PEER REVIEW HISTORY

ARTICLE DETAILS

TITLE (PROVISIONAL)	The effect of deinstitutionalisation for adults with intellectual disabilities on costs: a systematic review
AUTHORS	May, Peter; Lombard Vance, Richard; Murphy, Esther; O'Donovan, Mary-Ann; Webb, Naoise; Sheaf, Greg; McCallion, Philip; Stancliffe, Roger; Normand, Charles; Smith, V; McCarron, Mary

VERSION 1 - REVIEW

REVIEWER	Victoria Ratti Division of Psychiatry, University College London, UK
REVIEW RETURNED	26-Aug-2018

GENERAL COMMENTS	Thank you for the opportunity to review the manuscript "The effect of deinstitutionalisation for adults with intellectual disabilities on costs: a systematic review." The manuscript provided an interesting, well-written and concise read. The systematic review was generally methodologically sound and it is evident that the authors followed PRISMA guidelines. I believe, nonetheless, that the manuscript can be strengthened and I have provided specific comments for each section which I invite the authors to consider. Introduction This section is well written and concise and provides a valid rationale for the need to conduct economic evaluations for different accommodation types for people with intellectual disabilities (ID). Nevertheless, deinstitutionalisation has been ongoing for a number of decades and has been advocated in publications such as Better services for the Mentally Handicapped (1971). The majority of people with ID in countries such as the UK and Australia no longer live in institutions and now reside in the community. I believe the authors need to provide a stronger rationale for comparing the costs and outcomes of various accommodation options to institutions rather than comparing different residence types within the community, a topic which would appear more relevant to the current provision of care landscape. Although I agree with the authors that "the accurate estimation of resource use in providing services can inform budgeting,

workforce planning and organization of services", I believe that nowadays it would be more useful to have information on costs and outcomes of different accommodation types in the community such as supported living versus residential care homes. Even if the results of this review had shown that institutions were more cost-effective, where outcomes such as choice, self-determination and community participation have been demonstrated in previous research to be improved with the move to the community, increases in cost may be justified. Additionally, given that the great majority of the literature on deinstitutionalization is from the 80s-90s and that there have been previous reviews on the topic (e.g. Kozma, Mansell and Beadle-Brown, 2009, QoL; Walsh, Kastner and Reed, 2003, cost) I wonder if the authors need to make a stronger case why a systematic review is needed now.

That being said, in the UK, there are still approximately 2600 people with ID living in inpatient settings (Health and Social Care Information Centre, 2016) who are likely to move into the community, and more recent publications such as Building the right support (2015) still emphasise that people shouldn't be staying in inpatient units unnecessarily, suggesting that long stays are still a current issue. In many countries the process of deinstitutionalisation is far from complete and therefore information regarding costs is useful to inform current and future services. I think the authors could emphasize these points to provide a stronger rationale for examining the cost effects of deinstitutionalization and reiterate that there are still high numbers of individuals living in institutions in the present day.

At line 33 the abbreviation ID is given but the non abbreviated term intellectual disabilities is still largely used throughout the manuscript.

Method
The methodology section is well-written, clear and comprehensive. I do however have some queries regarding some of the methodological decisions made by the authors which are outlined below:

Types of participants: it is stated that participants were adults with ID. Did you use a specific criteria such IQ <70 to select studies or considered all studies where the authors had described participants as having ID?

Types of studies/reports: I found the choice to exclude cross-sectional studies comparing community-living and institutional arrangements surprising as various studies have used this methodology and provided useful information on costs. I believe it would be useful to justify this decision.

Database search: The authors do not report on supplementing their electronic database searches by hand-searching the references of selected papers, which is often also done when conducting systematic reviews. Was this step omitted?

A comprehensive description of the search strategy is provided in Appendix 1. Here I noticed that the search was not only focused on articles on cost but also on articles of QoL. This is not clear in the main article. Although it is mentioned in the results section that the numbers are reflective of a combined search of articles

	reporting on both costs and QoL, this comes a bit out of the blue and leaves the reader questioning why QoL focused studies were included when the focus of the review is on cost, and QoL studies are not mentioned at all in the present article. I think the authors need to be transparent about this in the search strategy section or conduct separate searches for cost and Qol articles. Assessment of methodological quality/risk of bias: The authors describe using CASP checklists to appraise the studies. CASP tools do not suggest a scoring system, so I was wondering how the authors determined moderate/high quality studies? Did they use a cut-off score/ % of "YES" answers? I believe, for clarity, that this should be specified and it may also be useful to add the main questions (not necessarily the prompts) of the CASP checklists in the Appendix. Main results. On page 17 line 11 it is stated that many participants moved from institutions to "larger institutional settings". Although these nursing homes/hospices are described as community settings it leaves one wondering if this was actually a form of deinstitutionalization. Table 2. I believe, where possible, it would be useful to report p values/effect size. On page 19 line 19-20 is stated that secondary analysis shows that accommodation accounts for 81-86% of community costs post-move. I think it may need to be specified or provide examples of which other variables were considered as expenditure/cost (e.g. day activities, health services etc.). I was unclear whether the authors conducted the secondary analysis or if these were reported in the original paper. Discussion On page 20 line 25-26 it is stated that there are significant associations between costs and both participant and residence characteristics. These however were not reported in the results section and therefore it is unclear what these characteristics were. The same phrase is reported in table 2 where I think these characteristics and their associations should be specified. Appendix. -Pages 36-37 on my PDF are a repetition of Appendix 3. This may be a mistake. -The PRISMA checklist is included in the Appendix but it is not mentioned in the main article. Using the PRISMA checklist to conduct/write the review could be added as a strength and evidence of the methodological rigor of the review. -I think including another systematic review in the appendix is unnecessary, particularly as the appendices of this second article are a repetition of the appendices of the article under review (cost review). I understand that the systematic search combined terms of both cost and QoL which were pooled and the authors may want to include an appendix with the references of the QoL articles that were retained. However I believe that the QoL review should be submitted as a separate article rather than being included in the current article's appendix. Ideally for reasons of replicability, i believe it would be more appropriate to conduct separate searches for the two reviews, one including only cost terms and the other including only QoL terms and I invite the authors to consider this.

	I have endeavoured to provide a comprehensive review and I hope the authors find my comments/suggestions useful. Comments to specific questions Question 4. Are the methods described sufficiently to allow the study to be repeated? I have addressed these points in the comments and these relate to the definition of ID used, the omission in the main article of the combination of cost and QoL search terms and how moderate/high quality was determined from CASP checklists. Question 10. Are the results presented clearly? For the most part results are presented clearly, however as stated in my comments I believe it would be useful to include p values and effect sizes where possible and also spell out the associations between cost and individual and residence characteristics. Also I think there needs to be more clarity regarding the secondary analysis results i.e. accommodation counted for 81% of cost, but what other variables were considered? And who conducted the analysis?

REVIEWER	Martin Knapp LSE UK None declared, except that I was a co-PI on one of the two studies discussed in this review paper, and the lead PI on the other.
REVIEW RETURNED	13-Sep-2018

GENERAL COMMENTS	Broadly speaking, this is a reasonably well-conducted systematic review. One limitation is that only one reviewer undertook quality assessment and data extraction (as the authors recognise). Two other limitations can be noted. One is that what the authors describe as ‘patient and public involvement’ does not look to be what I understand by the term. Perhaps I am missing something in the paper, but I assume from the omission of the explanation that this representative was not somebody with an intellectual disability. The second limitation is that the authors do not appear to have fully analysed or described the studies they selected. The study described as Hallam et al is the 12-year follow-up of a group of people leaving hospitals in England. The authors of this paper mention the earlier studies (baseline, 1-year and 5-year follow-ups) but miss elements in those earlier analyses in making their criticisms. As I understand the purpose of the review, it was not only focused on very long-term economic effects, so why were earlier findings ignored? For example, on p.20 the authors write ‘One-off costs associated with deinstitutionalisation also appeared not to be taken into account’. This is incorrect in relation to the English study. The book reporting the 1-year results includes a 23-page chapter on the financing of community care, and there is discussion around set-up costs and other challenges elsewhere in the book. I will mention other missing elements below. A second example is where the authors write (p.21) that ‘Details on the participants were similarly scant’. The English study gives

information on the following characteristics for sample members at baseline: gender, local authority of origin, severity of intellectual disability, skills (i.e. ADLs), behavioural issues, satisfaction with services, social interaction (quantity and self-reported quality), depression, psychosocial function, life satisfaction (Cantril's Ladder). I struggle to understand what further data are needed to persuade the authors that the details are not 'scant'.

The strategy adopted by the team not to specify ex ante what was meant by 'institution' or 'community' was the right one. As they say later in the paper (p.22) 'No ex ante definition of institutional community settings' was employed. However, the discussion of findings certainly employs an ex post judgement on what is meant by 'community', but without explicating what it is. For example, on p.17 they write that only one person across the two studies 'can be said certainly to have moved to community-living arrangements as understood in the 21st century', by which they mean independent living, I think. I would be happier with this statement if the authors explained whose view of 'community-living arrangements as understood in the 21st century' they are referring to here, and added a reference or two. If the only interpretation of 'community-living' is living independently, then I think their statement is misplaced.

Linked to this is the sentence 'An additional shared limitation [of the two included studies] was the use of the term 'community care'' (p.16). At the time these two studies were conducted, the term 'community care' reflected not only a policy position but also the position of advocates in the sector. Of course, a large residential home would not be seen as 'community', but are the authors arguing that supported lodgings, foster care and unstaffed group homes are really not considered to be community care today? It is fine for the authors to criticise terminology employed in research 20-30 years ago – that is to be expected in an area such as this – but it is a failing of the paper not to offer any clear alternative definition.

On p.12 the authors write 'None of the included studies evaluated both cost data and QOL'. The authors do not define what is meant by 'QOL', but I would have imagined that using a life satisfaction measure such as Cantril's Ladder and a measure of self-expressed satisfaction with living environment and life in general by individuals with intellectual disability would come reasonably close to 'QOL'. If I make a leap of assumption here, I did wonder if by 'QOL' the authors meant the sort of thing that most health economists understand by the term, which is health-related quality of life measure of the kind measured by tools such as EQ-5D. If so, then I can understand why they did not find any such studies, which might be because an HRQOL is unlikely to be prioritised among the outcomes of studies in the intellectual disability field. If by 'QOL' the authors mean concepts similar to those reflected in the search strategy, then at least one of these two included studies looks at 'QOL'.

The authors excluded studies that they call 'static comparisons', by which they mean studies that do not look at change over time but simply compare two or more different settings within a cross-sectional design. This may well have been sensible in order to make their reviewing task easier, but it does not necessarily help policy-makers. On p.21 they explain why they chose this strategy,

	including the fact that 'they did not evaluate the impact of a move in residential setting, and there are additional concerns about the robustness of such comparisons and unobserved confounding, particularly with routinely collected data'. But earlier on that same page, they note the need for evidence in relation to, for example, 'younger people with very complex needs and challenging behaviours'. Many of the latter group may never live in an institutional setting as defined in this review, and so confining a review to just moves from institutions will not provide information on such a group. Presumably, the authors therefore believe that a different review is needed, which feels something like a missed opportunity. Having gone to the effort of designing what generally looks like a good search strategy, it does seem a pity that potentially rich evidence has been discarded. I agree that there can be difficulties in the interpretation of evidence because of unobserved confounding and suchlike, but then this review could have provided a great service by highlighting the limitations of studies that were uncovered. On p.23 the authors write 'The level of detail gathered in those [two included] studies is not fit for today's policy purposes'. In part I think this is inevitable, but the reviewers are, in my judgement, simply wrong to use the justification that the two studies did not report sufficient detail. As noted earlier, the authors have failed to look at the evidence in the included studies, of which there were only two.

REVIEWER	Claire de Oliveira Institute for Mental Health Policy Research, Centre for Addiction and Mental Health, Canada
REVIEW RETURNED	27-Nov-2018

GENERAL COMMENTS	The effect of deinstitutionalisation for adults with intellectual disabilities on costs: a systematic review The objective of this study was to undertake a systematic literature review of the existing work that has examined the effect of deinstitutionalisation on costs of care for adults with intellectual disabilities. Unfortunately, very few moderate to high quality studies were retrieved, which limited the scope of the analysis. Nonetheless, the authors found two studies with conflicting findings. Overall, the manuscript would benefit from further detail. Also, given that the two studies retrieved provided conflicting results, it would be useful to understand whether the authors have an insight as to why this is the case. In addition, it would be good to discuss the clinical and policy implications of their findings. Abstract Intervention should be "from institution to residential setting" Primary and secondary outcomes measures – are costs to the health care system only considered? Search should be "from database inception to September 2017". Given the lack of studies, did the authors also check the references of relevant articles?

Results state that included studies examined people leaving NHS hospitals in the 1980s; however, at least one study examined the issue in the 90s. The authors should also clarify the type of costs in which there was a reduction/increase.

In the conclusion, the authors state that there are significant gaps in the evidence base – which gaps are these?

Strengths and limitations
“Two researchers independently reviewed returned studies for eligibility.” – Why is this mentioned as a strength? Isn’t this standard practice for systematic literature reviews? The same applies to “Included studies were additionally subject to quality assessment to minimise bias.” –again, this is standard practice.

One potential strength of this work would be the fact that the authors examined a topic that has not been covered before or the fact that this review examined an under-served population.

Initially it was not clear why cross-sectional studies were excluded; this was later clarified in the manuscript. Perhaps include a bit more detail in the abstract.

Introduction
The authors should provide a bit more background on the relevance of examining the economic outcomes of de-institutionalisation. The authors should also provide a better explanation as to why it is important to examine this patient population. In addition, the authors should provide a bit more description around how th3 population with ID has changed over time and the implications of this to the care they receive and, consequently, the costs of care.

Methods
Minor comment: while PICOS and PEOS are indeed acronyms, these are frameworks that help guide researchers undertaking literature reviews.

When describing the type of participants, there authors should define “intellectual disabilities.”

Type of outcomes. The primary outcome was economic effects; however, in practice, the authors were mostly interested in the cost effects. Furthermore, cost effects were defined broadly as any cost-consequence framework or resource use. Were the authors interested in resource use alone or resource use that could be quantified (i.e. expenditure)? This section was a bit difficult to understand.

Database search. One of the search terms (in the appendix) was 1915. What is meant by 1915? How was the grey literature search done? Was this done through a search engine and, if so, which one? For example, did the authors use OpenGrey and/or Grey Literature Report? A bit more detail around this would be helpful.

Screening citations. Do the authors have any additional information on consensus (i.e. consensus scores)?

Assessment of methodological quality/risk of bias. The authors state that one reviewer assessed the studies methodological quality and a second reviewer performed a rapid assessment. Should both reviewers have assessed the quality of all studies? Again, any information around level of consensus? To be included, studies were required to meet a minimum of moderate to high quality of rigour. How was this defined? What criteria were used?

Data extraction. How were the data extraction forms developed? Was jurisdiction included under “study setting”? Was “perspective” included in the form? Shouldn’t two reviewers have extracted the data and then come together to discuss?

Analytical measures. The authors were hoping to pool the data across studies but were not able to do so. This could be mentioned as limitation (that the studies retrieved did not enable this type of analysis). Instead, the authors present a narrative synthesis of the data using descriptive statistics and thematic analyses. Were any frameworks used to help with this?

Patient and public involvement. It was nice to see that the authors engaged a representative from the National Disability Authority of Ireland. But wouldn’t this be more of a policy advisor instead of actual PPI?

Results
The authors were not able to retrieve 32 studies. From the list, it seems these studies would cover both costs and quality of life. This should be stated. Was there no way that these studies could be retrieved? This is a fairly large amount. I was able to find at least 5 of those studies and would be happy to share those with the authors, if they are interested.

Discussion
The authors mention that the two studies retrieved are not representative of individuals with ID today. Are there any references that could be added to bolster this statement? Also, is there any chance that there may have been more recent studies published in the last year? The search was done until September 2017.

Do the authors have any idea of why the studies provided conflicting results? It would be important to provide some discussion around this. Also, there was no discussion round the clinical and/or policy implications of their findings.

The authors state that the review suffers from language publication bias. Moreover, they state that no work from the Nordic countries was found. However, some of this work may exist but purely offline. Is there any possibility that some of this work could be retrieved? This is a very minor comment as this may indeed be difficult to obtain.

Overall
Minor comment: the manuscript would benefit from some editing.

The reviewer provided a marked copy with additional comments. Please contact the publisher for full details.

VERSION 1 – AUTHOR RESPONSE

Reviewer(s)' Comments to Author:

Reviewer: 1

Reviewer Name: Victoria Ratti

Institution and Country: Division of Psychiatry, University College London, UK

Please state any competing interests or state ‘None declared’: None declared

Please leave your comments for the authors below

Thank you for the opportunity to review the manuscript “The effect of deinstitutionalisation for adults with intellectual disabilities on costs: a systematic review.”

The manuscript provided an interesting, well-written and concise read. The systematic review was generally methodologically sound and it is evident that the authors followed PRISMA guidelines.

I believe, nonetheless, that the manuscript can be strengthened and I have provided specific comments for each section which I invite the authors to consider.

Introduction

This section is well written and concise and provides a valid rationale for the need to conduct economic evaluations for different accommodation types for people with intellectual disabilities (ID).

1.1 Nevertheless, deinstitutionalisation has been ongoing for a number of decades and has been advocated in publications such as Better services for the Mentally Handicapped (1971). The majority of people with ID in countries such as the UK and Australia no longer live in institutions and now reside in the community. I believe the authors need to provide a stronger rationale for comparing the costs and outcomes of various accommodation options to institutions rather than comparing different residence types within the community, a topic which would appear more relevant to the current provision of care landscape.

>>> This choice of exposure variable reflected the interests of the Irish Department of Health, who commissioned this work to inform ongoing policy reforms. The policy of de-congregation was published in Ireland as recently as 2011, much later than many other western countries, and implementation has been much slower than anticipated. Thus the specific need for the review at this time to inform Irish policy implementation, and this is also the case in many other countries not as advanced in the process as the UK and Australia. In addition, the primary focus is on the ‘moving’ process and not on a comparison of the variety of different living arrangements which may be available.

To explain further this choice and why this work remains of interest we have added a paragraph to Discussion>Limitations (beginning: “This choice of intervention – the process of deinstitutionalisation, and not comparative analysis of outcomes living in one setting versus another - reflected the interests of the Irish Department of Health, who commissioned this work to inform ongoing policy reforms”).

1.2 Although I agree with the authors that “the accurate estimation of resource use in providing services can inform budgeting, workforce planning and organization of services”, I believe that nowadays it would be more useful to have information on costs and outcomes of different accommodation types in the community such as supported living versus residential care homes. Even if the results of this review had shown that institutions were more cost-effective, where outcomes such as choice, self-determination and community participation have been demonstrated in previous research to be improved with the move to the community, increases in cost may be justified.

>>>We agree that evaluative frameworks should not be interested purely in resource use but also whether changes in costs are good value in the context of changes in outcome, and we recognise this in the Introduction (“Economic evaluations comparing the costs and outcomes of different options….”) and the Conclusion (“It is critical that more studies are conducted to understand both how to best support this growing population in leading independent lives of their choosing…”).

Our eligibility criteria included cost-consequence analyses (e.g. cost-effectiveness, cost-utility analyses) that combine outcomes and costs to evaluate whether cost increases or decreases are justified in the context of changes in outcome. No such studies were identified and therefore the literature was not available to answer the question the reviewer has posed.

1.3 Additionally, given that the great majority of the literature on deinstitutionalization is from the 80s-90s and that there have been previous reviews on the topic (e.g. Kozma, Mansell and Beadle-Brown, 2009, QoL; Walsh, Kastner and Reed, 2003, cost) I wonder if the authors need to make a stronger case why a systematic review is needed now.

>>>Per response to 1.1, this review was commissioned by policymakers seeking the most recent evidence on this topic. While our review finds no new papers since 2003, it was reasonable to seek some given that this remains an ongoing process in many countries around the world. As well as providing an update on the state of the science, this review advances the work done previously in the scope of its systematic searches.

1.4 That being said, in the UK, there are still approximately 2600 people with ID living in inpatient settings (Health and Social Care Information Centre, 2016) who are likely to move into the community, and more recent publications such as Building the right support (2015) still emphasise that people shouldn’t be staying in inpatient units unnecessarily, suggesting that long stays are still a current issue. In many countries the process of deinstitutionalisation is far from complete and therefore information regarding costs is useful to inform current and future services. I think the authors could emphasize these points to provide a stronger rationale for examining the cost effects of deinstitutionalization and reiterate that there are still high numbers of individuals living in institutions in the present day.

>>>We have revised the second paragraph of the introduction (“While significant numbers of people have moved out of institutional settings over the last half century…”) and added the ‘Building the right support’ reference.

1.5 At line 33 the abbreviation ID is given but the non abbreviated term intellectual disabilities is still largely used throughout the manuscript.

>>>We have deleted this abbreviation and any other uses in the manuscript, using the non-abbreviated term throughout. We are happy to make further changes on instruction from the editors.

Method

The methodology section is well-written, clear and comprehensive. I do however have some queries regarding some of the methodological decisions made by the authors which are outlined below:

1.6 Types of participants: it is stated that participants were adults with ID. Did you use a specific criteria such IQ <70 to select studies or considered all studies where the authors had described participants as having ID?

>>>Added to Methods>Participants:

No pre-determined operational definition of intellectual disability was used; we followed author definition in the first instance and planned to resolve ambiguities through discussion and author contact if necessary.

1.7 Types of studies/reports: I found the choice to exclude cross-sectional studies comparing community-living and institutional arrangements surprising as various studies have used this methodology and provided useful information on costs. I believe it would be useful to justify this decision.

>>>See response to 1.1 above.

1.8 Database search: The authors do not report on supplementing their electronic database searches by hand-searching the references of selected papers, which is often also done when conducting systematic reviews. Was this step omitted?

>>>Added Abstract>Search

We searched MEDLINE, PsycINFO, CENTRAL, CINAHL, EconLit, Embase and Scopus to September 2017 and supplemented this with grey literature searches and hand searching of the references of eligible studies.

>>>Added Results>Database search

We reviewed references of two included studies and did not identify further eligible studies for inclusion.

1.9 A comprehensive description of the search strategy is provided in Appendix 1. Here I noticed that the search was not only focused on articles on cost but also on articles of QoL. This is not clear in the main article. Although it is mentioned in the results section that the numbers are reflective of a combined search of articles reporting on both costs and QoL, this comes a bit out of the blue and leaves the reader questioning why QoL focused studies were included when the focus of the review is on cost, and QoL studies are not mentioned at all in the present article. I think the authors need to be transparent about this in the search strategy section or conduct separate searches for cost and Qol articles.

>>>The original review was conducted for both QoL and costs. We report this at the Introduction ("We undertook a systematic review of the evidence on the effect of deinstitutionalisation on economic outcomes and on quality of life (QoL) for people with intellectual disabilities…").

The QoL review is also with the BMJ Open editors. We are happy for them to provide a copy on request. In the event that both are accepted for publication, we are happy to take specific guidance from editors on how the two reviews will be cross-referenced.

1.10 Assessment of methodological quality/risk of bias: The authors describe using CASP checklists to appraise the studies. CASP tools do not suggest a scoring system, so I was wondering how the authors determined moderate/high quality studies? Did they use a cut-off score/ % of "YES" answers? I believe, for clarity, that this should be specified and it may also be useful to add the main questions (not necessarily the prompts) of the CASP checklists in the Appendix.

>>>In anticipation of multiple possible study designs (e.g. economic evaluation, cohort study with cost as an outcome), we chose the CASP tool because it has been used previously in reviews, and tools have been developed for the varying study designs. Furthermore all CASP checklists cover the three main areas of validity, results and clinical relevance. The CASP tool offers a set of 10 questions to which the reviewer assigns Yes, No or Can't tell judgements. Those studies that receive all (or mostly) Yes assessments were judged to be of High quality (low risk of bias) and similarly, those receiving majority 'No' or Can't Tell assessments were considered of moderate or low quality.

We have added this explanation to Methods>Assessment of methodological quality/risk of bias and we have provided the results of this quality assessment in a table (Table 2).

We have added an Appendix 3 with CASP questions.

1.11 Main results. On page 17 line 11 it is stated that many participants moved from institutions to "larger institutional settings". Although these nursing homes/hospices are described as community settings it leaves one wondering if this was actually a form of deinstitutionalization.

>>>We share this observation and have acknowledged it in the Results (paragraphs starting "An additional shared limitation was the use of the term "community care". …").

Additionally place “community” in inverted commas through the reporting of results.

Finally, we have revised the opening sentence of Discussion>Key findings to further spell this out:

Lack of detail on the type of “community care” that participants moved to, as well as limitations in reporting of cost data and their characteristics, not only precludes meta-analysis but raises questions about the extent to which observed transitions are deinistutionalisation as understood in contemporary policy and practice.

1.12 Table 2. I believe, where possible, it would be useful to report p values/effect size.

On page 19 line 19-20 is stated that secondary analysis shows that accommodation accounts for 81-86% of community costs post-move. I think it may need to be specified or provide examples of which other variables were considered as expenditure/cost (e.g. day activities, health services etc.). I was unclear whether the authors conducted the secondary analysis or if these were reported in the original paper.

>>>Paired t-test results added from Hallam study. Secondary analyses were reported by the authors but since they did not relate directly to our objective we preferred not to include them in the table and instead deleted mention of these factors.

Discussion

1.13 On page 20 line 25-26 it is stated that there are significant associations between costs and both participant and residence characteristics. These however were not reported in the results section and therefore it is unclear what these characteristics were. The same phrase is reported in table 2 where I think these characteristics and their associations should be specified.

>>>This sentence has been deleted as part of revisions to the Results and Discussion, particularly with respect to Reviewer 2 Comments below.

Appendix.

1.14 -Pages 36-37 on my PDF are a repetition of Appendix 3. This may be a mistake.

>>This was an error. A new Appendix 3 has been added (see 1.10).

1.15-The PRISMA checklist is included in the Appendix but it is not mentioned in the main article. Using the PRISMA checklist to conduct/write the review could be added as a strength and evidence of the methodological rigor of the review.

>>>Addition to Discussion>Strengths and Limitations:

This study has followed best practice guidelines in systematic evidence reviews where possible, following the PRISMA guidelines.

1.16 -I think including another systematic review in the appendix is unnecessary, particularly as the appendices of this second article are a repetition of the appendices of the article under review (cost review). I understand that the systematic search combined terms of both cost and QoL which were pooled and the authors may want to include an appendix with the references of the QoL articles that were retained. However I believe that the QoL review should be submitted as a separate article rather than being included in the current article's appendix. Ideally for reasons of replicability, i believe it would be more appropriate to conduct separate searches for the two reviews, one including only cost terms and the other including only QoL terms and I invite the authors to consider this.

>>> The original search was conducted together (QoL and cost) and cannot be delineated after the fact; the submission of two separate reviews was made at the request of BMJ Open editors. See also 1.9: The QoL review is also with the BMJ Open editors and we are happy to take specific guidance from editors on how the two reviews will be cross-referenced.

I have endeavoured to provide a comprehensive review and I hope the authors find my comments/suggestions useful.

>>>Many thanks to the reviewer for their careful and considered comments. We consider the revised manuscript is much stronger as a result.

Comments to specific questions

1.17<i>Question 4. Are the methods described sufficiently to allow the study to be repeated?</i> I have addressed these points in the comments and these relate to the definition of ID used, the omission in the main article of the combination of cost and QoL search terms and how moderate/high quality was determined from CASP checklists.

>>>These points have been variously addressed as raised above.

1.18 <i>Question 10. Are the results presented clearly? </i> For the most part results are presented clearly, however as stated in my comments I believe it would be useful to include p values and effect sizes where possible and also spell out the associations between cost and individual and residence characteristics. Also I think there needs to be more clarity regarding the secondary analysis results i.e. accommodation counted for 81% of cost, but what other variables were considered? And who conducted the analysis?

>>>These points have been variously addressed as raised above.

Reviewer: 2

Reviewer Name: Martin Knapp

Institution and Country: LSE, UK

Please state any competing interests or state ‘None declared’: None declared, except that I was a co-PI on one of the two studies discussed in this review paper, and the lead PI on the other.

Please leave your comments for the authors below

Broadly speaking, this is a reasonably well-conducted systematic review. One limitation is that only one reviewer undertook quality assessment and data extraction (as the authors recognise). Two other limitations can be noted.

2.1 One is that what the authors describe as ‘patient and public involvement’ does not look to be what I understand by the term. Perhaps I am missing something in the paper, but I assume from the omission of the explanation that this representative was not somebody with an intellectual disability.

>>>We have revised the relevant section:

There was no involvement of any person with an intellectual disability or the wider public in this systematic review. A representative from the National Disability Authority of Ireland, an independent state body providing expert advice on disability policy and practice to the government and the public sector, participated in the design of the systematic search strategy to maximise relevance to current policy and practice.

2.2 The second limitation is that the authors do not appear to have fully analysed or described the studies they selected. The study described as Hallam et al is the 12-year follow-up of a group of people leaving hospitals in England. The authors of this paper mention the earlier studies (baseline, 1-year and 5-year follow-ups) but miss elements in those earlier analyses in making their criticisms. As I understand the purpose of the review, it was not only focused on very long-term economic effects, so why were earlier findings ignored?

>>>Both books were read in detail in the initial review and have been re-read in preparing this response to reviewers. The manuscript itself and this response reflect best attempts to interpret the reported work for the purposes of our review, and where errors or oversights have been made we welcome specific pointers. However, we are for the most part satisfied that the manuscript reflects the evidence for our review.

As our PICOS lays out, we are interested in studies that evaluated the effect of deinstutionalisation on costs/cost-effectiveness for adults with ID.

As worded, our Objective in the Abstract and Introduction is perhaps imprecise (speaking of “economic evidence”, a more nebulous concept). We have therefore revised the wording of Objective in both locations:

To review systematically the economic evidence on the costs and cost-effectiveness of deinstitutionalisation for adults with intellectual disabilities.

While the included studies were ground-breaking on their own terms and the books include large volumes of detail, only small amounts of these data are suited for addressing our objective. Essentially there are three difficulties in interpreting the papers and the books to our purposes.

First, the 'exposure' variable. While details are given on the characteristics of different accommodation types in Table 6.2 of 'Care in the community: Challenge and demonstration' (1992), the primary cost results in Hallam et al (Table 7) group all destinations together. The Beecham paper groups the NI participants by sector.

These are important limitations for this review's purposes in the context of (a) major variation in costs across settings, as the same study reports elsewhere, and (b) variation in the settings themselves – for example, nursing homes and hostels are widely understood to be institutions (see also 1.11 above).

Second, evaluating the significance of association between predictor and outcome. Paired t-tests are used in Hallam et al to evaluate the Table 7 data, but pooling of designations as one 'community' category has the same limitations mentioned in the previous paragraph. Where costs are disaggregated by destination (Table 13.10 in 'Challenge and demonstration' (1992); Table 7.4 in 'Five years on' (1994)) there are some very small sample sizes and no statistical tests reported. One follow-up in Table 13.11 in 'Challenge and demonstration' reports a statistically significant F statistic but it's not clear which head-to-head pairs are significantly different in post-hoc tests (e.g. Tukey HSD, Bonferroni, etc.).

Third, with respect to sample. While the books provide baseline data on the overall study sample, it is not clear who is in the sub-samples for cost analyses. For example, Table 5.1 in 'Challenge and demonstration' indicates a baseline sample of 529 people with ID, of whom 227 are described at one year. In 'Five years on', this one-year sample is retrospectively reported as 265 (Box 3.2, page 22) and dropping to 203 by the five-year follow-up, of whom 162 participants have data for all three timepoints. The corresponding cost analyses have 194 participants at one year (e.g. Table 13.10) and 159 at five years (Table 7.4). For the 12-year follow-up in Hallam et al. (2006), the baseline sample is described as 373, the one-year sample as 207 and the 12-year longitudinal sample is 103.

Nevertheless, we appreciate the feedback and we have made a series of revisions to Results and Discussion in respect of these points:

- Lack of clarity over destinations and the association with outcome do not reflect a lack of detail in the books but rather pooling and reporting of disparate destinations in ways that hinder interpretation;

- We have added some additional descriptive material to Table 1 from 'Challenges and demonstration' to describe the Hallam sample but concerns about the representativeness of the cost sub-samples when we have only whole-sample baseline data remain.

- We aim to reflect the results of the studies as reported at the time, but we interpret these results in the context of contemporary policy and practice challenges (e.g. consideration of nursing homes as "community" destinations, growing prominence of independent living as the policy ideal).

Specific responses to further points follow below.

2.3 For example, on p.20 the authors write 'One-off costs associated with deinstitutionalisation also appeared not to be taken into account'. This is incorrect in relation to the English study. The book reporting the 1-year results includes a 23-page chapter on the financing of community care, and there is discussion around set-up costs and other challenges elsewhere in the book. I will mention other missing elements below.

>>>We did not identify specific mention of one-off costs associated with deinstitutionalisation in ‘Challenge and demonstration’ or in Hallam et al., and as such it is not clear how these are factored into the results reported in the latter paper. However, we acknowledge the correction and have deleted the sentence.

2.4 A second example is where the authors write (p.21) that ‘Details on the participants were similarly scant’. The English study gives information on the following characteristics for sample members at baseline: gender, local authority of origin, severity of intellectual disability, skills (i.e. ADLs), behavioural issues, satisfaction with services, social interaction (quantity and self-reported quality), depression, psychosocial function, life satisfaction (Cantril’s Ladder). I struggle to understand what further data are needed to persuade the authors that the details are not ‘scant’.

>>>We have added the following to Main results>Description of included studies

in the supporting books the authors provide detailed baseline data (including gender, severity of intellectual disability, skills, behavioural issues, social interaction, depression, psychosocial function, life satisfaction) on the original recruited sample of people with intellectual disabilities (n=529) but it is not clear how representative is the sub-sample of 103 reported in the paper.

We have removed the sentence describing details as “scant” in a revised paragraph beginning, “Both studies report cost analyses for sub-samples drawn from larger studies.”

2.5 The strategy adopted by the team not to specify ex ante what was meant by ‘institution’ or ‘community’ was the right one. As they say later in the paper (p.22) ‘No ex ante definition of institutional community settings’ was employed. However, the discussion of findings certainly employs an ex post judgement on what is meant by ‘community’, but without explicating what it is. For example, on p.17 they write that only one person across the two studies ‘can be said certainly to have moved to community-living arrangements as understood in the 21st century’, by which they mean independent living, I think. I would be happier with this statement if the authors explained whose view of ‘community-living arrangements as understood in the 21st century’ they are referring to here, and added a reference or two. If the only interpretation of ‘community-living’ is living independently, then I think their statement is misplaced.

>>>We have deleted the section “as understood in the 21st century” and made revisions to how the Discussion is presented.

The essential point here is that we deliberately adopted a broad approach to defining the intervention to capture as many relevant studies as possible but older studies nevertheless have to be assessed for policy usefulness on today’s terms: evaluations of movement from long-stay hospitals to nursing homes do not assess deinstitutionalisation as currently understood, and interpretation occurs in this context.

2.6 Linked to this is the sentence ‘An additional shared limitation [of the two included studies] was the use of the term ‘community care’’ (p.16). At the time these two studies were conducted, the term ‘community care’ reflected not only a policy position but also the position of advocates in the sector. Of course, a large residential home would not be seen as ‘community’, but are the authors arguing that supported lodgings, foster care and unstaffed group homes are really not considered to be community care today? It is fine for the authors to criticise terminology employed in research 20-30 years ago – that is to be expected in an area such as this – but it is a failing of the paper not to offer any clear alternative definition.

>>>Per 2.2 above and the revised discussion, the point is not that none of the destinations studied qualify as community care in a 21st century context but that the effect of moving from institutions to these destinations is not isolated in the reported studies (but, rather, pooled with moves to destinations that do not qualify as community care in a 21st century context).

2.7 On p.12 the authors write ‘None of the included studies evaluated both cost data and QOL’. The authors do not define what is meant by ‘QOL’, but I would have imagined that using a life satisfaction measure such as Cantril’s Ladder and a measure of self-expressed satisfaction with living environment and life in general by individuals with intellectual disability would come reasonably close to ‘QOL’. If I make a leap of assumption here, I did wonder if by ‘QOL’ the authors meant the sort of thing that most health economists understand by the term, which is health-related quality of life measure of the kind measured by tools such as EQ-5D. If so, then I can understand why they did not find any such studies, which might be because an HRQOL is unlikely to be prioritised among the outcomes of studies in the intellectual disability field. If by ‘QOL’ the authors mean concepts similar to those reflected in the search strategy, then at least one of these two included studies looks at ‘QOL’.

>>>This sentence describes only the eligibility of studies for the QoL review (see 1.9 above) and this cost review. Revised this sentence to:

No study was eligible for both the QoL review and this economics review.

The scope of your study as it relates to outcomes, is described in the final sentence of Results>Key findings:

While both overall studies to which the cost papers were attached did examine client outcomes, no cost-consequence analysis or ratio is reported in either study.

2.8 The authors excluded studies that they call ‘static comparisons’, by which they mean studies that do not look at change over time but simply compare two or more different settings within a cross-sectional design. This may well have been sensible in order to make their reviewing task easier, but it does not necessarily help policy-makers. On p.21 they explain why they chose this strategy, including the fact that ‘they did not evaluate the impact of a move in residential setting, and there are additional concerns about the robustness of such comparisons and unobserved confounding, particularly with routinely collected data’. But earlier on that same page, they note the need for evidence in relation to, for example, ‘younger people with very complex needs and challenging behaviours’. Many of the latter group may never live in an institutional setting as defined in this review, and so confining a review to just moves from institutions will not provide information on such a group. Presumably, the authors therefore believe that a different review is needed, which feels something like a missed opportunity. Having gone to the effort of designing what generally looks like a good search strategy, it does seem a pity that potentially rich evidence has been discarded. I agree that there can be difficulties in the interpretation of evidence because of unobserved confounding and suchlike, but then this review could have provided a great service by highlighting the limitations of studies that were uncovered.

>>>Per 1.1, this research question was commissioned by policymakers in our country (Ireland). Naturally relevance to other countries will vary but we are hopeful that the work is useful internationally in the context of limited evidence of an underserved group.

An additional review would be necessary to examine the cross-sectional literature and given this feedback, we are considering developing a separate review of cross-sectional studies.

2.9 On p.23 the authors write ‘The level of detail gathered in those [two included] studies is not fit for today’s policy purposes’. In part I think this is inevitable, but the reviewers are, in my judgement, simply wrong to use the justification that the two studies did not report sufficient detail. As noted earlier, the authors have failed to look at the evidence in the included studies, of which there were only two. >>>See 2.3 above. We have revised and restructured the Discussion to better clarify the arguments here. In particular these are now laid out in three paragraphs in the Discussion, starting “There are three principal barriers to interpreting these results, the apparent inconsistency between their key findings, and the lessons for contemporary policy and practice…”

We thank the reviewer for their detailed consideration of this work.

Reviewer: 3

Reviewer Name: Claire de Oliveira

Institution and Country: Institute for Mental Health Policy Research, Centre for Addiction and Mental Health, Canada

Please state any competing interests or state ‘None declared’: None declared.

Please leave your comments for the authors below

The effect of deinstitutionalisation for adults with intellectual disabilities on costs: a systematic review

The objective of this study was to undertake a systematic literature review of the existing work that has examined the effect of deinstitutionalisation on costs of care for adults with intellectual disabilities. Unfortunately, very few moderate to high quality studies were retrieved, which limited the scope of the analysis. Nonetheless, the authors found two studies with conflicting findings. Overall, the manuscript would benefit from further detail. Also, given that the two studies retrieved provided conflicting results, it would be useful to understand whether the authors have an insight as to why this is the case. In addition, it would be good to discuss the clinical and policy implications of their findings.

Abstract

3.1 Intervention should be “from institution to residential setting”

>>>Per the main manuscript Methods> Types of intervention/exposure/comparators, we did not define ex ante what constituted a ‘residential setting’. As noted above in Reviewer Comment 1.1, and also our response to 2.3, some would challenge the destinations for some participants in included studies as representing a “residential setting”. We therefore prefer to leave this Abstract definition as it is.

3.2 Primary and secondary outcomes measures – are costs to the health care system only considered?

>>>Revised:

within any cost-consequence framework (e.g. cost-effectiveness analysis, cost-utility analysis) or resource use typically considered to fall within the societal viewpoint (e.g. cost to payers, service-users, families, informal care costs).

3.3 Search should be “from database inception to September 2017”. Given the lack of studies, did the authors also check the references of relevant articles?

>>>See 1.8 above.

3.4 Results state that included studies examined people leaving NHS hospitals in the 1980s; however, at least one study examined the issue in the 90s. The authors should also clarify the type of costs in which there was a reduction/increase.

>>>Amended:

between 1984 and 1992

3.5 In the conclusion, the authors state that there are significant gaps in the evidence base – which gaps are these?

>>>>Added:

Significant gaps in the evidence base were observable, particularly with respect to priority populations in contemporary policy: older people with intellectual disabilities and serious medical illness, and younger people with very complex needs and challenging behaviours.

Strengths and limitations

3.6 “Two researchers independently reviewed returned studies for eligibility.” – Why is this mentioned as a strength? Isn’t this standard practice for systematic literature reviews? The same applies to “Included studies were additionally subject to quality assessment to minimise bias.” –again, this is standard practice.

>>>First bullet revised:

Examining a topic that was not previously the subject of a systematic review, we searched seven databases of peer-reviewed literature evaluating returned studies using two independent reviewers.

3.7 One potential strength of this work would be the fact that the authors examined a topic that has not been covered before or the fact that this review examined an under-served population.

>>>Second bullet revised:

Identified evidence therefore represents state of the science on a pressing policy question for an underserved population.

3.8 Initially it was not clear why cross-sectional studies were excluded; this was later clarified in the manuscript. Perhaps include a bit more detail in the abstract.

>>>Fourth bullet revised:

Commissioned by policymakers to examine specifically the process of deinstitutionalisation, we did not include cross-sectional studies comparing outcomes for different populations in different settings.

Introduction

3.9 The authors should provide a bit more background on the relevance of examining the economic outcomes of de-institutionalisation. The authors should also provide a better explanation as to why it is important to examine this patient population. In addition, the authors should provide a bit more description around how th3 population with ID has changed over time and the implications of this to the care they receive and, consequently, the costs of care.

>>>We have clarified the continuing importance of deinistutionalisation in the revised paragraph beginning, “While significant numbers of people have moved out of institutional settings over the last half century.”

>>>We address changing ID population over time and growing support needs in the paragraph beginning “The population of people with intellectual disabilities is changing in important ways that must be accounted for in planning and provision of services…”.

>>>The Introduction as a whole is intended to establish why it is important to examine this patient population. We are happy to make further revisions on editor instruction.

Methods

3.10 Minor comment: while PICOS and PEOS are indeed acronyms, these are frameworks that help guide researchers undertaking literature reviews.

>>'acronyms' changed to 'frameworks'

3.11 When describing the type of participants, there authors should define "intellectual disabilities."

>>>See 1.6 above.

3.12 Type of outcomes. The primary outcome was economic effects; however, in practice, the authors were mostly interested in the cost effects. Furthermore, cost effects were defined broadly as any cost-consequence framework or resource use. Were the authors interested in resource use alone or resource use that could be quantified (i.e. expenditure)? This section was a bit difficult to understand.

>>>Paragraph revised:

Our pre-specified primary outcome of interest was economic effects. For purposes of the review, economic effects were defined broadly as any cost-consequence framework (e.g. cost-effectiveness analysis, cost-utility analysis) or resource use quantified as costs. We did not require that resource use reflect the literal cost of provision for the study to be included, but also considered eligibility based on other approaches to calculating expenditures, such as insurance programme charges, frequency utilisation combined with unit cost data. We considered eligible any perspective typically considered to fall within the societal viewpoint (e.g. cost to payers, service-users, families, informal care costs).

3.13 Database search. One of the search terms (in the appendix) was 1915. What is meant by 1915? How was the grey literature search done? Was this done through a search engine and, if so, which one? For example, did the authors use OpenGrey and/or Grey Literature Report? A bit more detail around this would be helpful.

>>>Added to Appendix 1:

1915 was included to identify US studies of the relevant Medicaid waiver to meet the needs of people who prefer to get long-term care services and supports in their home or community, rather than in an institutional setting (https://www.cms.gov/Outreach-and-Education/American-Indian-Alaska-Native/AIAN/LTSS-TA-Center/info/national-overview-1915-c-waivers.html).

>>>With respect to grey literature strategy, see Appendix 2:

The subject experts decided ex ante to search based on country and centres of disability studies (and not specific grey databases such as OpenGrey, OpenSIGLE and GreyNet) as these were deemed the strongest source of potentially relevant material.

3.14 Screening citations. Do the authors have any additional information on consensus (i.e. consensus scores)?

>>>No, we did not record consensus scores.

3.15 Assessment of methodological quality/risk of bias. The authors state that one reviewer assessed the studies methodological quality and a second reviewer performed a rapid assessment. Should both reviewers have assessed the quality of all studies? Again, any information around level of consensus? To be included, studies were required to meet a minimum of moderate to high quality of rigour. How was this defined? What criteria were used?

>>>See 1.10 above.

3.16 Data extraction. How were the data extraction forms developed? Was jurisdiction included under "study setting"? Was "perspective" included in the form? Shouldn't two reviewers have extracted the data and then come together to discuss?

>>>As already noted in Data analyses>Data extraction, we predesigned and piloted data extraction forms. Characteristics of economic evaluations to be recorded were evaluation type, perspective, time horizon, currency, discount rates, statistical models used and sensitivity analyses. In practice, no full economic evaluations were identified and most of these fields were redundant.

>>>While two reviewers at all stages of a systematic review is optimal, it is acceptable to pursue the approach that we report. This approach was decided ex ante in consultation with a systematic review methodologist (VS).

3.17 Analytical measures. The authors were hoping to pool the data across studies but were not able to do so. This could be mentioned as limitation (that the studies retrieved did not enable this type of analysis). Instead, the authors present a narrative synthesis of the data using descriptive statistics and thematic analyses. Were any frameworks used to help with this?

>>>The challenges in interpretation are now addressed in paragraphs beginning: "There are three principal barriers to interpreting these results, the apparent inconsistency between their key findings, and the lessons for contemporary policy and practice."

3.18 Patient and public involvement. It was nice to see that the authors engaged a representative from the National Disability Authority of Ireland. But wouldn't this be more of a policy advisor instead of actual PPI?

>>>See 2.1 above.

Results

3.19 The authors were not able to retrieve 32 studies. From the list, it seems these studies would cover both costs and quality of life. This should be stated. Was there no way that these studies could be retrieved? This is a fairly large amount. I was able to find at least 5 of those studies and would be happy to share those with the authors, if they are interested.

>>>We state at the start of Results>Database search that “The database search, which was a combined search of studies reporting on both cost and QoL, returned 25,853 citations for consideration against the review’s eligibility criteria…. “

Per 1.9 and 1.16 above, BMJ Open editorial requested separate reviews from this single search. The QoL review is also with the BMJ Open editors and we are happy to take specific guidance from editors on how the two reviews will be cross-referenced.

Discussion

3.20 The authors mention that the two studies retrieved are not representative of individuals with ID today. Are there any references that could be added to bolster this statement? Also, is there any chance that there may have been more recent studies published in the last year? The search was done until September 2017.

>>>References have been added in the relevant paragraph of the revised discussion.

>>>With respect to timeliness, please see 0.1 above.

3.21 Do the authors have any idea of why the studies provided conflicting results? It would be important to provide some discussion around this. Also, there was no discussion round the clinical and/or policy implications of their findings.

>>>See three paragraphs in the revised discussion, starting “There are three principal barriers to interpreting these results, the apparent inconsistency between their key findings, and the lessons for contemporary policy and practice.”

3.22 The authors state that the review suffers from language publication bias. Moreover, they state that no work from the Nordic countries was found. However, some of this work may exist but purely offline. Is there any possibility that some of this work could be retrieved? This is a very minor comment as this may indeed be difficult to obtain.

>>>We are not certain that such analyses exist and are consequently unaware of any means to retrieve this work.

Overall

3.23 Minor comment: the manuscript would benefit from some editing.

>>>We have made various revisions in respect to three sets of reviewer comments and are happy to make further revisions on editor request.

We thank the reviewer for their detailed consideration of this work.

VERSION 2 – REVIEW

REVIEWER	Victoria Ratti Division of Psychiatry, University College London
REVIEW RETURNED	21-Jan-2019

GENERAL COMMENTS	Thank you for the opportunity to review the revised manuscript "The effect of deinstitutionalisation for adults with intellectual disabilities on cost: a systematic review. The manuscript has been improved following reviewers' comments however there are still some questions/comments which I think need addressing. In general the methodology of this review was sound and generally well thought out. My main concern is not so much with the methodological rigour of the review but rather with the relevance of the subject and the findings. The authors have argued that this work reflects needs of the policy landscape in countries such as Ireland and other countries where deinstitutionalisation is still in early phases or isn't as advanced as in other countries. Perhaps this needs to be emphasised to provide a stronger rationale for the need of this review now. I also think the authors need to reflect on what these findings add? What do they mean and what are the implications? Although I think it was good review my main question is, was it necessary? I invite the authors to emphasise this point. Abstract Intervention: a move in residential settings. This is unclear as it can lead the reader to believe that the review includes studies where any move from any type of residence were considered. Although I understand that the authors did not want to provide a definition ex ante, the current statement does not suggest that it was from institutions to other settings and it is misleading. Introduction Given that in the responses to reviewers the fact that the work was commissioned by the Irish Department of Health is often used as a reason as to why certain decisions were made, the authors may want to be clear about it and introduce this earlier in the paper rather than in the discussion; furthermore in response 1.1. to the reviewers it was stated that this review reflects the needs of countries which are less advanced in terms of deinstitutionalisation (e.g. Ireland), perhaps this should me mentioned in the intro as a rationale for conducting the review. Method CASP: studies were considered high quality if all or mostly all criteria were marked as yes. This still does not allow replication what does mostly mean? 60%, 70%, 90% of criteria? All three reviewers noticed that cross-sectional studies were not included and I still believe that this is a limitation as rich information on costs could be extrapolated. The authors have justified their decision based on the work being commissioned by Irish policy makers for a specific purpose (i.e. deinstitutionalisation/move) (in the responses) and I believe this needs to be addressed a stronger case needs to be made to support this decision. Another reason for excluding cross-sectional studies was to limit possible confounding bias, although I agree that it could have made findings more difficult to interpreted, as mentioned by reviewer 2 this might have been a missed

	opportunity and I think a stronger case needs to be made about why they were not included. Discussion I think that the discussion is missing one important bit which is what do we do with these findings? Ok we need more research, but what is the point of these results now? Is the fact that we need more research the main conclusion of the study? The work was commissioned by policy makers, what is the main message? How does this help them now? I think there needs to be a bit more discussion around this, as at the moment it doesn't seem like there is any major conclusion/point to be made from this review.

VERSION 2 – AUTHOR RESPONSE

Thank you for the opportunity to review the revised manuscript "The effect of deinstitutionalisation for adults with intellectual disabilities on cost: a systematic review. The manuscript has been improved following reviewers' comments however there are still some questions/comments which I think need addressing.

In general the methodology of this review was sound and generally well thought out. My main concern is not so much with the methodological rigour of the review but rather with the relevance of the subject and the findings. The authors have argued that this work reflects needs of the policy landscape in countries such as Ireland and other countries where deinstitutionalisation is still in early phases or isn't as advanced as in other countries. Perhaps this needs to be emphasised to provide a stronger rationale for the need of this review now.

I also think the authors need to reflect on what these findings add? What do they mean and what are the implications? Although I think it was good review my main question is, was it necessary? I invite the authors to emphasise this point.

>>>We thank the reviewer for their comments. We deal specifically on a point-by-point basis below.

Abstract

Intervention: a move in residential settings.

This is unclear as it can lead the reader to believe that the review includes studies where any move from any type of residence were considered. Although I understand that the authors did not want to provide a definition ex ante, the current statement does not suggest that it was from institutions to other settings and it is misleading.

>>>Abstract intervention has been changed to read:

Deinstitutionalisation, i.e. the move from institutional to community settings.

Introduction

Given that in the responses to reviewers the fact that the work was commissioned by the Irish Department of Health is often used as a reason as to why certain decisions were made, the authors may want to be clear about it and introduce this earlier in the paper rather than in the discussion; furthermore in response 1.1. to the reviewers it was stated that this review reflects the needs of countries which are less advanced in terms of deinstitutionalisation (e.g. Ireland), perhaps this should me mentioned in the intro as a rationale for conducting the review.

>>>As we describe in the Introduction, large numbers of people living with intellectual disabilities, are still prevented from living in a place of their own choosing, even in those countries where the process of deinstitutionalisation is quite advanced [Page 6, paragraph 2].

Additionally, we have added a paragraph with Irish context and the background to the review commission on page 6, beginning:

In Ireland, a first wave of deinstitutionalization….

Method

CASP: studies were considered high quality if all or mostly all criteria were marked as yes. This still does not allow replication what does mostly mean? 60%, 70%, 90% of criteria?

>>>Clarification added page 10:

In a list with 11 categories, six 'Yes' verdicts was therefore sufficient for inclusion.

All three reviewers noticed that cross-sectional studies were not included and I still believe that this is a limitation as rich information on costs could be extrapolated. The authors have justified their decision based on the work being commissioned by Irish policy makers for a specific purpose (i.e. deinstitutionalisation/move) (in the responses) and I believe this needs to be addressed a stronger case needs to be made to support this decision. Another reason for excluding cross-sectional studies was to limit possible confounding bias, although I agree that it could have made findings more difficult to interpreted, as mentioned by reviewer 2 this might have been a missed opportunity and I think a stronger case needs to be made about why they were not included.

>>>As well as our general Irish context as highlighted above, we have added the following specific statement about commissioners' interests to our introduction (page 6):

Their interest was specifically in analysis of those moving residence, rather than in cross-sectional analysis of different people living in different settings.

We have also addressed our approach to cross-sectional studies on page 9:

To be consistent with the desire to understand the likelihood of increases in quality of life and in cost consequences over time we excluded studies that did not evaluate economic effects following a move, and cross-sectional studies comparing community-living and institutional arrangements for two different groups at a single point in time.

And again in the discussion (page 21):

This review was originally commissioned by policymakers to inform policy and cost projections in Ireland, which is in the relatively early stages of a comprehensive deinstitutionalisation compared to neighbouring countries.

Discussion

I think that the discussion is missing one important bit which is what do we do with these findings? Ok we need more research, but what is the point of these results now? Is the fact that we need more research the main conclusion of the study? The work was commissioned by policy makers, what is the main message? How does this help them now? I think there needs to be a bit more discussion around this, as at the moment it doesn't seem like there is any major conclusion/point to be made from this review.

We have added two paragraphs to the discussion:

Page 21: This review was originally commissioned by policymakers to inform policy and cost projections in Ireland, which is in the relatively early stages of a comprehensive deinstitutionalisation compared to neighbouring countries. Unfortunately, the results have limited relevance for those commissioners.

Page 22: The primary importance of our findings is that community care is not unambiguously less expensive than institutional care over time. Consistent with earlier non-systematic assessments of this issue, the data are inconclusive. Advocates sometimes argue that deinstitutionalisation is what economists call a dominant strategy, i.e. one that both reduces costs and improves outcomes. However well-intentioned, this position is not supported by the best available evidence. This finding in no way undermines the position that all people should be supported to lead lives in places of their own choosing, and our QoL results suggest that deinstitutionalisation is associated with significant benefits. Nevertheless, these benefits will not be realised without substantial resource commitments from government and other funding bodies.