

**SCIENTIFIC REVIEW OF THE 'BENCHMARKING AUTISM SERVICES EFFICACY'
(BASE) REPORT (2015)**

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1. Terminology and abbreviations

1.1. Terminology

Terminology is a contested issue in the autism educational sphere (e.g. Kenny, Hattersley, Molins, Buckley, Povey, & Pellicano, 2016). We do not use the term 'disorder' when describing people on the autism spectrum, as we see autism as a neurological difference rather than as a deficient or disordered way of being. Throughout this report we refer to 'autistic people' or 'people on the autism spectrum' to include all those who have a diagnosis of autism or any other autism spectrum condition. When quoting other published work and the BASE report we use the terminology used by the authors of the publications we quote.

1.2. Abbreviations

ABA	Applied Behaviour Analysis
ASD	Autism Spectrum Disorder
BASE	Benchmarking Autism Services Efficacy
CWA	Children With Autism
DENI	Department of Education Northern Ireland
EIBI	Early Intensive Behavioural Intervention
FE	Further Education
HE	Higher Education
HESA	Higher Education Statistics Agency
HSCT	Health and Social Care Trust
MCS	Millennium Cohort Study
NAC	National Autism Center
NAS	National Autistic Society
NCSE	National Council for Special Education
NICE	National Institute for Health and Care Excellence
NILT	Northern Ireland Life and Times
OECD	Organisation for Economic Co-operation and Development
OFMDFM	Office of the First Minister and Deputy First Minister
OU	Open University
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-Analyses
SIGN	Scottish Intercollegiate Guidelines Network
UCAS	Universities and Colleges Admissions Service
UNCRPD	United Nations Convention on the Rights of Persons with Disabilities

2. Executive summary

2.1. Introduction

This report conducts a rigorous and independent peer-review of the five separate Volumes of the 'Benchmarking Autism Services Efficacy,' (BASE) report by Dillenburger, McKerr, and Jordan (2015). The BASE report was produced by Professor Karola Dillenburger, from the School of Education's Centre for Behaviour Analysis, Queen's University Belfast, and is available online¹. The research was funded by the Office of the First Minister and Deputy First Minister (OFMDFM).²

The five Volumes of the BASE report are:

- a literature review (Volume 1);
- analysis of the Northern Ireland Life and Times Survey (Volume 2);
- secondary data analysis of existing Northern Ireland government datasets (Volume 3);
- the design and analysis of qualitative studies using focus groups, interviews and surveys (Volume 4);
- summaries and recommendations based on the findings of Volumes 1-4 (Volume 5).

Given the potentially important and far-reaching implications for policy and practice of the BASE report, the John and Lorna Wing Foundation³ commissioned Dr Karen Guldberg (University of Birmingham) and Professor Sarah Parsons (University of Southampton) to conduct a scientific review of its content. The aim of this scientific review was to report on the quality of the methodology employed, the subsequent reliability of the findings, and the validity of the recommendations.

¹ <http://www.qub.ac.uk/research-centres/CentreforBehaviourAnalysis/Research/BenchmarkingAutismServiceEfficacyBASE/>

² Since then, there has been a restructuring of Government Departments in Northern Ireland and many of the former functions of the OFMDFM now come under The Executive Office.

³ This scientific review reflects the opinion of the authors. The John and Lorna Wing Foundation is not responsible for any use which may be made of the information contained therein.

2.2. Research question and methodology

The key question addressed through this review was:

Are the conclusions of the BASE report (2015) scientifically grounded and based on a robust enough methodology to be used by policy makers to make decisions or take actions in respect of children, young people and adults with autism?

The peer review was planned and conducted according to the internationally recognised standard for scrutinising academic work intended for the public domain, i.e. using a methodology in line with the editorial process of high-quality academic journals. Dr Guldberg and Professor Parsons acted as Editors and invited two internationally recognised expert peer reviewers for each of the BASE Volumes 1-4. Reviewers were unknown to each other at the time, and were asked to comment on the strengths of the BASE report, the robustness of the methodology in the Volume they reviewed, as well as the resultant findings and recommendations. The Editors synthesised the reviewers' comments with their own assessments, summarising what could be learnt from the BASE (2015) report, evaluating the extent to which it adhered to high-quality research practices, as well as highlighting issues of concern. After completing a draft report, Dr Guldberg and Professor Parsons sent the report to two experts in the autism and disability field for peer review, as well as to the eight reviewers of Volumes 1-4 for feedback, before completing this final version.

2.3. Findings

The BASE report represents a substantial piece of work, utilising a wide range of methodological approaches to provide baseline data against which improvements to policy and practice can be compared. An impressive amount of data emerged from this mixed-methods study conducted over a number of phases. The overall study represents a significant effort on behalf of the authors, with a large range of literature consulted (Volume 1), sizeable samples (Volume

2), a comprehensive overview and analysis of secondary data sources (Volume 3), and with a welcome inclusion of autistic people and families in research (Volume 4).

The wide lens that the report takes on the issues relating to service provision (and related policies) for people on the autism spectrum, and their families, is welcome in a field that tends to overly concentrate on narrow views or specific approaches to intervention. The project had the potential to be wide-reaching and ground breaking in terms of its reach and scope and, therefore, to be genuinely informative for practice and policy.

Despite these strengths, there were serious limitations. The BASE project did not specify any research questions to guide and shape the research, nor did it define key terminology. Furthermore, there was no transparent discussion of the methodology chosen, neither for the mixed methods approach for the project as a whole, nor for individual phases of the research. There were tenuous links made between data, findings and recommendations; with statements that made causal leaps that were not grounded in the data. There was little evidence of systematic examination of the research evidence, nor of an assessment of its validity, quality, results, or relevance before using it to make recommendations for policy. In addition, the researchers failed to make their assumptions and influences clear. This lack of audit trail led reviewers to question many of the conclusions drawn throughout the separate volumes.

This independent review of the BASE report therefore concludes that the lack of objectivity, rigour and transparency in the BASE report overall mean that the findings and conclusions should not be taken at face value and should be re-evaluated in the light of the many concerns raised by reviewers.

2.4 Recommendations

In making recommendations for future research investigating the link between poverty and autistic people, we suggest that the following gaps and priorities emerge from this review:

- There needs to be an appropriately designed and conducted systematic literature review that is carried out to address core research questions relating to poverty and wellbeing of individuals with autism and their families.
- A survey should be conducted with service providers to understand whether there is indeed a failure to implement aspects of practice recommended or mandated in existing policy documents. Autistic people and their families should be included in this research from the outset.
- A clearly framed analysis should be commissioned of the Northern Ireland Life and Times (NILT) data (excluding ambiguously phrased questions) that addresses specific research questions and incorporates more sophisticated planned analyses in order to understand the possible links between independent and dependent variables.
- Further analysis should be undertaken of the secondary datasets. These should incorporate statistical significance for comparisons made, use a set of researchable questions to guide the analysis, with transparent explanation of where measures were taken from, and with coherent reporting of findings and careful presentation of figures and graphics.
- In order to fully understand and represent the voices of autism stakeholders, there needs to be a more robust and larger scale qualitative study that enables participation from a wider range of people. More stakeholders need to be included in qualitative methods that support a range of communication preferences and needs.
- There would be value in undertaking an audit of the range of training courses and resources that are currently available for staff, autistic people and their families in Northern Ireland, including those available by distance learning.

More generally, we hope the following specific recommendations will be helpful to The Executive Office in Northern Ireland when commissioning future research:

- Ensure there is a robust external peer review process for all reports

intended for the public domain.

- Provide clear guidelines for authors to ensure that all commissioned and published research reports adhere to best research practice by: disclosing conflicts of interest; being detailed about procedures for ethics review and approval for all appropriate aspects of the research; designing research to answer specific research questions and adequately specifying all necessary aspects of methodology, analysis and key terminology.

3. Scientific Review – full report

3.1. Introduction

This report conducts a rigorous and independent peer-review of the five separate Volumes of the Benchmarking Autism Services Efficacy, (BASE) report by Dillenburg, McKerr, and Jordan (2015). The BASE report was produced by Professor Karola Dillenburg, from the School of Education's Centre for Behaviour Analysis, Queen's University Belfast, and is available online⁴. The full title of the BASE report is: 'Helping the most vulnerable out of the poverty trap and reducing inequality: Policies, strategies, and services for individuals with Autism Spectrum Disorder, including intellectual and neurodevelopmental disabilities'.

The Volumes of the BASE report are: a literature review (Volume 1); analysis of the Northern Ireland Life and Times Survey (Volume 2); secondary data analysis of existing Northern Ireland government datasets (Volume 3); the design and analysis of qualitative studies using focus groups, interviews and surveys (Volume 4). Volume 5 provides summaries and recommendations based on the findings of Volumes 1-4.

The BASE report was one of many projects that were part of an open call for research by the Office of the First Minister and the Deputy First Minister (OFMDFM)⁵ in Northern Ireland to inform policy in relation to poverty and inequality⁶. Dillenburg et al. (2015) state that: "The BASE report provides baseline data regarding individuals with autism and it sets benchmarks against which the effect of the Autism Act (Northern Ireland) 2011 and associated Autism Spectrum Disorder (ASD) strategy can be measured" (Volume 5, p.5). The authors describe that "the primary purpose of the BASE Project was to establish

⁴ <http://www.qub.ac.uk/research-centres/CentreforBehaviourAnalysis/Research/BenchmarkingAutismServiceEfficacyBASE/>

⁵ Since then, there has been a restructuring of Government Departments in Northern Ireland and many of the former functions of the OFMDFM now come under The Executive Office.

⁶ . The open call was a programme of independent research and consequently the views expressed and conclusions drawn were those of the author and not necessarily those of OFMDFM.

how to help individuals with Autism Spectrum Disorder out of poverty by promoting social inclusion” (Vol.2, p.6) and they highlight that the BASE project started in 2012, before the Autism Strategy (2013-20) and Action Plan were launched in 2014 (Department of Health, Social Services and Public Safety, 2015). They, therefore, state that the findings are available to support the review of the first Action Plan (2013-2016) and inform the second Action Plan.

The ‘Benchmarking Autism Services Efficacy,’ (BASE) report by Dillenburg, McKerr, and Jordan (2015) potentially has far reaching public policy implications for the future of autism services in Northern Ireland. Mindful of this, Autism Northern Ireland (NI) sought assistance from the John and Lorna Wing Foundation to commission an independent review of the findings and recommendations of the report. The John and Lorna Wing Foundation agreed to commission this scientific review on the basis that:

- The Benchmarking Autism Services Efficacy,’ (BASE) report is a large and significant piece of work and should be subject to expert independent peer review.
- The Foundation would be the sole commissioner and funder of such a review. Without prejudice, the findings of such a review would be made available to Autism NI and to the official bodies in Northern Ireland.
- As part of the methodology, the scientific review of the ‘The Benchmarking Autism Services Efficacy’ (BASE) report would itself be subject to independent peer review by suitable experts.

The John and Lorna Wing Foundation is a charitable foundation supporting autism research. Lorna Wing established the Trust in her lifetime to continue her work in autism (see <http://www.johnandlornawingfoundation.org>). The Foundation commissioned Dr Karen Guldberg (University of Birmingham) and Professor Sarah Parsons (University of Southampton) to conduct a scientific review of its content (further information about the authors can be found in section 10.2). The aim of this scientific review was to report on the quality of the

methodology employed, the subsequent reliability of the findings, and validity of the recommendations. The key question addressed through this review was:

Are the conclusions of the BASE report (2015) scientifically grounded and based on a robust enough methodology to be used by policy makers to make decisions or take actions in respect of children, young people and adults with autism?

3.2. Methodology and procedure of the peer review

The methodology chosen for this scientific review was driven by a commitment to the peer review process as we see assessment of methodological and reporting quality as critical to adequately judging the credibility of a study's conclusions. In our view, peer review is integral to scholarly research as it promotes and maintains high standards in research, and is essential in order to check and improve quality. It is a critical part of the functioning of a scientific community, and acts as a quality control filter.

As the BASE report had not been subjected to peer review and had already been published, the task was to develop the most appropriate methodology for undertaking scientific review. We decided to plan and conduct the review according to the standards for scrutinising academic work intended for the public domain, i.e. using a methodology in line with the editorial process of high-quality academic journals.

Dr Guldborg and Professor Parsons acted as Editors and invited two internationally recognised expert peer reviewers for each of the BASE Volumes 1-4. Reviewers were unknown to each other, and were asked to comment on the robustness of the methodology in the Volume they reviewed, the strengths of the BASE report, as well as the resultant findings and recommendations.

The reviewers were selected to represent autism-specific knowledge where necessary, as well as methodological expertise from outside the autism field, in order to have a balance of views as well as some control against bias or conflict

of interests. Reviewers were paid a set fee for their work, in recognition of the time commitment, and they were given two months to complete the reviews. Once all reviews were received, the Editors synthesised the reviewers' comments with their own assessments, summarising what could be learnt from the BASE (2015) report, evaluating the extent to which it adhered to high-quality research practices, as well as highlighting issues of concern. After completing a draft report, Dr Guldberg and Professor Parsons sent the report to two experts in the autism and disability field for peer review, as well as to the eight reviewers of Volumes 1-4 for feedback, before completing this final version. All reviewers involved in undertaking the scientific review are listed in the acknowledgement section of the report (see section 10.3).

We approached the critical appraisal of the BASE report from the vantage point that scientific studies should acknowledge and build on other work in the field, back up claims with evidence and provide rigorous, systematic, objective, and accurate citations of extant work. Arguments should be evidence based, with a good overview of published studies in the field, and with transparency and precision in the conclusions reached. Critical appraisal should include, but not be limited to, assessing the appropriateness of the research design in meeting the aims of the research, the rigour of data-collection and analysis, whether there was a well-conducted and accurate sampling strategy, and the extent to which conclusions and recommendations were in line with findings (Grayson and Gomersall, 2003). It should be conducted ethically; there should be consideration of the relevance to inform practice or policy, as well as the use of appropriate and rigorous methods, and clarity and coherence of the reporting (Cohen and Crabtree, 2008).

In undertaking peer review of research, this report therefore followed the conventions of scientific review by making an assessment about whether:

- the research questions and aims of the study are clear;
- the methodology is clearly explained and described, and robustly justified in terms of answering those research questions, aims and objectives;

- there is coherence between the research questions, methodology, findings and discussion; and
- there is an appropriately critical analysis of the literature.

In order that all reviewers were supported to make a focused assessment about the quality of the BASE report, the Editors created proformaes to structure comments and feedback (see Appendix 1). In each proforma, reviewers were given the aims of the scientific review and the central research question. They were all asked to comment on the originality of the Volume they were reviewing and its relationship to the literature; methodology and the results, and implications for research, practice and/or society. They were also asked to make general comments and to give an overall assessment.

Given that the four Volumes of the BASE report were based on different methodologies, specific questions needed to be asked for each Volume to determine quality. Thus, Volume 1 was a Literature Review, so one of the sub-questions for Volume 1 asked whether the Volume demonstrated an adequate understanding of the relevant literature in the field and cited an appropriate range of literature sources. As the BASE report authors indicated they had conducted a systematic literature review, another sub-question focused on whether the literature review had been conducted in a rigorous and systematic manner.

Volume 2 of the BASE report employed a very different methodology. This Volume analysed data from the Northern Ireland Life & Times (NILT) questionnaire 2012 and the authors had designed the questions for the section they analysed. Reviewers were therefore asked to examine the validity and balance of the questions posed for the autism section and the use of the data in relation to the stated aims and objectives of the BASE review.

3.3. Limitations of the scientific review

The limitations and challenges for peer review in academic research are well known and continue to be debated (e.g. Bohannan, 2013; Lee, Sugimoto, Zhang &

Cronin, 2013). Despite these limitations however, a majority of researchers remain supportive of the principles of peer review and the need for researchers in cognate fields to check, monitor, and improve the standards of each other's work (Mulligan, Hall, & Raphael, 2013). There are different ways in which this review could have been conducted. Double-blind review is considered by many to be the best (less prone to bias) way of completing peer review (Mulligan et al., 2013). However, this was not possible in the current context because the BASE review was already published and in the public domain. Consequently, we adopted a single-blind peer review approach, which is less common in the social sciences and humanities, but more common in life sciences, physical sciences and engineering (Lee et al., 2013).

The single-blind approach is critiqued because there is potentially more scope for bias (gender, institution, seniority etc.) when the author is known to reviewers (Lee et al., 2013). Due to these reasons researchers generally prefer double-blind to single-blind reviews (Mulligan et al., 2013). Single-blind reviews are also critiqued because there could be a tendency for reviewers to be overly critical if they feel that their identity remains anonymous.

Consequently, to help to strengthen our overall approach, we offered all reviewers the opportunity to review a draft copy of the report prior to finalising and to make any changes to the text, based on their own comments and having had the opportunity to read others'. While this cannot guard against bias towards the authors, this process allows reviewers to see their comments in the overall context of the review and to check whether they feel an appropriate balance has been achieved in the report. We also asked reviewers whether they were happy to be named as contributors to the review as a further check on balance and fairness. All reviewers were happy to be named as contributors.

An additional limitation of our peer-review approach could be the lack of a systematic and standardised approach to the review criteria we gave to reviewers. That is, we could have used established protocols for structuring reviewers' evaluations and feedback such as adopting the GRADE quality criteria

for evidence, within a Cochrane systematic review framework (for example). While such systems have advantages in terms of focusing review comments on standardised and specific aspects of research studies, they also come with very clear epistemological assumptions regarding the hierarchies of methodological designs. They are also developed specifically for reviewing primary research in healthcare contexts, with a clear focus on evaluating the strength or weight of evidence relating to the particular outcomes of healthcare interventions. These features makes them problematic for evaluating research that comes from a range of disciplines and takes a range of methodological approaches (Hammersley, 2001), such as in the BASE review.

In addition, while there are frameworks for reviews of evidence that accommodate a range of research designs (e.g. from the EPPI-Centre in the UK (eppi.ioe.ac.uk), such frameworks are designed for conducting systematic reviews for collating and synthesising evidence on particular issues or outcomes. Our objective here was not to collate and synthesise primary evidence *per se*, rather it was to review the collation, synthesis, analysis and interpretation of primary evidence as reported within the BASE review. Consequently, it was more appropriate for us to take a peer review approach to reviewing the BASE reports rather than a systematic review approach using structured protocols.

3.4. Structure of the report

The main comments of the reviewers are summarised below for each of the Volumes in sequence. Reviewers often made additional, more minor, suggestions for improvements to the clarity and coherence of the information presented in each Volume, but we have not included all of these details here as the BASE report is already published and cannot respond to the suggestions from reviewers. Therefore, we focus only on the main lines of critique. The following sections also include some additional observations made by the Editors. We then provide a concluding paragraph that draws together overall comments on, and conclusions to, the main research question posed:

Are the conclusions of the BASE report (2015) scientifically grounded and based on a robust enough methodology to be used by policy makers to make decisions or take actions in respect of children, young people and adults with autism?

We end the report by making some brief recommendations for how research in this area could be strengthened in order to provide a robust and dependable evidence base for informing and influencing policy.

4. Review of BASE project (Vol. 1) literature review - Dillenburg, McKerr, & Jordan, March 2015

4.1. Introduction

Volume 1 of the BASE report consists of a literature review comprising two parts:

(1) Section 1 is described as being a “systematic review of research literature on ASD and poverty and social inclusion” (p.8) framed around the eight “most autism relevant” benchmarks of the United Nations Convention on the Rights of Persons with Disabilities (UNCRPD, 2006).

(2) Section 2 is a “summary review of ASD related reports, policies, and initiatives that have been published in Northern Ireland since 2002” (p.8).

On the opening page of the report (p.3) this Volume is described as a **“comprehensive literature review** using a systematic approach on outcomes for individuals with autism and the policies designed to improve those outcomes” (emphasis in original).

Reviewer A notes some strengths of the review, namely that:

... the reviews are very wide ranging and incorporate both primary research evidence (part 1) and 'grey' literature (part 2). This is a major undertaking and is clearly the product of a great deal of work from the authors who are led by a recognised expert in the field. The report draws from a large number of appropriate literature sources.

Reviewer B comments that the review in Section one:

... provides a potentially interesting approach to the analysis of what we already know in relation to global benchmarks and people with ASD ... [but] ... while there are strengths to utilising UN criteria to look at the [research]

evidence base, this required some thinking through in relation to the approach to the policy review.

Overall, however, both reviewers raised a number of significant concerns about Volume 1, specifically relating to:

- a. a lack of clarity and detail about the methodology employed for searching and selecting literature;
- b. omissions of key studies, and details of studies, in the field;
- c. a lack of objectivity, including misrepresentation of others' research, and a strong bias towards Applied Behaviour Analytic (ABA) approaches, specifically early-intensive applied behaviour analytic interventions;
- d. a lack of clear trace between conclusions and recommendations, and the evidence reported;
- e. a high level of self-citation from the authors.

We provide more details about each of these concerns in the following sections covering the methodology, analysis and findings, and conclusions of the report.

4.2. Methodology

The review of research evidence in Section 1 is described explicitly as *systematic*. In research methodology, the term 'systematic' carries a very specific set of meanings, which extend beyond the normal expectations of literature reviews to be clear, rigorous and transparent with regard to the searching and summarising of literature. Therefore, if using the term 'systematic' researchers always need to be fully cognisant of its connotations and implications. For example, according to Punch and Oancea (2014; p.134):

The term 'systematic review' has ... a further, more technical sense. It refers to a particular approach to inquiry, the development of which was strongly stimulated by the evidence-base movement in professional practice and policy ... In this sense, systematic reviews of the literature can be research projects in their own right: they seek to answer specific

research questions via explicit and rigorous methods, and they communicate their findings to a range of audiences.

Cohen, Manion, and Morrison (2013; p. 342) provide a very helpful summary of what such a “particular approach to inquiry” in a systematic review should comprise:

Systematic reviews are a refinement of general research synthesis by being more rigorous and less ‘narrative’ in character; they require the use of techniques to minimize bias, they follow protocols and criteria for searching for relevant primary, usually empirical, studies, their inclusion and exclusion, the standards for acceptable methodological rigour, their relevance to the topic in question, the scope of the studies included, team approaches to reviewing in order to reduce bias, the adoption of a consistent and clearly stated approach to combining information from across studies, and the careful, relevant conclusions and recommendations drawn.

What is clear from these definitions and descriptions is that there are very specific steps that should be followed and reported when undertaking a systematic review, such that an as-objective-as-possible view is achievable in the searching, analysis, and synthesis of the literature. This is vital for providing a replicable, reliable, and trustworthy evidence base for informing policy and practice. Unfortunately, the reviewers report that most – if not all – of these steps appear to be absent from Volume 1 (Section 1) of the BASE report, which casts significant doubt on the objectivity, reliability and robustness of the findings. Specifically:

- the methods undertaken are not sufficiently transparent to give the reader confidence that a systematic literature review has taken place;
- there is no specific research question, or set of research questions, that guide the review of the literature;

- the full and complete list of search terms, together with specific Boolean operators, is not adequately specified;
- there is no information provided to demonstrate that PRISMA or other systematic review guidelines were followed;
- there is no inclusion of a flow chart to show the number of identified studies that were initially found, and then included or rejected at different stages of the review process, as would typically be expected in a systematic review;
- readers are not informed of how many papers were consulted, nor about any inclusion or exclusion criteria that were applied;
- there is no indication or discussion of the quality of research and how such judgements were made by the team i.e. there is no actual analysis of the papers included in the review;
- there is no information provided regarding whether or how team decisions about the relevance and scope of the nature of the evidence were made;
- the databases used are listed, but no date parameters are given and so it is unclear how inclusive Section 1 of the research review is.

Overall, therefore, the review of the literature in Section 1, Volume 1 of the BASE report cannot claim to be systematic. Any suggestion that the review is methodologically systematic is misleading and should be treated with significant caution in terms of reliability and objectivity.

Section 2 of Volume 1 is described as a “summary review” (p.8) and provides an analysis of a number of autism reports, policies and initiative documents “published in Northern Ireland since 2002” (p.8). While the technical requirements for this part of the methodology are not the same as for a systematic review, there still needs to be a clear rationale for the inclusion or exclusion of particular reports. Again, this is important to ensure objectivity and to guard against potential biases in the selection and interpretation of material.

There are some areas of clarity and detail in Section 2 that were missing from Section 1, notably the authors specify the number of reports identified in their search in Section 2 and there is some information provided regarding the designation of these documents (e.g. ASD specific, Education non-specific; p.50), which improves transparency. In addition, there is a specific timeline provided for the policy review search, numerical tallies for the different types of report, and a rationale provided for the order of their presentation.

However, as Reviewer B notes, Section 2 of the report is still somewhat “opaque” with regard to methodology:

... the relationship between the two parts of the review is unclear. The policy review includes reports that are funded by autism charities, largely based around the collection of survey data and, hence, providing some confusion between what are policy documents and what are empirical studies. The rationale for also making reference to additional research literature in the policy/report section is not clear, although these appear to be limited to additional material on ABA [Applied Behavioural Analysis]. In addition to those reports concerning NI, it draws on other countries in the UK as well as the Republic of Ireland and it includes generic policy reports that are relevant to autism.

In short, the rationale for the inclusion or exclusion of documents in this section is unclear, and the remit for inclusion certainly seems to be much wider than reports “published in Northern Ireland since 2002”. Additionally, while some self-citation is to be expected given that the authors are active in the field, there are approximately 30 different references featuring the lead author alone, which is arguably rather excessive as it represents over 10% of all the cited references in Volume 1. A more transparent and appropriately systematic approach to the searching and selection of literature would have enabled the authors to control for this issue.

4.3. Analysis

There appears to be no clear analysis provided or justification reported for how the judgements about the current status of recommendations in policy reports (Table 1, p.14) were made. Specifically, the authors use a green, amber, red traffic light system to indicate whether policy and practice recommendations from the reports reviewed have been implemented fully, partially, or not at all (respectively). However, it is unclear whether these judgements were based solely on the authors' opinions, or via consultation with relevant professional agencies. Without knowing this, it is difficult to assess just how important their findings are, or indeed whether they represent the actual state of play in the field. This is a shame because there is a potentially important key message contained within Section 2 about the lack of progress 'on the ground' with respect to the implementation of policy and practice recommendations from other reports. If this is indeed the case on the ground then this is certainly something that needs to be fully investigated and transparently reported.

There is also a lack of clear analysis in Section 2 in order to show how the "key themes" (p.91) that were derived from the review were developed; studies are primarily reported in a descriptive manner. A mapping of evidence to themes would have been helpful here (e.g. to help the reader to understand which evidence sources have contributed to each theme/recommendation).

4.4. Findings

The lack of clarity and detail regarding the methodology of both sections of the literature review, as outlined above, raises questions about possible omissions of studies for informing the findings, and omissions of the details of studies, and the reasons for these. As Reviewer A notes:

... without knowing more about exactly how the authors conducted the reviews (including, for example, inclusion and exclusion criteria that were applied), one might unfairly accuse the authors of having missed some key publications (when in fact these were identified but rejected for valid reasons).

This point underscores the importance of clearly planning, undertaking, and reporting each step of a literature review so that decisions about inclusion or exclusion are transparent. As it stands, the review seems to omit some major publications in the field and the rationale for this is unclear, for example:

- Reports from the National Autistic Society (NAS) that are directly pertinent to the remit of the report (e.g. 'B is for Bullied', Reid & Batten, 2006; and 'We've got great expectations', Reid & Ayriss, 2011) appear to have been omitted, despite other publications by this group having been included (e.g. 'A* is for Autism', Stewart, 2012) in Section 2 of the report;
- In Section 1, Wong et al.'s (2014; 2015) review of evidence-based practices for ASD is omitted. Parsons et al.'s (2009) international literature review of evidence of best practice in the provision of education of persons with ASD, commissioned by the National Council for Special Education (NCSE) in Ireland, and subsequent publications from the report (Parsons et al., 2011; Guldberg et al., 2011) are also omitted;
- The National Institute for Health and Care Excellence (NICE) Autism Quality Standard (QS51, 2014) is also missing, despite other guidelines and reports by NICE being referenced in Volume 1.

In addition, the reporting of some aspects of the literature is more nuanced than others, particularly with respect to reporting the context of the research. For example, healthcare costs are quoted without acknowledging that the data is from the USA; elevated divorce rates are drawn from a Californian website; the reported cost of raising a child with disabilities draws on a study of high functioning autism only. While there is an indication of the scale of the research for some studies, it is unclear for others. This gives the impression that all studies make an equal contribution. Other sections provide a more critical analysis: for example, the difficulties of comparing percentages in school to Further Education (FE) and Higher Education (HE) and the challenges of diagnosing mental health difficulties.

There are also some mismatches with respect to the main text of the report and the bullet points that summarise the Findings (pp.8-12) in the Executive Summary. Generally, the summary text ignores more positive aspects found in the literature, strengths that might be built on for future. Examples include the ways in which the telephone-based service has improved accessibility in the job centre (p.26) and the success of targeted employment programmes, including job satisfaction of employees and positive experiences of employers (p.30). There are also other surprising omissions from the Executive Summary, for example, the summary bullets on Adequate Living Standards do not reflect the page and a half (pp.25-26) discussing these issues as people with autism grow into adulthood. It therefore downplays the issue of eligibility for benefits.

Overall, without clarity regarding the decisions made for the inclusion and exclusion of literature, and without sufficient contextualisation of the studies discussed, it is not possible for readers to be confident about the scope, range and quality of the evidence base analysed and presented and, therefore, the validity of the conclusions and recommendations that follow from the literature review.

4.5. Conclusions

As Reviewer A notes above, there may be very good and understandable reasons for including some of the literature and policy documents while excluding others. In other words, it might be reasonable to give the authors the benefit of the doubt regarding whether and how bias was recognised and controlled for in the conduct of the literature review. However, other aspects of the document suggest that there is a bias with regard to how key citations have been reported and interpreted which, in turn, casts doubt on any implied objectivity (via the labelling of a 'systematic' methodology) in the literature review.

For example, the authors state that, "failure to provide effective early intensive behaviour analysis-based intervention has serious lifetime economic consequences" (p.13) and cite Knapp, Romeo and Beecham (2009) to support this assertion. The Knapp et al. (2009) paper provides an analysis of the societal

costs of autism and states there is a need to evaluate interventions (without stating which ones specifically) “for both effectiveness and cost-effectiveness” (p.332). They do not mention early intensive behaviour analysis-based intervention specifically and certainly do not endorse it, or supply a conclusion that could be interpreted as indicated in the BASE review. Specifically, Knapp et al. (2009) conclude from their analysis:

... given that the autistic spectrum includes a number of disorders and a wide range of needs, symptoms and characteristics, it is likely that a wide range of behavioural, educational and medical interventions could be required in order to meet some or all individual needs. (p.332)

Similarly, the statement is made in Volume 1 of the BASE report (p.20) that a “... lack of intensive behavioural interventions has been linked with extremely poor long-term outcomes” (Howlin et al., 2014). The claim that the Howlin et al. (2014) study drew the conclusion that there were detrimental long-term effects because their cohort did not receive early behavioural intervention is a serious misrepresentation of the points made by Howlin et al. (2014). Howlin et al. (2014) clearly acknowledged that there is research available in which *claims have been made* for significant IQ and long-term outcomes as a result of early behavioural interventions. Secondly, they made the point that the cohort they studied had not had access to this particular approach. They did not draw a causal link between the outcomes for their cohort and the fact that this cohort did not receive early behavioural interventions, and hence the conclusion drawn about this in Volume 1 of the BASE report is not warranted.

This leads to concerns regarding the objectivity of the review. Specifically, there appears to be a strong bias towards Applied Behaviour Analysis (ABA) in general and, specifically, early intensive behaviour analysis-based intervention relative to other forms of intervention. Having said this, there is no definition of Applied Behaviour Analysis, so it is unclear whether developmental ABA approaches such as the Early Start Denver Model (Rogers and Dawson, 2009), for example, are included when the authors refer to ABA.

The bias towards ABA is first evident in the executive summary of Volume 1, in which ABA is mentioned several times, and continues throughout the report. While intervention approaches based on ABA principles have good empirical support as evidence-based practices (e.g. Estes et al., 2015; Wong et al., 2014; Dawson et al., 2010), there are important questions still to be resolved about the clinically effective details of such interventions (Romanczyk, Callahan, Turner & Cavalari, 2014) and the idiosyncratic nature of evidence in the field (see Kasari and Smith, 2016 for further discussion of this).

Moreover, these are not the only evidence-based approaches to intervention in the field. For example, there is good evidence from a number of well controlled trials of alternative interventions, such as those based on improving early adult child interaction and joint attention (e.g. Green et al., 2015; Kasari, Gulsrud, Paparella, Hellemann & Berry, 2015). Reiterating this in a recent systematic review of the literature, Bond et al. (2016) recognised the good evidence for early intervention for pre-school children based on behavioural principles, but also recognise interventions that focus on play and joint attention, as well as those that are peer-mediated. Bond et al. (2016) concluded that for pre-school children and school-aged children there should be a range of intervention options available:

A range of options are required, which can be tailored to child and family needs (p.5, pre-school provision and intervention).

The evidence provided in the systematic review and guidance strands indicates that a range of provision types and intervention strategies are needed (p.5, school-aged ASD provision and interventions).

Similarly, the second edition of the comprehensive *New Zealand Autism Spectrum Guideline* (Ministries of Health and Education, 2016) reaches the same conclusion:

A single approach or solution is unlikely to ever meet the needs of all learners with ASD. Educators need to choose approaches that fit the individual and their settings, and need to be skilled in providing a range of interventions.

The Scottish Intercollegiate Guidelines Network (SIGN) also issued an updated version of its guidance on the *Assessment, diagnosis and interventions for autism spectrum disorders* in 2016, based on a systematic review of evidence. One of its main conclusions with regard to early intensive behavioural intervention (EIBI) was:

While it has been shown that EIBI is superior to no intervention or treatment as usual, the evidence does not warrant the provision of universal EIBI. It does however justify rigorous cost-effectiveness research, which currently is of poor quality and does not support wide application of this approach. (p.24)

While Bond et al. (2016), the New Zealand Guideline (2016), and the SIGN guideline (2016) were all published after the publication of the BASE report, earlier comprehensive reviews of the literature had reached similar conclusions (Parsons et al., 2009; Ministries of Health and Education, 2008; SIGN, 2007), though none of these sources are cited in the BASE review. For example, Parsons et al. (2009) concluded:

... the studies included in this review that have robustly explored the comparative benefits of intensive behavioural interventions with other approaches (Section 3.1.2) report, on balance, mixed findings with respect to the superiority of one approach over another. Based on these findings, ... it is clear that a range of interventions (eclectic provision) should continue to be funded and provided for families ... It is also important to acknowledge that this conclusion does not exclude the usefulness of intensive behavioural approaches for some children learning specific skills. (pp.114-115)

The main point is that where systematic review methodologies have adopted a broad search for literature (i.e. across the range of interventions available) there tends to be a more balanced set of conclusions drawn from the evidence. These more balanced conclusions recognise and support the role of ABA principles, and early intensive behavioural interventions specifically, in promoting successful intervention approaches for particular skills for some children, while also recognising that there are other approaches to intervention that are successful. This overall balance, or weight of evidence, in the field is not reflected in the recommendations drawn in the BASE report, which refer exclusively to ABA (recommendations 4 and 5, p.92), viz:

4. ***Effective, evidence-based interventions*** of international standards based on the science that has been identified as providing the most rigorous data for evidence-based interventions, applied behaviour analysis (ABA); rather than an eclectic mix of interventions, that are not based on evidence.

5. ***Expertise of staff delivering services*** including re-training of existing staff (e.g., fully utilising/commissioning internationally approved training in applied behaviour analysis and autism offered at local Universities) and/or the targeted employment of Board Certified Behaviour Analysts.

Given the lead author's status as a well-known advocate of ABA, this suggests a lack of objectivity in interpreting the evidence and making recommendations from it. It could be that the weight of evidence in the field changed dramatically between the literature search of Volume 1 and publication of the BASE review, and the literature searches and publication of the 2016 reviews cited above, but we think this interpretation is highly unlikely. The substantial disparity between the conclusions of Volume 1 of the BASE review and those of other wide-ranging, international reviews of the evidence in the field is noteworthy and, in the words of one of the reviewers "*troubling*".

In alignment with these observations, there is also a lack of clear trace between the conclusions and recommendations of the report and the evidence presented from the literature review. While the literature review text makes a broader point about the levels of dissatisfaction by parents of their child's educational placement, when this is reported in the Executive Summary the point is made specifically in relation to ABA. Moreover, the opening paragraphs of the authors' conclusions section of Volume 1 of the BASE report (pp.90-91) focus on early intensive behavioural interventions, together with the failure to provide interventions based on ABA principles, thereby providing considerable prominence for these concluding statements in the report. However, the prominence given to these conclusions, relative to others within the report, does not seem to follow from either the policy review in Section 2 or from the summary of key findings in the literature review in Section 1.

The conclusion for the review puts forward eight important recommendations, although the links between these and the findings of the literature and policy reviews are not transparent. Although there is no 'thematic mapping', some of these recommendations are clearly drawn from the two reviews that preceded them. For example, the need for a comprehensive database, financing and transparency of budgets, and improvement of transition processes reflect the substantive findings of the report well. The literature review also indicated that bullying, exclusion and lack of post-school provision were central findings, but only the latter was addressed in the recommendations. It would have been useful if the summary bullets had been utilised to show the reader the link between the evidence and the recommendations. These recommendations would also have benefitted from some expansion, as some were clearer than others. The recommendation for the improvement of transition processes and post-19 provision gives no sense of what is needed, for example. The recommendation to use ABA and to train staff in ABA goes beyond the evidence presented in the report.

4.6. Main conclusions from the review of Volume 1

There are some important messages contained within the report about some of the existing strengths and positive experiences of provision in Northern Ireland, and there is value in applying the framework of the UNCRPD (2006) to examine the evidence base. There is a wide range of literature included in this Volume, representing a significant undertaking by the authors, which is to be respected.

However, the independent reviewers of this Volume raised substantial concerns about the overall quality of the methodology employed, which leads to questions regarding the reliability of the findings, and the validity of the conclusions and recommendations drawn. While being described as a systematic review, Section 1 of Volume 1 did not employ the planned, transparent and sequential steps that characterise systematic review methodology. Consequently, the methodology is not replicable, and cannot be judged to have controlled for bias. In both sections of the report there needed to be a clear and transparent approach to the analysis of the material contained within the research literature and policy documents, however, no such analysis was reported. There also needed to be a much clearer contextualisation of all studies and reports discussed, as well as a transparent audit trail between the findings and the recommendations. The listing and description of policies and recommendations without a contextualising narrative inevitably leads to a disconnection with the conclusions. Our overwhelming main conclusion from the reviews of Volume 1 is, therefore, that the work presented is not sufficiently robust or reliable to inform policy and practice.

5. Review of BASE project (Vol. 2) NI Life and Times (NILT) Survey Autism Module - Dillenburger, McKerr, & Jordan, September 2013

5.1. Introduction

Volume 2 of the BASE report presents the findings from the Northern Ireland Life and Times (NILT) Survey Autism Module “to survey public attitudes, knowledge and awareness of autism (n=1200)” (p.3). As with the other Volumes, the main purpose of the BASE report is reiterated: “The primary purpose of the BASE Project was to establish how to help individuals with Autism Spectrum Disorder out of poverty by promoting social inclusion” (p.6). With specific reference to the content and purpose of Volume 2, the authors (p.6) state that:

The NILT (2012) survey first ever autism module (n=1204) offered a baseline against which the impact of new autism legislation, policies, and strategies can be measured.

The NILT Autism Survey Module comprised 17 questions in total asking respondents about their knowledge, experiences, and attitudes towards autism, including prevalence, causes, treatments, aspirations, and support.

Reviewer C states:

The report is, in my assessment, appropriately evidenced given the research design and data and does not, for the most part, make inferences that are not warranted by the analyses presented.

Indeed, the use of an existing survey methodology that includes a systematically sampled group is a notable strength of this Volume of the BASE report. The large sample size obtained similarly lends weight and credibility to the findings presented. Reviewer D also commented on these strengths:

This [the overall research design] seems impressive and piggy-backs a national survey undertaken by an experienced and independent (of the research team) research organisation...the recruitment and sampling seem solid and in keeping with standards that would be expected of surveys of this kind ... It is broadly an interesting piece of work which offers an impressive dataset that gives an insight in public views of autism in NI.

However, both reviewers also make a number of observations that raise concerns about the framing and interpretation of the survey questions, and they also make some recommendations for improving the reporting of the methodology of this Volume. These suggestions relate to the precision of the reporting, clarity of presentation, as well as the basis for some of the claims made.

5.2. Context and rationale

Firstly, there were no specific research questions stated for the survey, which means that the basis for the specific construction of the questions is rather vague, and any analysis would remain necessarily exploratory. While exploratory approaches to data collection are acceptable in general, "... surveys are not well suited to carrying out exploratory work ... [because this would be] inefficient and ineffective" (Robson, 2002; p.234). A stronger approach would be to aim to answer specific research questions such that particular analyses and comparisons can be planned *a priori* (Cohen et al., 2011). Overall, while the data presented have good external validity through being sampled in a representative way, this feels like a missed opportunity to more fully explore the issues relating to autism knowledge and awareness amongst the general public because planned, interpretive comparisons were not reported.

Secondly, there is a lack of definition of key terminology, and a lack of evidence provided for the central assumption underpinning the report. Poverty is clearly a central concept for the entire BASE report, and Volume 2 mentions poverty and the "poverty trap" (e.g. p.28) a number of times. However, there is an opening assumption in Volume 2 that individuals with autism need to be helped "out of

poverty” (p.8) and that they are subject to a “poverty trap” (p.8). However, definitions of both terms are not provided and nor, in this Volume, is any evidence that supports the central claim that autistic individuals are indeed living in poverty.

In addition, the central concept of social inclusion is not defined or conceptualised in this Volume. Reviewer D in particular flagged this lack of definition as problematic, since social inclusion is a very broad concept that can be interpreted and defined in different ways:

The challenge of such a piece of research is that there are no doubt different views of what ‘social inclusion’ might look like. Put very simply, this is often characterised as approaches which focus upon the individual (in this case, the autistic person) and those which focus upon broader context (e.g. public attitudes, employment opportunities). So, these are ‘individually-focussed’ and ‘socially-focused’ interventions – and it is important to recognise that these can co-exist. A researcher who is interested in the public’s knowledge of autism with an interest in promoting social inclusion must engage in all these different views – and this is a difficult task.

The survey reported in Volume 2 attempts to engage with a range of perspectives (e.g. public awareness, views of different types of individually-focused interventions, views of equality of opportunity). Nevertheless, it would have been very helpful in terms of clarity (and probably would have improved the quality of the research) if the different perspectives had been more explicitly unpicked. By not doing this there are, in my opinion, some sections of the report which are ambiguous and analysed from a relatively narrow perspective.

This reviewer highlights particular sections where such a narrow perspective is of particular concern (causes of “co-occurrence” on p.23, and “treatment”, p.25), and we include more detail about these issues in the relevant sections below.

5.3. Methodology

An important point relates to the internal validity of the survey, i.e. the construction of the questions and how this may (unintentionally) bias, conflate, or confuse responses. For the most part, much of the interview schedule used in the survey seems straightforward and easy to understand, but there are places where the survey contained some ambiguity, which could have created potential difficulties when questioning the public. Usually, when describing survey methodology, a pilot study should be carried out to check that the questions are fit for purpose and clearly understood by target respondents. One would also expect a clear explanation to be provided of how questions were revised or refined as a result of the pilot study (Robson, 2002). A pilot study of the survey questions was conducted. This is good, but the only information provided in Volume 2 about the pilot is this statement:

Before being used in the main NILT Survey, the questions were pilot tested with 60 participants from the general public (p.10).

Therefore, it would be helpful to have more information about the pilot study, for example, to describe how the respondents were selected, the characteristics of the pilot sample (and how that related to the population), whether the participants understood the questions being asked, and what changes to the questions, if any, were made on the basis of the pilot findings.

The extent to which pilot participants understood the questions being asked is an especially important point, given that there are specific concerns about the clarity of some of the questions included in the survey. First, given the central aim of the survey and headline finding relating to overall awareness of autism amongst the general public, it is worth noting that by providing respondents with a definition of autism and then asking them if they have ever heard of autism may serve to inflate estimates of awareness. In addition, the question about awareness of autism asks about autism, Autism Spectrum Disorder, and Asperger Syndrome in the same question. This makes it impossible to ascertain whether there are differences in awareness between these different labels and,

indeed, whether respondents even recognised these labels as describing cognate difficulties. In other words, there is a lack of specificity regarding what respondents were aware of (or not).

Similarly, question 15(a) “Autism is a life-long disability and you can do nothing about it”, is double-barrelled. It is unclear, therefore, whether the respondent is agreeing with the notion of the “lifelong disability” or that “nothing can be done”. This distinction is important in terms of the interpretations made and conclusions drawn from this data (see Section E below).

Second, Reviewer D highlighted particular concerns relating to two main sections of the survey:

1. *‘Co-occurring diagnosis’*: Question 7 (p.53), reflected in Appendix 23, 24, 25 and 26; and
2. *‘Treatment and support for autism’*: Questions 9 (p.55) and 15(a) (p.59), reflected in Appendix 27, 28, 29, 30, 31, and 48.

Co-occurring diagnosis – first, the overall framing of the question as posed in the survey and as presented in the main body of the report are out of alignment. The question (p.53) asks: “Could you tell me whether you think people with autism have ... [e.g.] a type of learning disability?” (our emphasis). This is, in essence, a question about *definition*, i.e. whether autism is defined as *being* a learning disability. The question does not ask anything about co-occurrence and yet the main report labels the data in response to this question as: “Figure 16 – Perceived probability of co-occurrence of autism with learning disability” (p.23). This is misleading. If the question had been about co-occurrence it would, we suggest, have been phrased differently e.g. Could you tell me whether you think that people with autism are also likely to have ... [e.g.] a learning disability? The problems with the interpretation of this data are expanded further in Section E below.

'Treatment and support for autism' – the questions about 'treatment' (and its interchangeability with the word 'intervention') are also somewhat ambiguous, for example, to what extent is it likely that the general public will have a clear sense of what 'behavioural interventions' are or what 'other non-drug related therapies and supports' are? Moreover, it is unclear what 'effective in helping people with autism' really means. The targeted outcome of the treatments / interventions is not explicitly stated in the line of questioning which means it is unclear what participants are judging 'effective in helping' to be. Arguably, the stem question "How much do you agree or disagree with the following statements about the **treatment** of autism?" (original bold, p.55) suggests that the interventions are focused upon 'correcting' or 'curing' autism as a condition. Nevertheless, the language that follows ('effective in helping people with autism') has wider implications. Overall, this ambiguity in questions means that it is unclear how respondents are interpreting the language of treatment and intervention, and on what basis they judge the treatments to be effective.

Overall, the sampling part of the methodology was strong through being part of a standardised survey instrument. While all surveys are prone to recruitment bias (i.e. the extent to which those participants who take part in a survey are different from those who do not), the team have undertaken weighting to compensate for this. This could have been strengthened further in relation to calculating the response rate. There was a recommendation from Reviewer C that the formulae from the American Association for Public Opinion Research (n.d.) for calculating response rates should be used. This is because these formulae standardise the reporting of response rates and improve the transparency of quality judgements because they control for nonresponse bias in more robust ways than traditional measures.

5.4. Analysis

Linked to the point in Section A (concepts and definitions) regarding the absence of any specific research questions to guide the survey design there is, as a corollary, a rather descriptive approach taken to the analysis of the data. Much of the data is presented according to descriptive summaries for each of the

questions. While this is a reasonable thing to do in terms of presenting findings simply for a wide audience, it does mean, as also noted earlier, that there is a missed opportunity for building more interpretive and meaningful pictures of the data. There was some attempt to do this via the use of a logistic regression model in relation to some of the background and demographic variables from the survey (e.g. Gender, Higher Education, Ethnicity), but these are not used across all appropriate questions and there is no further cross-tabulation of survey responses to establish possible links between responses. For example, those who know, and do not know, someone with autism may have differed in important ways in their responses to some of the questions. As Robson (2002; p.235) notes, a survey can “ ... provide explanations of the phenomena studied and the patterns of results obtained ... [but] what is required is a sophisticated analysis of the detailed pattern of correlations.” Unfortunately, such an analysis was not presented here, which means that useful explanations for, and interpretations of, the data are difficult to deduce.

As a minimum, then, it is necessary to include some introduction to, and description of, the logistic regression model and how this relates to what is presented in the text. At the moment, the rationale for including the regression model is not clear e.g. why was the summary of background variables shown in Appendix 6 not also applied to other survey responses?

Reviewer C also felt that the references to the odds ratios from the model in the text (e.g. ethnic minorities were 12 times less likely...) would be confusing to readers without additional explanation because these refer to ratios of odds not probabilities. For example, someone looking at Figure 2 could wonder how a ratio of 12 is derived for this comparison. Additionally, the inclusion of an ethnic minority comparison group is questionable, given that it is based on only 25 respondents and presumably combines quite disparate ethnic groups.

It would be clearer and more transparent to provide detail of how respondents answered the open questions, including whether they were transcribed verbatim by the interviewer, or coded in situ by coders to a response frame. Moreover, the

use of these open responses in this survey cannot properly be described as a 'qualitative approach' given the way that the data are analysed and presented, i.e. using frequency counts to describe the data.

5.5. Findings

The structure of the results is broadly clear, generally taking the parts of the interview schedule section-by-section. However, there would have been greater clarity in the reporting of the data had the authors used the actual wording of questions in the tables of the main report. Given the ambiguity in the wording of two main sections of the survey, as outlined above, there is some concern as to how this ambiguity has been handled in the presentation and interpretation of the results. Expanding further on the two main sections outlined in Section B:

'Co-occurring diagnosis': (p.23) As noted above, the authors present the responses as a probability of a co-occurrence of other 'diagnoses' (e.g. "Figure 16: Perceived probability of co-occurrence of autism with learning disability"). This is presented as evidence that the public do, or do not, understand the nature of co-occurrence in ASD (p.39). However, as already explained, this was not the question posed to respondents and the question could be interpreted as being about the definition of autism, rather than about co-occurrence. Nevertheless, despite this ambiguity, it is clear that only one interpretation of the data is presented, relating only to co-occurrence. Given that specific research questions are not stated for the survey, and there is slippage between actual question wording and the presentation of data, this suggests that there could be some bias in the reporting and interpretation of the findings.

'Treatment and support for autism': (p.25) As noted, there is also ambiguity in the phrasing of the questions about what is meant by the terms treatment/intervention, and about how to judge the treatments to be effective. In terms of the latter point, the authors appear to assume effective interventions are those which help people with autism to achieve "optimal outcomes" (p.25) which is linked to no longer meeting "diagnostic ASD criteria" (p.27) i.e. to remove or reduce autism-related behaviour. This is highly problematic because

being 'effective in helping people with autism' could mean so many things beyond removing behaviours that characterise the diagnosis. Nevertheless, 'no longer meeting diagnostic ASD criteria' is the only interpretation being placed upon this data by the authors. In the words of Reviewer D: "*this is unhelpful and invalid*".

5.6. Conclusions

The problems identified in Section B above, relating to the double-barrelled phrasing of the question: 'Autism is a life-long disability and you can do nothing about it', are compounded in the interpretation of the data and the conclusions drawn. Specifically, the authors claim that: "The NILT Survey respondents were more optimistic about outcomes for individuals with autism, with more than half disagreeing that autism was a lifelong disability" (p.42). First, it is impossible to know whether respondents were focusing on the 'life-long disability' or the 'do nothing about it' part of the question when giving an answer. Disagreeing with the latter does not automatically mean disagreeing with the former.

Second, the authors clearly suggest that disagreeing that autism is a lifelong disability is a welcome finding; in addition to the previous quote they say: "The idea that autism is necessarily life long was not supported" (p.39). This is a very surprising stance to take given the decades of scientific research that has firmly established autism as a pervasive (lifelong) developmental condition (e.g. Magiati, Tay & Howlin, 2014; Howlin, Moss, Savage & Rutter, 2013; Piven & Rabins, 2011). To suggest otherwise is very misleading and not based on the leading scientific evidence regarding long-term outcomes in adulthood.

Furthermore, there were a number of claims in Volume 2 that Reviewer C indicated were not supported by the data presented, specifically: "...half of the general population knew someone personally with autism and clearly others had heard about autism from those people" (p.16). Reviewer C suggests that "*this seems to be a reasonable assumption but goes beyond what is evidenced by the data.*"

Question 6 of the autism module in the survey asked: “How common do you think autism is in our society today? Would you say that 1 in 100, 1 in 1,000 or 1 in 10,000 people have autism?” Respondents were shown a card with each of these response options, plus a ‘don’t know’ category. There is, therefore, a 33% chance of guessing the correct answer (1 in 100). With reference to the results shown in Figure 10 on p.19, the 38% who chose the correct response of 1 in 100 may not reflect firm knowledge. Consequently, it is not reasonable to conclude from this data that “38% of the public were aware of this increase [in prevalence rates]” (p.38).

On p.36 the authors conclude that:

Overall, issues such as finding employment, mental health problems, dependency on parents, and individual differences were mentioned infrequently by the respondents, and this was likely a reflection of the lack of focus on these issues by the UK media (Huws & Jones, 2010).

While this may seem intuitively appropriate, this conclusion is not warranted by the data presented because the survey did not ask about UK media exposure or awareness. Similarly, the conclusion that “These findings indicated that local and international efforts to improve autism awareness amongst the public have been successful” (p.43) is not supported by the data presented because the survey did not ask any questions about where and how respondents had learned about autism. The linked conclusion that: “These findings clearly indicate that there is no need for a general ‘autism awareness campaign’ in Northern Ireland, i.e., precious resources would be wasted as many people would be targeted unnecessarily” (p.36), likewise is not supported by the evidence. Indeed, this is a somewhat naïve statement since it should be the case that levels of public awareness need to be maintained; awareness is not something which is ever-present, or can be taken for granted, once achieved.

5.7. Main conclusions from the review of Volume 2

Overall, the findings from the *Northern Ireland Life and Times (NILT) Survey Autism Module* reported in Volume 2 of the BASE report has good external validity through being sampled systematically via a well-established and respected survey methodology. While there are some areas where the reporting of the procedure and the data could be improved and clarified, the survey module was completed by a large number of respondents, meaning that there is much interesting data reported in this Volume from a sizeable sample.

However, given the size of the sample, the analysis of the survey data is exploratory or descriptive at best, and so there is a missed opportunity to present a more robust and interrelated picture of the data. There is the potential to plan and search for patterns in the data based on empirical evidence and/or theoretical frameworks, which would provide stronger insights into potential causal relationships between variables. This would help to move beyond the rather simplistic reporting of the responses to each question as at present in order to develop a more conceptually informed and coherent approach. For example, returning to the point raised by one of our reviewers regarding social inclusion and the differences between ‘individually-focused’ and ‘socially-focused’ support, there is the potential to group variables in ways that would allow a targeted analysis into attitudes towards these concepts, and the background characteristics of respondents that may relate to those attitudes.

Finally, questions were raised through our review about the poor internal validity of at least some of the survey questions, specifically relating to autism as a lifelong disability, the co-occurrence of autism with other conditions, and the treatment and support for people with autism. These questions were ambiguously phrased and so, as a minimum, the data relating to these questions must be treated with substantial caution. In particular, these findings must not be used to bolster a broader point suggesting the public need educating about which interventions work and which do not. There may be value in the aspiration to improve knowledge amongst the public about the relative quality of interventions, but this report does not provide meaningful data which supports a

position about the relative value or preference for one type of intervention over another. There were also other places in the report where there was some overreach between the evidence presented and the conclusions drawn. This means that it is difficult to make distinctions between conclusions that we may have more confidence in and those where we have less confidence (due to overreach and/or threats to internal validity). Our main recommendation, then, is that readers should draw their own conclusions from the data presented for each question from the NILT Autism Survey Module, rather than relying on the interpretations of the data provided by the authors of the BASE report.

6. Review of BASE project (Vol. 3) secondary data analysis - Dillenburg, McKerr, & Jordan, March 2014

6.1. Introduction

With specific reference to the content and purpose of Volume 3, Dillenburg, McKerr and Jordan (2014; p.3) state that: Volume 3 of the BASE report “represents secondary data analysis of all relevant NI governmental and related departmental etc. datasets focusing on education, employment and poverty.” In this Volume, the main purpose of the BASE report is reported in the introduction as being: “...to provide baseline data for individuals with autism against which the effect of the Autism Act (NI) and associated Autism Spectrum Disorder (ASD) strategy can be measured.” (p.3). In the discussion section of the Volume, the aims are outlined as being to use “a large number of existing datasets with the aim to enhance understanding of the extent, distribution and causes of inequality and social exclusion in Northern Ireland society and the consequences of policies and actions aimed at their reduction” (p.112).

In outlining the justification for undertaking secondary data analysis, Dillenburg et al. (2014) highlight that although strategies to support individuals with autism and their families have been developed in Northern Ireland these have not been based on a prior baseline of assessment of need. Meanwhile, there are large datasets in existence and these have not been subjected to autism-specific secondary data analysis. Furthermore, Dillenburg et al., (2014) highlight that the research in this Volume is in line with guidance from the United Nations General Assembly (2012) that encourages member states to “undertake to collect appropriate information, including disaggregated statistical and research data, on ASD, developmental disability (DD) and associated disabilities” (p3).

Reviewer E notes the key strengths of Volume 3, namely that:

As the first comprehensive secondary data analysis of children and families affected by ASD in NI, this report is both timely and a very welcome addition to the acknowledged sparse evidence base.

Reviewer F also notes strengths of Volume 3 as follows:

A key strength of the report is its systematic approach to searching for secondary data that adequately informs understanding of the life experiences and opportunities of children and young people with ASD and the relationship between ASD and indicators of disadvantage. In conducting the search for secondary data sources, the researchers are also able to identify important gaps in the data particularly regarding post compulsory education and training opportunities.

Both reviewers acknowledge the wealth of good informative research included in this report and the work that has gone into it. The strengths of this Volume are discussed further below alongside methodological and reporting issues that are also apparent.

6.2. Context and rationale

If we turn to the full title of the BASE project it is clear that the focus of the research is on ‘Helping the most vulnerable out of the poverty trap and reducing inequality’. This means that poverty is a central concept for the project and so should be defined very carefully and precisely. Given that Volume 3 has the power to establish an evidence-base for inequality in relation to people on the autism spectrum, it is especially important that such key terminology is defined. However, there is no definition of poverty provided in the report which means that it is not clear how the concept of poverty was operationalised statistically in the analyses and, therefore, whether the data presented address this core concern. The OECD (2010) has a clear definition: ‘the poverty rate is the ratio of the number of people who fall below the poverty line and the total population; the poverty line is here taken as half the median household income’ (n.p.n.). In fact, “The UK government, the European Union and many other countries use 60

per cent of median household income as the poverty ‘threshold’” (Mack, 2016; n.p.n). However, this definition is not provided in the report.

This point matters because claims are made for the data that may indicate relative disadvantage for families that include people on the autism spectrum, but this may not equate to poverty according to the standard definition (Mack, 2016). An example from the text helps to illustrate this point. On page 16 of Volume 3, Dillenburger et al. (2014) state that:

With regard to poverty, MCS data revealed that families of CWA [children with ASD] had significantly lower incomes (up to 12% lower) and experienced considerably more unemployment (up to 20% more) than families of children without ASD (COA).

While the household income data from the Millennium Cohort Study uses equivalised income as a way of comparing households (this is a measure of household income that is adjusted for the size and composition of the household; Eurostat, 2014), this is not identical to the median household income threshold that indicates poverty. As Carter, Imlach Gunasekara and Blakely (2013) state: “while relative income inequality... and poverty usually travel together, they are not completely in step” (p.26).

There is considerable debate and discussion in the academic literature about the measures that are used to discuss and define poverty (e.g. Kuypers & Marx, 2016; Logue & Callan, 2016; Perry, 2002), the detail of which is beyond the scope of this report. Nevertheless, the key message is that definitions matter in this area and there is a need for precision in the claims being made. As a minimum, clear definitions needed to be provided in Volume 3 of the BASE report, including a brief rationale for their use, as well as some acknowledgement that there are debates about how measures of household income should be used and interpreted in relation to poverty.

Alongside clear definitions of key terms, Volume 3 of the BASE report also needed to provide clear research questions for driving the analysis and framing the findings and conclusions. Johnson (2014) highlights that the development of research questions should be the first step in secondary data analysis, before identification of the dataset/s and subsequent analysis. Whilst the objectives of the study were broadly outlined at the beginning of Volume 3, these were very broad objectives and specific research questions were not stated. This resulted in a lack of clarity about what the researchers wanted to find out from the analysis of each dataset. A clear set of research questions would have given the reader a better, more transparent, sense of what the research team wanted to find out from each stage of analysis. An example of this, given by Reviewer F, was that it was unclear why attainment data did not feature in the analysis of the school census, especially with regard to proportions of autistic pupils reaching benchmark levels in mainstream schools.

6.3. Methodology

Key issues in secondary data analysis relate to: sampling the data that will be used, interpreting the secondary data, cross-analysing data and making recommendations regarding the data (Goodwin, 2012). It is an empirical approach, so the focus of investigation and the research questions should guide the method, the collection, analysis and interpretation of the study (Creswell, 2009). Bearing in mind these points, the reviewers noted that appropriate criteria were used for the selection of the datasets; the quality of the datasets were robust; a range of experts were consulted, and the datasets selected were also appropriate for addressing the broad aims of this study. Furthermore, the range of organisations and people contacted and followed-up was applauded and the 'snowballing' to identify potentially hidden datasets were well described. Providing information across the lifespan from young children to adulthood is a good way of presenting the data. Both reviewers commented that it appears that all relevant data sources have been utilised in this report and that the reasons for considering which data sources were considered but excluded from analysis were outlined very clearly.

Both reviewers were confident that the sampling decisions were adequate. Reviewer F states that:

The selection of datasets appears to have been undertaken rigorously and the methods of analysis appear to be objective. I would personally have liked to have read more detail about the nature of the variables available in the some of the datasets (the level of detail provided by the research team does vary from dataset to dataset) including information about samples. For each dataset, key findings are summarized effectively in part one of the report with links drawn to the wider literature which places the findings in context.

As data quality is of utmost importance in secondary data analysis (Saunders, et al., 2011), Reviewer F gave some detailed comments on the quality of some of the datasets (see Table 1).

Table 1: Quality of data sets utilised in Volume 3, as reported by Reviewer F

The data set	Quality and focus	Design of study and relevance to this study
Millennium Cohort Study (MCS)	The best quality data on the early life trajectories of a nationally representative sample of young people who were born in 2000.	Its extensive coverage means that it is well placed to provide contextual information about the life experiences and life chances of this group of young people and their families. It is invaluable in providing data on diagnosis and support through the child's early years.
NI school census	As a census dataset, coverage is for the whole school population, with data disaggregated by different types of SEN and extensive coverage of attainment-related variables along with data on school attendance, as well as proxy measures of socioeconomic status. Issues of inconsistency of the statementing processes and other limitations to the data are recognised	This is the best quality data for providing a national picture of prevalence of ASD among children in school as well as school attendance and academic outcomes.
Young person's behaviour and attitudes survey	This dataset is specific to NI and only one sweep (2010) gathered data on ASD. As a cross-sectional sample study with relatively low school level response rates and incomplete coverage of all types of schools (no special schools were included for example) the YBPAS is less useful than the pupil census or MCS.	Despite weaknesses it nevertheless provides a useful comparison of findings with the other surveys and some details on pupil's attitudes and experiences.
School Leavers' Survey	Data for this survey were provided at aggregate level only, so this limits any analysis. Also, the SLS does not include leavers from independent and special schools.	Data on school leavers' trajectories is important and it is unfortunate that more detailed individual level data were not available to the researchers (data confidentiality issues notwithstanding).
Further Education (FE)	This is a very useful source of data. The issues with the way this dataset is administered and data is recorded point to some of the challenges of working with secondary data, but these are beyond the control of the research team.	The researchers acknowledge the gaps in research with young people in FE. This is particularly crucial given the aims of the present study.
Higher Education (HE)	Data providing good coverage of disabilities appears to have been available from HESA (with important caveats about self-declaration).	The reviewer did not really get a sense of the scale and scope of the data that the researchers were able to gather from HESA.

Reviewer F suggested further comparative analysis would have been useful in relation to FE and some of the datasets (such as the MCS) would have facilitated this. However, the reviewer recognises that this could be beyond the scope of the study, as it would have required significant additional resource for analysis. Reviewer F also highlights that limited detail was provided about the nature of the employment data that was available to the researchers. There is also no indication given in the Volume about whether or not any of this data had been purchased, or if the resources had been available, whether there was any data that the researchers could have purchased that would augment these analyses. For example, data from UCAS and HESA might have provided further detail on participation and engagement in Higher Education. The incorporation of such additional data is certainly a consideration for future research in this area.

Reviewer E noted that there needs to be greater clarity about *where and who the measures were obtained from*. There were inconsistencies in how measures were collected, with no information about some of the sampling decisions. For example, in reading the Executive Summary, it was unclear which data sources provided which information and whether it was UK or NI based. More specifically, it was unclear why autism prevalence was presented for the UK and not for NI. Reviewer E argues that prevalence and/or sample sizes of those with autism in each dataset would be useful to include in the Executive Summary.

6.4. Analysis

Although the research questions might vary from dataset to dataset, and would be constrained by the data that is available, Reviewer F highlights that stating the research questions explicitly would have given a clearer focus to the analysis section. This lack of focus for the analysis also made it more difficult to determine whether the most appropriate analytical techniques were used. Ostensibly, the largely descriptive analysis is appropriate but there might have been opportunities for more complex multivariate analysis had this been indicated by the research questions.

In analysing secondary data, it is also crucial to be transparent about *the points at which measures were taken*. For example, Reviewer E notes that prevalence of autism was measured at ages 5, 7 and 11 in the original research. However, when cross-tabulating children with and without autism with other survey variables, it was unclear when the age of the autism measure was taken for this study. Reviewer E therefore questions whether the measure is a cumulative or a cross-sectional measure. Similarly, autism by Organisation for Economic Co-operation and Development (OECD) income was measured at age 9-months, 3, 5, 7, and 11 years. It needs to be made absolutely clear which measure was reported in the BASE study.

6.5. Findings

In relation to the findings reported in Volume 3, both reviewers highlight that this is a valuable and detailed piece of research. The main findings are presented by dataset, key data is largely well presented and important findings are signposted, making it relatively easy to follow. Within each dataset findings are organised by themes with links to the wider literature or other sources of data, as appropriate.

Reviewer F highlights that although the analysis is largely descriptive it is thorough and appears appropriate for the data that is available. Reviewer E notes that there was, nevertheless, a lot of information that could at times be hard to digest as the information was dense, particularly when describing each of the datasets. It would help if it was made clearer that the 'Introduction' was an 'Introduction to the dataset' and thus the section could be skipped without detriment to understanding the 'Key Findings' section.

While the use of themes to organise the findings was useful, some sections lacked a clear narrative (including concluding remarks) to help the reader navigate the various sections. A good example is Section 3.2. on 'schooling for children with ASD'. This presents the findings for the Annual School Census (p.18- p.23). In the absence of clear research questions, the order of each section was not clear, nor how they linked together (it might have been as simple as

using effectively numbered paragraphs, but this would have helped the narrative structure of the piece).

Another presentational point relates to the fact that the section headings are inconsistent in both the methods and the results sections, where headings are listed either by dataset or by topic or phase. This makes it somewhat confusing both to understand the content and qualities of the different datasets that were being used, and their link to the different themes that the researchers have adopted. At the end of the presentation of Findings it lacked a section to draw together the key themes from the analysis across the different datasets. This was a missed opportunity to draw together key findings and communalities from across the various data sets. The Executive Summary is also not very clear in terms of structure. It starts by saying the report is structured by age stage, but the key findings are presented by topic. It would benefit from following the same flow as the 'Main findings by dataset' section and the main age-based order of the body of the report.

Reviewer E highlights a crucial point related to the importance of significance testing *"the report would benefit greatly from some significance testing to some of the findings from the administrative data sources if they are to inform policy."* When using DENI School Census (4.2), School Leavers (4.4), FE (4.5) or HE (4.6) data, the results were presented as a descriptive profile and significance between groups or variables was not tested. Reviewer E acknowledges that results should not be interpreted as important only if statistical significance is achieved, as significance is very dependent upon sample size. However, when results are discussed with phrases such as "much higher" or "slight preference" or "slightly higher" this can be misleading in the absence of significance tests. Sometimes a 1% or even 0.5% difference is discussed in terms of a big difference (p.58), but at other times not so (e.g. Figure 5.8 where a 1% difference between children with and without autism is described as "very similar" (p.91). Reviewer E stresses that the results really do need to have statistical testing applied in order to interpret the scale of any differences meaningfully.

Relatedly, caution and careful attention should also be given to how very small differences are discussed and presented graphically. Graphics are used very fully in the report for reporting the data, but some of these are potentially misleading as the scales are sometimes presented to exaggerate very small differences. Given that we do not know if the differences shown are statistically significant, much caution is needed when interpreting the data. Two specific examples illustrate this point:

- Figure 2.4 (Prevalence of Asperger's Syndrome for cohorts of pupils over time; p.59) has a scale from 0.0-1.2 with an increase from 0.8 to 1.02 being described as a "substantial peak" (p.58).
- The scale on Figure 2.10 (Attendance in Primary Schools; p.65) also only goes from 92.5 to 95.5; actual differences between groups are no more than 1.3%.

Overall, the figures in the section on prevalence of autism and Asperger Syndrome need a lot more care and attention, legends are also often incomplete and sizes of graphs fluctuate (e.g. Figure 2.9). To be comparable, Figures 2.3 (p. 58) and 2.5 (p. 60) need to be on the same scale, likewise all graphs in Figures 2.8 (p. 63) and 2.9 (p. 64).

Reviewer F suggests that it would have been clearer to present more abridged tables within the text rather than in the appendix, where it is more difficult to find them. Reviewer F also suggested that she would also have liked to have seen more use of N (sample size) in the main data tables, rather than in the appendix, especially where sample size is small.

6.6. Conclusions

Although there were no findings from the various data sources or in the recommendations about applied behaviour analysis-based interventions, there is mention of these in the section on the Millennium Cohort Study (MCS) (p.16) and in the Discussion section on page 114. These paragraphs are out of place as they do not link back to any of the data or the findings in Volume 3. Related to this

point, there seems to be some overreach with regard to some interpretations of the data. For example, Dillenburger et al. (2014; p.16; our emphasis) state that:

According the [sic] MCS data, the behavioural symptoms of ASD were the main reasons for support needs, while the child's learning difficulties **caused concern** for only about one fifth of the families. This was interesting given that it is generally reported that 50-70% of persons with ASD have co-occurring learning disabilities; yet, according to the secondary data analysis of the MCS, behavioural symptoms associated with ASD evidently caused more concern than co-occurring learning disabilities.

First, this statement seems to come from the following data, reported on p.39:

Most parents of CWA (64%) said that autism/Aspergers, i.e., behavioural issues related to ASD, was the reasons for their support needs (Appendix 1.30). Fewer than one fifth of the parents of CWA (19%) stated that the child had support needs as a result of their learning difficulties (Appendix 1.31).

However, this statement comes from prevalence data of children with special educational needs/additional support needs due to autism/Aspergers (p.151), which says nothing about whether or not the reported support needs “caused concern” for families.

Second, Dillenburger et al. (2014) then discuss a study by Howlin, Savage, Moss, Tempier, and Rutter (2014) – not related to the MCS – to state that a “recent study of long-term outcomes paints a depressing picture” (p.16) and imply that this is because applied behaviour analysis-based interventions were not available to Howlin et al.'s participants. The key argument put forward is that positive results from such interventions will not be replicated in the UK because they are not routinely available to service providers. The Howlin et al. (2014) study is misrepresented again (see comments on Volume 1) as the authors of the

BASE report claim that Howlin et al., (2014) predicted that the positive results reached in the US would not be achieved in the UK due to the fact that early intensive behaviour analysis-based interventions are not statutory in the UK.

6.7. Main conclusions from the review of Volume 3

The value in Volume 3 lies in its comprehensive overview of secondary data sources. Overall, the analysis is appropriate and some useful substantive findings have emerged that could inform policy and practice in Northern Ireland. The report provides important findings in relation to the fact that more children with autism live in families that have a greater experience of poverty and/or financial hardship, and more live in families that do not work or where a least one parent does not work. More children with autism miss school, compared to other children, and they also have less positive experiences of school.

Various stakeholders may be aware and supportive of inclusive education policies but the experience of children with autism at school shows that the challenges of successfully educating them in mainstream schools are yet to be fully met. Parents also need to be supported to find ways (back) into employment. Acknowledging the lack of a reliable adult prevalence rate, Volume 3 draws attention to the likely increase in the adult prevalence rates going forward given the increases that are being observed in the child population. This will undoubtedly have an impact on a wide range of services and have financial implications.

A further strength of Volume 3 lies in its identification of gaps in current data collection, and highlighting opportunities for better data collection. Recommendations for gaps in data collection are detailed and specific to the different surveys. In addition, there are separate recommendations made for data collection protocols for GPs and these are helpful.

That said, the empirical analysis was largely descriptive and would have been strengthened by being directed by a set of researchable questions. This could have enabled the researchers to explore further interactions between ASD

prevalence and other key variables. There were also key issues that need to be addressed to strengthen the presentation of the analysis and these include the need to:

- explain which measures were taken and whether they referred to UK or NI data;
- be more transparent and consistent about how very small differences were discussed and presented graphically;
- report the findings more coherently; and
- present Figures more accurately.

Therefore, despite some of the strengths of Volume 3, some caution needs to be exercised with regard to the direct interpretation and presentation of Findings in the report. Most importantly, as Reviewer E states: *“the report would benefit greatly from some significance testing to some of the findings from the administrative data sources if they are to inform policy.”*

7. Review of BASE project (Vol. 4) qualitative data analysis - Dillenburg, McKerr, & Jordan, March 2015.

7.1. Introduction

Volume 4 presents the findings from the qualitative phase of the BASE Report. With reference to the content and purpose of Volume 4, Dillenburg et al. (2015) state:

The present volume adds a qualitative perspective to the project by exploring the experiences of individuals with autism, their parents or carers and the staff (both public and private sector) who are tasked with supporting service users affected by autism. Volume 4 reports on how autism policies and strategies translate into services that aim to help vulnerable individuals and their families out of the poverty trap and social exclusion (p.6).

The findings draw on an online survey completed by a range of professionals (n=798), with a number of those professionals also participating in individual interviews. Furthermore, service user perspectives were drawn from individual interviews, focus groups and surveys. Focus groups included eight children with autism who attended secondary schools. Ten students who were enrolled in Further and Higher Education completed online surveys. Semi-structured interviews were also conducted with adults with autism (n=5) and parents of children with autism (n=14).

Reviewer G highlights some strengths of the Volume, namely that:

Overall this is a thorough and generally well written volume which reports on a survey and small number of qualitative interviews, giving some helpful information and recommendations about awareness and training about autism for professionals and the experiences of education, health and social care services by some young people and adults affected by autism and their families.

Reviewer H makes the following comments on the strengths of Volume 4:

Qualitative research on the life experiences of people with autism and their family members who use (or reject/cannot access) services is a worthwhile project. ... I think the inclusion of the views and experiences of adults with autism is a strength and I liked the way each chapter finished with a summary of key points, as this helped maintain the report's focus and coherence.

There are different criteria for judging quality in qualitative research (Oakley, 2000), and these are often dependent on discipline or theoretical background (Sandelowski et al., 1997). This has resulted in arguments that different qualitative methods need to be appraised in different ways (Kuper et al., 2008). Noting this, Reviewer H acknowledges that while there is some debate around indicators of quality in qualitative research, the following features from Mays and Pope (2000) provide a useful framework. These include that research should have a relationship with theory; be credible to the participants and readers; and have rich description of cultural and contextual features. In addition, researchers should critically reflect on their own research; there should be evidence of searching for counter-evidence; and there should be transparency and discussion of the extent to which the findings are transferable to other settings. Based on Reviewer H's thorough review of Volume 4, this suggested framework therefore informs our approach to the review of this Volume.

7.2. Context and rationale

Reviewer H highlights that there is no explicit theoretical engagement in Volume 4, in terms of either existing theory or theory building. The aim of the BASE project is to generate a baseline picture of autism awareness and training among professionals and the experiences of autism diagnosis, support, education and employment among people with autism and their families. The research is not used to generate theory about what the researchers learn is going on (a similar lack of clarity regarding conceptual orientation was also discussed in relation to the reviews of Volume 2). Instead, statements about what most or many of the

participants thought or experienced are proffered in a descriptive way, and recommendations made. There is the implicit assumption (working theory) that early diagnosis will lead to treatment, which will reduce autistic behaviour, and which will reduce the chances of the person and their family experiencing poverty. This train of causation is not subjected to critique in this Volume or explored with any of the participants. There is also an implicit assumption of a causal relationship between training in autism awareness and interventions and positive outcomes for the persons with autism which is, again, not evidenced or critiqued in the report.

In addition, as Reviewer G points out, there is an assumption that all children and adults with autism face a life of vulnerability, poverty and social exclusion. We recognise that the aim of the study is to help people with autism access the range of benefits and services they may need and that children and young people with autism and their families are far more likely to experience low incomes and social isolation (like most disabled children's families). Yet a concept like vulnerability is a loaded one and should, as Reviewer G points out, be problematised, as it implies a deficit model of disability rather than one which also emphasises the resources that families have (economic, social, agentic, cultural) in addition to the need to be supported by outside agencies. This point also relates to the lack of definition of poverty noted in the review of Volume 3, and poverty trap in the review of Volume 2.

Reviewer G also comments on the fact that there were a few occasions when the language used in the report was not quite appropriate, given the subject matter and the focus on the social (as opposed to the medical) nature of the research i.e. the 'Policies, strategies, and services for individuals with Autism Spectrum Disorder...'. For example, on page 9, (also on p.77 and p.115) education professionals are reported to like the "quirkiness of their [people on the autism spectrum] personalities". Although this is in quotation marks, the reviewer highlights that this comment, probably unintentionally, could reflect an attitude towards young people and adults with autism that they are 'different' from those working with them and could be seen as figures of fun. This could be interpreted

as an 'othering' of those who experience autism, which could come across as patronising and disrespectful.

Both reviewers highlight that Volume 4 does not have its own literature review, but cross-references to the literature in Volume 1 (which has substantial concerns regarding research quality and objectivity, see the review of Volume 1). Reviewer H highlights the importance of grounding the work in the extensive research literature on the experiences of people with disabilities in general and autism in particular, or their families and the people who work with them:

This needs to build on published accounts already available in the literature and to be reported independently of research on the knowledge, awareness and training of professionals (also a worthwhile project). Case studies, which genuinely explore the complexity of particular lived situations and bring the perspectives of service users and providers together, would also be worthwhile. The research reported here does not manage these things though it does contribute some useful insights into the situation for individuals in Northern Ireland.

Given that grounding in this extensive literature is missing, the findings and recommendations are, therefore, unable to discuss the experiences of participants in the BASE research in the context of other published findings.

7.3. Methodology

The second chapter of Volume 4 ('Introduction and research methodology' pp.17-25) is very short and does not explain the methodological rationale for the study in sufficient detail, for example, there is no explanation for why a combination of questionnaires, focus groups and interviews are chosen. A case study appears amongst the data but that is not outlined at the start of the Volume, neither is there any discussion of case studies in the methodology section. It is unclear why questionnaires are described as qualitative data. This leads Reviewer G to question whether this volume can accurately be described as

qualitative research or would be more accurately described as a 'mixed methods' study:

Given that qualitative research is concerned with in-depth data about people's views and experiences, and that at least one chapter (3) of this volume is drawn from a survey of 798 professionals, compared to 40 interviews, I am not sure it is accurate to call this volume a 'Qualitative study'.

There are also gaps in the rationale for the sampling. There is a need for a more detailed account of this rationale in order to make claims for transparency and validity. These gaps are as follows:

- There is no rationale for holding a survey for a large number of professionals in comparison with such a comparatively small number of young people and adults on the autism spectrum and their parents. How could this substantial difference in relative sampling impact on the findings? As a minimum, it is vital to explain this choice and to contextualise the Findings accordingly (including reference to concomitant limitations).
- It is not clear why interviews took place only with education professionals, rather than including other health and social care professionals too.
- Of the Health and Social Care Trust (HSCT) staff surveyed, the largest proportion (35%) came from the South-Eastern region. It is not clear why this might be the case or what the implications may be for the study's findings and recommendations.
- There is no explanation or detail provided for the selection of the employers' case studies, relative to those not selected.
- Eight children/young people were interviewed in focus groups: why was this number chosen or arrived at? Why was the focus on young people aged 15-16 – was there a particular rationale for this age group? What implications does this have for the study's findings?

We recognise that there may be good justification for these decisions, but they are not made explicit to the reader. Although participation in research (whether it is a survey or interview) is (or should be) always voluntary, it is still possible to select a sample, purposively in terms of size and variables and have a rationale for this.

In terms of methodological transparency, the report and technical information in the appendices make clear the sample size and the basic methods used: questionnaires (primarily with professionals), focus groups with the pupils and individual semi-structured interviews with the parents and adults with autism, plus some professionals. As already discussed above, the rationale for the different methods for the different participant groups is not transparent, nor is the bias in sampling towards professionals compared to service users. The interview schedules, focus groups and survey questions are available in the appendices and this greatly improves transparency around data generation. However, the report itself does not always make clear that a theme in the data, e.g. need for training in independent living skills, arises because a direct question about this is asked. Reviewer H was clear that a very close reading of the appended material was needed to establish what should, with proper reflexivity, have been transparent in the report.

It would be helpful to have a section somewhere in the methodology section or in the appendices, which explains any challenges in doing the interviews and obtaining informed consent from participants, especially those with autism and associated learning difficulties. The interview schedules are quite structured and include quite a few closed questions, requiring yes and no questions before elaborating further. It would have been useful to explain to the readers whether these questions worked well or whether the more open questions (more commonly used in qualitative research) such as 'How?' and 'Tell me about' questions, worked better. A section discussing some of these methodological issues would be useful for the reader and add to the report's (and the methods used) credibility.

In relation to the importance of a rich description of context as a quality indicator in qualitative research (Mays & Pope, 2000), this only really applies to the 'case studies' of businesses offering work experience where the interview basis for the data includes rich contextual detail. However, these are not case studies in the typical qualitative research sense of gathering multiple perspectives and using multiple methods to explore and describe the particularity and complexity of the case (e.g. Cohen et al., 2011; Yin, 1994), and so this is another example where defining the particular approach to this data collection is important and necessary.

7.4. Analysis

Of considerable concern in judging this as a report of piece of qualitative research is that the authors share no information on the data analysis process. The only comment on the analysis is that themes emerged from the data. The analysis appears to have been deductive, with little scope in the way the questions were posed for themes to emerge beyond the authors' frame of reference, or for inductive processes to be applied. As Reviewer H concludes: *"It is hard for the reader to have trust in the interpretations in the report with so little attention given to the analytic procedure."*

It would also have been more helpful to state the actual numbers of service users who were claimed to think or experience specific things, in the light of the small size of the sample. For example, the authors refer to 'most' or 'many' parents saying or thinking a particular thing, but there were only 14 parents included and they may have differed in important ways from parents who did not come forward. As Reviewer H notes: *"There is no implication that this is an intention of dishonesty, rather a lack of reflexivity"*. This is something we say more about in Section 7.6. below.

In contrast to the information provided for the case studies, the pupils' school contexts are not described beyond one being mainstream secondary and one being for pupils with special educational needs. This leaves readers largely

unable to judge the relevance of the views and experiences of the pupils for other settings. Similarly, readers are told very little about the life circumstances of the parent participants and of the adults with autism beyond their age, gender and employment status. It would have been much more meaningful if there had been (for example) pen portraits to get a sense of individuals or family units; instead, despite the very small numbers involved in the qualitative aspects of the data collection (i.e. not including the survey), data are aggregated into thematic trends, which implies a degree of generalisability which is not warranted by the sampling or the data.

7.5. Findings

There are a number of limitations in Volume 4 that reduce the external validity of the findings of the report. First, the sample size of service users is very small, and they participated via different methods (each with strengths as well as limitations). While rich qualitative data from small samples can be informative for other settings beyond those studied, the data would need to align with the quality criteria discussed here in order to do so. We have already indicated that this is not the case with the data and background information reported in Volume 4.

Second, none of the five adults on the autism spectrum who took part in semi-structured interviews were diagnosed at school (p.12) which is likely to mean their experiences would be different from those children and young people on the autism spectrum who are diagnosed at school age/pre-school age.

Third, Dillenburger et al. (2015) state that “Individuals whose first language was not English, or who used an alternative communication strategy (such as sign language or the Picture Exchange Communication System) were invited to take part, where unpaid facilitators were available” (p.133). It is welcome that the recruitment strategy was not constrained to those who may be more articulate and able to verbally express their views, however, it is unclear whether the diversity of communication preferences and needs of people on the spectrum was represented here. The authors comment that “Group discussions were lively,

articulate and relaxed, and the children talked easily about their wide range of interests and hobbies” (p.24), suggesting that it was only the more articulate children who were included. In addition, stipulating that only ‘unpaid facilitators’ could be included would have had an effect on who would be likely to come forward, namely those who have the social and financial capital to do so (e.g. Hammel et al., 2015).

In summary, the small numbers of service users included in this Volume, coupled with limitations in making generalisations from their experience raises questions about “*whether there was sufficient engagement with service users to come to conclusions about what might apply in other settings or to make recommendations*” (Reviewer H).

7.6. Main conclusions from the review of Volume Four

Reviewer H raises a concern about the lack of reflexivity shown throughout this Volume, which has important repercussions for the conclusions that may be drawn from the report. As is often the case in research, definitions and understandings of key terminology are debated and, sometimes, contested (and this is why the use of specific definitions is so important) but we use here the definition of reflexivity by Archer (2007; p.4) to illustrate the core idea: “... the mental ability [of people] to consider themselves in relation to their (social) contexts and vice versa.” In research terms, this (generally speaking) means that researchers need to be self-aware and critically examine how their own professional and personal identities impact on the research they conduct and engage with. Reporting conflicts of interest (see review of Volume 5) is a rudimentary and technical part of this, but reflexivity should go much deeper to examine where existing assumptions and preferences may filter or shape research choices and interpretation. The lack of clear research questions, rationale for methodological choices, or acknowledgement of limitations throughout Volumes 1-3 of the BASE report are indications that reflexivity has not been adequately addressed.

There is further evidence of this in Volume 4 where Reviewer H notes that:

The report authors' personal and intellectual biases toward the need for early behaviour analytic intervention are not made plain at the outset, as quality criteria would demand. Yet these permeate the report, repeatedly finding their ways into the findings and recommendations despite the participants not raising this issue themselves. There is no reflection on how the research questioning and methods selected influenced the data presented; the report merely presents these as truths unadulterated by the subjectivities of the researchers.

Indeed, there are tenuous links made between some of the findings and the recommendations. For example, The Executive Summary does not make the relationship between the findings/ recommendations and the data generated in the study clear. As with Volume 1, for example, there is a lack of trace or audit trail about the data or information that informs the recommendations. There are four recommendations regarding diagnosis; four recommendations regarding training; five recommendations regarding education and intervention; and seven recommendations regarding employment and quality of life. Most readers would infer, as the report implies, that these recommendations are based on the data from Volume 4. However, as Reviewer H notes:

Sometimes this [relationship] is the result of the study data showing that there might be an issue with something and the study authors presenting their preferred solution to it, based on their other work, knowledge of the wider issues or preference.

To illustrate, Dillenburger et al. (2015; p.54) state in the section about diagnosis that: "It is clear that Trusts are finding it difficult to resource the increasing demand for adult autism diagnosis and adult services". They base this finding on the information they then show in parentheses: "(e.g., all of the adults with autism who participated in BASE Project interviews (n=5) received their diagnosis recently)". A recommendation based on this observation seems to have been translated into a need for outsourcing: "A more expedient solution may be to outsource triage and diagnosis to the voluntary or private sector"

(recommendation #4, p.15, and p.123), and yet this recommendation does not appear to come from the data at all. Of course, the causal assumption between age of diagnosis and services lacking resources is also problematic since the data do not show this. This is a good example where reference to the wider research literature would have been helpful to illustrate the heterogeneity of experiences, as well as complex reasons for later diagnosis (e.g. Jones, Goddard, Hill, Henry & Crane, 2014).

One of the recommendations made, which has clear implications for practice and policy, (p.9 and p.65) is that diagnosis should be made early, and that the 'watch and wait' strategy should be discontinued. This is despite the report demonstrating that some parents and professionals are concerned about diagnosing too early as a result of labelling implications (pp.55-56). Thus, the recommendation does not clearly follow from the data that was reported on this issue. As a minimum, there is a need to critically reflect on this issue more fully, perhaps by adding a caveat that not all parents or professionals agree with this strategy and, therefore, diagnosis should be reached in partnership and negotiation between all parties, including the child.

Another example of over-claiming is the sentence: "having autism made children vulnerable to bullying in mainstream schools more so than in special schools" (p.118). In actuality, parents of just two children talked about bullying and the children talked about media hype regarding bullying making them apprehensive about school transition. Nonetheless, there is a recommendation that, "Awareness programmes should be offered to all pupils at school to promote inclusion and reduce bullying; autism related training should be integrated during initial teacher training" (p.16 and p.124).

Whilst the reviewers and Editors would not disagree with the value of awareness-raising programmes, our point is that this recommendation is not based in a sound analysis of adequate data. The real problem for the authors of the BASE report is that by not providing a very clear audit trail between data, findings, and recommendations, doubt is cast on the quality and interpretation of

all of the conclusions. This is a shame as there are welcome and appropriate recommendations made in the report that are important for policy makers and practitioners. The difficulty lies in knowing the extent to which these are appropriately balanced and evidence-based from good quality data.

Overall this Volume reports clearly on the findings from the survey, interviews and focus groups conducted, giving some helpful information and recommendations about awareness and training about autism for professionals and the experiences of education, health and social care services by some autistic young people and adults, and their families. Qualitative research on the life experiences of people with autism and their family members who use services is a worthwhile project and the inclusion of adults with autism is a strength of the study.

Serious concerns were nevertheless raised about the quality of the research/research report in Volume 4. A more detailed explanation of the methodology, such as rationale for the methods and sample chosen, a section on limitations and some checking of the tables, would strengthen the robustness of the report. However, additional concerns relate to lack of reflexivity, lack of transparency, insufficient contextual detail, inadequate reporting of analytic process, over-claiming, and a poor relationship between the data and the conclusions and recommendations. We agree with the conclusions of Reviewer H who says: *"This is a very poor foundation on which to build policy"*. Overall, we conclude that the findings and recommendations of Volume 4 of the BASE report are not scientifically grounded, and are not based on a robust enough methodology to be used by policy makers to make decisions or take actions in respect of children, young people and adults with autism.

8. Review of BASE project (Vol. 5) final project report - Dillenburg, McKerr, & Jordan, March 2015.

8.1. Introduction

The Benchmarking Autism Services Efficacy (BASE) project was successful as part of an OFMDFM call (2011) to inform policy in relation to poverty and equality. The aims and purpose of the BASE Project have alternately been described in the different Volumes as being:

- “to establish how to help individuals with Autism Spectrum Disorder out of poverty by promoting social inclusion” (Vol.2, p.6), and
- “to provide baseline data for individuals with autism against which the effect of the Autism Act (NI) and associated Autism Spectrum Disorder (ASD) strategy can be measured” (Vol.3, p.3).

Volume 5 is described as a “triangulation” (p.14) of the findings from the four preceding Volumes of the BASE project, summarising “the process and outcome record of the BASE project” (p5). It opens with the executive summary that outlines the main components of the different Volumes of the BASE report. This starts by giving an abbreviated summary of findings, in which each finding is listed as a bullet point, with the findings cross-referenced with the relevant source Volume in parenthesis after the bullet point. Dillenburg et al. (2015) state that “... a number of issues were identified that need to be addressed to help families affected by autism out of poverty and inequality” (p 7). The four areas identified and prioritised by the authors as having the greatest positive impact for individuals with autism and families are:

1. **Autism awareness and staff training** with a focus on primary health care, first responders, public sector front-line staff, and employers in both the public and private sectors, whilst autism skills training in schools should adhere to international best practice.
2. **Early intervention support for children with autism** focusing on delay in diagnosis, discontinuation of the watch and wait approach, cost saving

analysis should be undertaken for early intensive behavioural analysis-based interventions to see if they are cheaper than other interventions and results of this should guide implementation strategies; school attendance should be monitored.

3. **Training and employment for young people and adults with autism** highlighting that FE and HE attendance should be monitored, employment rates for adults should be monitored and supported, and adult advisory services should be expanded.
4. **Families of individuals with autism** recommending that employment rates in families affected should be monitored and return to paid employment should be supported. A note in brackets highlights that this can happen through facilitating childcare and providing good quality early intensive behavioural analysis intervention for preschool aged children; benefit uptake should be monitored and information about financial support should be disseminated, futures planning should be integrated in support packages. A final recommendation to undertake a cost savings analysis is added to the end of the Executive Summary.

8.2. The scientific review

Critical appraisal is “the process of systematically examining the research evidence to assess its validity, results and relevance before using it to inform a decision” (Hill & Spittlehouse, 2001, p.1). Ways of assessing quality include that research should be conducted ethically, and it should be of relevance to inform practice or policy. Appropriate and rigorous methods should be used with clarity and coherence of reporting (Cohen & Crabtree, 2008). In addition, the Cochrane collaboration state that one of the core criteria for assessing the quality of research is researcher bias:

Believability in this context refers to the ability and efforts of the researcher to make his or her influence and assumptions clear and to provide accurate information on the extent to which the findings of a research report hold true. However, it is the actual audit trail provided by

researchers that allows for an in-depth evaluation of a study (Hannes et al., 2011, p.2).

These points are informative in providing a framework for the concerns of the Editors in relation to Volume 5. While there are a number of strengths associated with the BASE project as a whole, as noted at the start of the preceding sections that discuss each Volume in detail, we nevertheless note a number of concerns regarding Volume 5 relating to the context and rationale, methodology, analysis and interpretation, and the conclusions.

8.3. Context and rationale

In any research, it is crucial that researchers declare a conflict of interest. Most peer-reviewed journals expect authors to declare a conflict of interest and to be transparent about any competing interests they may have. Whilst this often relates to financial interests, 'conflict of interests' extends beyond this and includes broader interest. It is not that such a declaration should stop a paper or research report being published, but readers should be made aware of it so that they can draw their own conclusions about the context in which the research was planned and conducted, and the lens through which interpretations and recommendations are made. The Committee on Publication Ethics (n.d.) have produced guidelines that state that:

conflicts of interest comprise those that may not be fully apparent and which may influence the judgement of authors, reviewers, or editors. They have been described as those which, when revealed later, would make a reasonable reader feel misled or deceived (p.44).

Given that the authors promote a particular approach to early intervention in autism, which they are strong proponents of, and they are involved in the design, delivery and promotion of University-based courses, which they actively recommend in the report, a section on conflict of interest is crucial. For example, the authors refer specifically to the Pre-graduate course at University of Ulster in the School of Psychology; the Undergraduate-level Open University (OU) course

which is jointly run with the Queen's University Belfast School of Education; and they refer to the Postgraduate (Queen's University Belfast School of Education and University of Ulster School of Psychology) course. Indeed, they specifically recommend (p.7) that "for staff in schools and multi-agency autism intervention teams ... it is crucial that local University courses are fully included in the training menu". According to the Queen's University Belfast website⁷, Professor Dillenburger runs the MSc in ASD and continues to teach on the online ABA course there, for example.

Given that Volume 5 is a synthesis of the main findings and ideas presented in Volumes 1-4, it is clear that the issues relating to the lack of definitions of core concepts is replicated here. For example, poverty, poverty trap, vulnerability, and social exclusion are all used without any definition. The failure to define these key terms means that it is difficult to come to a confident conclusion that the BASE report addresses its key aims.

The authors make a statement that the UNCRPD underpinned the rights-based rationale of the BASE project research, but this rationale is not made explicit in terms of informing the readers about how it underpinned the work. Readers are left unclear as to how it influenced the research questions, the methodology, or the analysis. Furthermore, there is no identification of specific research questions across any of the Volumes, and without research questions, it is difficult to determine whether the methodology chosen was appropriate for the task. Given that research questions are not listed, the findings are consequently not linked back to the research questions. It is therefore difficult to determine whether the aims of the study have been met. As a result, readers are left with a set of bullet points and findings that appear to have little grounding in the data in some places and without a conceptual framework overall.

⁷ <http://pure.qub.ac.uk/portal/en/persons/karola-dillenburger%2845704146-89fc-4d3f-8e6d-81e6620eced4%29.html>

8.4. Methodology

The range of methodologies utilised across Volumes 1-4 have already been critically appraised in the preceding sections of this report and so their relative strengths and limitations need to be borne in mind, and will not be repeated here. Nevertheless, given the range of methodologies and methods used in the BASE report, it is surprising that Volume 5 gives no explanation or discussion of the overall methodology of the BASE project. The authors state that this Volume triangulates findings from the previous volumes, so one would expect a transparent overview of how the different methodologies of the various volumes contributed to the overall project methodology, and the relative weight given to each (with appropriate justification). The failure to do so leaves the report open to the criticism that some findings and recommendations may have been selected and prioritised over others without clear explanation. This certainly seems to be the case in relation to the reporting of the 'key findings', which is covered in Section 8.5 below.

We also note that there is a lack of detail and discussion about ethics – both in terms of the ethical issues inherent in the research as well as full disclosure of all ethics protocols and procedures. All research should give prominence to its ethical approach. Adherence to ethical guidelines and approaches is covered in Volume 4 of the BASE report and examples of information sheets are provided. It is stated there that the research was reviewed and approved by Queen's University Belfast. However, there is no mention of ethics in Volumes 2, 3 and 5.

8.5. Findings

The lack of transparency regarding the triangulation of methods and subsequent analyses mean that the findings are presented in such a way that it is difficult to get an overview of them. No justification was given as to why these particular findings, rather than others, were considered important, nor was the process for their selection as 'key findings'. As reviewers, we also had difficulty gaining a coherent overview of the findings because they were presented as a set of bullet points and lacked a clear linking narrative. As a result, we inserted the findings listed in the BASE Volume 5 report (pages 5 and 6) into a Table, where we

provide an overview of the relative balance of reported 'key findings' from each Volume of the BASE review.

As can be seen from Table 2, there were few 'key findings' taken from Volumes 2 (n=3) and 4 (n=1), and higher numbers of findings from Volumes 1 (n=8) and 3 (n=9) selected by the authors from each volume. We are surprised that only one finding was listed from Volume 4, which drew upon the views and experiences of stakeholders, including individuals on the autism spectrum. We are concerned that their voices appear to be largely omitted from the key findings in the final report. Findings from Volume 2 are also quite limited, and the findings that are listed, have no evidence base in the data from the research in Volume 2 (see section 5.6.).

Table 2: Summary of the findings listed in the Executive Summary of the (Vol. 5) BASE report.

Volume 1	Volume 2	Volume 3	Volume 4
2% of children have autism; adult prevalence is unknown.	Autism awareness is high in the general population (82%), 51% know someone with autism personally.	Up to 3.5% of 11-year-old children are thought to have autism.	In-service training in autism is very basic.
Local Universities' comprehensive autism training and research was omitted in the autism strategy/action plan.	Knowledge about autism is good in the general population.	Identification of vulnerability/autism diagnosis is protracted and delayed; watch and wait approach in some trusts means autism diagnosis can take more than two years.	
The cost of autism per lifetime is estimated at £0.9-1.5. million in the UK.	Attitudes towards people with autism are positive.	Children with autism miss school 8-13 days more than other children.	
The total cost of autism is £34 billion in the UK.		Educational attainment is lower for children with autism (20-40 percentage points lower than other children).	
Cost of bringing up a child with autism is up to 6 times greater than for other children.		Exact employment figures for adults with autism are not known in NI.	
Early intensive behaviour analysis-based interventions (evidenced as effective interventions) are not available in the statutory sector.		Unemployment is high in families with autism and many families give up employment to care for their child.	

Children with autism are frequently excluded from (20%) and bullied (20%) in school.		Income is up to 12% lower in families with autism.	
Employment figures for adults with autism are low (15%).		Families with autism are likely to live in deprived areas (6 percentage points higher than other families).	
		Income is up to 12% lower in families with autism.	

We have highlighted previously that there was omission of studies in the field in section 4.4. of Volume 1. Misrepresentation and omission of other studies in the field is also evident in Volume 5 and this warrants discussion. Firstly, this relates to the way the National Autism Center (NAC) report is quoted in Volume 5, Dillenburger et al. (2015) state that:

There is extensive and unequivocal evidence (National Autism Center, 2015) that early intensive behavioural interventions (i.e. based on the scientific discipline of Applied Behaviour Analysis; ABA) can enhance the quality of life of individuals with ASD and their families (BASE project Vol. 1) and lead to optimal outcomes (Fein et al., 2013; Orinstein et al., 2014) (p.24).

The National Autism Center, (NAC; 2015) report cited was a systematic review of behavioural and educational peer-reviewed intervention literature. For children and young people under 22, the NAC (2015) authors found 14 interventions that “have been thoroughly researched and have sufficient evidence for us to confidently state that they are effective” (p.41). EIBIs were classed as an established intervention with a strong evidence base, leading to increased play skills and academic readiness amongst other skills. However, a number of other interventions were also listed as having an established intervention base, including language training, modelling, and naturalistic teaching strategies. Therefore, the established interventions included behavioural approaches but were not limited to those.

The statement in the BASE report that links this ‘extensive and unequivocal’ evidence with enhanced quality of life for people with autism and their families, leading to optimal outcomes seriously over-stretches the findings from the NAC (2015) research. The NAC (2015) report lists very specific skills and behaviours that are evidenced as having been affected by specific intervention approaches. It does not draw the conclusion that such interventions lead to optimum outcomes and increased quality of life. In line with the review of Volume 1 of the BASE report, this partial representation of another report in the field casts further

doubt on the objectivity of the findings of the BASE review and concomitant recommendations made.

Furthermore, describing this evidence as ‘unequivocal’ is overstating the case. In the past decade, a number of systematic international literature reviews have examined the evidence base for different interventions and approaches for pupils with autism. As also discussed in the reviews of Volume 1 of the BASE report, these literature reviews have found that there is no evidence to suggest that a single intervention will meet the needs of all learners, or that any single intervention or technique stands out from the others as superior for a majority of children (Parsons et al., 2011; Odom et al., 2010; Wong et al., 2015). The NICE (2014) guidelines in the UK also recommend a range of approaches be provided for supporting children with ASD (p.27). Indeed, we reiterate one of the conclusions from the SIGN (2016) guidelines that is also covered in the review of Volume 1, *viz*:

While it has been shown that EIBI is superior to no intervention or treatment as usual, the evidence does not warrant the provision of universal EIBI. It does however justify rigorous cost-effectiveness research, which currently is of poor quality and does not support wide application of this approach. (p.24)

Relatedly, the claim on page 12 that EIBIs are a recognised mechanism that have the potential to help autistic people and their families out of the ‘poverty trap’ is at best a controversial claim needing specific justification and it is out of place in this list of otherwise generally accepted (although not very precise) statements.

In addition, the authors make the claim that failure to provide effective EIBI has serious lifetime economic consequences and they cite Knapp et al. (2009) to back up this point. This claim has already been discussed and critiqued in Section 4.5 of the Volume 1 review above, which concludes that this is a misrepresentation of the Knapp et al. (2009) analysis. Specifically, Knapp et al. (2009) actually concluded:

More, generally, given that autistic spectrum includes a number of disorders and a wide range of needs, symptoms and characteristics, it is likely that a wide range of behavioural, educational and medical interventions could be required in order to meet some or all individual needs. (p.332).

The Howlin et al., (2014) study is also quoted extensively throughout other volumes of the BASE report. We here refer to the way in which study is discussed in Volume 5 (page 24). The paragraph starts by making the statement that “Without early intensive behaviour analysis-based interventions the outlook is bleak” (p.24). This comment is unhelpful, especially for families, as it is not supported by empirical evidence. As covered in Section 4.5 of Volume 1 of the review, the authors go on to make the point that the Howlin et al., (2014) study, in their 40-year follow-up of adults whom they had diagnosed in the 1970s, many of their participants had attended special schools as children but none had access to early intensive behavioural programmes. The authors of the BASE report argue that:

Howlin et al. (2014) go on to assert that only ‘some’ people in the UK believe that intensive, early behavioural programmes are effective, despite the overwhelming international evidence in their favour (e.g., Surgeon General, 1999; NAC, 2015) and Howlin et al.’s own findings that lack thereof has detrimental long-term consequences (Howlin, Goode, Hutton, & Rutter, 2004, Howlin et al., 2014).

In Volume 1, we highlight that Howlin et al. (2014) do not make a causal connection between the outcomes for these adults and the notion that these adults did not have access to early intensive behavioural interventions as children, neither do they draw the conclusion that there were detrimental long-term effects because the cohort did not receive early behaviourist intervention. The other point made by Howlin et al. (2014) are that early behavioural approaches are claimed by some to have significant impact on IQ and long term-outcome. This is an acknowledgement that there is research available in which

claims have been made for significant IQ and long-term outcomes as a result of early behaviourist intervention.

Moreover, given that one of the key criteria in assessing quality of research is researcher bias (Hannes et al., 2011), we note with concern that this Volume has a clear bias toward Applied Behaviour Analytic approaches, specifically early intensive behaviour analysis-based interventions. This bias is not made clear in the outset. The authors may have good reasons for the bias, but this should be declared to the readers, and arguments put forward as to why and how the authors have arrived at their conclusions. Such arguments should refer to the broader evidence base in the field, even if to disagree with it. Evidence of bias includes the high prominence given to promoting early intensive behaviour analysis-based intervention approaches, with few mentions of other approaches and interventions.

Table 3 highlights some key quotes promoting early intensive behaviour analysis-based interventions. In addition to the quotes in Table 3, early intensive behavioural analysis-based interventions are mentioned 12 times in the Baseline, Benchmarks and Recommendations diagrams on pages 36-46. Given the doubts raised by the reviewers about the overall quality of the research conducted for the BASE report, and the lack of a clear audit trail between data, findings, and recommendations, the basis for these claims is highly questionable.

Table 3. Quotes and page numbers where early intensive behavioural analysis-based interventions are mentioned

Quotes	Page Numbers
Early intensive behaviour analysis-based interventions (evidenced as effective interventions) are not available in the statutory sector.	Page 6
Cost-saving analysis should be conducted for early intensive behaviour analysis-based interventions against other options (i.e. treatment as usual in NI) and results of this analysis should guide implementation strategies.	Page 8
<i>'Availability of early intensive behaviour analysis-based interventions'</i> is listed as a mechanism that has the potential for helping people with autism	Page 11

and their families out of the poverty trap.	
Furthermore, the 2012 NILT survey found that over half of the general public (58%) realised that autism is not necessarily a 'lifelong' disability and there was strong support for evidence based behavioural interventions (77%), especially in the early years.	Page 17
These findings confirm international findings evidencing that diagnosis is possible much earlier than presently practiced in Northern Ireland, thus allowing for early intensive behavioural interventions or parent training to start much earlier (Dawson et al., 2012).	Page 20
Without early intensive behaviour analysis-based interventions, the outlook is bleak.	Page 24
There is extensive and unequivocal evidence (National Autism Center, 2015) that early intensive behavioural interventions (i.e. based on the scientific discipline of Applied Behaviour Analysis (ABA) can enhance the quality of life of individuals with ASD and their families (BASE project Vol. 1) and lead to optimal outcomes (Fein et al., 2013; Orinstein et al., 2014).	Page 24
Evidently, early intensive behaviour analysis-based interventions lead to positive outcomes (BASE project Volume 1) and thus constitute a crucial mechanism to help vulnerable people with autism out of poverty and inequality.	Page 25
Despite the fact that Applied Behaviour Analysis (ABA)-based interventions were recommended by the Task Group Report on autism (DENI, 2002), these interventions are not available through the HSC Trusts or the Department of Education.	Page 26
More recent NICE guidelines for challenging behaviour (Guideline 11, 2015) explicitly include behavior analysts in the list of relevant professionals (2.1.4; 6.4.3 and 14.1.5). Therefore, given that challenging behaviours are common in children and adults with autism, ABA –based interventions and appropriately qualified staff (www.bacb.com) should be available within the statutory sector.	Page 27
The delay in onset of interventions, caused by delayed diagnosis, means that most children with autism in NI miss invaluable opportunities in pre-school years (i.e. exploiting brain plasticity through early ABA-based interventions cf., Dawson, 2008) and consequently, potential long-term savings to the tax payer (up to 1 million across a single lifetime) are missed (Buescher et al., 2014; Motiwala et al., 2006).	Page 28
'Early intervention' in Northern Ireland is mostly brief and variable (e.g., a once off 1-3 hour visit), while internationally recognized best practice (i.e.,	Page 32

early intensive behaviour analysis-based intervention) is not available in the statutory sector.	
Make available good early years childcare tailored for children with ASD and early intensive behaviour analysis-based intervention.	Page 36
Teachers should be encouraged to undertake advanced training in autism and ABA to University-based Masters level; Board Certified Behaviour Analysts (BCBA) should be included in education teams.	Page 42 and 43
Early intensive applied behaviour analysis-based interventions can lead to optimum outcomes and enhanced quality of life, potentially saving up to 1 million across a lifetime.	Page 56
There is ample evidence that applied behaviour analysis interventions are linked to optimal outcomes and this body of evidence has grown over the last 45 years.	Page 55

8.6. Conclusions and recommendations

The authors state that benchmarks and recommendations were developed on the basis of BASE project findings, but there is no clear outline or description of how the recommendations build on the findings. It would have been much more useful to outline in a transparent way to the readers how the findings informed the baselines that were chosen, and therefore guided the recommendations that emerged from these findings. The reviews of Volumes 1-4 have also highlighted a tenuous relationship between data, findings and recommendations

Overall, throughout the report that informs the recommendations, there is also a high number of self-citations. There are 87 references listed in Volume 5. Out of those, the lead author references herself 19 times in the reference list as first author and seven times as second author, leading to self-citation 26 times, which is effectively very close to a third of all references. In the report itself, the author cites herself or previous volumes of the BASE report 89 times (this includes footnotes). Whilst we acknowledge that the authors are active in the field and that a certain amount of self-citation is to be expected, we think these figures indicate an overreach in self-citation and an under-citation of other important work in the field (see Section 8.5 on under reporting and misrepresentation of other research).

Finally, it is widely expected that research reports should include a section on the limitations of research to acknowledge that authors have thought critically about the research problem, and have assessed the methodology and methods used to address this problem. These should be delivered through critical appraisal and interpretation of impact. Sections on limitations should usually exist in the main body of the report, and typically towards the end while drawing main conclusions. In the BASE report, a section on limitations is found in Appendix 7 of Volume 5 (p. 84). The description of study limitations is brief and does not cover the full report. Bias in the self-selection of participants is acknowledged but the bias of the authors not. Indeed, the authors state:

The BASE Project research built on extensive previous research conducted by members of the multidisciplinary BASE Project team. Previous projects spanned

over 20 years in the field of autism research and included significant numbers of collaborations with national and international colleagues and service users from many different settings. This experience meant that some potential limitations were avoided from the outset.

As limitations are meant to focus on influences that the researcher cannot control, and that might place restrictions on the methodology and conclusions, this sentence is out of place in a section outlining limitations. The paragraph merely promotes the authors' standing in the field and does not engage critically with the study limitations of this specific research.

8.7. Main conclusions from the review of Volume 5

Volume 5 draws together ('triangulates') findings from the four Volumes of a study aiming to address an important gap in research, namely the investigation of the link between poverty and autism, with a focus on how to help families out of poverty. This is a highly relevant and much needed topic. An impressive amount of data has emerged from this mixed-methods study conducted over a number of phases. The overall study represents a significant effort on behalf of the authors, with a large range of literature consulted (Volume 1), sizeable samples (Volume 2), a comprehensive overview and analysis of secondary data sources (Volume 3), and with a welcome inclusion of autistic people and families in research (Volume 4).

However, despite these strengths, there are serious concerns about the scientific rigour of the BASE project. Volume 5 lacks identification of specific research questions and definitions of terminology. Furthermore, there is no transparent discussion of the methodology chosen, and there are tenuous links between data, findings and recommendations. Statements are made which make causal leaps that go way beyond the evidence. There is no evidence of systematic examination of the research evidence, nor of an assessment of its validity, results or relevance before using it to inform recommendations for policy (Hill & Spittlehouse, 2001). Most importantly, the researchers fail to make their assumptions and influences clear. This lack of audit trail has led all reviewers to question many of the conclusions drawn.

9. Summary, conclusions and recommendations

We conclude that the lack of objectivity, rigour and transparency in the BASE report overall mean that the findings and conclusions should not be taken at face value and should be re-evaluated in the light of the wide-ranging concerns raised by reviewers.

In relation to **future research** in investigating the link between poverty and autistic people, we suggest that the following gaps and priorities emerge from this review:

- There needs to be an appropriately designed and conducted systematic literature review that is carried out to address core research questions relating to poverty and wellbeing of individuals with autism and their families.
- A survey should be conducted with service providers to understand whether there is indeed a failure to implement aspects of practice recommended or mandated in existing policy documents. Autistic people and their families should be included in this research.
- A clearly framed analysis should be commissioned of the NILT data (excluding ambiguously phrased questions) that addresses specific research questions and incorporates more sophisticated planned analyses in order to understand the possible links between independent and dependent variables.
- Further analysis should be undertaken of the secondary datasets. These should incorporate the suggestions of the reviewers, namely to provide statistical significance for comparisons made, to use a set of researchable questions to guide the analysis, with transparent explanation of where measures were taken from, and with coherent reporting of findings and careful presentation of figures and graphics.
- In order to fully understand and represent the voices of autism stakeholders there needs to be a more robust and larger scale qualitative study that enables participation from a wider range of people. More stakeholders need to be included in qualitative methods that support a range of communication preferences and needs.
- There would be value in undertaking an audit of the range of training courses and resources that are currently available for staff, autistic people and their

families in Northern Ireland, including those available by distance learning.

More generally, we hope the following specific **recommendations** will be helpful to the The Executive Office in Northern Ireland in relation to **commissioning future research**:

- Ensure there is a robust external peer review process for all reports intended for the public domain.
- Provide clear guidelines to ensure that all commissioned and published research reports adhere to best research practice by disclosing conflicts of interest, being detailed about procedures for ethics review and approval for all appropriate aspects of the research, and that the research is designed to answer specific research questions, with all necessary aspects of methodology and analysis, and key terminology, adequately specified.

10. Additional information

10.1. Conflicts of interest statement

Karen Guldberg and Sarah Parsons worked together on a systematic review of evidence for educational interventions for children, young people, and adults with autism, commissioned by the National Council for Special Education (NCSE) in Ireland. The report, and subsequent publications, (Parsons et al., 2009, 2001; Guldberg et al., 2011) are cited in the present report. Karen Guldberg runs the Autism Children Distance Learning programme at University of Birmingham. We allude to different courses being available for professionals in the scientific review.

10.2. The authors

Karen Guldberg is Reader in Autism Studies, University of Birmingham and Director of the Autism Centre for Education and Research (ACER). Dr Guldberg conducts real-world research in the classroom, through implementing distinctive methodological approaches to the study and development of autism educational provision, pedagogy and practice. She has led and been involved in a number of research projects working in partnership with schools, practitioners and parents. Further information about Dr Guldberg can be found at:

<https://www.birmingham.ac.uk/staff/profiles/education/guldberg-karen.aspx>

Sarah Parsons is Professor of Autism and Inclusion, and Director of Research for Southampton Education School, at the University of Southampton. She has significant research experience in disability related projects and particular interests in the use of innovative technologies for children with autism, evidence-based practices in autism, and research ethics. Further information about Professor Parsons can be found at:

<https://www.southampton.ac.uk/education/about/staff/sjp1e10.page>

10.3. List of reviewers and acknowledgements

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Dr Abigail Knight, UCL Institute of Education.

Professor Melanie Nind, University of Southampton.

Dr Samantha Parsons, University College London.

Professor Jill Porter, University of Reading.

Professor Emma Smith, University of Warwick.

Professor Patrick Sturgis, University of Southampton

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Appendices

Appendix 1: BASE Review Proformas

BASE Review Proforma Vol. 1

UNIVERSITY OF
BIRMINGHAM



UNIVERSITY OF
Southampton

Proforma

'The Benchmarking Autism Services Efficacy,' (BASE) report by Dillenburg, et al. (2015), consists of five separate Volumes, and was produced by the School of Education at Queen's University, Belfast and funded by Northern Ireland's Office of the First Minister and Deputy First Minister.

The aim of this scientific review is to reach an objective view on the methodology and scientific soundness of the findings and recommendations in respect of the expressed aims of the project. The key question is whether or not the data obtained and interpreted by the authors would lead logically to the conclusions drawn by them, or whether there was overreach where conclusions went beyond the data.

The overarching question for the scientific review is: "Are the conclusions of the BASE report (2015) scientifically grounded and based on a robust enough methodology to be used by policy makers to make decisions/actions in respect of children, young people and adults with autism?"

With this in mind, we would be grateful if reviewers would address the following questions:

Originality and relationship to Literature
Please comment on the comprehensiveness and completeness of literature cited, noting any key omissions, selectivity, imbalance or evidence of any misinterpretations of the studies cited; and provide an analysis of the proportion of author self-citations and how this fits with standard scientific practice. Does Volume One demonstrate an adequate understanding of the relevant literature in the field and cite an appropriate range of literature sources? Is any significant work ignored?
Methodology
To what extent is the literature review conducted in a rigorous and systematic manner? Are the methods employed appropriate?

Results

Are results presented clearly and analysed appropriately? Do the conclusions adequately tie together the other elements of the paper?

Implications for research, practice and/or society

Does Volume One identify clearly any implications for research, practice and/or society? Are these implications consistent with the findings and conclusions of the publication?

General

Please comment on any strengths of the approach taken in this Volume to the collation, analysis, interpretation, presentation, and discussion of evidence. Please use this space to comment on any other aspects of the report as you think appropriate and not already covered under the above points.

Overall Assessment

Please provide a few sentences to summarise your overall impression of the quality of the Volume that you have reviewed.

Implications for research, practice and/or society Does the publication identify clearly any implications for research, practice and/or society? Are these implications consistent with the findings and conclusions of the publication?
General Please comment on any strengths of the approach taken in this Volume to the collation, analysis, interpretation, presentation, and discussion of evidence. Please use this space to comment on any other aspects of the report as you think appropriate and not already covered under the above points.
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With this in mind, we would be grateful if reviewers would address the following questions:

Originality and relationship to Literature
Does the 'Discussion section' of Volume Three demonstrate an adequate understanding of the relevant literature in the field and cite an appropriate range of literature sources? Is any significant work ignored?
Methodology
Volume Three of the BASE report analyses a set of nine separate data sets in Northern Ireland. Please comment on the criteria for choosing those particular datasets; how existing government datasets were utilised and interpreted and address whether the study represents an objective scientific analysis that adhered to best practice conventions in the analysis of secondary data.
Results
Are results presented clearly and analysed appropriately? Do the conclusions adequately tie together the other elements of the paper?

<p>Implications for research, practice and/or society</p> <p>Does the publication identify clearly any implications for research, practice and/or society? Are these implications consistent with the findings and conclusions of the publication?</p>
<p>General</p> <p>Please comment on any strengths of the approach taken in this Volume to the collation, analysis, interpretation, presentation, and discussion of evidence. Please use this space to comment on any other aspects of the report as you think appropriate and not already covered under the above points.</p>
<p>Overall Assessment</p> <p>Please provide a few sentences to summarise your overall impression of the quality of the Volume that you have reviewed.</p>

Proforma

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The aim of this review is to reach an objective view on the methodology and scientific soundness of the findings and recommendations in respect of the expressed aims of the project. The key question is whether or not the data obtained and interpreted by the authors would lead logically to the conclusions drawn by them, or whether there was overreach where conclusions went beyond the data.

The overarching question for the scientific review is: "Are the conclusions of the BASE report (2015) scientifically grounded and based on a robust enough methodology to be used by policy makers to make decisions/actions in respect of children, young people and adults with autism?"

With this in mind, we would be grateful if reviewers would address the following questions:

Originality and relationship to Literature
Does Volume Four demonstrate an adequate understanding of the relevant literature in the field and cite an appropriate range of literature sources? Is any significant work ignored?
Methodology
Does the qualitative study follow conventional standards for qualitative research? Has the work been carried out to an acceptable standard in terms of transparency, authenticity, and credibility? Are the findings reported in a fair and reasonable manner? Are the questions for participants framed in a clear and open way? Do the findings and conclusions drawn follow clearly and logically from what was asked?
Results
Are results presented clearly and analysed appropriately? Do the conclusions adequately tie together the other elements of the paper?

Implications for research, practice and/or society

Does the publication identify clearly any implications for research, practice and/or society? Are these implications consistent with the findings and conclusions of the publication?

General

Please comment on any strengths of the approach taken in this Volume to the collation, analysis, interpretation, presentation, and discussion of evidence. Please use this space to comment on any other aspects of the report as you think appropriate and not already covered under the above points.

Overall Assessment

Please provide a few sentences to summarise your overall impression of the quality of the Volume that you have reviewed.