

PEER REVIEW HISTORY

BMJ Paediatrics Open publishes all reviews undertaken for accepted manuscripts. Reviewers are asked to complete a checklist review form and are provided with free text boxes to elaborate on their assessment. These free text comments are reproduced below.

ARTICLE DETAILS

TITLE (PROVISIONAL)	Cost of assessing a child for possible autistic spectrum disorder? An observational study of current practice in child development centres in the United Kingdom.
AUTHORS	Male, Ian; Galliver, Mark; Gowling, Emma; Farr, William; Gain, Aaron

VERSION 1 - REVIEW

REVIEWER	Hudson, Lee Great Ormond St Hosp and UCL Inst Child Health UK Competing interests: I have none to declare.
REVIEW RETURNED	06-Jul-2017

GENERAL COMMENTS	<p>It is on an important topic - and I think unique in terms of identifying the costs of assessing for autism assessments in the UK in CDC. I'm not aware of this having been done before or at least published, The authors have gone to a great amount of effort to collect important data. I have no concerns over the ethics.</p> <p>However, I wonder and if the editors have considered using a health economist as well to look at this paper. It is an economic paper. If they haven't I recommend so.</p> <p>I believe this would be interesting, and pertinent for this journal. However I do think it needs some revision before being published - I think it deserves publication. Most of my critique here is about content and detail. A lot of work has gone into this, and the authors should be congratulated.</p> <p>I have made individual comments on the sections as I have gone through it below, but as a summary, my main issues with the paper as it is currently written are:</p> <ol style="list-style-type: none"> 1. I don't think the authors explain why they have done this study in terms of why it is important and why it is should be published in the introduction. Why is it so important? And what question were they asking. They haven't told the story of why it is important. They should look to what they have highlighted in "what this study adds" and make the introduction more succinct from the beginning. 2. It's not clear from my reading what % of CDCs did they approach in total and is the entirety of the UK? But moreover the methods section is insufficient in describing what they did. 3. The bringing together of the importance of their data isn't clear in the discussion about the implications and how the paper is rounded off.
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	<p>4. I think some of the sections belong in other sections, moreover there should be a introduction, methods, results, discussion, conclusion format. I think this gets blurred. I've tried to point this out as I have gone along.</p> <p>Introduction.</p> <p>In paragraph one page 4, as well as building up an accurate picture of the child, are there also criteria?</p> <p>The introduction discusses the NICE guidelines in particular suggested members of the team. The authors could be clear here about whether there are mandatory members of teams or just suggestions as this is relevant to their argument about costs and the implications they delineate in the discussion about team make-up</p> <p>The authors should also explain the options for assessments in school aged children nationally - for example CAMHS vs CDC and where this crosses over. How does this vary? This is important as they have looked at differences in makeup of teams who see pre school and post school teams. The authors are writing to a general audience here - they might not be aware of why one would be referred to one or another. Many readers won't know were CDC centres are, what they are and how common they are (which is important for the denominator for the data they present)</p> <p>I think the objectives section should be a last paragraph of an introduction. The authors use a number of points that belong in the discussion not the intro here - for example "the results will help inform" - this is an implication of data point that should be in the discussion unless it was an aim, in which case it needs rephrasing. "It is important to be aware..." this belongs in the limitations section of the discussion.</p> <p>Did the authors have a hypothesis in their study? This should be at the end of the introduction.</p> <p>Design should really be methods</p> <p>The methods section isn't detailed enough. If word count is an issue, I'd suggest trimming down the introduction. There needs to be more detail here to help the reader understand. I have a number of questions that should be in here:</p> <ul style="list-style-type: none"> - How were teams contacted and who were contacted - i.e. how were they recruited. -How many times were they contacted -What efforts were made to chase those that did not reply -What were the dates of the assessments <p>The methods section needs a more detailed analysis section (as a paragraph within the methods), even though the data is mostly presented as responses. For example explaining that medians were used for non normally distributed distributions and IQR. The cost analysis method should be in this section too</p> <p>I also suggest a figure for the summary of questions asked, with the</p>
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	<p>actual survey provided as supplemental data. This is important to see exactly what and how they asked for additional critique, and also so their study can be repeated in other settings if required.</p> <p>Results</p> <p>This is appropriately succinct and I like the figures. However a number of points</p> <p>It's unclear to me where all of these centres were and how many total (I know they say 60%). This speaks to my point in the methods also.</p> <p>All of the medians presented should have an accompanying IQR or range.</p> <p>Second sentence should read "A median of 110 children WERE assessed"</p> <p>I think related disorders (or do they mean associated disorders) should be broken down.</p> <p>There are some potential missing opportunities for analysis to make this more interesting in the results. For example, in the number of professionals used by age of child in the graph, were there statistical differences (t-test or Mann-Whitney would be helpful between groups). Was there a difference in amount of professionals needed or time taken to assess and presence of "related disorders" - this is important in terms of explaining and defending costs in such complex children.</p> <p>Importantly, was there an association between time taken and the number of professionals or types of professionals involved in assessments? For example did using more expensive professionals mean shorter assessments</p> <p>Was there an association between team makeup, cost and % who were given a diagnosis? For example some teams may have accepted a lower threshold child with ASD, others might have been more certain.</p> <p>Do know who responded in each centre?</p> <p>Discussion</p> <p>I think the authors could be more focused here on the "so whats". They have found important data, and have highlighted what they add in the "what this study adds" and they provide a good critique, but I think the end of the discussion needs refining. The authors make compelling arguments about savings made from early intervention as a balance for what sounds like an expensive assessments. My suggestion would be that this is more focused and the points about long term costs to society, and savings are brought together. At the moment the long term costs are tagged onto what seems like a limitations section. The authors finish by providing some solutions. They spill into interventions and after care. I don't think this works in this paper, valid points as they are. I think they should be cut, and the focus on solutions should be how to make it cheaper, or defend why it is expensive. Data on families perceptions of assessments would be more valid here than interventions. As much as I agree with their points.</p> <p>I have an issue with describing this study as retrospective. From what I have read, the researchers asked each unit what they did for assessments in general - if this isn't the case then this needs making</p>
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	<p>clearer in the methods. In saying this I mean - haven;t the researchers asked centres how much time they would usually spend on an assessment? That isn't retrospective really. I think to be clearer about this, the methods section needs to be clearer - as discussed above.</p> <p>I don't like the word roughly. Just state the range or the mean. Approximately is better perhaps.</p> <p>Some comment should be made on the fact that only 50% received a diagnosis. How did this vary between centres?</p> <p>In the discussion, the authors provide a very detailed limitation and self-critique. But I think they can add in more positive strengths so that this paper is seen to be relevant. 60% response rate in a survey for example is a good result!</p>
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REVIEWER	<p>Crocombe, Juli Director of Clinical Services and Research Caudwell Children UK Competing interests: None</p>
REVIEW RETURNED	18-Aug-2017

GENERAL COMMENTS	<p>A very relevant and much needed piece of evidence to inform commissioning of ASD diagnostic services.</p> <p>Can you say that most centres in the UK adopt a 2-stage process when only 12 out of 20 centres completed the survey? Is it not most centres that responded?</p> <p>Is it numbers of individuals from each discipline or number of hours from each discipline in Fig one?</p> <p>More graphs / figures to show the breakdown and spread of costs across disciplines and centres would be helpful to inform our understanding.</p>
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REVIEWER	<p>Absoud, Michael Evelina London Children's Hospital, Guy's and St Thomas' NHS Foundation Trust, King's Health Partners Academic Health Science Centre, London, UK. Competing interests: None</p>
REVIEW RETURNED	28-Aug-2017

GENERAL COMMENTS	<ul style="list-style-type: none"> - This is a valuable and timely cross sectional retrospective multicentre clinician survey study, ascertaining clinician cost for an assessment of autism I have some minor comments: - The introduction would benefit from introducing ASD as a definition (as per DSM5 and ICD11 beta), instead of describing 'Aspergers syndrome' which was part of the previous DSM4 definition. - The study does not reference other costs which would contribute to the assessment including room costs; assessment material costs including forms; report time and administration costs; secretarial costs; feedback follow up costs. The manuscript would benefit from highlighting these costs, and perhaps providing an estimate.
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	<p>- Although the manuscript introduction rightly stresses that 'this team also needs to be able to recognise possible alternative explanations of a child's social communication difficulties, such as language disorder, and identify co-morbidities, including attention deficit hyperactivity disorder (ADHD)', there is no description on how many children would have had such an assessment. The DSM5 ASD criteria stress that adaptive functioning, presence or absence of specifiers (such as presence of intellectual disability, genetic disorder) should be described. Investigations related to children being assessed for ASD (if they have a comorbid neurologic symptoms/disorder) and their associated costs were not described. These limitations should be considered as they likely contribute to the cost of assessment.</p> <p>- The study would benefit from attempting to describe how much commissioners paid for assessment in each locality, as per payment by results.</p> <p>- The study does not refer to the fact that some children may underdoing more than one neurodevelopmental assessment before final diagnostic classification. Assuming this data was not collected as the study focussed on initial assessment, the manuscript would benefit from highlighting this.</p>
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VERSION 1 – AUTHOR RESPONSE

Comments		Page(s)	Paragraph (s)	Line(s)	Author Response
Reviewer 1					
1.	However, I wonder and if the editors have considered using a health economist as well to look at this paper. It is an economic paper. If they haven't I recommend so.	Title page	1	29-30	Although not an author we did take advice from a health economist (Prof heather Gage) who has joined us formally for a follow on study to this one). She is now acknowledged in acknowledgements.
2.	1. I don't think the authors explain why they have done this study in terms of why it is important and why it is should be published in the introduction. Why is it so important? And what question were they asking. They haven't told the story of why it is important. They should look to what they have	2	1	47-51 Also 103-110 and through	Altered paragraphs in opening introduction (in red in tracked

	highlighted in “what this study adds” and make the introduction more succinct from the beginning.			hout introdu ction	version)
3.	2. It's not clear from my reading what % of CDCs did they approach in total and is the entirety of the UK? But moreover the methods section is insufficient in describing what they did.	3	1	117-121	Paragraph one method section insertion in red
4.	3. The bringing together of the importance of their data isn't clear in the discussion about the implications and how the paper is rounded off.	6,7	1-5	194-197 199-203 208-220 223-225 238-245 251-256	Multiple changes and additions
5.	4. I think some of the sections belong in other sections, moreover there should be a introduction, methods, results, discussion, conclusion format. I think this gets blurred. I've tried to point this out as I have gone along.				Added throughout paper as suggested
6.	Introduction In paragraph one page 4, as well as building up an accurate picture of the child, are there also criteria?	2	1	37-43	Inserted more detail
7.	The introduction discusses the NICE guidelines in particular suggested members of the team. The authors could be clear here about whether there are mandatory members of teams or just suggestions as this is relevant to their argument about costs and the implications they delineate in the discussion about team make-up	2	2	56-62	Clarification as advised
8.	The authors should also explain the options for assessments in school aged children	2,3	3,1	64-78	Clarification

	nationally - for example CAMHS vs CDC and where this crosses over. How does this vary? This is important as they have looked at differences in makeup of teams who see pre school and post school teams. The authors are writing to a general audience here - they might not be aware of why one would be referred to one or another. Many readers won't know where CDC centres are, what they are and how common they are (which is important for the denominator for the data they present)				as advised
9.	I think the objectives section should be a last paragraph of an introduction. The authors use a number of points that belong in the discussion not the intro here - for example "the results will help inform" - this is an implication of data point that should be in the discussion unless it was an aim, in which case it needs rephrasing. "It is important to be aware..." this belongs in the limitations section of the discussion.	3,4	4,1	99-108	All moved as advised
10.	Did the authors have a hypothesis in their study? This should be at the end of the introduction.	4	1	108	Detail added
11.	Design should really be methods	4	N/A	116	Altered as per request
12.	The methods section isn't detailed enough. If word count is an issue, I'd suggest trimming down the introduction. There needs to be more detail here to help the reader understand. I have a number of questions that should be in here:	4	2,3,4	118-121 126-129 132-135 137-140	More detail added
13.	- How were teams contacted and who were contacted - i.e. how were they recruited. -How many times were they contacted -What efforts were made to chase those that did not reply -What were the dates of the assessments	4	3	126-129	See comment 12 above
14.	The methods section needs a more detailed analysis section (as a paragraph within the methods), even though the data is mostly	5	1	137-140	Detail added

	presented as responses. For example explaining that medians were used for non-normally distributed distributions and IQR. The cost analysis method should be in this section too				
15.	I also suggest a figure for the summary of questions asked, with the actual survey provided as supplemental data. This is important to see exactly what and how they asked for additional critique, and also so their study can be repeated in other settings if required.	5	N/A	142	Added
	Results This is appropriately succinct and I like the figures. However a number of points				
16.	It's unclear to me where all of these centres were and how many total (I know they say 60%). This speaks to my point in the methods also.	5	N/A	156	Figure 1 added with map showing site distribution
17.	All of the medians presented should have an accompanying IQR or range.				Inserted throughout paper as suggested
18.	Second sentence should read "A median of 110 children WERE assessed"	5	2	150	Changed as suggested
19.	I think related disorders (or do they mean associated disorders) should be broken down	5	2	154	Additional clarity added
20.	There are some potential missing opportunities for analysis to make this more interesting in the results. For example, in the number of professionals used by age of child in the graph, were there statistical differences (t-test or Mann-Whitney would be helpful between groups). Was there a difference in amount of professionals needed or time taken to assess and presence of "related disorders" - this is important in terms of explaining and defending costs in such complex children.	6	1	168-176	More detail added e.g. correlational analysis
21.	Importantly, was there an association between time taken and the number of professionals or types of professionals involved in assessments? For example did using more expensive professionals mean	6	1	175-176	Analysis added

	shorter assessments				
22.	Was there an association between team makeup, cost and % who were given a diagnosis? For example some teams may have accepted a lower threshold child with ASD, others might have been more certain.	6	1	169-173	Analysis added
23.	Do you know who responded in each centre?	5	3	163-165	Detail added
24.	<p>Discussion</p> <p>I think the authors could be more focused here on the “so whats?”. They have found important data, and have highlighted what they add in the “what this study adds” and they provide a good critique, but I think the end of the discussion needs refining. The authors make compelling arguments about savings made from early intervention as a balance for what sounds like an expensive assessments. My suggestion would be that this is more focused and the points about long term costs to society, and savings are brought together. At the moment the long term costs are tagged onto what seems like a limitations section. The authors finish by providing some solutions. They spill into interventions and after care. I don’t think this works in this paper, valid points as they are. I think they should be cut, and the focus on solutions should be how to make it cheaper, or defend why it is expensive. Data on families perceptions of assessments would be more valid here than interventions. As much as I agree with their points.</p>	7	1	208-220	More broad discussion points have been cut back – financial implications elaborated upon e.g. lines 208-220. Order of paragraphs changed in discussion to emphasise impact and importance of findings
25.	I have an issue with describing this study as retrospective. From what I have read, the researchers asked each unit what they did for assessments in general - if this isn’t the case then this needs making clearer in the methods. In saying this I mean - haven’t the researchers asked centres how much time they would usually spend on an assessment? That isn’t retrospective really. I think to be clearer about this, the methods section needs to be clearer - as discussed above.				Changed to “observational” study throughout paper and in title. See also points 12-15

26.	I don't like the word roughly. Just state the range or the mean. Approximately is better perhaps.	7	1	215	Amended
27.	Some comment should be made on the fact that only 50% received a diagnosis. How did this vary between centres?	6	3	194-197	Comment added on amount of diagnosis occurring
28.	In the discussion, the authors provide a very detailed limitation and self-critique. But I think they can add in more positive strengths so that this paper is seen to be relevant. 60% response rate in a survey for example is a good result!	8	1	238-245	Detail added on the implications of the study
Reviewer:					
29.	Can you say that most centres in the UK adopt a 2-stage process when only 12 out of 20 centres completed the survey? Is it not most centres that responded?				Implication removed – tone of results moderated
30.	Is it numbers of individuals from each discipline or number of hours from each discipline in Fig one?	16-19	N/A	N/A	Amended to be clear – number of individuals in figure 1 clarified. Additional figures 4,5,6 added
31.	More graphs / figures to show the breakdown and spread of costs across disciplines and centres would be helpful to inform our understanding.				Added as above point 30
Reviewer: 3					
32.	- The introduction would benefit from introducing ASD as a definition (as per DSM5 and ICD11 beta), instead of describing 'Aspergers syndrome' which was part of the previous DSM4 definition.	2	1	37-43	Detail added as suggested
33.	- The study does not reference other costs which would contribute to the assessment including room costs; assessment material costs including forms; report time and administration costs; secretarial costs; feedback follow up costs. The manuscript	7 7	1 2	209-220 223-225	Detail added as suggested

	would benefit from highlighting these costs, and perhaps providing an estimate.				
34.	- Although the manuscript introduction rightly stresses that 'this team also needs to be able to recognise possible alternative explanations of a child's social communication difficulties, such as language disorder, and identify co-morbidities, including attention deficit hyperactivity disorder (ADHD)', there is no description on how many children would have had such an assessment. The DSM5 ASD criteria stress that adaptive functioning, presence or absence of specifiers (such as presence of intellectual disability, genetic disorder) should be described. Investigations related to children being assessed for ASD (if they have a comorbid neurologic symptoms/disorder) and their associated costs were not described. These limitations should be considered as they likely contribute to the cost of assessment.	4	1	107	Additional clarity added
35.	The study would benefit from attempting to describe how much commissioners paid for assessment in each locality, as per payment by results.	8	1	240-245 93-100	Additional information added. The introduction also already explained that commissioning was by block contract, indeed part of reason for the paper was to inform potential tariff or PBR
36.	The study does not refer to the fact that some children may undergo more than one neurodevelopmental assessment before final diagnostic classification. Assuming this data was not collected as the study focussed on initial assessment, the manuscript would benefit from highlighting this.	4	1	107	Added sentence for clarity