sup> Drahota et al. (2011) op.cit.

<sup>209</sup> Tonge, B., Brereton, A., Kiomall, M. et al. (2005). Effects on parental mental health of an education and skills training program for parents of young children with autism: a randomized controlled trial. *Journal of American Academy of Child and Adolescent Psychiatry*. 45, 561-569.

<sup>210</sup> Lemmi, V., Trachtenberg, M., Knapp, M. (2016) paper in preparation.

## Conclusions

Anxiety experienced by autistic children and young people can have serious repercussions for those individuals, their parents and other carers, including stress, other mental health problems, school and later employment difficulties. CBT-based interventions have been found to help to alleviate the anxiety symptoms, and some studies also point to other benefits, for example for parental wellbeing and tasks. All of the studies to date have had relatively short follow-up periods, and it is not known how durable these benefits are in the months following therapy.

The available evidence on CBT comes from a range of randomised trials and other studies that focus on high-functioning autistic young people with anxiety problems that are already established (i.e. we did not find any preventative studies). There is no way of knowing whether the evidence would generalise to autistic young people with learning disabilities, but we suspect not readily, though one small study has researched visual adaptations of CBT for anxious autistic adults<sup>211</sup>.

The one study that we found of telehealth-based CBT was quite modest in scale and scope but offered encouraging evidence. In the longer-term, there ought to be considerable potential

to make greater use of technology-delivered therapies such as CBT, but at the moment robust, generalisable evidence is still awaited.

Almost all of those studies of CBT come from outside the UK, and there is thus the familiar question about transferability. However, the findings from CBT studies in wider non-autistic contexts have been found to replicate across different high-income countries, and we would expect the same to apply to the autism-specific evidence. Interestingly, most studies look at CBT that is not necessarily delivered by fully trained clinical psychologists, which should make those approaches more feasible and affordable but may not enhance their autism specificity.

Although there have been very few economic studies of CBT, the model built by NICE to inform its clinical guideline development offered evidence of cost-effectiveness. Our new analyses broadened the NICE model to incorporate parent effects, and showed that the economic case for CBT-based approaches is even stronger. As with many of the interventions we looked at in NAP, there are important cross-overs from one domain of health or wellbeing to others. It would therefore be helpful to see more research on combinations of interventions such as CBT linked to supported employment programmes or social skills training.

<sup>211</sup> Ekman, E., Hiltunen, A.J. (2015). Modified CBT using visualization for Autism Spectrum Disorder (ASD), anxiety and avoidance behaviour - a quasi-experimental open pilot study. *Scandinavian Journal of Psychology*, 56, 641-648.

# EMPLOYMENT SUPPORT<sup>212</sup>

## Target population

Autistic young people and adults without learning disabilities.

## Context

Few autistic adults in the UK are in paid employment. A 2008 estimate for England suggested that only 15% are in full time employment, while 66% are not working<sup>213</sup>. A more recent survey of autistic people paints a similarly bleak picture with no real change<sup>214</sup>. Data from other countries consistently show that fewer than a third of autistic adults are employed<sup>215</sup>. Even among those achieving work, employment is likely to be part-time or in low-skill, low-paid occupations<sup>216</sup>.

Unemployment is both widespread and long-term. Among autistic adults aged 55 or over, 41% have been unemployed for more than 10 years. Among autistic people out of work, 59% do not believe or do not know if they will ever be employed<sup>217</sup>.

Maintaining employment can also be problematic for autistic people<sup>218</sup>. Nine out of ten job losses for disabled people relate to social skills deficits, which poses a particular problem for autistic adults<sup>219</sup>. They may experience multiple challenges in accessing and maintaining employment, such as difficulties in understanding nuances in job advertisements and interviews, difficulties in travelling to work because of sensory overload, lack of support before and during work, as well as employer discrimination<sup>220,221</sup>.

Nevertheless, and despite these challenges, 70% of autistic people without learning disabilities would like to work, as would 65% of people with learning disabilities (including autistic people)<sup>222</sup>.

While the challenges of increasing employment are substantial, so too are the potential rewards. Over 60% of autistic adults are financially dependent on their families<sup>223</sup>, and the productivity loss for an autistic adult without learning disabilities has been estimated at £21,797 per annum (2011 price levels). For an autistic adult with learning

<sup>212</sup> Case study prepared by Valentina Lemmi, Martin Knapp, Eric Koh, Philip Noden and Margaret Perkins.

<sup>213</sup> Rosenblatt, M. (2008). *I Exist: The message from adults with autism in England*. London: National Autistic Society.

<sup>214</sup> National Autistic Society (2016). *Too much information*. <http://www.autism.org.uk/get-involved/tmi.aspx>.

<sup>215</sup> Taylor, J.L., Seltzer, M.M. (2011). Employment and post-secondary educational activities for young adults with autism spectrum disorders during the transition to adulthood. *Journal of Autism and Developmental Disorders*, 41, 566-574.

<sup>216</sup> Levy, A., Perry, A. (2011). Outcomes in adolescents and adults with autism: a review of the literature. *Research in Autism Spectrum Disorders*, 5, 1271-1282.

<sup>217</sup> Bancroft, K., Batten, A., Lambert, S., Madders, T. (2012). *The Way We Are: Autism in 2012*. London: National Autistic Society.

<sup>218</sup> Taylor, J. L., Henninger, N.A., Mailick, M. R. (2015). Longitudinal patterns of employment and postsecondary education for adults with autism and average-range IQ. *Autism*, 19, 785-793.

<sup>219</sup> Strickland, D.C., Coles, C. D., Southern, L. B (2013). JobTIPS: a transition to employment program for individuals with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, 43, 2472-2483

<sup>220</sup> Forsythe, L., Rahim, N., Bell, L. (2008). *Benefits and employment support schemes to meet the needs of people with an Autistic Spectrum Disorder*. London: National Audit Office.

<sup>221</sup> Baldwin, S., Costley, D., Warren, A. (2014). Employment activities and experiences of adults with high-functioning autism and Asperger's disorder. *Journal of Autism and Developmental Disorders*, 44(10), 2440-2449.

<sup>222</sup> Townsley, R., Robinson, C., Williams, V. et al. (2014). *Employment and Young People with Autistic Spectrum Disorders: An Evidence Review*. Cardiff: Welsh Government.

<sup>223</sup> Rosenblatt (2008) op. cit.

disabilities the estimated loss is £25,644<sup>224</sup>. Of course, employment has a positive impact not only on an individual's income, but also and foremost on their quality of life, cognitive functioning, and well-being<sup>225</sup>.

In the right environment, autistic people can demonstrate particular strengths such as intense focus and attention to detail, which can result in higher productivity when compared with the neurotypical population<sup>226,227</sup>. They may also perform well in socially isolated or repetitive jobs which are often avoided by others<sup>228</sup>, although this should be a personal choice and not a societal assumption. The potential value of autistic employees has recently been recognised by some high-profile companies launching recruitment campaigns specifically targeting autistic people<sup>229</sup>. However, when receiving assistance in seeking employment, most autistic individuals rely on pan-disability employment programmes which may not be well suited to their needs<sup>230</sup>.

Autistic people are more likely to be employed if they come from households with a higher family income, higher parental education, if they are male, have higher social skills, an absence of learning disability, higher educational qualifications, have received career counselling in school and received post-secondary vocational training<sup>231,232</sup>.

## Intervention

Employment support interventions vary. They comprise a range of programmes that aim to enable autistic people to gain and maintain employment. Traditional arrangements were usually train and place programmes, offering training and supported (often closed) employment options. Over the past 20 or more years, interest has focused on place and train programmes that offer on-the-job training and support.

Many of these interventions share common features:

- consideration of the individual's strengths and interests when matching employers and employees;
- vocational training involving structured techniques such as behavioural therapies, natural support or simulation;
- a job coach to provide individualised training and workplace support;
- involvement of families, partners, carers, employers and colleagues to provide long term, comprehensive support and follow up to ensure job retention<sup>233</sup>.

<sup>224</sup> Buescher, A.V., Cidav, Z., Knapp, M., Mandell, D. S. (2014). Costs of autism spectrum disorders in the United Kingdom and the United States. *JAMA Pediatrics*, 168, 721-728.

<sup>225</sup> Walsh, L., Lydon, S., Healy, O. (2014). Employment and vocational skills among individuals with autism spectrum disorder: Predictors, impact, and interventions. *Review Journal of Autism and Developmental Disorders*, 1, 266-275.

<sup>226</sup> Wehman, P., Smith S.C. (2009). *Autism and the transition to adulthood: success beyond the classroom*. Baltimore: Brookes Publishing Company.

<sup>227</sup> Hendrie, D., Falkmer, M., Falkmer, T. et al. (2016). *Autism in the workplace: maximising the potential of employees on the autistic spectrum*. Bankwest Curtin Economics Centre.

<sup>228</sup> Knapp, M., Romeo, R., Beecham, J. (2009). Economic cost of autism in the UK. *Autism*, 13, 317-336.

<sup>229</sup> [www.bloomberg.com/news/articles/2015-06-02/autistic-coders-get-jobs-as-microsoft-sap-woo-software-sleuths](http://www.bloomberg.com/news/articles/2015-06-02/autistic-coders-get-jobs-as-microsoft-sap-woo-software-sleuths).

<sup>230</sup> Forsythe, L., Rahim, N., Bell, L. (2008). *Benefits and employment support schemes to meet the needs of people with an autistic spectrum disorder*. London: Inclusion Research and Consultancy.

<sup>231</sup> Walsh, L., Lydon, S., & Healy, O. (2014). Employment and vocational skills among individuals with autism spectrum disorder: Predictors, impact, and interventions. *Review Journal of Autism and Developmental Disorders*, 1, 266-275.

<sup>232</sup> Chiang, H.M., Cheung, Y.K., Li, H., Tsai, L.Y. (2013). Factors associated with participation in employment for high school leavers with autism. *Journal of Autism and Developmental Disorders*, 43, 1832-1842.

<sup>233</sup> Westbrook, J.D., Nye, C., Fong, C.J. et al. (2012). Effectiveness of adult employment assistance services for persons with autism spectrum disorders. *Campbell Systematic Review* 5, 1-67.

Employment support is, however, currently received by only 10% of autistic people, whereas 53% would like to have access to it<sup>234</sup>.

## Effectiveness

There is a small but consistent body of literature indicating that employment support programmes for autistic people have been effective in helping them gain and retain competitive employment<sup>235</sup>. Most studies have some methodological limitations, but two are of particular interest.

The only UK study was a pilot investigation of the effects of *Prospects* (a supported employment scheme funded by the then Department for Education and Employment in collaboration with the National Autistic Society)<sup>236</sup>. The intervention focused on high-functioning autistic people. At the end of the 2-year study period those using the *Prospects* service were significantly more likely to be employed (63% compared to 25%) than those getting usual services. The supported employment group also spent a greater proportion of time at work (27% compared to 12%) and had obtained higher level jobs than the control group. When followed up, 13 of the 19 employed adults from the original sample who found employment were still employed 7-8 years later. Overall, two-thirds of young adults had found employment, mostly with permanent contracts and involving administrative,

technical or computing work. Those individuals who received employment support experienced a rise in salaries, contributed more in taxation and claimed fewer benefits<sup>237</sup>.

A recent randomised controlled trial of *Project Search with ASD supports (PS-ASD)* - a very intensive 9-month job internship programme developed at Cincinnati Children's Hospital - found that 88% of the participants in the intervention group acquired employment compared with 6% of participants in the control group (who were provided with educational support)<sup>238</sup>. A subsequent retrospective study of administrative records showed that the PS-ASD group achieved higher wages and had better employment retention rates than a similar group of autistic adults who received supported employment only<sup>239</sup>.

## Economic case

In 2012, NICE published a clinical guideline providing evidence-based recommendations. Their systematic review identified only the Mawhood and Howlin study<sup>240</sup> as meeting the quality criteria to inform an economic evaluation of the cost-effectiveness of employment support<sup>241</sup>. The economic evaluation carried out by NICE assumed that the benefits of employment for autistic people were the same as the benefits for the general population, and concluded that the supported

<sup>234</sup> Bancroft et al. (2012) op.cit.

<sup>235</sup> National Collaborating Centre for Mental Health. (2012). *Autism: recognition, referral, diagnosis and management of adults on the autism spectrum*. London: British Psychological Society.

<sup>236</sup> Mawhood, L. Howlin, P. (1999). The outcome of a supported employment scheme for high-functioning adults with autism or Asperger syndrome. *Autism*, 3, 229-254.

<sup>237</sup> Howlin, P., Alcock, J., Burkin, C. (2005). An 8 year follow-up of a specialist supported employment service for high-ability adults with autism or Asperger syndrome. *Autism*, 9, 533-549.

<sup>238</sup> Wehman, P.H., Schall, C.M., McDonough, J. et al. (2014). Competitive employment for youth with autism spectrum disorders: early results from a randomized clinical trial. *Journal of Autism and Developmental Disorders*, 44, 487-500.

<sup>239</sup> Schall, C.M., Wehman, P., Brooke, V. et al. (2015) Employment interventions for individuals with ASD: the relative efficacy of supported employment with or without prior Project SEARCH training. *Journal of Autism and Developmental Disorders*, 45, 3990-4001.

<sup>240</sup> Mawhood & Howlin (1999) op.cit.

<sup>241</sup> Mavranzeouli, I., Megnin-Viggars, O., Cheema, N. et al. (2014). The cost-effectiveness of supported employment for adults with autism in the United Kingdom. *Autism*, 18, 975-984.

employment intervention was cost-effective. The cost-effectiveness ratio was calculated to be £5600 per quality-adjusted life year (QALY), which is well below the NICE threshold for recommending treatments (£20,000 to £30,000 per QALY).

Further analysis as part of the National Autism Project extended the NICE analysis, in particular to look beyond the health and social care perspective that NICE always adopts in its analyses. By taking additional account of the productivity gains arising from supported employment (including the types of jobs that people secured) and also from carers' increased productivity, the intervention was found to be even more cost-effective. Indeed, the further analysis showed that supported employment was not only more effective but also *cost-reducing*<sup>242</sup>. Individuals who received employment support would have been better off as a result.

## Conclusions

Few autistic people are in paid employment, yet when it is secured it can confer financial as well as many non-financial benefits. Supported employment schemes take account of individual strengths and preferences, provide individualised training and workplace support through a job coach, and involve a range of stakeholders (including families, partners, carers, employers

and work colleagues). For autistic people they may also include structured therapeutic techniques. Research shows that supported employment can be successful, although - while the research conclusions are consistent and encouraging - they are based on a small number of small-scale studies targeting a subgroup of autistic people.

There is also a strong economic case for supported employment, whether by reference to NICE thresholds (which only take into account the consequences in the health and social care system) or by looking at the broader societal perspective which also measures the productivity gains. Supported employment has benefits for autistic people, including economic benefits.

There is a need to examine whether the outcomes from studies to date can be replicated on a larger scale and - where appropriate - to a progressively wider subgroup of autistic people.

<sup>242</sup> Koh, E., Lemmi, V., Knapp, M. (2016) paper in preparation.

# HEALTH CHECKS<sup>243</sup>

## Target population

Autistic children and adults with or without learning disability.

## Context

Many autistic people experience poor health and poor healthcare, causing pain, distress, premature mortality and above-average suicide risk. There is a particular danger that mental health problems go unrecognised and untreated.

A UK population-based psychiatric study found that 70% of autistic children aged 10-14 years reported one or more co-occurring psychiatric disorders and 40% had two or more<sup>244</sup>. A US study compared autistic and non-autistic children and found that the autistic group had higher rates of language disorder, major depressive disorder, psychosis, anxiety (in particular, multiple anxiety disorders, specific phobias, agoraphobia, social phobia, and obsessive-compulsive disorder) and encopresis elimination disorder<sup>245</sup>. Treating mental health problems might improve a child's functioning and increase the efficacy of autism-related behavioural interventions<sup>246</sup>.

Compared to the UK general population, suicide rates are higher among adults with a diagnosis of

Asperger's syndrome, and are even higher than for people with psychotic illness or with one or more medical illnesses<sup>247</sup>. Asperger adults with depression are more likely to report suicidal ideation and suicide plans or attempts than Asperger adults without depression<sup>248</sup>. Depression is potentially a problem for many autistic people given that it often results from social isolation or exclusion, and unemployment. Similar evidence comes from another recent Swedish study: high-functioning autistic adults are more than nine times more likely to commit suicide than the general population. This same study also found that suicide rates are lower for adults with learning disability (about twice the population average) but increase tremendously when these individuals have co-occurring mental and behavioural disorders (21 times higher)<sup>249</sup>.

In the same study, overall mortality rates were more than twice as high among autistic adults than in the general population with the rate being especially high for adults with low-functioning autism (six times more likely), and indeed slightly higher still for females<sup>250</sup>. A number of disorders were identified as particularly increasing the risk of mortality (with some differences by gender): epilepsy, congenital malformations, mental and behavioural disorders, respiratory system disorders, circulatory system disorders and digestive system disorders<sup>251</sup>.

<sup>243</sup> Case study prepared by Valentina Lemmi, Martin Knapp and Marija Trachtenberg.

<sup>244</sup> Simonoff, E., Pickles, A., Charman, T. et al. (2008). Psychiatric disorders in children with autism spectrum disorders: prevalence, comorbidity, and associated factors in a population-derived sample. *Journal of the American Academy of Child & Adolescent Psychiatry*, 47, 921-929.

<sup>245</sup> Joshi, G., Petty, C., Wozniak, J. et al. (2010). The heavy burden of psychiatric comorbidity in youth with autism spectrum disorders: A large comparative study of a psychiatrically referred population. *Journal of Autism and Developmental Disorders*, 40(11), 1361-1370.

<sup>246</sup> McDougle, C. J., Stigler, K. A., Posey, D. J. (2003). Treatment of aggression in children and adolescents with autism and conduct disorder. *Journal of Clinical Psychiatry*, 64, 16-25.

<sup>247</sup> Cassidy, S., Bradley, P., Robinson, J. et al. (2014). Suicidal ideation and suicide plans or attempts in adults with Asperger's syndrome attending a specialist diagnostic clinic: a clinical cohort study. *Lancet Psychiatry*, 1, 142-147.

<sup>248</sup> *ibid.*

<sup>249</sup> Hirvikoski et al. (2016) *op.cit.*

<sup>250</sup> *ibid.*

<sup>251</sup> *ibid.*

One major contributor to these poor health outcomes is poor healthcare. A survey by the Westminster Commission on Autism found that three-quarters of respondents (autistic people, parent-advocates and professionals) believed that autistic people receive ‘worse’ or ‘much worse’ healthcare than neurotypical people<sup>252</sup>. A similar proportion felt that health professionals ‘rarely’ or ‘never’ understand autism and how it affects someone’s physical and mental health. Evidence on actual patterns of healthcare use by autistic people compared to the rest of the population is not readily available in the UK, but a large-sample US study revealed significantly higher use of accident and emergency services for psychiatric and non-psychiatric reasons and due to injuries, compared to non-autistic people. Comparatively, visits due to injuries were highest for falls, self-harm and suicidal ideation<sup>253</sup>.

## Intervention

Health checks aim ‘to reduce morbidity and mortality through earlier detection and treatment of diseases and risk factors for diseases’<sup>254</sup>, and include preventative health screening for at-risk groups and periodic health examinations for people with previously diagnosed conditions. Health checks may be offered in residential or community settings by medical professionals, such as general practitioners, dentists and nurses.

## Effectiveness

There have been a small number of evaluations of health checks for autistic people or for people with learning disabilities (including some who are autistic).

An intervention based on the *Treatment and Education of Autistic and related Communication-handicapped Children (TEACCH)* model (see the **Social Skills** case study for more detail) was structured into five 20-minute sessions over 3 weeks (including training) to facilitate a 10-component dental assessment programme. Structured teaching strategies used visual information (photographs, plastic models, dental brushes, dentist material, toys) and auto-modelling using computers and cameras. The intervention was administered by a dentist specialised in special needs. A small uncontrolled before-after study in Spain showed an increase in compliance and number of dental assessment components reached<sup>255</sup>.

*The Sensory Adapted Dental Environment (SADE)* intervention aims to reduce sensory-related distress and anxiety in autistic children facing dental treatment. It is based on both the multisensory environments and sensory integration theories, with the treatment environment adapted in visual, auditory, and tactile ways (darkened room, music and a butterfly-like wrap providing a ‘hugging’ sensation). A randomised controlled trial of a pilot intervention in the US found that SADE reduced sensory-related distress and anxiety in both autistic and neurotypical children<sup>256</sup>. The trial found that

<sup>252</sup> Westminster Commission on Autism (2016). *A Spectrum of Obstacles An Inquiry into Access to Healthcare for Autistic People*. London: National Children’s Group.

<sup>253</sup> Vohra, R., Madhavan, S., Sambamoorthi, U. (2016). Emergency department use among adults with autism spectrum disorders (ASD). *Journal of Autism and Developmental Disorders*, 46, 1441-1454.

<sup>254</sup> Krogsbøll LT, Jørgensen KJ, Grønhøj Larsen C et al. (2012) General health checks in adults for reducing morbidity and mortality from disease. *Cochrane Database of Systematic Reviews*, 10: CD009009.

<sup>255</sup> Orellana, L. M., Martínez-Sanchis, S., Silvestre, F. J. (2014). Training adults and children with an autism spectrum disorder to be compliant with a clinical dental assessment using a TEACCH-based approach. *Journal of Autism and Developmental Disorders*, 44, 776-785.

<sup>256</sup> Cermak, S.A., Duker, L. I. S., Williams, M. E. et al. (2015). Sensory adapted dental environments to enhance oral care for children with autism spectrum disorders: a randomized controlled pilot study. *Journal of Autism and Developmental Disorders*, 45, 2876-2888.

SADE increased the duration of dental cleaning but reduced the need both for staff supporting the children and for anaesthesia, which could have resource implications, although an economic evaluation was in fact not conducted.

There have been a few studies of health checks - both nurse-led and GP-led – that have focused on the overall group of people with learning disabilities, including some who are autistic but who generally are not separated out in the analyses.

One intervention in Scotland looked at a *nurse-lead health check* for adults with learning disabilities which aimed to identify at-risk conditions and general health needs, and to monitor chronic conditions. Nurses with experience in learning disabilities reviewed an individual's primary care records and then assessed overall health (physical and mental, developmental problems, behavioural issues) using a semi-structured assessment instrument, physical examinations and blood tests. This process lasted about 4 hours. The nurse then discussed the results with a GP, sent a summary report and recommendations to the individual's own GP and also made a direct referral to the learning disability team if needed. An initial evaluation looked at 50 adults with learning disabilities and 50 matched controls; it found better detection and treatment of new health needs, health monitoring, and health promotion compared to usual care treatment after one year<sup>257</sup>. A subsequent cluster randomised trial found no significant difference in newly detected

health needs but that the health-check group had significantly more health monitoring needs met than the standard-care group<sup>258</sup>.

An alternative is a *GP-lead health check*. NHS Wales introduced the first national learning disabilities health check scheme in 2006, and in 2008, NHS England introduced a financial incentive to primary care teams to offer people with learning disabilities an annual health check<sup>259</sup>. A longitudinal cohort study<sup>260</sup> showed an increase in health assessments, health action plans, and referrals compared to non-incentivised practices over the first three years of implementation of the programme. Factors found to reduce the likelihood of attendance for a health check were being a woman, being young and living in a more socially deprived area.

In Australia, a *Comprehensive Health Assessment Programme (CHAP)* has been trialled. A carer of someone with learning disabilities will complete a detailed medical history in a CHAP booklet, pass it to the GP to review the medical history and assess the individual's health, and then a health action plan is developed jointly between GP, carer and patient. The CHAP contains information on at-risk and poorly managed conditions. A randomised controlled trial in Australia showed that CHAP increased detection of new health needs and health promotion over the year following CHAP assessment compared to usual care for adults with learning disabilities living in supported accommodation<sup>261</sup>. An associated study showed that GPs found it

<sup>257</sup> Cooper, S.A., Morrison, J., Melville, C. et al. (2006). Improving the health of people with intellectual disabilities: outcomes of a health screening programme after 1 year. *Journal of Intellectual Disability Research*, 50, 667-677.

<sup>258</sup> Cooper, S.A., Morrison, J., Allan, L.M. et al. (2014). Practice nurse health checks for adults with intellectual disabilities: a cluster-design, randomised controlled trial. *Lancet Psychiatry*, 1, 511-521.

<sup>259</sup> McConkey, R., Taggart, L., Kane, M. (2015). Optimizing the uptake of health checks for people with intellectual disabilities. *Journal of Intellectual Disabilities*, 19, 205-214.

<sup>260</sup> Buszewicz, M., Welch, C., Horsfall, L. et al. (2014). Assessment of an incentivised scheme to provide annual health checks in primary care for adults with intellectual disability: a longitudinal cohort study. *Lancet Psychiatry*, 1, 522-530.

<sup>261</sup> Lennox, N., Bain, C., Rey-Conde, T. et al. (2007). Effects of a comprehensive health assessment programme for Australian adults with intellectual disability: a cluster randomized trial. *International Journal of Epidemiology*, 36, 139-146.

helpful to have a structured tool to guide and strengthen their practice, but that one barrier to implementation was the additional time needed for the assessment. Another difficulty was a lack of collaboration from support workers due to limited capacity or motivation<sup>262</sup>.

The CHAP approach was explored further for community-dwelling adults with learning disabilities, now in comparison with a *health diary*. The latter is a health education intervention to encourage collection of personal health information and to teach self-advocacy skills. The Ask health diary has four sections: 'All about me' (personal and health information on the individual); 'Health Advocacy Tips' (information and advice on medical consultations for the person with learning disabilities and carer); 'For the Doctor' (information and advice on medical consultations for clinical staff); and 'Medical Records' (a detailed medical history). A randomised controlled study in Australia compared the health diary and CHAP, finding the latter to be better in terms of increased health promotion, disease prevention and case-finding activity<sup>263</sup>.

Another randomised controlled trial evaluated the Ask health diary and CHAP combined, now delivered as a school-based intervention, compared to usual care for adolescents with learning disabilities. The intervention was better

at triggering healthcare activities (checking of vision, hearing, blood pressure and weight) but did not improve identification of new diseases<sup>264</sup>. A parallel paper reported that adolescents were more knowledgeable and responsible for their health, and carers more knowledgeable and able to support the adolescent<sup>265</sup>.

## Economic case

The two studies of nurse-led health checks in Scotland described earlier both included economic evaluations. The earlier matched-control study found the cost of care to be lower for adults who received the health check, mainly due to lower costs of unpaid care<sup>266</sup>. The later randomised trial found that the intervention 'dominated' usual care treatment: it was both more effective and cheaper<sup>267</sup>.

There have also been economic evaluations of the CHAP intervention in Australia described earlier. Analyses of administrative data alongside the RCT for the study of adults with learning disabilities found that public sector costs over one year for those receiving CHAP were not statistically different from costs for those receiving usual care<sup>268</sup>. The study of a school-based intervention for adolescents with learning disabilities (CHAP plus a health diary) found that there was no difference in GP or hospital use between the groups<sup>269</sup>.

<sup>262</sup> Lennox, N., Brolan, C. E., Dean, J. et al. (2013). General practitioners' views on perceived and actual gains, benefits and barriers associated with the implementation of an Australian health assessment for people with intellectual disability. *Journal of Intellectual Disability Research*, 57, 913-922.

<sup>263</sup> Lennox, N., Bain, C., Rey-Conde, T. et al. (2010). Cluster randomized-controlled trial of interventions to improve health for adults with intellectual disability who live in private dwellings. *Journal of Applied Research in Intellectual Disabilities*, 23, 303-311.

<sup>264</sup> Lennox, N., McPherson, L., Bain, C. et al. (2016). A health advocacy intervention for adolescents with intellectual disability: a cluster randomized controlled trial. *Developmental Medicine & Child Neurology* 58, 1265-1272.

<sup>265</sup> McPherson, L., Ware, R. S., Carrington, S. et al. (2016). Enhancing self-determination in health: results of an RCT of the Ask project, a school-based intervention for adolescents with intellectual disability. *Journal of Applied Research in Intellectual Disabilities*, forthcoming.

<sup>266</sup> Romeo, R., Knapp, M., Morrison, J. et al. (2009). Cost estimation of a health-check intervention for adults with intellectual disabilities in the UK. *Journal of Intellectual Disability Research*, 53, 426-439.

<sup>267</sup> Cooper et al. (2014) op.cit.

<sup>268</sup> Gordon, L. G., Holden, L., Ware, R. S. et al. (2012). Comprehensive health assessments for adults with intellectual disability living in the community: weighing up the costs and benefits. *Australian Family Physician*, 41, 969-972.

<sup>269</sup> Lennox et al. (2016) op.cit.

## Conclusions

Many studies have provided evidence that autistic people have poorer health than the neurotypical population, in part because of poorer access to healthcare. Action is urgently needed to tackle these health inequities. Regular health checks offer one way to support health promotion and to ensure reasonably prompt illness detection.

We could, however, find very little evidence at all that was specific to autistic people, and there is clearly a pressing need to explore this area through well-conducted research. One particular emphasis should be on how to improve access to healthcare for high-functioning autistic people, especially because common experiences among this group – for example, social isolation and unemployment – predispose them to mental health problems such as depression. Anxiety is also prevalent, in part because of sensory-sensitivity and bullying. Interventions that improve awareness among key professionals of the risk of suicide among autistic people, and that take action to prevent such acts, should be explored.

Where autism-specific interventions have been tried – such as in relation to dental checks for children – the evidence is broadly encouraging. Success depends in part on a child's neuropsychological profile, especially their learning disabilities and behaviours<sup>270</sup>.

In contrast, there are some well-designed studies of health checks for people with learning disabilities,

and within that population will be autistic people. These health checks can be nurse-led or GP-led, and Australian studies have explored a comprehensive health assessment programme. The evidence suggests that more health needs are indeed identified, and that these various programmes of health checks are cost-effective – indeed, quite possibly cost-saving, even in the relatively short term. They ought to establish better ways of working that can also generate longer-term health improvements. Using a health diary might additionally promote self-advocacy and thence better health outcomes<sup>271</sup>.

Health checks for people with learning disabilities have been promoted for a few years now in parts of the UK. Analysis of administrative data for 2013/14 in England found that 65% of GP practices provided GP-led health checks, and 44% of people with learning disabilities known to practices received the intervention<sup>272</sup>. In Northern Ireland it was found for the same year that 84% of practices provided GP-led health checks, and 64% of people with learning disabilities known to practices received the intervention<sup>273</sup>. There is clearly still some distance to travel to achieve a truly comprehensive service: factors found to be negatively associated with attendance by individuals invited to attend for a health check are living independently and living in a more deprived area. What is needed now is collection of data on health checks specifically for autistic people<sup>274</sup>.

<sup>270</sup> Du, R.Y., Yiu, C. C., Wong, V. C. et al. (2015). Autism developmental profiles and cooperation with oral health screening. *Journal of Autism and Developmental Disorders*, 45, 2758-2763.

<sup>271</sup> Carrington, S., Lennox, N., O'Callaghan, M. et al. (2014). Promoting self-determination for better health and wellbeing for adolescents who have an intellectual disability. *Australasian Journal of Special Education*, 38, 93-114.

<sup>272</sup> Public Health England (2014). *The Uptake of Learning Disability Health Checks 2013 to 2014*. London: PHE.

<sup>273</sup> McConkey et al. (2015) op.cit.

<sup>274</sup> Westminster Commission on Autism (2016) op.cit.

# PERSONALISED CARE AND SUPPORT<sup>275</sup>

## Target population

Autistic children and adults with or without learning disability.

## Context

There is obviously enormous heterogeneity in any population in terms of individual characteristics, social and economic circumstances, health status, aspirations, preferences and much more. That is certainly the case for the autistic population. It means that different autistic people and their families are likely to need and to want different types and levels of support. The need for personalised responses to needs and preferences was emphasised in our discussion of overarching principles. The effects of individualised programmes of support have also been noted in some of the other case studies (such as those on **Social skills, Assistive technologies, Parent training and Employment support**). Here we describe evidence on some other ‘personalised’ approaches.

The broader policy context for this case study is the emphasis from successive UK and English governments on ‘personalisation’ of social and healthcare (and indeed other areas of public policy such as housing, education and pensions, although in slightly different guises) with the aim of giving service users (‘consumers’) more direct choice over how their needs are met, and more control over their lives. This approach has appeal across the political spectrum. It reflects the observation that individuals generally want greater opportunity for self-determination (to participate, choose, take control), and also the (testable) argument that empowering

people in this way leads to more responsive care and support systems, and thereby to better outcomes and greater cost-effectiveness. It mimics some aspects of market-like discipline. It might also encourage family and community action, and by giving support to disadvantaged groups it could lead to fairer public systems.

For people with learning disabilities, there were earlier initiatives to move towards more individualised support arrangements, as exemplified by such approaches as normalisation and social role valorisation, which gained strong support in some quarters during the period when the old asylums and other long-stay institutions were being closed and new community care arrangements were needed to replace them.

## Intervention

Personalised care and support includes interventions for autistic individuals and individuals with learning disability tailored to individual needs, strengths and preferences, and aiming to improve autonomy and quality of life. They include a wide range of interventions targeting different groups of people.

## Effectiveness

There have been a few evaluations of different types of personalised care and support for autistic people or people with learning disability.

### Active support

*Active support* is a person-centred model of support aiming to improve participation in meaningful

<sup>275</sup> Case study prepared by Valentina Iemmi and Martin Knapp.

activities and relationships for people with learning disability (including autistic people)<sup>276</sup>. The approach was initially designed for use in community-based residential group homes and involved developing processes and training staff in seven areas: a system of ‘routines and rhythms’ to assure the accomplishment of everyday activities; ‘activity protocols’ to break down activities into single components to assure consistency; ‘activity support plans’ to plan activities and the staff required for them; ‘community contacts monitoring’ to monitor community participation; ‘individual plans’ to target outcomes that are both realistic and meaningful for the individuals and their carers; ‘opportunity plans’ to develop through practice already acquired skills; and ‘structured teaching plans’ to develop long-term skills that are meaningful for the person.

A more recent conceptualisation of the approach, sometimes referred to as *person-centred active support*, has shifted the focus to enabling relationships between the person providing and the person receiving support in any settings in which people with learning and developmental disabilities are supported (e.g. at home, at school, at work, in the community)<sup>277</sup>. The four key elements are: ‘Every moment has potential’; ‘Little and often’; ‘Graded assistance to ensure success’; and ‘Maximising choice and control’.

In line with a previous review<sup>278</sup>, a more recent systematic review found promising improvements in engagement in meaningful activities for individuals receiving active support<sup>279</sup>. A recent UK observational study evaluated three types of skilled support: active support, PBS and SPELL (see below)<sup>280</sup>, suggesting that active support may be the ‘core indicator’ of skilled support, leading to better participation in meaningful activities for people with severe to profound learning disability and complex needs.

### **Structure, Positive (approaches and expectations), Empathy, Low arousal, Links (SPELL)**

SPELL is the National Autistic Society’s framework for understanding and responding to the needs of autistic individuals<sup>281</sup>. It can be applied in any settings and focuses on five core elements: ‘structure’ to provide predictability, reduce anxiety and promote independent action; ‘positive approaches and expectations’ to promote autistic individuals to learn new skills, have new experiences and to increase their self-esteem; ‘empathy’ to understand how autistic people experience the world; ‘low arousal’ by adapting the environment in order to address sensory sensitivities and facilitate calm and focused support; and ‘links’ with families, carers, schools, and public agencies to achieve consistency and social inclusion.

<sup>276</sup> Totsika, V., Toogood, S., Hastings, R.P. (2008). Active support: development, evidence base, and future directions. *International Review of Research in Mental Retardation*, 35, 205-249.

<sup>277</sup> Mansell, J., Beadle-Brown, J. (2012). *Active support: enabling and empowering people with intellectual disabilities*. London: Jessica Kingsley Publishers.

<sup>278</sup> Stancliffe, R.J., Jones, E., Mansell, J., Lowe, K. (2008). Active support: A critical review and commentary. *Journal of Intellectual and Developmental Disability*, 33, 196-214.

<sup>279</sup> Hamelin, J.P., Sturmey, P. (2011). Active support: a systematic review and evidence-based practice evaluation. *Intellectual and Developmental Disabilities*, 49, 166-171.

<sup>280</sup> Beadle-Brown, J., Leigh, J., Whelton, B. et al. (2015). Quality of life and quality of support for people with severe intellectual disability and complex needs. *Journal of Applied Research in Intellectual Disabilities*, 29, 409-421.

<sup>281</sup> Beadle-Brown, J., Roberts, R., Mills, R. (2009). Person-centred approaches to supporting children and adults with autism spectrum disorders. *Tizard Learning Disability Review*, 14, 18-26.

### Positive behavioural support (PBS)

PBS is a person-focussed framework of care that seeks to improve quality of life and decrease concerning behaviours in people with learning disabilities<sup>282</sup>. PBS is 'a multicomponent framework (a) for developing an understanding of the behaviour displayed by an individual, based on an assessment of the social and physical environment and broader context within which it occurs; (b) with the inclusion of stakeholder perspectives and involvement; (c) using this understanding to develop, implement and evaluate the effectiveness of a personalised and enduring system of support; and (d) one that enhances quality of life outcomes for the focal person and other stakeholders'<sup>283</sup>.

PBS is built upon theory and an evidence base that is not autism specific<sup>284</sup>: understanding the functions of concerning behaviour, primary use of applied behaviour analysis and secondary use of other evidence-based approaches. It relies on specific values: stakeholder participation, quality of life and inclusion, using a constructional approach to prevent and reduce concerning behaviour. It is a data-driven approach, usually comprising an initial functional assessment, followed by multicomponent interventions, monitoring and evaluation. Multicomponent interventions vary according to individual needs and personal circumstances, and

may include management of concerning behaviour, functional communication training and self-care<sup>285</sup>. Similarly variable is the length of a PBS intervention, ranging from a few months to a few years.

PBS interventions are delivered by multidisciplinary teams including applied behavioural analysis (ABA)-trained professionals (clinical psychologists or behavioural analysts) and support workers. They can be delivered at home or in schools, health centres and residential care facilities. A UK trial that compared 'specialist behaviour therapy' (including PBS and ABA) with usual care for people with mild to severe learning disabilities showed improvement in concerning behaviours at 6 months, maintained at the 2 year follow-up<sup>286,287</sup>. Another UK trial, currently underway, is assessing the impact of PBS training for staff and paid carers working with people with mild to severe learning disabilities compared to treatment as usual<sup>288</sup>. A small UK non-controlled before-after study showed improvement in concerning behaviours and in carer ability to cope, although caution is needed due to the small sample size and design<sup>289</sup>.

### Circles of support

A Circle of Support 'is a group of people who meet together on a regular basis to help somebody accomplish their personal goals in life'<sup>290</sup>. The person at the centre of a Circle may have disabilities or other

<sup>282</sup> Carr E.G., Dunlap G., Horner R.H., et al. (2002) Positive behavior support: evolution of an applied science. *Journal of Positive Behavior Intervention* 4, 4–16.

<sup>283</sup> Gore N., McGill P., Toogood S., et al. (2013). Definition and scope for positive behavioural support. *International Journal of Positive Behavioural Support* 3, 14–23.

<sup>284</sup> *ibid.*

<sup>285</sup> National Collaborating Centre for Mental Health (2015). *Challenging Behaviour and Learning Disabilities: Prevention and Interventions for People with Learning Disabilities Whose Behaviour Challenges*.

<sup>286</sup> Hassiotis A., Robotham D., Canagasabay A. et al. (2009). Single-blind, controlled trial of a specialist behavior therapy team for challenging behavior in adults with intellectual disability. *American Journal of Psychiatry*, 166, 1278–1285.

<sup>287</sup> Hassiotis A., Canagasabay A., Robotham D. et al. (2011). Applied behaviour analysis and standard treatment in intellectual disability: 2-year outcomes. *British Journal of Psychiatry*, 198, 490–491.

<sup>288</sup> Hassiotis, A., Strydom, A., Crawford, M. et al. (2014). Clinical and cost effectiveness of staff training in Positive Behaviour Support (PBS) for treating challenging behaviour in adults with intellectual disability: a cluster randomised controlled trial. *BMC Psychiatry*, 14, 219.

<sup>289</sup> Reid, C., Sholl, C., Gore, N. (2013). Seeking to prevent residential care for young people with intellectual disabilities and challenging behaviour: examples and early outcomes from the Ealing ITSBS. *Tizard Learning Disability Review*, 18, 171–178.

<sup>290</sup> [circlesnetwork.org.uk/index.asp?level=0z114z115&parent\\_id=115](http://circlesnetwork.org.uk/index.asp?level=0z114z115&parent_id=115).

characteristics that makes it hard for them to achieve their life goals on their own, and the Circle acts like a personalised community. The person at the centre of the Circle should be in charge, although sometimes this does not happen (out of choice). Members of the Circle are unpaid; they can include family members, friends and other community supporters. The dominant philosophy permeating all Circles is that they see 'people as individuals who feel they need support in order to take more control over their own lives. A Circle properly facilitated is empowering to all of the individuals involved and, unlike many service systems, does not reinforce dependence'<sup>291</sup>.

A very small English exploratory study of Circles of Support for five adults with moderate to profound learning disabilities, one autistic, tentatively suggested that there had been success in enabling each 'focus person' to live a more independent life with better social care outcomes than had previously been achieved through more traditional support arrangements (which would probably have been long-term residential care placements)<sup>292</sup>. The Circles appeared to harness community resources to promote social inclusion and improve well-being for these five individuals.

### Personal budgets and direct payments

Personal budgets were piloted in social care in England more than a decade ago and rolled out nationally in 2008. These are individualised calculations of the care budget for an individual who is assessed by their local authority as meeting both social care need and financial criteria. The individual can, if they wish, take this as a *direct payment* and pay for their own care directly, and quite often will

then employ a personal assistant (PA) to support them. There are also now *personal health budgets* for some people in the NHS<sup>293</sup>, as well as a pilot scheme to evaluate integrated personal budgets that pool funds to cover both the health and social care needs of an individual. Local support organisations in either the statutory and voluntary sectors provide advice and a range of specific services such as help with employing a PA.

Individuals are supported to take varying degrees of control – depending on their abilities and preferences – over the (public) funds that would otherwise have been spent through conventional channels on their care and support and where decisions about that spending would be taken by care professionals. These budgets – whether directly held or managed by the local authority or a care provider on the user's behalf – in principle represent a highly individualised way to deliver services and support that people feel they need and want, subject to monitoring by a care professional.

A randomised trial of personal budgets in England involved almost 1000 adult users of social care services, comparing support in the usual way with the opportunity to hold a personal budget. At the time of the study these were called 'individual budgets'. A number of 'need groups' participated in the study. For the learning disabilities subgroup, which included autistic people (although separate analyses were not carried out for that subgroup) there were mixed results. Individuals and their carers often found the personal budgeting processes to be stressful, particularly when there were delays between assessment and putting the budget in place. On the other hand, compared to other groups

<sup>291</sup> Ibid.

<sup>292</sup> Wistow, G., Perkins, M., Knapp, M. et al. (2016). Circles of support and personalization: exploring the economic case. *Journal of Intellectual Disabilities*, 20, 194-207.

<sup>293</sup> Turner, S., Giraud-Saunders, A. (2014) *Personal health budgets: including people with learning disabilities*. Bath: NDTi.

of adult social care users, people with learning disabilities had more resources allocated to leisure or social participation activities, which was likely to contribute to higher levels of well-being<sup>294,295</sup>. An accompanying qualitative study interviewed 130 service users to paint a fuller picture of what a personal budget might offer, and how it might help to personalise care and support<sup>296</sup>.

A quasi-experimental study looked at a pilot programme of personal health budgets, finding them broadly effective and cost-effective<sup>297</sup>.

The voluntary organisation In Control, in collaboration with Lancaster University and Think Local Act Personal (a consortium of organisations), has monitored the roll-out of personal social care and health budgets, and the experiences of holders and families through a series of surveys using the personal outcomes evaluation tool (POET). The most recent survey found that the most common ways that individuals used their personal health budgets was on care and support services (60%), personal assistants (48%), community and leisure services (27%) and equipment (25%)<sup>298</sup>. Most respondents reported that having a personal health budget had a positive impact on quality of life, independence and arranging support. More than half reported a positive impact on relationships with people paid to support them, on friendships, physical and mental health. Carers frequently

reported that having a personal health budget reduced stress, improved their own quality of life, and the choice and control they experienced. These personal health budget were most commonly managed through direct payments (37%).

## Economic case

Some of these ‘personalised’ interventions have been examined from an economic perspective. We are not aware of published economic evidence on active support but there is currently a UK study underway, looking at people with severe learning disabilities and complex needs; it will report in 2017<sup>299</sup>.

The UK trial of specialist behaviour therapy described earlier<sup>300</sup> included an economic evaluation. It showed that health and social care costs appeared to decrease after 6 months for people with mild to severe learning disabilities receiving this therapy, although the difference was not statistically significant. No difference in costs was found at the 2-year follow-up either<sup>301</sup>. An economic evaluation is included in an ongoing UK trial<sup>302</sup> of PBS training for staff and paid carers.

An exploratory economic study alongside a very small UK non-controlled before-after study<sup>303</sup> of PBS at school showed improvements in concerning behaviours and social and communication skills of

<sup>294</sup> Glendinning, C., Challis, D., Fernández, J-L. et al. (2008). Evaluation of the Individual Budgets Pilot Programme, SPRU, PSSRU and SCWRU, York and London.

<sup>295</sup> Netten, A., Jones, K., Knapp, M. et al. (2012). Personalisation through Individual Budgets: does it work and for whom? *British Journal of Social Work*, 42, 1556-1573.

<sup>296</sup> Moran, N., Glendinning, C., Wilberforce, M. et al. (2013). Older people’s experiences of cash-for-care schemes: evidence from the English Individual Budget pilot projects. *Ageing and Society*, 33, 826-851.

<sup>297</sup> Forder, J., Jones, K., Glendinning, C. et al. (2012). Evaluation of the Personal Health Budget Pilot Programme. Department of Health, London.

<sup>298</sup> Hatton, C., Waters, J. (2015) Personal health budget holders and family carers: the POET surveys 2015. In *In Control*, Lancaster University and TLAP.

<sup>299</sup> Led by Julie Beadle-Brown and Jennifer Beecham.

<sup>300</sup> Hassiotis et al. (2009) op.cit.

<sup>301</sup> Hassiotis et al. (2011) op.cit.

<sup>302</sup> Hassiotis et al. (2014) op.cit.

<sup>303</sup> Lemmi, V., Knapp, M., Jackson Brown, F. (2016). Positive behavioural support in schools for children and adolescents with intellectual disabilities whose behaviour challenges: an exploration of the economic case. *Journal of Intellectual Disabilities*. DOI: 10.1177/1744629516632402

children and adolescents with learning disabilities. The cost of PBS was estimated at £36,405 per child per year (2012/13 price levels). Two other small economic studies have been undertaken in the UK. One estimated the cost of PBS in the community for two illustrative case studies of adolescents with learning disabilities and concerning behaviour<sup>304</sup>, and the other looked at PBS for adults<sup>305</sup>. These three exploratory studies together suggest the potential of PBS for cost savings from supporting people with learning disabilities and concerning behaviour because the intervention appeared to make it possible for them to stay living in the community rather than have to move to costly residential accommodations<sup>306</sup>.

The small Circles of Support study described earlier also concluded that this individualised approach to identifying and responding to needs and preferences – with the active involvement of the individual for whom the Circle is established – can help to avoid expensive long-term residential placements<sup>307</sup>.

The evaluation of the national pilot programme of personal social care budgets (known as individual budgets at the time of the pilot) found that social care costs were slightly lower for people with learning disabilities with a personal budget compared to the group getting usual care (£359 versus £390). Looking at both social care and health costs, personal budgets were found to have the potential to be cost-effective when considering how well social care needs were being met, but implementation delays in the pilot sites undermined these gains. Looking at the other main outcome measure (psychological well-being), standard care arrangements looked slightly more cost-effective than personal budgets<sup>308</sup>.

## Conclusions

We have looked at a number of interventions that seek to personalise care and support for individuals, although some of the interventions described in other case studies also aim to tailor services to individual circumstances and strengths and (to some extent) to individual preferences. As noted above, most of the evidence, which looks broadly positive, is not autism-specific, but relates to a wider population of people with learning disabilities or with concerning behaviours. We therefore do not know that the findings from those studies apply specifically to the distinctive autistic subgroup. There are also concerns strongly expressed by some autistic people and their families and carers that some behavioural therapies (and this would include ABA and PBS) are unethical, trying to force people to adopt (or stop) certain behaviours, even when it causes considerable discomfort or even pain, and therefore can cause harm.

Personalised approaches may require more staff inputs and organisational change, and so, initially at least, could look costly. Circles of Support need commitments from a range of people but in this case unpaid. The economic evidence is tentative but again broadly positive. It is not yet clear from the research literature whether there are long-term effectiveness gains and economic benefits. What *does* seem clear is that approaches that properly recognise individuality, respond creatively to offer real (and if necessary adequately supported) opportunities for people to take more control of their lives are surely to be encouraged.

<sup>304</sup> Lemmi, V., Knapp, M., Reid, C. et al. (2016). Positive behaviour support for children and adolescents with learning disabilities and behaviour that challenges living in the community: an exploratory economic evaluation. *Tizard Learning Disability Review*, 21, 169-180.

<sup>305</sup> Lemmi, V., Knapp, M., Saville, M. et al. (2015). Positive behavioural support for adults with intellectual disabilities and behaviour that challenges: an initial exploration of the economic case. *International Journal of Positive Behavioural Support*, 5, 16–25.

<sup>306</sup> Lemmi, V., Knapp, M., Gore, N. et al. (2016). What is standard care for people with learning disabilities and behaviour that challenges and what does it cost? *British Journal of Learning Disabilities*, published online.

<sup>307</sup> Wistow et al. (2016) op.cit.

<sup>308</sup> Glendenning et al. (2008) op.cit.

# ASSISTIVE DEVICES AND TECHNOLOGIES<sup>309</sup>

## Target population

Autistic children and adults with and without learning disability.

## Context

Many autistic people have a strong interest in technologies that has been linked to clarity in demands (and absence of social demands), immediate, predictable and repeatable responses, use of visual cues, and diminished sensory stimuli<sup>310</sup>. Technologies may be used to support functional skills such as interventions aiming to improve activities of daily living, social participation, communication skills, spatial and temporal planning and recognition of emotions.

Among the new technologies for autistic people that have emerged over recent years are augmentative and alternative communication technologies, computer-based technologies (e.g. educational and recreational), medical devices (e.g. biofeedback), and sensory aids (e.g. noise-cancelling headphones, coloured-lens glasses).

## Interventions

The World Health Organization offers this helpful definition: 'Assistive devices and technologies are those whose primary purpose is to maintain or improve an individual's functioning and

independence to facilitate participation and to enhance overall well-being. They can also help prevent impairments and secondary health conditions'<sup>311</sup>. Assistive technologies for autistic people are often designed as aids to develop social, behavioural and adaptive skills.

There are also several technology tools designed to help autistic people to communicate, often referred to as augmentative and alternative communication systems (AAC). Broadly speaking, AAC refers to strategies to compensate for poor (or absent) verbal skills, but have also demonstrated benefits for developing social, behavioural, academic and other skills. AAC systems encompass a variety of techniques from low-tech sign language and printed material to high-tech speech-generating devices, picture exchange communications systems, and eye-gaze systems.<sup>312,313</sup>

## Effectiveness

### Computer-assisted instruction (CAI)

Computer-assisted instruction interventions are designed to assist with developing social, academic and behavioural skills in both classroom and home settings using everyday information communication technology (ICT) devices such as DVD players, television, desktop or laptop computers.

<sup>309</sup> Case study prepared by Valentina Lemmi, Jackie Damant and Martin Knapp.

<sup>310</sup> Grynszpan, O., Weiss, P.L.T., Perez-Diaz, et al. (2014). Innovative technology-based interventions for autism spectrum disorders: a meta-analysis. *Autism*, 18, 346-361.

<sup>311</sup> [who.int/disabilities/technology/en/](http://who.int/disabilities/technology/en/).

<sup>312</sup> Ganz, J.B., Earles-Vollrath, T.L., Heath, A.K. et al. (2012). A meta-analysis of single case research studies on aided augmentative and alternative communication systems with individuals with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, 42(1), 60-74.

<sup>313</sup> Royal College of Speech and Language Therapists (2013). *Augmentative and Alternative Communication (AAC) support in Scotland: A review of the research literature and cost benefit analyses*. NHS Education for Scotland.

*The Transporters* is an animated DVD series involving eight characters representing different vehicles with human faces, each performing specific physical functions. There are 15 episodes of 5-10 minutes each, portraying 15 key emotions. The programme also includes interactive quizzes to reinforce emotional learning, and a parent user guide. Children are asked to watch a minimum of 15 minutes per day for four weeks. Parents are encouraged to use the guide to help their child complete the quiz, and are asked to keep a daily record of episodes watched.

Four randomised controlled trials (RCT) involving children aged 4-8 years have been conducted. A UK-based three-arm trial (autistic children with intervention, autistic children without intervention, and autistic and non-autistic children without the intervention) demonstrated significant improvements in emotion comprehension and recognition skills for autistic children of average IQ who had watched *The Transporters* compared to the autistic group who did not watch the programmes<sup>314</sup>. An Australian two-arm RCT involving autistic children with below average IQ found improvements in both basic emotion recognition and recognition of expressions of anger, but with limited retention of any acquired

skill at follow-up<sup>315</sup>. Another Australian study found that children improved significantly in terms of social behaviour after watching both *The Transporters* and *Thomas the Tank Engine*, suggesting both formats have value<sup>316</sup>. Finally, an Israeli study of autistic children of average IQ in an RCT confirmed positive effects of *The Transporters*<sup>317</sup>.

*The Junior Detective Training*<sup>318</sup> is another CAI programme which combines a computer game, small group sessions and parent training to teach children about emotion recognition, emotion regulation and social interaction. An RCT involving 7-12-year old children with a diagnosis of Asperger syndrome demonstrated greater improvement in social skills and emotion regulation (e.g. anxiety and anger management) in the intervention group, and this difference was sustained at 6-week and 5-month follow-ups. There were no significant differences in emotion recognition.

*The FaceSay* computer game featuring avatars of real people is designed to teach children specific social skills<sup>319</sup>. Children use a mouse or touch-screen, and appropriate use of the game and technology is rewarded with praise or a small snack. An RCT which separately compared autistic

<sup>314</sup> Golan, O., Ashwin, E., Granader, Y. et al. (2010). Enhancing emotion recognition in children with autism spectrum conditions: An intervention using animated vehicles with real emotional faces. *Journal of Autism and Developmental Disorders*, 40, 269-279.

<sup>315</sup> Williams, B.T., Gray, K.M., Tonge, B.J. (2012). Teaching emotion recognition skills to young children with autism: a randomised controlled trial of an emotion training programme. *Journal of Child Psychology and Psychiatry*, 53, 1268-1276.

<sup>316</sup> Young, R.L., Posselt, M. (2012). Using the transporters DVD as a learning tool for children with autism spectrum disorders (ASD). *Journal of Autism and Developmental Disorders*, 42, 984-991.

<sup>317</sup> Gev, T., Rosenan, R and Golan, O (2016). Unique Effects of The Transporters Animated Series and of Parental Support on Emotion Recognition Skills of Children With ASD: Results of a Randomized Controlled Trial. *Autism Research* doi: 10.1002/aur.1717.

<sup>318</sup> Beaumont, R., Sofronoff, K. (2008). A multi-component social skills intervention for children with Asperger syndrome: The Junior Detective Training Program. *Journal of Child Psychology and Psychiatry*, 49, 743-753.

<sup>319</sup> Hopkins, I.M., Gower, M.W., Perez, T.A. et al. (2011). Avatar assistant: improving social skills in students with an ASD through a computer-based intervention. *Journal of Autism and Developmental Disorders*, 41, 1543-1555.

children who were low-functioning (LFA) or high-functioning (HFA) to matched control groups reported significant improvements in emotion recognition and social interactions in LFA children who used *FaceSay* compared to the LFA control group. HFA children who used *FaceSay* showed significant improvements in emotion recognition, social interactions and facial recognition compared to their own control group.

*Let's Face it!* (LFI!) is a series of seven computer games designed to develop children's face-processing skills such as recognising changes in facial configuration, features and expression. An RCT in the US found significant improvement in analytic and holistic face-processing skills in the group using LFI! compared to the control group<sup>320</sup>.

Other programmes targeting social and emotional development have also demonstrated positive effects on emotion recognition<sup>321,322,323,324</sup>. A review concluded that CAI is effective in teaching children to discern basic emotions and facial expressions as well as more abstract social skills such as verbal irony<sup>325</sup>.

## Academic Skills

*TeachTown: Basics* is a CAI for developing academic skills. It consists of daily 20-minute computer game sessions followed by 20-minute group activity. An RCT conducted with 47 autistic children aged 3-6 years found that those using *TeachTown: Basics* improved in overall measures of language and cognition. The intervention group also showed significant mastery of lessons across four learning domains (receptive language, social understanding, life skills and academic skills). Using the programme for longer led to greater gains, suggesting that CAI is an effective tool for autistic children in a classroom environment<sup>326</sup>. One evidence review concurred: despite limited evidence, CAI is a promising tool for improving academic skills in autistic children<sup>327</sup>. On the other hand, another systematic review concluded that the effects of CAI on literary skills were inconsistent due to variability in autism diagnoses of participants and the wide range of skills taught at the schools<sup>328</sup>.

<sup>320</sup> Tanaka, J.W., Wolf, J.M., Klaiman, C. et al. (2010). Using computerized games to teach face recognition skills to children with autism spectrum disorder: The Let's Face It! program. *Journal of Child Psychology and Psychiatry*, 51, 944-952

<sup>321</sup> Faja, S., Webb, S.J., Jones, E. et al. (2012) The effects of face expertise training on the behavioral performance and brain activity of adults with high-functioning autism spectrum disorders. *Journal of Autism and Developmental Disorders*, 42, 278-293.

<sup>322</sup> Golan, O., Baron-Cohen, S. (2006). Systemizing empathy: Teaching adults with Asperger syndrome or high-functioning autism to recognize complex emotions using interactive multimedia. *Development and Psychopathology*, 18, 591-617.

<sup>323</sup> Ryan, C., Ní Charragáin, C. (2010). Teaching emotion recognition skills to children with autism. *Journal of Autism and Developmental Disorders*, 40, 1505-1511.

<sup>324</sup> Bölte, S., Feineis-Matthews, S., Leber, S. et al. (2002) The development and evaluation of a computer-based program to test and to teach the recognition of facial affect. *International Journal of Circumpolar Health*, 61, 61-68.

<sup>325</sup> Sansosti, F.J., Mizenko, M.L., Krupko, A. (2014). The use of computer-based technologies to increase the academic, behavioral, and social outcomes of students with autism spectrum disorders in schools: considerations for best approaches in educational practice, Chapter 4 in: Sansosti, F.J. et al (editors), *Innovative Technologies to Benefit Children on the Autism Spectrum*. Hershey, PA: IGI Global.

<sup>326</sup> Whalen, C., Moss, D., Ilan, A.B. et al. (2010). Efficacy of TeachTown: Basics computer-assisted intervention for the intensive comprehensive autism program in Los Angeles unified school district. *Autism*, 14, 179-197.

<sup>327</sup> Sansosti et al. (2014) op.cit.

<sup>328</sup> Ramdoss, S., Mulloy, A., Lang, R. et al. (2011). Use of computer-based interventions to improve literacy skills in students with autism spectrum disorders: A systematic review. *Research in Autism Spectrum Disorders*, 5, 1306-1318.

## Behavioural Skills

CAI shows promise for developing behavioural skills, especially functional skills and becoming more independent<sup>329</sup>. Examples include a sticker chart programme that helped reduce tantrums and self-harming behaviours<sup>330</sup>, and a computerised social story programme that decreased pushing behaviour<sup>331</sup>.

## Communication skills

A review of speech-generating devices (SGD) and picture-based systems – the most commonly used AAC for autistic children - found 24 eligible studies, 20 investigating interventions for communication skills, two for effects on social skills, two for academic skills and five for concerning behaviours<sup>332</sup>. Meta-analyses indicated that AAC systems can have positive effects on all skills for autistic people, although the impact of AAC was greater on communication skills than on other skill categories. Another review – of computer-based interventions (CBI) - concluded that the effects are only modestly positive, due to the heterogeneity of autistic participants and to the complexity of the emotional and social skills targeted<sup>333</sup>. This review further suggested that the combination of CBI and tutoring is as effective as face-to-face instruction for autistic children.

## Mobile devices

Growing everyday reliance on mobile ICT devices such as smartphones and tablet computers and the expansion of mobile internet networks (e.g. 3G and 4G connectivity) have greatly encouraged exploration of the use of mobile applications (or apps) as skill-development aids for autistic people.

The *Findme* app is designed to help autistic children to follow social cues and to identify other people's needs<sup>334</sup>. The game is set in a shop where characters point and look at various objects, and progresses in complexity as the social cues become subtler. A UK RCT with 54 children aged under 6 showed that children with varying levels of ability were consistently engaged with the game, and parents also highly rated the app. There were, however, no significant improvements in real-world communication skills.

A *job-coaching app* to support autistic adults in the workplace includes task reminders, task lists, picture prompts, task-sequencing prompts, behavioural self-management adaptations, way-finding tools and a communication tool to contact support workers<sup>335</sup>. Adults who used the app with an Apple iPod Touch needed significantly fewer hours of face-to-face support in the first 24

<sup>329</sup> Sansosti et al. (2014) op.cit.

<sup>330</sup> Soares, D.A., Vannest, K.J., Harrison, J. (2009). Computer aided self-monitoring to increase academic production and reduce self-injurious behavior in a child with autism. *Behavioral Interventions*, 24, 171–183.

<sup>331</sup> Mancil, R.G., Haydon, T., Whitby, P. (2009). Differentiated effects of paper and computer assisted Social Stories™ on inappropriate behaviour in children with autism. *Focus on Autism and Other Developmental Disabilities*, 24, 205–215.

<sup>332</sup> Ganz et al. (2012) op.cit.

<sup>333</sup> Ramdoss, S., Machalicek, W., Rispoli, M. et al. (2012). Computer-based interventions to improve social and emotional skills in individuals with autism spectrum disorders: A systematic review. *Developmental Neurorehabilitation*, 15, 119-135.

<sup>334</sup> Fletcher-Watson, S., Petrou, A., Scott-Barrett, J. et al. (2016). A trial of an iPad™ intervention targeting social communication skills in children with autism. *Autism*, 20, 771-82.

<sup>335</sup> Gentry, T., Kriner, R., Sima, A. et al. (2015). Reducing the need for personal supports among workers with autism using an iPod touch as an assistive technology: delayed randomized control trial. *Journal of Autism and Developmental Disorders*, 45, 669-684.

weeks of employment compared to participants who did not use the app. There was no significant difference in functional job performance between intervention and control groups.

There are a number of other apps for mobile devices that have been developed to support autistic people, but at the time of our review we did not find evaluations of them.

### Augmentative and Alternative Communication

Increasingly, AAC apps on mobile devices are used as communication aids for autistic people. The *AppsforAAC* website<sup>336</sup> currently lists 265 apps, 55 of which are free and 105 of which cost less than £8. One description of available AAC apps identified seven categories: text to speech, which converts text to spoken word; symbols in grid system, where each symbol activates a spoken word or phrase; word predictor, where words and spelling are suggested while typing; phrases, which suggests set phrases or symbols; eye pointing, designed for people who communicate with eye direction; photo story, where users can take a photo and add speech to relay the story; and picture exchange communication apps<sup>337</sup>.

Reviews of AAC on mobile devices report mixed results. One review of communication apps for mobile devices (e.g. tablet computers) and their effects on autistic people included 15 studies, 10 investigating the effects on communication skills and five the impacts on behaviour<sup>338</sup>. Speech-

generating device (SGD) apps were found to have positive effects on communication skills and behaviours, and they were also preferred over other systems such as picture-exchange communication and sign language. Another review also concluded that AAC systems can improve communication skills, but noted that mainstream mobile devices, such as tablets, are not universally accepted as aids for accessing AAC<sup>339</sup>. People with physical limitations reported challenges with using the manual functions and some preferred traditional technology aids, such as Lightwriter<sup>340</sup>, which they found to be more robust and accessible.

Many observers of this area suggest that mobile devices have the potential to surpass other technologies due to their portability, pervasive availability, relative affordability and broad appeal to autistic children and adults, parents and instructors alike<sup>341,342,343</sup>. Experts we spoke to enthused about the future role of tablet computers in autism. One autism researcher and campaigner argued that ‘tablets are immeasurably more flexible and offer more potential than anything gone before: all previous have been comprehensively superseded now. I have a Google news feed on iPads and autism, and hardly a day goes by without an encouraging story [about tablets. They are highly] recommended!’ On the other hand, the design of off-the-shelf (and comparatively cheap) devices may not suit people with physical or sensory disabilities or they may be introduced prematurely before proper assessment of individual needs<sup>344</sup>.

<sup>336</sup> [appsforaac.net/](http://appsforaac.net/), accessed on November 25, 2016.

<sup>337</sup> Bradshaw, J. (2013). The use of augmentative and alternative communication apps for the iPad, iPod and iPhone: an overview of recent developments. *Tizard Learning Disability Review*, 31-37.

<sup>338</sup> Alzrayer, N., Banda, D.R., Koul, R.K. (2014). Use of iPad/iPods with individuals with autism and other developmental disabilities: a meta-analysis of communication interventions. *Review Journal of Autism and Developmental Disorders* 1: 179.

<sup>339</sup> Bradshaw (2013) op.cit.

<sup>340</sup> [www.toby-churchill.com/products/lightwriter-sl40-connect/](http://www.toby-churchill.com/products/lightwriter-sl40-connect/), accessed on November 25, 2016.

<sup>341</sup> Bradshaw (2013) op.cit.

<sup>342</sup> Alzrayer et al. (2014) op.cit.

<sup>343</sup> Fletcher-Watson et al. (2015) op.cit.

<sup>344</sup> Bradshaw (2013) op.cit.

'Traditional' lower-tech aids may more suitable for some people who need help with communication, and often will be cheaper<sup>345,346</sup>.

It has also been suggested that encouraging the extensive use of ICT devices by autistic children could be counterproductive to building the social and emotional awareness skills needed to interact with other people<sup>347</sup>. The NICE guideline also noted that too much exposure to television or a computer screen (e.g. having a screen in the bedroom) could interfere with sleeping patterns<sup>348</sup>.

## Economic case

Evidence on the cost-effectiveness of assistive devices and technologies is sparse and of modest quality. There is no doubt that many interventions cost very little, such as a number of apps: Model Me Going Places is free, Social Skill Builder lite costs \$2.99, and Conversation Social Stories created by Touch Autism costs \$3<sup>349,350</sup>.

One US trial of iPod use in job coaching – summarised earlier - estimated savings of \$379 over 12 weeks and \$2025 over 24 weeks, suggesting that this work-related assistive technology could save money for public vocational rehabilitation services<sup>351</sup>.

The Royal College of Speech and Language Therapists commissioned cost-benefit analyses of augmentative and alternative communication support for children with cerebral palsy and autistic

children. This was a modelling study based on available evidence at the time, with outcomes measured in terms of quality-adjusted life years (QALYs) valued at the threshold value associated with NICE guideline discussions. One calculation was that every £1 invested in high-tech AAC generated QALYs with a monetary value of £1.79. Improved communication skills would be expected to confer benefits in terms of better quality of life for both the autistic person and family members, but the quantification and monetary valuation of benefits in this study relies on several quite strong assumptions. Although we are unsure about the precise estimates generated by this study, particularly the lifetime costs, the sensitivity analyses provide reassurance that there is likely to be an economic case<sup>352</sup>.

## Conclusions

It is in the area of assistive and related technologies where, not surprisingly, development of new interventions is most rapid. Also not surprisingly, evidence from evaluations has not kept up. Consequently, while technologies for autistic people often look promising, evidence from robust evaluations remains quite limited, with a predominance of small design and case studies. Nevertheless, the studies that have been conducted to date report broadly positive effects.

Most of this research has taken place outside the UK, but we would expect most of the effectiveness evidence to be transferable *in*

<sup>345</sup> Iacona, T., Lyon, K., West, D. (2011). Non-electronic communication aids of people with complex communication needs. *International Journal of Speech Language Pathology*, 13, 399-410.

<sup>346</sup> Mirenda, P. (2001). Autism, augmentative communication and assistive technology: what do we really know? *Focus on Autism and Other Developmental Disabilities*, 16, 141-151.

<sup>347</sup> Fletcher-Watson et al. (2015) op.cit.

<sup>348</sup> NICE (2013). CG170s, <https://www.nice.org.uk/guidance/cg170>, accessed on November 25, 2016

<sup>349</sup> Alzahrani, N.S. (2014). Meta-analysis of the effectiveness of using assistive technology to enhance social skills with students diagnosed with autism, graduate thesis, Lamar University-Beaumont, Texas.

<sup>350</sup> AppsforACC

<sup>351</sup> Gentry et al. (2015) op.cit.

<sup>352</sup> Royal College of Speech and Language Therapists (2013) op.cit.

*principle* to UK contexts, although this should always be considered carefully, as we have noted previously. One challenge could be a lack of appropriate infrastructure to support autistic children and adults in the use of new technologies: schools may not be geared up to use some technologies, either because they do not have the hardware or because staff are not suitably trained. The same is likely to be the case in many workplaces. Health and care staff may not have the right skills either.

This infrastructural issue would be particularly pertinent with respect to the transferability of cost-effectiveness findings, although we did not find much such evidence in our search. Implementing some of these technologies would require additional expenditure on staff training and possibly capital investment.

Some of the technologies that are being developed, especially apps on mobile

devices, cost very little. Given that many such interventions appear to be able to improve social, communication, academic or behavioural skills - at least in the short term - then this would potentially affect the support that individuals need. We might therefore expect the economic case to be strong, although as yet there have been few economic evaluations. However, one of the experts we spoke to warned of 'pseudo-efficiencies', where the novelty effect bolsters positive findings.

Understanding the enduring effects of ICT use on skills development requires more long-term research, and there has been little such research to date. Many of the evidence reviews in this area emphasise this same point: we do not know whether autistic people continue to use these assistive and other technologies beyond the short term, and we do not know what, if any, long-term benefits will flow.

# ANTI-STIGMA CAMPAIGNS AND ANTI-BULLYING INTERVENTIONS <sup>353</sup>

## Target population

Autistic children and adults with or without learning disability.

## Context

Limited awareness of autism and negative attitudes towards autistic people can have a number of bad consequences, including social isolation, stigma discrimination, and bullying, each with potential knock-on effects on health, especially mental health, and quality of life.

Public awareness of autism in the UK is high (99.5% of recently surveyed individuals have heard about autism), but 16% of surveyed autistic people and their families think this does not translate into *meaningful understanding* of autism<sup>354</sup>. For example, surveyed families reported that members of the public often get annoyed when their child is having a tantrum or cannot be controlled<sup>355</sup>. Lack of awareness can be a contributor to social isolation. A study of young people receiving special education services in the US found that those who are autistic were more likely to never see friends, never get called by friends, never get invited to activities and be socially isolated when compared to

young people with other needs. Social isolation was greater for those autistic young people with lower conversation ability and poorer functional skills<sup>356</sup>. A cross-sectional study in the US found that loneliness was associated with increased risk of depression and anxiety, lower life satisfaction and poorer self-esteem, even after controlling for symptoms of autism. More and better quality friendships were associated with decreased loneliness<sup>357</sup>.

A report from the National Autistic Society a few years ago found that 63% of young autistic people have experienced bullying at school. The rate was higher in secondary school (75%) and particularly for those with a diagnosis of Asperger's or high-functioning autism (82%)<sup>358</sup>. Bullying can take many forms: direct aggressive behaviour such as physical intimidation and verbal threats, and indirect aggression such as exclusion and rejection<sup>359</sup>. Some non-UK studies have found that young autistic people experience more victimisation than young people with learning disabilities or neurotypical children<sup>360,361</sup>. The situation is complicated: one school-based study in the US, based on parent and teacher reports, found that 46% of autistic adolescents had been the victims of bullying, 15% the perpetrators, and 9% both victims and perpetrators<sup>362</sup>. However, another

<sup>353</sup> Case study prepared by Valentina Lemmi, Martin Knapp, Marija Trachtenberg, Margaret Perkins and Dylan Watts.

<sup>354</sup> National Autistic Society (2016). Too much information. <http://www.autism.org.uk/get-involved/tmi.aspx>.

<sup>355</sup> *ibid.*

<sup>356</sup> Orsmond, G.I., Shattuck, P.T., Cooper, B.P. et al. (2013). Social participation among young adults with an autism spectrum disorder. *Journal of Autism and Developmental Disorders*, 43, 2710–2719.

<sup>357</sup> Mazurek, M.O. (2014). Loneliness, friendship, and well-being in adults with autism spectrum disorders. *Autism*, 18, 223–232.

<sup>358</sup> Bancroft, K., Batten, A., Lambert, S., Madders, T. (2012). *The Way We Are: Autism in 2012*. London: National Autistic Society.

<sup>359</sup> Evans, C.B., Fraser, M., Cotter, K.L. (2014). The effectiveness of school-based bullying prevention programs: A systematic review. *Aggression and Violent Behavior*, 19, 532–544.

<sup>360</sup> Zeedyk, S.M. (2014). Bullying of youth with autism spectrum disorder, intellectual disability, or typical development: victim and parent perspectives. *Research in Autism Spectrum Disorders*, 8, 1173–1183.

<sup>361</sup> Kloosterman, P., Kelley, E., Craig, W. et al. (2013). Types and experiences of bullying in adolescents with an autism spectrum disorder. *Research in Autism Spectrum Disorders*, 7, 824–832.

<sup>362</sup> Sterzing, P., Shattuck, P., Narendorf, S. et al. (2012) Prevalence and correlates of bullying involvement among adolescents with an autism spectrum disorder. *Archives of Pediatrics and Adolescent Medicine*, 166, 1058–1064.

US study describes the difficulties that autistic adolescents attending special schools may have in distinguishing between bullying and non-bullying situations<sup>363</sup>. A recently published systematic review and meta-analysis of school bullying and autistic young people found 17 studies of sufficient quality to be considered. The pooled analyses generated prevalence estimates for perpetration of bullying (10%), victimization (44%) and both (16%)<sup>364</sup>. Compared to their ‘typically developing’ peers, school-aged autistic young people were at significantly greater risk of school victimisation in general, as well as verbal bullying.

Studies of bullying in a general population show clear effects on mental health and emotional wellbeing, self-harm and suicide<sup>365</sup>. The effects on mental health can extend for a very long time: a recent UK study showed that children bullied at ages 7 or 11 had higher levels of depression, anxiety disorders and suicidal ideation at ages 23 and 50; they also had poorer social relationships, more economic hardship, and lower quality of life at age 50<sup>366</sup>. Those who were frequently bullied in childhood were more likely to use mental health services, both in childhood and adolescence and in midlife<sup>367</sup>.

In adulthood, poor awareness of autism might close off employment opportunities.

Through interviews and surveys it appears that autistic people face employer discrimination<sup>368</sup>. Other barriers to employment include lack of understanding about what beneficial supports could be provided in the workplace<sup>369</sup>. As described in the *Employment support* case study, few autistic people are able to find full-time paid employment. The recent NAS survey shows that these difficulties are experienced by autistic people of all ages; for example, 41% of those aged 55 years or older had not worked for 10 or more years<sup>370</sup>. Fewer than half of all autistic people surveyed believe they will ever be employed<sup>371</sup>, and 60% of people are financially reliant on their families<sup>372</sup>.

Stigma is at the root of many of these difficulties. In fact, stigma affects all aspects of an autistic individual’s life, including their public or social life, experiences at school and work. Feeling stigmatised is associated with psychological distress. A study of people with learning disabilities found that, among those reporting feeling stigmatized, there was a positive association with psychological distress, a negative association with quality of life, and increased use of services, including contacts with the police<sup>373</sup>. The same study found that feeling stigmatised was associated with unwillingness to attend some services, which would then potentially damage their longer-term health. As the Westminster

<sup>363</sup> van Roekel, E., Scholte, R., Didden R. (2010). Bullying among adolescents with autism spectrum disorders: prevalence and perception. *Journal of Autism and Developmental Disorders*, 40, 63–73.

<sup>364</sup> Maïano, C., Normand, C.L., Salvas M.C. et al. (2016). Prevalence of school bullying among youth with autism spectrum disorders: a systematic review and meta-analysis. *Autism Research*, 9, 601-615.

<sup>365</sup> van Geel, M., Vedder, P., Tanilon, J. (2014). Relationship between peer victimization, cyberbullying, and suicide in children and adolescents: a meta-analysis. *JAMA Pediatrics*, 168, 435-42.

<sup>366</sup> Takizawa, R., Maughan, B. & Arseneault, L. (2014). Adult health outcomes of childhood bullying victimization: evidence from a five-decade longitudinal British birth cohort. *American Journal of Psychiatry*, 171, 777-784.

<sup>367</sup> Evans-Lacko, S., Takizawa, R., Brimblecombe, N. et al. (2016) Childhood bullying victimisation is associated with use of mental health services over five decades: a longitudinal nationally representative cohort study. *Psychological Medicine*, published online.

<sup>368</sup> Forsythe, L., Rahim, N., Bell, L. (2008). Benefits and employment support schemes to meet the needs of people with an autistic spectrum disorder. London: National Audit Office.

<sup>369</sup> Baldwin, S., Costley, D., Warren, A. (2014). Employment activities and experiences of adults with high-functioning autism and Asperger’s disorder. *Journal of Autism and Developmental Disorders*, 44, 2440-2449.

<sup>370</sup> National Autistic Society (2016) op.cit.

<sup>371</sup> Bancroft, K., Batten, A., Lambert, S and Madders, T. (2012) *The Way We Are: Autism in 2012*. London: National Autistic Society.

<sup>372</sup> Rosenblatt, M. (2008) *I Exist: The message from adults with autism in England*. London: National Autistic Society.

<sup>373</sup> Ali, A., King, M., Strydom, A., Hassiotis, A. (2015). Self-reported stigma and symptoms of anxiety and depression in people with intellectual disabilities: findings from a cross sectional study in England. *Journal of Affective Disorders*, 187, 224-231.

Commission found from their own recent survey, use of healthcare services was sometimes hindered by the feeling that healthcare professionals ‘rarely’ or ‘never’ understand autism and how it affects their physical and mental health<sup>374</sup>.

Stigma can also affect family members and other carers. In the UK, families whose children have any type of disability feel socially isolated<sup>375</sup>, leading to poor mental health, including anxiety and depression, and high use of GP services, counselling and medication. In this sample, 21 % reported that social isolation caused substantial difficulties with family life. Half the sample felt that discrimination and stigma contributed to their social isolation. Autism-specific evidence comes mainly from outside the UK, and differences in cultural context mean that the findings might not be transferable. In Hong Kong, stigma was associated with poorer caregiver psychological wellbeing<sup>376</sup>. Interviews with parents in an Australian study found that experienced stigma was greater for parents of children described as aggressive compared to those who are more passive<sup>377</sup>. Feelings of stigma were higher for Israeli parents of autistic children compared to parents of children with learning or physical disabilities<sup>378</sup>.

## Interventions

Awareness campaigns aim to tackle

misconceptions, increasing public understanding of autism. Anti-stigma campaigns aim to tackle stigma, improving public knowledge, attitudes and behaviour of the public towards autistic people, and thereby reduce discrimination. Those campaigns may include social marketing (billboards, radio and television commercials, social media), public education of key population groups, and public relations efforts with key stakeholders. Campaigns may target the general public or specific groups of the population, such as school pupils, healthcare professionals or the police. They may operate at national or local level, such as schools, healthcare facilities and workplaces. Anti-bullying interventions can take a number of forms, including education on bullying in schools and online and telephone support (e.g. Child Line)<sup>379</sup>.

There are a number of initiatives being taken to tackle bullying<sup>380</sup>, but we are not aware that these important efforts have yet been evaluated.

## Effectiveness

The United Nations chose 2nd April as *World Autism Awareness Day*<sup>381</sup> in 2007, and in some countries (including the UK), a week at around that date has been designated *World Autism Awareness Week*<sup>382</sup>. Some years earlier – in the 1970s - April was designated National Autism Awareness Month in the US. A small study based

<sup>374</sup> Westminster Commission on Autism (2016). *A Spectrum of Obstacles: An Inquiry into Access to Healthcare for Autistic People*. London: National Children's Group.

<sup>375</sup> Contact a Family (2011). *Forgotten families: the impact of isolation on families with disabled children across the UK*. York: Contact a Family.

<sup>376</sup> Mak, W.W., Cheung, R.Y. (2008). Affiliate stigma among caregivers of people with intellectual disability or mental illness. *Journal of Applied Research in Intellectual Disabilities*, 21, 532-545.

<sup>377</sup> Gray, D. E. (2002). 'Everybody just freezes. Everybody is just embarrassed': Felt and enacted stigma among parents of children with high-functioning autism. *Sociology of Health & Illness*, 24, 734-749.

<sup>378</sup> Werner, S., Shulman, C. (2015). Does type of disability make a difference in affiliate stigma among family caregivers of individuals with autism, intellectual disability or physical disability? *Journal of Intellectual Disability Research*, 59, 272-283.

<sup>379</sup> Cantone, E., Piras, A.P., Vellante, M. et al. (2015). Interventions on bullying and cyberbullying in schools: a systematic review. *Clinical Practice and Epidemiology in Mental Health*, 11, 58-76.

<sup>380</sup> For example, see <https://www.ambitiousaboutautism.org.uk/understanding-autism/young-people-with-autism-make-a-nose-about-bullying-through-myvoice>; <http://www.autismtogether.co.uk/>; and <http://www.modelmekids.com/bully.html>.

<sup>381</sup> [un.org/en/ga/search/view\\_doc.asp?symbol=A/RES/62/139](http://un.org/en/ga/search/view_doc.asp?symbol=A/RES/62/139).

<sup>382</sup> [autism.org.uk/get-involved/world-autism-awareness-week.aspx](http://autism.org.uk/get-involved/world-autism-awareness-week.aspx).

on Google Trend data concluded that online search interest in autism responded positively to *National Autism Awareness Month* and televised reports on autism between 2004 and 2014<sup>383</sup>.

The Ad Council, a leading producer of public service communications in the US, launched *Autism Awareness* in 2006 as a national campaign aiming to increase knowledge about autism, including advertisements in print, radio, television, internet and social media<sup>384</sup>. Another American campaign is *Learn the signs: act early (LSAE)*; it targets parents, healthcare professionals, teachers and childcare providers to improve early identification and support for autistic children and those with other developmental disorders<sup>385</sup>. Based on formative research and behaviour change theory, LSAE comprises a social marketing campaign with many online and other facets and a 24/7 live call centre. A non-controlled before-after study three years after its launch showed the positive impact of LSAE in improving parents' knowledge of early symptoms and early support for autistic children, and improving healthcare professionals' attitudes and intended behaviours towards education of parents and referral to early support<sup>386</sup>.

In the UK, both the National Autistic Society and the Autism Alliance have launched national awareness campaigns. *Too Much Information (TMI)*, launched by the NAS in 2016, aims to increase knowledge of autism in the general population, students and teachers, and employees and employers<sup>387</sup>. It has a social marketing

component and public education components for students and teachers during Schools Autism Awareness Week, and for employees and employers through the Access Award, which promotes autism-friendly venues. TMI is currently being evaluated and recent figures suggest that it is having an impact on awareness of the difficulties that autistic people face in public spaces.

The Autism Alliance launched *Connect to Autism* in England in 2015 to improve knowledge and attitudes in settings such as public transport, shops and leisure centres<sup>388</sup>. It has four components: an *autism charter* that can be signed by organisations committing to receive free training and to become autism-friendly; a *champion network* of individuals with a national profile committing to increase awareness of autism through their networks and participation; *national chains* committing to identify six pilot venues to sign the Autism Charter; and local activities in eight pilot areas across the UK to commit local communities and venues to include autistic people. In Northern Ireland, the IMPACT Award was created with similar aims<sup>389</sup>.

There have been a number of local initiatives. The *Psychoeducation group for autism spectrum understanding and support (PEGASUS)* for autistic young people (aged 9-14) and their parents was found to improve knowledge of autism and awareness of their unique strengths and difficulties<sup>390</sup>. An anti-stigma intervention for a general school population in the US improved knowledge and attitudes about autism, but had

<sup>383</sup> DeVilbiss, E.A., Lee, B.K. (2014). Brief report: trends in US National autism awareness from 2004 to 2014: the impact of national autism awareness month. *Journal of Autism and Developmental Disorders*, 44, 3271-3273.

<sup>384</sup> [adCouncil.org/Our-Campaigns/Health/Autism-Awareness](http://adCouncil.org/Our-Campaigns/Health/Autism-Awareness).

<sup>385</sup> [cdc.gov/ncbddd/actearly/](http://cdc.gov/ncbddd/actearly/).

<sup>386</sup> Daniel, K. L., Prue, C., Taylor, M. K. et al. (2009). 'Learn the signs. Act early': A campaign to help every child reach his or her full potential. *Public Health*, 123, e11-e16.

<sup>387</sup> [autism.org.uk/get-involved/tmi.aspx](http://autism.org.uk/get-involved/tmi.aspx).

<sup>388</sup> [autism-alliance.org.uk/Connect-to-Autism](http://autism-alliance.org.uk/Connect-to-Autism).

<sup>389</sup> [autismni.org/impact-award.html](http://autismni.org/impact-award.html).

<sup>390</sup> Gordon, K., Murin, M., Baykaner, O. et al. (2015). A randomised controlled trial of PEGASUS, a psychoeducational programme for young people with high-functioning autism spectrum disorder. *Journal of Child Psychology and Psychiatry*, 56, 468-476.

less effect on behavioural intentions towards their autistic peers<sup>391,392</sup>. Other US evaluations have shown positive benefits of anti-stigma training for college students<sup>393</sup> and first responders in emergencies<sup>394</sup>. In Northern Ireland, a short training course on autism for youth leaders changed attitudes and perceptions, in a context where community groups such as scouts, sports organisations and youth clubs seemed reluctant to enrol autistic young people<sup>395</sup>.

While evidence specifically focused on attitudes towards autism is gradually accumulating, there is more plentiful evidence for anti-stigma efforts in the mental health field<sup>396</sup>. Whilst not directly relevant perhaps, there still may be useful lessons to draw.

The same sentiment applies to anti-bullying interventions. In the general context, there have been many studies, and some helpful systematic reviews<sup>397,398,399</sup>. To what extent the findings from those broader studies would apply to autistic people is unclear.

## Economic case

Stigma, poor awareness and bullying can have adverse economic consequences. The effects on mental health can lead to poorer academic results and employment prospects, with impacts on national productivity. There can be higher use of health and social care services and more contacts with police. It might therefore be expected that successful interventions that raise awareness, reduce stigma and discrimination or prevent bullying would have the potential to be cost-effective. However, we have not found any autism-specific studies.

There has been research in the wider context that can be mentioned, and it may be that findings from these broader studies have some relevance in the autism field. In relation to mental health problems, for example, anti-stigma campaigns have been found to be potentially cost-effective and low-cost. An economic modelling study of the *Time to Change* campaign in England suggested that it cost relatively small amounts to change intended behaviour, knowledge and attitudes<sup>400</sup>. Similar results were previously found for the *See Me* campaign in Scotland<sup>401</sup>.

<sup>391</sup> Staniland, J.J., Byrne, M.K. (2013). The effects of a multi-component higher-functioning autism anti-stigma program on adolescent boys. *Journal of Autism and Developmental Disorders*, 43, 2816-2829.

<sup>392</sup> Ranson, N.J., Byrne, M.K. (2014). Promoting peer acceptance of females with higher-functioning autism in a mainstream education setting: A replication and extension of the effects of an autism anti-stigma program. *Journal of Autism and Developmental Disorders*, 44, 2778-2796.

<sup>393</sup> Gillespie-Lynch, K., Brooks, P.J., Someki, F. et al. (2015). Changing college students' conceptions of autism: An online training to increase knowledge and decrease stigma. *Journal of Autism and Developmental Disorders*, 45, 2553-2566.

<sup>394</sup> Kelly, E., Hassett-Walker, C. (2015). The training of New Jersey emergency service first responders in autism awareness. *Police Practice and Research*, 1-12.

<sup>395</sup> McConkey, R., Mullan, A., Addis, J. (2011) Promoting the social inclusion of children with autism spectrum disorders in community groups. *Early Child Development and Care*, 182, 827-835.

<sup>396</sup> Mehta, N., Clement, S., Marcus, E. et al. (2015). Evidence for effective interventions to reduce mental health-related stigma and discrimination in the medium and long term: systematic review. *British Journal of Psychiatry*, 207, 377-384.

<sup>397</sup> Ttofi, M.M., Farrington, D.P. (2012). Risk and protective factors, longitudinal research, and bullying prevention. *New Directions for Youth Development*, 133, 85-98.

<sup>398</sup> Evans et al. (2014) op.cit.

<sup>399</sup> Cantone et al. (2015) op.cit.

<sup>400</sup> Evans-Lacko, S., Henderson, C., Thornicroft, G., McCrone, P. (2013). Economic evaluation of the anti-stigma social marketing campaign in England 2009-2011. *British Journal of Psychiatry*, 202, s95-s101.

<sup>401</sup> McCrone, P., Knapp, M., Henri, M., McDaid, D. (2010). The economic impact of initiatives to reduce stigma: demonstration of a modelling approach. *Epidemiologia e Psichiatria Sociale*, 19, 131-139.

In relation to anti-bullying interventions, there is evidence from a small number of modelling studies that school-based programmes can be both effective and cost-effective, even when they only measure short term impacts<sup>402,403,404</sup>. We would anticipate that these findings would be generalisable at least to some extent to the autism context.

## Conclusions

Low levels of awareness of autism and what it can mean, negative attitudes and stigma, and aggressive or passive bullying can all have serious consequences for autistic people and their families, both in the short term and over many years.

Studies from a few countries have shown that there are interventions that can begin to tackle these pervasive challenges, although most such studies are relatively weak in terms of design, sample size and length of follow-up. The evidence base for policy or practice efforts in this area therefore remains modest. Moreover, some of the better studies tend to look at broader anti-stigma and anti-bullying interventions – i.e. not focused solely on the experiences of autistic people – and so we must remain hesitant about how readily their findings can be transferred to the autism context.

When looking for economic evidence, we were only able to find studies that looked at broader anti-stigma and anti-bullying interventions, with nothing specifically relating to autism.

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<sup>402</sup> Knapp, M., McDaid, D., Parsonage, M. (2011) *The Economic Case for Mental Health Promotion and Mental Illness Prevention*. London: Department of Health.

<sup>403</sup> Beckman, L., Svensson, M. (2015). The cost-effectiveness of the Olweus Bullying Prevention Program: results from a modelling study. *Journal of Adolescence*, 45, 127-137.

<sup>404</sup> McDaid, D. et al. (2016) *Commissioning Cost-Effective Prevention Services for Mental Health and Wellbeing*. London: Public Health England.

# PHARMACOLOGICAL INTERVENTIONS<sup>405</sup>

The potential role of pharmacological interventions in autistic people has been explored in numerous clinical reports and clinical trials. The targets of individual interventions have focussed largely on either core symptoms of autism or the improvement of common co-occurring mental health problems.

Pharmacological interventions for core symptoms among children and young people were reviewed by NICE in 2013<sup>406</sup>. To recommend an intervention, NICE required favourable evidence from two or more randomised controlled trials (RCTs) that were double-blind, placebo-controlled and included at least 20 participants per treatment arm. On this basis, no pharmacological interventions for core symptoms were recommended. Furthermore, specific injunctions were given *against* the use of secretin, and chelation or hyperbaric oxygen therapy, for which evidence of harm was identified.

There are reports from a number of clinical trials suggesting possible benefits for a range of other pharmacological interventions but none have reached the evidence standard required by NICE. For example, early RCTs with small sample sizes suggested possible benefit of the selective serotonin reuptake inhibitors (SSRIs) for restricted and repetitive behaviours in autistic people, but a subsequent, carefully controlled and large RCT failed to find any evidence of change compared to

placebo<sup>407</sup>. Over the next decade, particularly as the genetic mechanisms underpinning autism are elucidated, novel and repurposed molecules will continue to be tested to evaluate their effect on core symptoms and it will be important to develop reliable, sensitive and meaningful measures of change.

Up to three-quarters of autistic people experience additional mental health problems. Amongst children and young people, the most common are: anxiety, attention deficit hyperactivity disorder (ADHD) and irritable/noncompliant behaviour (concerning behaviour). Amongst autistic adults, depression and anxiety may be the most common. The reasons for these high rates of co-occurring mental health problems are not well understood and it may be that the underlying biological mechanisms are different in the autistic population, which could require different interventions. Therefore, it is necessary that pharmacological interventions are independently trialled amongst autistic people. While SSRIs have proven benefit for anxiety disorders amongst neurotypical youth and adults, there are at present no RCTs evaluating their efficacy for anxiety disorders amongst the autistic population.

With respect to ADHD, there is RCT evidence for the benefits of methylphenidate<sup>408</sup>, atomoxetine<sup>409</sup> and guanfacine<sup>410</sup> amongst autistic youths, although the latter two drugs have only been

<sup>405</sup> Case study prepared by Emily Simonoff.

<sup>406</sup> National Collaborating Centre for Mental Health, (2013). The management and support of children and young people on the autism spectrum. <http://www.nice.org.uk/nicemedia/live/13297/63401/63401.pdf>.

<sup>407</sup> Williams, K., Wheeler, D. M., Silove, N., Hazell, P. (2004). Selective serotonin reuptake inhibitors (SSRIs) for autism spectrum disorders (ASD). Cochrane Database of Systematic Reviews, 14(1), CD004677.

<sup>408</sup> RUPP Autism Network. (2005). Randomized, controlled, crossover trial of methylphenidate in pervasive developmental disorders with hyperactivity. *Archives of General Psychiatry*, 62, 1266-1274.

<sup>409</sup> Harfterkamp, M., van de Loo-Neus, G. et al. (2012). A randomized double-blind study of atomoxetine versus placebo for attention-deficit/hyperactivity disorder symptoms in children with autism spectrum disorder. *Journal of the American Academy of Child & Adolescent Psychiatry*, 51, 733-741.

<sup>410</sup> Scahill, L., McCracken, J. et al. (2015). Extended-release guanfacine for hyperactivity in children with autism spectrum disorder. *American Journal of Psychiatry*, 172, 1197-1206.

evaluated in one double-blind RCT each. Effect sizes are variable, ranging from 0.3 to 1.0, and significant for all three treatments, but there is insufficient evidence to make comparisons among the three drugs.

There is substantial RCT evidence that atypical antipsychotic medication, including risperidone<sup>411,412</sup> and aripiprazole<sup>413</sup>, reduces severe concerning behaviour in autistic young people. A comparison of risperidone alone and its use combined with a parent training intervention showed that the combination intervention led to greater behavioural improvement with borderline significant lower doses of medication<sup>414</sup>. The atypical antipsychotics can have significant adverse effects, including weight gain, glucose intolerance and high prolactin levels, and therefore should be used with caution and careful monitoring. NICE recommended that concerning behaviour should initially be addressed with a psychological intervention, and atypical antipsychotics should be considered only when a behavioural intervention does not lead to sufficient improvement or where the behaviour is too severe to initiate a psychological intervention.

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<sup>411</sup> McCracken, J.T., McGough, J. et al. (2002). Risperidone in children with autism and serious behavioral problems. *New England Journal of Medicine*, 347, 314-321.

<sup>412</sup> RUPP Autism Network. (2002). Risperidone in children with autism and serious behavioral problems. *New England Journal of Medicine*, 347, 314-321.

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# APPENDIX C:

## National workshops

Devolution in the UK has led to approaches to autism provision in each of the four nations which may have similar aims, but have different processes and legislative frameworks. Here we summarise the policy landscape in each nation and some of the main points related to our recommendations that emerged from the workshops. Fuller versions of these analyses with appropriate references will be published on the NAP website ([www.nationalautismproject.org](http://www.nationalautismproject.org)).

### England

#### Background

The statutory guidance under the Autism Act (2009) sets out what local authorities and the NHS have to do to improve services for autistic adults. These include a diagnostic pathway in every area, training for key professionals, local plans and a local autism lead. Local implementation of the strategy is monitored through self-assessment while at high level the strategy is overseen by a cross-departmental Adult Autism Programme Board, led by the Department of Health.

NICE Guidelines set out local pathways to diagnosis and the types of support that should be available post-diagnosis. Autistic children with special educational needs (SEN) are entitled to an assessment to find out what they or their family might need from children's social services, but this does not automatically entitle them to provision. Children may receive an Education, Health and Care plan (EHC Plan) that looks at their needs across education, healthcare and social services. In education and health there are specific statutory duties to make the provision available, but in social care, there is no equivalent duty. The Department for Education recently committed to including

autism in Initial Teacher Training while training of existing teachers is through the Autism Education Trust. As yet only 25% of the teaching population have completed this.

While the organisational aspects of the strategy are largely in place, there are gaps in training, data collection, diagnosis waiting times and post-diagnostic support. Despite the implementation of the Children and Families Act, there are still delays in getting the right support for children in school. Based on prevalence it has been estimated that only around 75% of autistic children are recognised by the education system and that a high rate of exclusion of autistic children persists.

The existence of autism legislation at a time when budgets at local level are under increasing pressure has ensured a focus on autism that would not have been possible without the Act. It has been essential to getting autism moved up the agenda and to ensure the development of local services. Even so, local implementation and change has been patchy – as evidenced by the most recent Public Health England survey of local authorities and their partners - and the wider pressures on social care budgets are a significant challenge.

There are opportunities in the next few years to improve outcomes for autistic children and adults through work that the Department of Health and NHS England is doing on diagnosis, the Mandate to NHS England, Transforming Care and the employment Green Paper.

#### Recommendations: present policy and new initiatives

The National Autistic Society is campaigning for more consistent identification and diagnosis in

children especially considering that the average age of diagnosis (Recommendation 1, R1) has been unchanged at 5 years for the previous decade despite greater awareness and better training. NICE guidance on adult diagnosis is hampered by the lack of good adult diagnostic tools and data on waiting times. The Westminster Commission has made a number of recommendations on this topic.

The evidence base for interventions (R2) is weak but efforts are underway to address some of the deficiencies. Autistica is mapping current research spending against the James Lind Alliance priorities and the Economic and Social Research Council is continuing its Shaping Autism Research programme. Cost-effectiveness though is an area of need - even early intervention has not been assessed for cost reduction. The Department of Health is funding work to look at costs and outcomes for autistic people without learning disabilities but cost-effectiveness is for the future. The Transforming Care programme (see R5) may be assessed for economic impact.

Barriers to access (R4) exist at all life-stages and can be summed up by lack of training, lack of data and lack of communication. In education, the plethora of formal and informal sources of information is a hindrance to getting the right support. Healthcare access is hindered by the lack of an autism flag on medical records. However, the government's commitment to halve the disability employment gap is an exploitable opportunity and the charities are using this to promote employment initiatives.

Autistic people are more likely to be the victims of crime (especially financial abuse) than any other disability group (R6). The Time to Change initiative is aimed at discrimination and stigmatisation of people with mental health issues and the National Autistic Society's Too Much Information campaign is improving public understanding of autism. Death by indifference has been highlighted by recent events at Southern Health NHS Trust.

Peer support and mentoring are likely to be key in helping better transitions (R7). It is unfortunate that educational support continues to age 25 but mental health services transition from child to adult support at age 19. Employment support is very localised but the Department of Work and Pensions is consulting on support for disabled people.

Better coordination across sectors (R8) should result from the use of education, health and care plans for those children in need of specialist support but analysis of the impact of EHCs is still awaited. For adults, Sustainability and Transformation Plans (STPs) are intended to bring health and care together at a local level under NHS leadership but implementation has been patchy.

Very few data are collected on autism and the needs and outcomes of autistic people (R9).

Some of the research issues raised at the workshop have been included in the section on Research Recommendations.

## Northern Ireland

### Background

The Northern Ireland Autism Strategy (2013-20) and Autism Action Plan (2013-16) are underpinned by the Autism Act (Northern Ireland) 2011 which protects autistic people from discrimination by public services and mandates the implementation of a cross-departmental strategy for autism that is lifelong. Responsibility lies with the Department of Health but this all-age strategy has a legislative requirement for all departments to work together according to 11 themes: awareness, accessibility, children, young people and family, health and well-being, education, transitions, employability, independence, choice and control, access to justice, being part of the community, participation and active citizenship. An Autism Strategy Research and Advisory Committee (NIASRAC) assists and informs departments with research and best practice findings and the University of Ulster will add to the research focus with the launch in 2017 of the Autism Research Hub.

The restructuring of the Health and Social Care sector in Northern Ireland following the Bengoa report (announced in October 2016) will reshape and revise the delivery of services on the basis of population needs rather than with the aim of maintaining services which are not sustainable in the long term.

The strengths of the autism strategy in Northern Ireland lie in its roots and ownership, with community action and lobbying underpinning well-

thought out legislation, good cross-department responsibility and the introduction of a research committee to advise and inform on evidence and practice. A self-assessment in September 2015 identified that progress had been made in some areas of the strategy including training, advice, access to travel, employment and streamlining diagnosis. However, its critical weakness lies in the lack of a ring-fenced budget for autism. This results in an inability to scrutinise and evaluate cost-benefits and outcomes for the autistic community and to fully address the requirements of the legislation.

### Recommendations: present policy and new initiatives

Currently, diagnosis (R1) is seen as the gateway to support but demand for assessment of children is outstripping the capacity of the system. In the new post-Bengoa framework, which could take 2-3 years to be implemented, the focus will be on needs assessment by a multidisciplinary team and diagnosis will follow for those assessed as having special needs such as autism. In principle, the idea of basing provision on needs first rather than on the diagnostic label is attractive, but will require some adjustment such as retraining of professionals. Furthermore, the Health and Social Care Trusts will need to be held accountable for implementation and the 2-3 year wait for this to take place will put huge strain on existing arrangements.

The Centre for Autism in Middletown is a cross-border agency that promotes best practice

with a particular emphasis on practice-based evidence (R2). It publishes research bulletins and conducts its own research. The Autism Strategy Research Advisory Committee (NIASRAC) also has a role in promoting use of evidence-supported interventions. However, no economic analyses of interventions (R3) have been completed.

Through the efforts of Autism NI in particular, there have been a number of initiatives aimed at helping autistic people access services (R4), and providing training to alter attitudes such as its IMPACT Awards. As with improving access, it has fallen to the voluntary sector to introduce schemes like IMPACT which require the neurotypical to take responsibility for improving the environment (R5) for autistic people, for example, at work.

Mencap NI coordinates the efforts on fighting discrimination (R6), including hate crime, but this is of course not specific for autism nor particularly relevant for those autistic people without a learning disability. Otherwise, the voluntary sector efforts such as those described above are relevant.

The new post-Bengoa framework will operate across the Health, Communities and Education departments and should deal better with some of the transitions important for autistic people (R7). There is certainly provision in NI for coordination between sectors to be effective (R8).

The Autism Act (NI) 2011 mandates the delivery of a cross-departmental Autism Strategy and Action Plan. The implementation panel is made up from representatives from all government departments and the NIASRAC members come from a broad selection of stakeholders. The Children's Services

Cooperation Act (NI) 2015 mandates cross-departmental cooperation on children's services.

There is provision in place for gathering data on autism (R9). The Department of Health in 2012 held pre-consultation engagement workshops across NI capturing very rich data which has not been fully exploited. Furthermore, the Autism Act (NI) 2011 mandates the five Health and Social Care Trusts to gather data on autism to share across Trusts and to inform the NI Strategy for autism. However, present information systems are not adequate to capture autism data reliably even on incidence, but the new framework will be underpinned by a new information strategy which should help to address the problem.

## Scotland

### Background

The Scottish Strategy for Autism is a ten-year plan launched in 2011 by the Scottish Government as an alternative to the Autism (Scotland) Bill following its failure to pass through parliament. The Strategy made 26 recommendations for delivery of services to the autistic community, responsibility for which lies with the Directorate of Population Health in collaboration with the Learning Disabilities Policy Team and other government departments. In 2013 the Government produced the Menu of Interventions which provided information on the challenges faced by autistic people and the types of services that might address these issues.

A priority action plan was subsequently developed that looked at gaps in the strategy and progress since the launch. The priorities were reflected

in an Autism Outcomes Framework based on four outcomes - a healthy life, independence, choice and control, and active citizenship - which then became the Scottish Government's implementation plan for 2015-17. This plan identified previous shortcomings that will be familiar to those in other parts of the UK: barriers for access to services, to community facilities, to education, employment and social activities; the need for better understanding and knowledge of autism among professionals; better transition from school to adult life.

Legislation to implement health and social care integration came into force on 1 April 2016.

This brings together NHS and local council care services under one partnership arrangement for each of 31 areas. It is hoped that this will have a positive effect on delivery of support services for autism. In addition, the Scotland Act 2016 devolved new powers to the Scottish Government for employment support which will result in new services for disabled people and long term health conditions in 2017.

### **Recommendations: present policy and new initiatives**

The Autism ACHIEVE Alliance (AAA) is a multi-disciplinary collaboration to investigate and improve waiting times in the diagnosis of autism (R1). Most services for children work well but slowly, while adult services are more variable - many people throughout Scotland who do not have a co-occurring condition may have lengthy delays to a diagnostic assessment. Even so, the National Diagnosis Assessment Service from Scottish Autism finds that large numbers of

young adults are coming for assessment, mostly high-functioning. The implication is that timely identification and diagnosis is still problematic for this group.

The Menu of Interventions (R2), recommended by the Scottish Autism Strategy, provides guidance for 14 categories of 'ASD Challenge' to define the types of interventions to be considered, the service provider, the referral path and the desired outcomes. The National Training Framework for autism has been developed for all sectors of the workforce and could address several of our recommendations.

In 2014 Scottish Autism established The Centre for Practice Innovation to promote research minded approaches to practice and to encourage the academic community to support the concept of practice-based evidence through the development of new methodologies.

In education, there is a presumption of inclusion in mainstream schools, and through the Autism Toolbox, the government provides a resource to support autistic children in mainstream schools. The Right Click Programme from Scottish Autism is another example of national activity to support informed interventions: it offers advice and support for parents and carers, particularly in the period following a diagnosis, and has recently been extended to provide a dedicated resource for women. In 2017, a series of St Clements' Practical Autism videos offering advice for parents, teachers and support workers will be promoted by the Scottish government.

The most promising basis for evaluating the

economic case (R3) is the Microsegmentation Study funded by the Scottish government whose aim is to identify the escapable costs of autism, that is, those which would not be incurred with early appropriate intervention. While this work will not be able to provide definitive cost-benefit analysis of interventions, it will identify where the costs are and what targets would offer the best chance of success.

Healthcare access for autistic people is an issue (R4). There is specific training in autism for GPs in Scotland, but there are no data on uptake at present. However, the Royal College of GPs has identified autism as a clinical priority. In education, many autistic children do well in mainstream, but the presumption of inclusion may not be the best solution for some. Special school provision is severely limited in the numbers that can be admitted compared with the potential need.

In 2017, the government will set up its own employment schemes to help the unemployed and the disabled, an opportunity to address barriers to access in this sector. This is not without its downsides - increasing opportunities for employment may have to deal with the difficulty of terminating the employment of the disabled under current employment and disability legislation. Employment may increase stress on the home life of autistic people and attitudes of employment services as to the capabilities of people with learning difficulties need correcting.

Key transitions (R7) such as from school to further education or employment would work better if the current mechanisms were applied more fully. The Children and Young People (Scotland) Act legislates

for a Named Person or State Guardian to advise parents and carers and help them access services. This service may well help with transitions, although there is the view that this top-down policy undermines the parent, and the money might be better spent on the working partnerships between local authorities and families already in place under the Get It Right For Every Child (GIRFEC) policy. The future of this initiative is in doubt following a Supreme Court ruling that it is in conflict with the right to private and family life.

The integration of health and social care should or could address some of the cross-sector issues (R8) but implementation at local level will be the issue. In education the GIRFEC principle should drive better coordination.

Scotland has a number of systems for gathering, holding and managing information (R9). The Information Services Division is part of NHS Scotland and holds health administrative datasets on many areas of health including mental health. The Scottish Learning Disabilities Observatory holds data on both learning disability and autism, including information on prevalence, health, education, housing and employment. The Farr Institute and the Public Health Observatory are other data systems available in Scotland. There is need for a proper national database to maximise use of collected data.

## Wales

### Background

In 2008 Wales became the first country to implement a national autism strategy. Since

then over £15m of ring-fenced funding has been allocated to support the resulting action plan which underwent a 'refresh' in 2013/14. Almost all this funding has been used to support public sector initiatives focused on capacity-building and coordination at national, regional and local levels, assessment and diagnostic facilities, advice and information. A notable additional achievement was the fundraising effort by the charitable sector to establish the Wales Autism Research Centre in Cardiff University and the appointment of the UK's first named professorial chair in autism as its Director in 2010. In 2015 an interim enhancement to the autism action plan was announced, addressing better assessment and diagnostic services for children, and a new all-Wales National Integrated Autism Service was launched in 2016 and will be rolled out over the next 3 years.

The strengths of the Welsh Government's ASD Strategic Action Plan derive from its community-based roots and its national engagement. The commitment to ring-fenced funding has been sustained from 2008-16 and there have been significant developments such as the appointment of autism leads in all local authorities, and information and training materials developed on a wide range of topics. However, a 'whole nation' approach which includes the third sector as key partners, and an objective examination of outcomes and cost-benefits from delivery of the plan, is not in place. The Welsh government rejected calls for an Autism Act, believing that other current developments will support autistic people effectively. However, without such legislation, government initiatives lack statutory force resulting in an inability to require local authorities to implement the strategy to the full.

### **Recommendations: present policy and new initiatives**

Service provision is varied and in some areas a diagnosis (R1) is needed to unlock services. Despite the new Care and Support (Wales) Act 2015, there is still assessment of needs based on the presence or absence of learning disability which means that people are not offered needs assessments appropriately, and personal independence payment assessments are often refused if behavioural traits are not evident during interview.

In parallel with the new integrated autism service, the Together for Children and Young People initiative contains a neurodevelopmental work stream which draws health boards together and creates a new pathway for autistic children. Built into this is a data-capture mechanism within the Wales Community Care Information System (WCCIS) that will track interventions and their outcomes (R2).

In education, the presumption is that children will go into mainstream schools with some specialist provision in units or otherwise in dedicated schools. Choice is parent-driven and there are many home-schooled children. The number of exclusions of autistic children and those with learning disability is rising whilst other exclusion rates are falling.

Information on adult intervention is very thin. Decisions about interventions vary across the country and while there is a huge need to understand what autistic people think works for them, outcome surveys have been trialled but

are finding that autistic people really struggle to answer them.

From pilot projects run ahead of the launch of the integrated service it is clear that line-management structures are the key to success, but no economic data (R3) have been collected. Work in Gwent to bring people back from out-of-county placements and support them locally produced both life improvements and reduced costs. There are partnership boards between local authorities and health, and some work has been done on 'right-sizing' services, for example, looking at local rather than out-of-area provision.

Generic mental health services are not designed or suitable for autistic people and GPs can be a barrier (R4) particularly where autism training has not been taken up. Getting a diagnosis can be very lengthy, but under the new initiative (see above) it will be possible for parents to self-refer to a single point of contact. Parents can also ask their local authority for a carers assessment. An initiative was launched in 2016 to establish supported employment agencies in North and South Wales for learning disabled people and for autistic 16-25 year olds. This provided 6 months of government-funded employment and help from job coaches. Autism Spectrum Connections Cymru has had some success in getting people into employment.

Sensory and language issues are recognised in Wales, but anxiety issues (R5) less so. More girls are now being diagnosed than in the past and more men and boys are being seen with a 'female' presentation of autism i.e. autism that is well-disguised. There may be a link with transgender issues. Standards for new build social housing are good and provide a low-stress environment for

autistic people, but school environmental standards need attention.

Discrimination (R6) on the grounds of IQ is still common. Children's services receive recurrent funding while adult services do not, and treatment within the criminal justice system is inadequate, with poor awareness of autism. Parents of autistic children can be vulnerable to unwarranted social service intervention if they are honest in telling social services about challenges with their children.

The transition (R7) out of education service in Wales is moving to age 25 but given the current lack of provision up to age 19 there is no great expectation of change. There is an advisory service for SEN children at age 13 which looks at future education and social care needs. The Social Services and Well Being Act in Wales covers both children and adults so in theory transitions should be easier but there is a lack of capacity and a lack of understanding in the system which results in poor outcomes.

There are health and social care integration initiatives (R8) but education is often left out. Local authority autism leads are not consistently placed – some are in health, some in social care and some in education.

Concerning information systems, the National Centre for Mental Health Research at Cardiff underpins clinical research in Wales. There were two mapping exercises on autism in 2009 and 2011 but these did not use the same databases. There may be information on people receiving mental healthcare and the Waterloo Foundation is funding a new children's database as part of a study of 800 children with overlapping diagnoses and needs.

# APPENDIX D:

## People

### Strategy Board

Dr Elizabeth Vallance JP PhD (Chair)  
 Professor Sir James Drummond Bone FRSE FRSA  
 Dr Richard Horton BSc MB FRCP FMedSci  
 Professor Martin Knapp PhD  
 Dr Dinah Murray PhD  
 Dr Dame Bridget Ogilvie AC DBE FRS  
 Dr Ian Ragan MA PhD (Project Director)  
 Professor Emily Simonoff MD FRCPsych  
 Dame Stephanie Shirley BSc CEng FREng

### Expert Group

Professor Simon Baron-Cohen PhD FBA  
 Professor Julie Beadle-Brown MA PhD AFBPS  
 CPsychol  
 Professor Sir Adrian Bird CBE FRS FRSE  
 Professor Terry Brugha MD(NUI) FRCPsych  
 Caroline Bulmer BSc Msc MBAOT  
 Professor Tony Charman PhD  
 Dr Yo Dunn PhD  
 Professor Jonathan Green MA FRCPsych  
 Professor Francesca Happé FBA  
 Professor Pat Howlin PhD FBPS  
 Professor Mark Johnson PhD FBA  
 Dr Andrew Long MA MB FRCP FRCPCH  
 FAcadMED  
 Richard Mills  
 Professor Declan Murphy MD FRCPsych  
 Sue Osborn FRCA  
 Dr Jeremy Parr MBChB MRCPCH MD  
 Professor Liz Pellicano PhD

Carol Povey  
 Jude Ragan OBE (Honorary Expert/Advisor)  
 Professor Ilina Singh  
 Dr Catriona Stewart PhD  
 Reverend Professor John Swinton PhD BD RMN  
 RNMH  
 Dr Mark Tricklebank PhD DSc FBPhS  
 Dr Simon Wallace PhD

### Autistic Advisory Panel

Dr Larry Arnold PhD  
 Lenah Buckle MA  
 Ann Harber  
 Dr Wenn Lawson PhD  
 Dr Damian Milton PhD  
 Dr Dinah Murray PhD  
 Katie Nurse  
 Dr Catriona Stewart PhD  
 Dr Yo Dunn PhD

Short biographical sketches of the members of the Strategy Board, Expert Group and Autistic Advisory Panel can be found on the NAP website:

[www.nationalautismproject.org](http://www.nationalautismproject.org)

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Many other people – professionals, autistic people, family members, carers and researchers – have given us the benefit of their expertise, for which we are very grateful.

# APPENDIX E:

## Endorsements

The following organisations support the recommendations and conclusions of this report:

Ambitious about Autism  
All Party Parliamentary Group on Autism  
AT-Autism  
Autism Alliance  
Autism Education Trust  
Autism NI  
Autism Research Trust  
Autism Spectrum Connections Cymru  
Autistica  
Centre for Mental Health  
Children and Young People's Mental Health Coalition  
I CAN  
Kingwood Trust  
Mental Health Foundation  
Mental Health Network of the NHS Confederation  
MQ  
National Autistic Society  
NASEN  
Research Autism  
Royal College of Psychiatrists  
Scottish Autism  
Style Acre  
Wales Autism Research Centre  
Westminster Commission on Autism

“ Our work has found that a great deal more could and should be done to generate evidence to shape policy and improve practice in autism ”

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