

## PEER REVIEW HISTORY

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### ARTICLE DETAILS

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| <b>TITLE (PROVISIONAL)</b> | A Realist Evaluation of Autism ServiCe Delivery (RE-ASCeD): Which diagnostic pathways work best, for whom and in what context? Findings from a rapid realist review. |
| <b>AUTHORS</b>             | Abrahamson, Vanessa; Zhang, Wenjing; Wilson, Patricia; Farr, William; Reddy, Venkat; Parr, Jeremy; Peckham, Anna; Male, Ian  |

### VERSION 1 – REVIEW

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| <b>REVIEWER</b>        | Davidovitch, M<br>Maccabi sherutei briut |
| <b>REVIEW RETURNED</b> | 23-May-2021                              |

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| <b>GENERAL COMMENTS</b> | <p>This is an important article that discusses the right way to diagnose autism. The article deals with all the stages - from referral, triage, diagnosis, summary talk and further recommendations. Suggestions were discussed for performing an effective and professional diagnosis. In an era where there is such a sharp increase in the number of children and adolescents diagnosed with autism, the system often collapses under the requirements. An effective method for the entire diagnostic process, without professional shortcuts, is the right way to deal with the many requirements.</p> <p>I was sorry to read that the article deals solely with the diagnostic system in England about all the positive things but also the difficulties of the medical system in the country. Although there is an update from articles from other countries, the main focus is on the English system, and it is not certain that this will be of interest to other countries, where the systems work differently. Despite this there are important points of worldwide interest.</p> <p>I was amazed that one of the high quality points is an evaluation process that lasts up to a whole year! (Page 5, Line 8). The main benefit of the article is in the recommendations given in Supplement 3. It is not clear to me why the main points of these recommendations, which are the important part of the article, are not included in the article itself (in a more concise way).</p> <p>In a number of places it appears that someone who diagnoses autism may not be familiar with the differential diagnosis of autism. The very idea of having a diagnosis focused only on autism greatly raises the rate of autism.</p> <p>It was further stated that the clinician does not always have to perform a clinical diagnosis on his own, and can only rely on the impression of the parents and the educational staff. In my opinion this is a clinically incorrect way. The diagnosis should always be based on clinical judgment and of course also take into account reports from the child's natural environment (page 42, Line 270).<br/>Minor revisions:</p> |
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|  | Page 6 (20%percent)<br>Page 41, line 27 instead of a psychiatrist should be SALT... |
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| <b>REVIEWER</b>        | Sanders, Benjamin<br>Oregon Health & Science University, General Pediatrics |
| <b>REVIEW RETURNED</b> | 19-Jun-2021   |

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| <b>GENERAL COMMENTS</b> | <p>Page numbers noted here refer to the numbers at the top of each page, e.g. "Page 4 of 49"</p> <p>This study conducts a rapid realist review of literature on Autism detection and intake methods in the UK with the goal of identifying promising ways to improve the process. Authors write well and do an excellent job of putting the problem in context of service needs, explaining and justifying their use of realist review, describing their meticulous methods, and clearly organizing results and discussion in an engaging way. While this manuscript is very strong overall, I suggest a number of revisions that I hope will make it even more consistent with realist review, easier to read, and useful to its readers.</p> <p>This review is organized based on the RAMESES guidelines cited by authors<sup>1</sup>.</p> <ol style="list-style-type: none"> <li>1. Title identifies document as realist review.</li> <li>2. Abstract covers background, objectives, search strategy, selection method, appraisal, analysis &amp; synthesis, main results, implications for practice.</li> <li>3. Introduction rationale is present. Comments:       <ol style="list-style-type: none"> <li>a. P. 4 lines 56 – 59 "However, robust evidence is needed..." While this sentence makes perfect sense on its own, it might have slightly better conceptual flow if it alludes to the "for whom and in what circumstances" conditions in the previous and following paragraphs. For example, authors could add a phrase to make it read "However, robust evidence is needed to identify which models in which contexts have the most significant potential..."</li> </ol> </li> <li>4. Introduction objectives and focus are stated. Comments:       <ol style="list-style-type: none"> <li>a. P. 5 lines 5-8 "High quality...calendar year." This reads a lot like a Methods section. Please clarify that in this preliminary study you will not be comparing interventions to NICE guidelines.</li> <li>b. p. 5 line 12 "across the UK to determine works best" should read "across the UK to determine what works best".</li> <li>c. p. 5 lines 16-20 I'm going to be looking for clear definitions of "pathways of autism diagnostic and support services," "service user groups." Also, are service user groups defined ahead of time, or just based on available literature (a priori definition could uncover gaps in literature)? And, what *are* the differing service needs of these different service groups?</li> <li>d. #3: ~what aspects influence how models operate~ Are the "models" the same as aforementioned "pathways"? Please clarify and/or use more consistent terminology.</li> <li>e. Also, if this question is specifically designed to address the "Context" element of Realist Review, should it not also ask what aspects of *community* or other extra-organizational factors influence how models operate?</li> </ol> </li> <li>5. Methods – changes in proposed review process (cited with a link) are denied, see top of p. 6.</li> <li>6. Methods – rationale for realist synthesis given in first paragraph of Methods. Comments:       <ol style="list-style-type: none"> <li>a. p. 5 line 45-49 "In using a rapid realist review (RRR) approach we worked backwards from the intended outcomes (efficient, high quality and family friendly service delivery) as identified in our research questions." The 3 research questions</li> </ol> </li> </ol> |
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above do not explicitly name efficiency, quality, or family friendliness [family-centeredness in the US]. There is also the earlier mention of the triad “[what] works best, for whom and in what circumstances” Please help the reader organize or reconcile these two triads and 3 research questions? For example, maybe “working backwards” means question #1 is outcomes, #2 is mechanisms, and #3 is contexts?

b. p. 5 line 50 “stakeholder” should read “stakeholders”

c. p. 5 lines 45-53. This is the first reference to a definition of Rapid Realist Review, with citation #17. However the term RRR is used from the start of the paper. While I agree with putting details and references about realist review and RRR in the Methods in general, I’m confused as to the meaning of and distinction between the terms until the sentence “RRR is explicitly designed...” For those of us less familiar with these methods, please clarify (earlier in the paper) the significance of this term RRR in distinction from realist review, if you feel it is important to use both terms.

d. Ethics review not mentioned. While this is a policy paper, the authors emphasize the importance of parent and family input in the review, and so I would suggest authors mention any institutional (ethics) review for the study, or justify its absence.

7. Methods – exploratory scoping of the literature is described and justified (p. 5 lines 33-37, p. 6 lines 13-15).

8. Methods – Searching processes are described and justified, including sources, search terms, exclusion/inclusion criteria, example search in Supplement 1, and flow diagram in Fig 1. Comments:

a. Supplement 1: Medline search strategy is great illustration of approach. Limits set in items 35 & 37 refer to years 2011 – 2020, whereas methods describe on p. 6 line 25 dates of 2011 – 2019. Maybe Medline’s limits are exclusive and your Methods description is inclusive? May help to clarify.

b. p. 7 Table 1: I can infer that the OR between the last two inclusion criteria are grouped together, but Boolean operator precedence suggests otherwise, with AND taking precedence over OR. Maybe put these last two in parentheses or somehow otherwise clarify this?

c. p. 8 Section titled “Patient and public involvement” uses the abbreviation “PPI.” Please define the abbreviation by adding it in parentheses at its first appearance in the text.

d. P. 8 line 41 “...when synthesizing & interpreting the data (Stage 5).” Stage 5 is not explained here, or referenced elsewhere as far as I can tell. Does this refer to Step 4 of Pawson’s Key steps in realist review?2

9. Methods – selection and appraisal of documents is described and justified. Paragraph on p. 7 lines 54 “Mapping the sources to test...” is particularly illustrative to me.

10. Methods – Data extraction is given cursory mention (p. 7 lines 40-47), citing references 18 and 19 as guides to readers interested in more detail.

11. Methods – analysis & synthesis processes described including iterative consultation with expert stakeholders. Constructs analyzed are described under “Search Methods” section p. 6 lines 25-30. Synthesis further described in 1st paragraph of Results (see comment 14.a. below).

12. Results – flow diagram is figure 1, including sources, reasons for exclusion. This figure is referenced and described in the Methods section instead of Results, and I agree with authors’ decision to do so because it reads well.

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|  | <p>13. Results – document characteristics are given in detail in Supplement 2.</p> <p>14. Results – main findings are described as Programme Theories 1 through 7 in Results, Table 2, Figure 2, and Supplementary Document 3. Comments:</p> <p>a. P. 8 line 53 “Our focus...” through p. 9 line 3 “...ways of addressing it.” I would suggest that this helpful information be in the Methods section instead of Results. Specifically, along with other descriptions of “constructs analyzed” as described in Wong’s RAMESES Table 1 Methods item 11.</p> <p>b. P. 9 line 17 This sentence suggests that mislabeling is more likely with early referral? Maybe there is something I don’t understand about primary care and Autism detection in the UK, but how are there “consequences of mislabeling” if a GP simply places a referral to a diagnostic evaluation resource based on risk or concern for Autism? Wouldn’t the labeling task fall upon the diagnostician to which the family is referred? From a primary care perspective, early referral for evaluation of possible cases is more important than the GP’s level of certainty about a child’s likelihood of having an ASD. This is the function, for example, of validated ASD screening tools in augmenting the primary care provider’s sensitivity to possible cases of ASD. Consequences such as unnecessary stress for parents when a normal child is referred have been described, but are not mislabeling.</p> <p>c. P. 9 line 19 “...parents concerns” should read “parents’ concerns”</p> <p>d. P. 9 line 19 “...because they were often...” pronoun is an ambiguous reference, better to read “...because parents were often...”</p> <p>e. These Programme Theories don’t describe the varied contexts that I expected to see in a realist review. See comments in next section 15.</p> <p>15. Discussion – findings summarized, although objectives &amp; research questions as stated in the introduction are not explicitly addressed. Specifically:</p> <p>a. Objective as stated at end of Introduction, “...explore evidence about Autism diagnostic pathways that have been adopted across the UK to determine what works best, for whom and in what circumstances” seems only partially addressed in discussion, mainly by describing “what works best.” There is no explicit naming of circumstances under which approaches may be best suited (as in research question #3). The detailed labeling with C-M-O in Supplement 3 is helpful, but these contexts seem to be missing from the results and discussion. I’m looking for something in the discussion like “For those communities in which [Context] exists, a [Mechanism/Approach] would be appropriate.” Furthermore, I don’t see mention of Pawson’s Step 4.c. using “‘contradictory’ evidence to generate insights about the influence of context.” Please justify your approach or modify the manuscript to include these elements.</p> <p>b. In addition, the “for whom” of this paper’s results &amp; discussion only distinguishes various stakeholder roles (provider, parent) but does not distinguish sub-populations among these, such as region, race, first language, or socioeconomic status (the “different service user groups” of intro research question #1). These would likely be important contextual factors, so authors should at least mention their absence in the literature evaluated, if that is the case.</p> <p>c. As per the introduction, I was anticipating more explicit mention of service quality, timeliness, and family-friendliness in the</p> |
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|  | <p>results and discussion. Please include this or clarify what the reader should expect.</p> <p>16. Discussion – strengths &amp; limitations described.<br/>Comments:</p> <p>a. There is no comment on the overall strength of the evidence, at least not in so many words. Please describe your approach, or explain your approach.</p> <p>17. Discussion – Comparison with existing literature is largely absent.<br/>a. While this study uses as its data literature that summarizes Autism diagnostic pathways, the discussion does not prominently discuss literature that corroborates study findings overall. An exception is the Strengths &amp; Limitations section describing similar themes from international literature. Please comment on your decision not to make literature comparison more explicit.</p> <p>18. Discussion – this paper describes the first stage in a policy evaluation project and so implications appropriately consist of topics to explore further.</p> <p>19. Discussion – funding information provided.</p> <p>References</p> <p>1. Wong G, Greenhalgh T, Westhorp G, Buckingham J, Pawson R. RAMESES publication standards: realist syntheses. <i>BMC Medicine</i>. 2013;11(1):21.</p> <p>2. Pawson R, Greenhalgh T, Harvey G, Walshe K. Realist review - a new method of systematic review designed for complex policy interventions. <i>Journal of Health Services Research &amp; Policy</i>. 2005;10(1_suppl):21-34.</p> |
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### VERSION 1 – AUTHOR RESPONSE

| Review 1, Dr Davidovitch – comments  | Response  |   |
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| <p>This is an important article that discusses the right way to diagnose autism. The article deals with all the stages - from referral, triage, diagnosis, summary talk and further recommendations. Suggestions were discussed for performing an effective and professional diagnosis. In an era where there is such a sharp increase in the number of children and adolescents diagnosed with autism, the system often collapses under the requirements. An effective method for the entire diagnostic process, without professional shortcuts, is the right way to deal with the many requirements.</p> | <p>We agree with and thank the reviewer for these comments.</p> | - |

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| <p>I was sorry to read that the article deals solely with the diagnostic system in England about all the positive things but also the difficulties of the medical system in the country. Although there is an update from articles from other countries, the main focus is on the English system, and it is not certain that this will be of interest to other countries, where the systems work differently. Despite this there are important points of worldwide interest.</p> | <p>A good point which we debated at length. However, this study was funded by NHS England who required a specific focus on England and within a tight timeframe. Therefore, we used a rapid realist review (rather than a 'full' one) and excluded international literature. When further phases of the study have been completed, we plan to publish a comparative commentary of the current state of the system in England with other countries. Meanwhile, we think that this realist analysis of the current evidence on the diagnostic pathway in England will be of interest internationally.</p> | <p>-</p>    |
| <p>I was amazed that one of the high quality points is an evaluation process that lasts up to a whole year! (Page 5, Line 8).</p>  | <p>While we have defined timely as lasting no more than one calendar year, we agree with your comment but were being realistic given current demands on services.</p>   | <p>-</p>    |
| <p>The main benefit of the article is in the recommendations given in Supplement 3. It is not clear to me why the main points of these recommendations, which are the important part of the article, are not included in the article itself (in a more concise way).</p>   | <p>An important point which we discussed at length. We explored expanding Table 2 (PTs &amp; sources) or providing an additional table with key points from Supplement 3. In realist terms the findings, or CMO statements (Supplement 3), are expressed as 'If... then' statements because they are findings from the literature. As such, they need to be treated with caution and we need to test and refine them in the next phase of the study.</p>  | <p>-</p>    |
| <p>In a number of places it appears that someone who diagnoses autism may not be familiar with the differential diagnosis of autism. The very idea of having a diagnosis focused only on autism greatly raises the rate of autism.</p>   | <p>This is description of current approaches (not only in the UK), that often focus on autism 'yes or no', and even tools such as ADI and ADOS are designed to look specifically at autism. We agree that this is not ideal and have already suggested a neurodevelopmental approach as described by NICE and in our original programme theory, which we have added the method.</p>   | <p>Pg 5</p> |

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| <p>It was further stated that the clinician does not always have to perform a clinical diagnosis on his own, and can only rely on the impression of the parents and the educational staff. In my opinion this is a clinically incorrect way. The diagnosis should always be based on clinical judgment and of course also take into account reports from the child's natural environment (page 42, Line 270).</p> | <p>We think this refers to CMO 3b, Supplement 3: Many psychiatrists and paediatricians rely on the reports and observations of other professionals to inform their decisions while some, particularly educational psychologists, prefer to carry out their own observations within educational or home settings (C). This is valuable but time consuming; one solution (O) may be for professionals to only do observational assessment (M) if there are discrepancies between school and home reports.</p> <p>This is not suggesting that a clinician can diagnose solely based on other's observations, simply that they trust the observations of colleagues as part of the decision-making process ('to inform their decisions'). UK thinking recommends multidisciplinary assessment, not a single clinician working in isolation, and that those involved should triangulate information from parental history, educational settings and observation/direct assessment. This concurs with international thinking, although several countries have accepted single practitioner assessment as necessary given demand outstripping capacity.</p> | <p>CMO 3b, Suppl. 3 (p45)</p> |
| <p>Minor revisions:</p> <ul style="list-style-type: none"> <li>- Page 6 (20%percent)</li> <li>- Page 41, line 27 instead of a psychiatrist should be SALT...</li> </ul>   | <p>Thanks, corrected to 20%.</p> <p>We think this refers to CMO 3a, Supplement 3: 'Similarly, incorporating questions previously undertaken by psychiatrists into the parent interview (M) will free up time for psychiatrists to focus on complex diagnoses (O)', the implication being that SALTs focus on the less complex diagnoses. Apologies if this is not what is meant (the page numbers do not tally).</p>   | <p>Pg6<br/>Pg41</p>           |
| <p>Reviewer 2 Dr Sanders</p>  |  |                               |

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| <p>This study conducts a rapid realist review of literature on Autism detection and intake methods in the UK with the goal of identifying promising ways to improve the process. Authors write well and do an excellent job of putting the problem in context of service needs, explaining and justifying their use of realist review, describing their meticulous methods, and clearly organizing results and discussion in an engaging way. While this manuscript is very strong overall, I suggest a number of revisions that I hope will make it even more consistent with realist review, easier to read, and useful to its readers. This review is organized based on the RAMESES guidelines cited by authors<sup>1</sup>.</p> <ol style="list-style-type: none"> <li>1. Title identifies document as realist review.</li> <li>2. Abstract covers background, objectives, search strategy, selection method, appraisal, analysis &amp; synthesis, main results, implications for practice.</li> </ol> <p><sup>1</sup>Wong G et al. RAMESES publication standards: realist syntheses. BMC Medicine. 2013;11(1):21.</p> | <p>We thank the reviewer for his comprehensive and helpful comments.</p> | <p>-</p>    |
| <ol style="list-style-type: none"> <li>3. Introduction rationale is present. Comments:             <ol style="list-style-type: none"> <li>a. P. 4 lines 56 – 59 “However, robust evidence is needed...” While this sentence makes perfect sense on its own, it might have slightly better conceptual flow if it alludes to the “for whom and in what circumstances” conditions in the previous and following paragraphs. For example, authors could add a phrase to make it read “However, robust evidence is needed to identify which models in which contexts have the most significant potential...”</li> </ol> </li> </ol>  | <p>Thanks, we have added this on the bottom of p3.</p>                   | <p>Pg 3</p> |



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| <p>4. Introduction objectives and focus are stated. Comments:</p> <p>a. P. 5 lines 5-8 “High quality...calendar year.” This reads a lot like a Methods section.</p> <p>b. Please clarify that in this preliminary study you will not be comparing interventions to NICE guidelines.</p> <p>c. p. 5 line 12 “across the UK to determine works best” should read “across the UK to determine what works best”.</p> <p>d. p. 5 lines 16-20 I’m going to be looking for clear definitions of “pathways of autism diagnostic and support services,” “service user groups.” Also, are service user groups defined ahead of time, or just based on available literature (a priori definition could uncover gaps in literature)? And, what are the differing service needs of these different service groups? Are the “models” the same as aforementioned “pathways”? Please clarify and/or use more consistent terminology.</p> <p>e. Also, if this question is specifically designed to address the “Context” element of Realist Review, should it not also ask what aspects of community or other extra-organizational factors influence how models operate?</p> | <p>a) We would like to keep the definition placed beside our focus.</p> <p>b) We have clarified with an added phrase to start this sentence: Realist reviews do not seek to compare interventions, rather they present evidence as programme theories...’</p> <p>c) Thanks, have added the missing word (‘what’).</p> <p>d) Much of the UK literature uses care pathways and models interchangeably. We have removed use of the term model and added a definition of pathway (pg3). Re service users: the first line of the introduction states: ‘children and young people (CYP) diagnosed with autism spectrum disorder (autism)’ and Supplement 1 has the full list of search terms. Re the differing service needs of different groups – a good point, but there was little differentiation in the literature (see 15-16 below); the next stage of the full study will elucidate differences in the case study sites.</p> <p>e) Interesting point, thank you. In realist terms, we are seeking to identify relevant contextual issues and, as you say, community and organisational issues are likely to be important contextual issues that enabler or trigger mechanisms. However, most studies lacked contextual detail and we did not want to make assumptions. Please see points/changes outlined in 15-16 below.</p> | <p>Pg5</p> <p>Pg5</p> <p>Pg5</p> <p>Pg3</p> <p>-</p> |
| <p>5. Methods – changes in proposed review process (cited with a link) are denied, see top of p. 6.</p>   | <p>–</p>   | <p>-</p>   |

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| <p>6. Methods – rationale for realist synthesis given in first paragraph of Methods.</p> <p>a. p. 5 line 45-49 “In using a rapid realist review (RRR) approach we worked backwards from the intended outcomes (efficient , high quality and family friendly service delivery) as identified in our research questions.” The 3 research questions above do not explicitly name efficiency, quality, or family friendliness [family-centeredness in the US]. There is also the earlier mention of the triad “[what] works best, for whom and in what circumstances” Please help the reader organize or reconcile these two triads and 3 research questions? For example, maybe “working backwards” means question #1 is outcomes, #2 is mechanisms, and #3 is contexts?</p> <p>b. p. 5 line 50 “stakeholder” should read “stakeholders”</p> <p>c. p. 5 lines 45-53. This is the first reference to a definition of Rapid Realist Review, with citation #17. However the term RRR is used from the start of the paper. While I agree with putting details and references about realist review and RRR in the Methods in general, I’m confused as to the meaning of and distinction between the terms until the sentence “RRR is explicitly designed...” For those of us less familiar with these methods, please clarify (earlier in the paper) the significance of this term RRR in distinction from realist review, if you feel it is important to use both terms.</p> <p>d. Ethics review not mentioned. While this is a policy paper, the authors emphasize the importance of parent and family input in the review, and so I would suggest authors mention any institutional (ethics) review for the study, or justify its absence.</p> | <p>a. Thank you, this picks up another area we debated. We decided against repeating the initial programme theory (in the protocol paper) and summarised with the phrase ‘efficient , high quality and family friendly’ but as this is causing confusion we have deleted the phrase and added the initial PT. We have also expanded the explanation of a realist approach, at the start of the method section, to explain that it always starts with an initial theory of what should work and what outcomes would be expected of a complex intervention.</p> <p>b. Corrected, thanks.</p> <p>c. The key difference is around the timeframe, not the rigour, and the steps are the same. We have added a sentence to this effect in the first paragraph of the method.</p> <p>d. Thanks, good point. In England, Research Ethics Committee approval is not required for PPI because the parents (and families) are acting as research partners rather than research participants: ‘they are not acting in the same way as research participants. They are acting as advisers, providing valuable knowledge and expertise based on their experience of a health condition, and/or use of NHS/social care...’*. We have added a sentence to this effect under the introduction to the Method (top of p5: Ethical approval was not required because stakeholders were acting as research advisers, not participants).</p> <p>*INVOLVE/HRA (2016, updated 2021) Public involvement in research and research ethics committee review. <a href="https://sites.google.com/nih.ac.uk/pi-standards/home">https://sites.google.com/nih.ac.uk/pi-standards/home</a></p> | <p>Pg4-5</p> <p>Pg5</p> <p>Pg4-5</p> <p>Pg5</p> |
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| <p>7. Methods – exploratory scoping of the literature is described and justified (p. 5 lines 33-37, p. 6 lines 13-15).</p>  | <p>–</p>  | <p>-</p>   |
| <p>8. Methods – Searching processes are described and justified, including sources, search terms, exclusion/inclusion criteria, example search in Supplement 1, and flow diagram in Fig 1. Comments:</p> <p>a. Supplement 1: Medline search strategy is great illustration of approach. Limits set in items 35 &amp; 37 refer to years 2011 – 2020, whereas methods describe on p. 6 line 25 dates of 2011 – 2019. Maybe Medline’s limits are exclusive and your Methods description is inclusive? May help to clarify.</p> <p>b. p. 7 Table 1: I can infer that the OR between the last two inclusion criteria are grouped together, but Boolean operator precedence suggests otherwise, with AND taking precedence over OR. Maybe put these last two in parentheses or somehow otherwise clarify this?</p> <p>c. p. 8 Section titled “Patient and public involvement” uses the abbreviation “PPI.” Please define the abbreviation by adding it in parentheses at its first appearance in the text.</p> <p>d. P. 8 line 41 “...when synthesizing &amp; interpreting the data (Stage 5).” Stage 5 is not explained here, or referenced elsewhere as far as I can tell. Does this refer to Step 4 of Pawson’s Key steps in realist review?</p> | <p>a. The reason for 2020 date in the Medline search strategy is because Medline includes a small number of in-process citations ahead of publication date (Medline in-Process &amp; Other Non-indexed Citations). Medline enables pre-publication material to be searched beyond the current date range, so it seemed prudent to use this option to identify any forthcoming relevant papers. A brief note after the search strategy (supplement 1) has been added to explain this.</p> <p>b. The comments about inclusion criteria (p.7 Table 1) relate to the use of Boolean operators and on the guidance of our information specialist, we have removed them. The criteria are now listed in the same way as the exclusion criteria.</p> <p>c. Corrected</p> <p>d. Apologies - this is Stage 5 of our protocol paper and was unnecessary so we have deleted the reference to it.</p> | <p>Suppl. 1 (p28 of 52)</p> <p>Pg7</p> <p>Pg8</p> <p>Pg8</p> |

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| <p>9. Methods – selection and appraisal of documents is described and justified. Paragraph on p. 7 lines 54 “Mapping the sources to test...” is particularly illustrative to me.</p> <p>10. Methods – Data extraction is given cursory mention (p. 7 lines 40-47), citing references 18 and 19 as guides to readers interested in more detail.</p> <p>11. Methods – analysis &amp; synthesis processes described including iterative consultation with expert stakeholders. Constructs analyzed are described under “Search Methods” section p. 6 lines 25-30. Synthesis further described in 1st paragraph of Results (see comment 14.a. below).</p> | <p>-</p> <p>We have referred the reader to our protocol paper and other references. We did not expand due to the word limit/duplicating the protocol paper.</p> <p>Under synthesis &amp; refinement we’ve added the sentence: both synthesis and refining the evidence involved substantial discussion of ‘contradictory’ evidence, or unintended outcomes (overlaps with 15a)</p> <p>-</p> | <p>-</p> <p>Pg7</p> <p>Pg7</p> |
| <p>12. Results – flow diagram is figure 1, including sources, reasons for exclusion. This figure is referenced and described in the Methods section instead of Results, and I agree with authors’ decision to do so because it reads well.</p>  | <p>-</p>  | <p>-</p>                       |
| <p>13. Results – document characteristics are given in detail in Supplement 2.</p>  | <p>-</p>  | <p>-</p>                       |

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| <p>14. Results – main findings are described as Programme Theories 1 through 7 in Results, Table 2, Figure 2, and Supplementary Doc 3.</p> <p>a. P. 8 line 53 “Our focus...” through p. 9 line 3 “...ways of addressing it.” I would suggest that this helpful information be in the Methods section instead of Results. Specifically, along with other descriptions of “constructs analyzed” as described in Wong’s RAMESES Table 1 Methods item 11.</p> <p>b. P. 9 line 17 This sentence suggests that mislabeling is more likely with early referral? Maybe there is something I don’t understand about primary care and Autism detection in the UK, but how are there “consequences of mislabeling” if a GP simply places a referral to a diagnostic evaluation resource based on risk or concern for Autism? Wouldn’t the labeling task fall upon the diagnostician to which the family is referred? From a primary care perspective, early referral for evaluation of possible cases is more important than the GP’s level of certainty about a child’s likelihood of having an ASD. This is the function, for example, of validated ASD screening tools in augmenting the primary care provider’s sensitivity to possible cases of ASD. Consequences such as unnecessary stress for parents when a normal child is referred have been described, but are not mislabeling.</p> <p>c. P. 9 line 19 “...parents concerns” should read “parents’ concerns”</p> <p>d. P. 9 line 19 “...because they were often...” pronoun is an ambiguous reference, better to read “...because parents were often...”</p> <p>e. These Programme Theories don’t describe the varied contexts that I expected to see in a realist review. See comments in next section 15.</p> | <p>a. We have moved this to the first paragraph of the method.</p> <p>b. The sentence ‘Although professionals had to balance early referral against the consequences of mislabelling...’ does not mean that GPs are giving a diagnosis but that they balanced early referral against later referral, for fear of a child being misdiagnosed. i.e. GPs balanced referring a child (potentially to a ‘yes/no’ autism service) against waiting because of concern that if the child is labelled when very young, other diagnoses could be overlooked e.g. overlap in non-verbal 2 year old between autism and language disorder. However, as this was a point of debate within the team, we have simplified in the text.</p> <p>c. Corrected</p> <p>d. Corrected</p> <p>e. Please see below</p> | <p>Pg4</p> <p>Pg8</p> <p>Pg 8</p> |
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| <p>15. Discussion – findings summarized, although objectives &amp; research questions as stated in the introduction are not explicitly addressed. Specifically:</p> <p>a. Objective as stated at end of Introduction, “...explore evidence about Autism diagnostic pathways that have been adopted across the UK to determine what works best, for whom and in what circumstances” seems only partially addressed in discussion, mainly by describing “what works best.” There is no explicit naming of circumstances under which approaches may be best suited (as in research question #3). The detailed labeling with C-M-O in Supplement 3 is helpful, but these contexts seem to be missing from the results and discussion. I’m looking for something in the discussion like “For those communities in which [Context] exists, a [Mechanism/Approach] would be appropriate.” Furthermore, I don’t see mention of Pawson’s Step 4.c. using “‘contradictory’ evidence to generate insights about the influence of context.” Please justify your approach or modify the manuscript to include these elements. In addition, the “for whom” of this paper’s results &amp; discussion only distinguishes various stakeholder roles (provider, parent) but does not distinguish sub-populations among these, such as region, race, first language, or socioeconomic status (the “different service user groups” of intro research question #1). These would likely be important contextual factors, so authors should at least mention their absence in the literature evaluated, if that is the case.</p> <p>b. As per the introduction, I was anticipating more explicit mention of service quality, timeliness, and family-friendliness in the results and discussion. Please include this or clarify what the reader should expect.</p> | <p>a. The stages of data analysis/synthesis and interpreting/refining the evidence (with stakeholders) both involved substantial discussion of ‘contradictory’ evidence, or unintended outcomes. We have added a sentence to that effect (both synthesis and refining the evidence involved substantial discussion of ‘contradictory’ evidence, or unintended outcomes) but given the word limit and full description in the protocol paper, we have not elaborated on the process.</p> <p>We have added a sentence to ‘study limitations’ around the limited description of process and context, including ‘who’, or sub-analysis by region, race, first language, socioeconomic status et al. Please see 16 below.</p> <p>b. Thank you for the comment. We have reviewed the findings and think that the outcomes are covered, as far as the literature allowed. The next stage is to test the PTs where we will be able to elucidate with more certainty what service qualities enable these aspects and in what context.</p> | <p>Pg7</p> |
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| <p>16. Discussion – strengths &amp; limitations described. Comments:</p> <p>a. There is no comment on the overall strength of the evidence, at least not in so many words. Please describe your approach, or explain your approach.</p>  | <p>We have added to study limitations that a common limit was the lack of information appertaining to care pathway processes (i.e. specific details of how things were carried out) and contextual issues (as above). As we included all study types, it is difficult to comment collectively beyond this.</p>  | <p>Pg14</p>       |
| <p>17. Discussion – Comparison with existing literature is largely absent.</p> <p>a. While this study uses as its data literature that summarizes Autism diagnostic pathways, the discussion does not prominently discuss literature that corroborates study findings overall. An exception is the Strengths &amp; Limitations section describing similar themes from international literature. Please comment on your decision not to make literature comparison more explicit.</p> | <p>We have moved the comments in Strengths &amp; Limitations section describing similar themes from international literature to the discussion and expanded.</p> <p>As mentioned above, this study was funded by NHS England who required a specific focus on England. However, when further phases of the study have been completed, we plan to publish a comparative commentary of the current state of the system in England with other countries. Meanwhile, we hope that this realist analysis of the current evidence on the diagnostic pathway in England will be of interest internationally.</p> | <p>Pg13</p>       |
| <p>18. Discussion – this paper describes the first stage in a policy evaluation project and so implications appropriately consist of topics to explore further.</p> <p>19. Discussion – funding information provided.</p>  | <p>-</p> <p>-</p>   | <p>-</p> <p>-</p> |

**VERSION 2 – REVIEW**

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| <b>REVIEWER</b>        | Davidovitch, M<br>Maccabi sherutei briut |
| <b>REVIEW RETURNED</b> | 08-Sep-2021                              |

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| <b>GENERAL COMMENTS</b> | No comments |
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| <b>REVIEWER</b>        | Sanders, Benjamin<br>Oregon Health & Science University, General Pediatrics |
| <b>REVIEW RETURNED</b> | 14-Oct-2021   |

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| <b>GENERAL COMMENTS</b> | I apologize for my delay in response to your revisions. I am very satisfied with author's thoughtful response to my questions and concerns and those of Reviewer 1. I would like to leave a few final comments for author's consideration. I don't think they will greatly change the structure or content of the paper, and so I don't need a response to them. [Editors: I checked the Yes box to "would you be willing to review a revision" simply to offer my services should |
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|  | <p>you consider it necessary.] Thank you for sharing and discussing your work with me.</p> <p>Introduction</p> <p>A.<br/>My prior comment 4d, "I'm going to be looking for clear definitions of ...'service user groups.'" And authors' response, "Re service users: the first line of the introduction states: 'children and young people (CYP) diagnosed with ASD'"</p> <p>For clarity, I suggest you add the phrase "for this service user group" to the last sentence of Intro first paragraph, to read "The NHS Long Term Plan highlighted the need for research to identify the most effective ways to improve timely access to diagnosis for this service user group whilst maintaining high-quality assessment."</p> <p>B.<br/>And further on in comment 4d, "Also, are service user groups defined ahead of time, or just based on available literature (a priori definition could uncover gaps in the literature)?" While I understand you plan to address this need in subsequent stages of the study, my comment is more about setting reader expectations as to what you will do and report upon in this paper:</p> <p>Research question 1 asks "How do various pathways...address the differing needs of different service user groups..." This leads the reader to believe that you will at least attempt to find information on different service user groups. While authors may have found "little differentiation in the literature" preventing any subgrouping of service users, I find it insufficient to mention this in the Limitations section only. This is because question 1 states that you will at least look for these different service user groups. However, I don't think you ever say something like "we looked for comparisons by demographic, etc subgroups but found none," in your results or discussion? I would suggest you either add such a statement or change question 1 to say something like "How do various pathways of ASD diagnostic and support services address the needs of service users..."</p> <p>PT1: Listening and recognition<br/>Line 4 "...balance but 'was perceived to be..." This single quote before "was" doesn't seem to have a closing single quote to match it later on.</p> <p>Discussion<br/>4th paragraph 2nd line: "health oprovision" should read "health care provision" or similar.</p> <p>Thank you! :)</p> |
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## VERSION 2 – AUTHOR RESPONSE

Reviewer 2: minor revisions, Oct 2021

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| <p>Thank you for your thoughtful comments overall, and for these minor revisions which we have addressed as follows:</p>   |   |
| <p>Introduction<br/>A.<br/>My prior comment 4d, “I’m going to be looking for clear definitions of ...’service user groups.’” And authors’ response, “Re service users: the first line of the introduction states: ‘children and young people (CYP) diagnosed with ASD’”<br/>For clarity, I suggest you add the phrase “for this service user group” to the last sentence of Intro first paragraph, to read “The NHS Long Term Plan highlighted the need for research to identify the most effective ways to improve timely access to diagnosis for this service user group whilst maintaining high-quality assessment.”</p>  | <p>Agree, we have added the phrase as suggested.</p>  |
| <p>B.<br/>And further on in comment 4d, “Also, are service user groups defined ahead of time, or just based on available literature (a priori definition could uncover gaps in the literature)?” While I understand you plan to address this need in subsequent stages of the study, my comment is more about setting reader expectations as to what you will do and report upon in this paper:<br/><br/>Research question 1 asks “How do various pathways...address the differing needs of <u>different service user groups</u>...” This leads the reader to believe that you will at least attempt to find information on different service user groups. While authors may have found “little differentiation in the literature” preventing any subgrouping of service users, I find it insufficient to mention this in the Limitations section only. This is because question 1 states that you will at least look for these different service user groups. However, I don’t think you ever say something like “we looked for comparisons by demographic, etc subgroups but found none,” in your results or discussion? I would suggest you either add such a statement or change question 1 to say something like “How do various pathways of ASD diagnostic and support services address the needs of service users...”</p> | <p>Interesting point and upon further reflection we have amended the research question so it reads more clearly and reflects your point: How do various pathways of autism diagnostic and support services address the differing needs of service users and what contexts and mechanisms affect their ability to do so?</p> |
| <p>PT1: Listening and recognition<br/>Line 4 “...balance but ‘was perceived to be...” This single quote before “was” doesn’t seem to have a closing single quote to match it later on.</p>   | <p>Thanks, well spotted! We have added the missing quote mark.</p>  |

Discussion

4th paragraph 2nd line: "health oprovision" should read "health care provision" or similar.

Thank you, corrected.