

## 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

<b>TITLE (PROVISIONAL)</b>	Patient safety vulnerabilities for children with intellectual disability in hospital: A systematic review and narrative synthesis
<b>AUTHORS</b>	Mimmo, Laurel; Harrison, Reema; Hinchcliff, Reece

### VERSION 1 - REVIEW

<b>REVIEWER</b>	Thomas, Megan Blackpool Teaching Hospitals NHS Foundation Trust UK Competing interests: None
<b>REVIEW RETURNED</b>	08-Oct-2017

<b>GENERAL COMMENTS</b>	This is a very important topic and a well written and clear review of the identified literature. My only concern is whether all the literature was identified by the search terms shown. While considered not a helpful term in many parts of the world, mental retardation is still a term commonly used in America. Developmental delay is another commonly used term particularly in young children, and learning disability or learning difficulty are other terms. In addition it may have been helpful to look at the articles where the term complex medical conditions was used as this term does often encompass children with ID.
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<b>REVIEWER</b>	Ahmad, Alia The Children's Hospital & the Institute of Child Health Lahore Pakistan Competing interests: Paediatrics, Paediatric Haematology/Oncology. Infection Control and prevention, Psycho-social Issues, Palliative care
<b>REVIEW RETURNED</b>	20-Oct-2017

<b>GENERAL COMMENTS</b>	1- Extent and severity of ID Intellectual Disability and its impact on specific safety risks and outcome could have been elaborated more. 2- Majority of the studies included Down Syndrome Children, the characteristics of other children included not described and whether how many having physical disabilities along with ID. 3- The procedures, in addition to cardiac surgeries in Down Syndrome children, requiring hospitalizations not mentioned .
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### VERSION 1 – AUTHOR RESPONSE

#### Response to Reviewer 1:

1. Whether all the literature was identified by the search terms shown. While considered not a helpful

term in many parts of the world, mental retardation is still a term commonly used in America. Developmental delay is another commonly used term particularly in young children, and learning disability or learning difficulty are other terms.

Response:

The search strategy for Medline and CINAHL included the MeSH terms, Intellectual Disability and Developmental Disabilities, see Appendix 1, with the explode function, and the scope notes for these terms include Mental Retardation and Developmental Delay respectively. For completeness search strategies used for the other databases – Embase, PsycINFO, Scopus and Web of Science – have been updated by including ‘Mental Retardation’ and ‘Developmental Delay’ as additional search terms. The date range for the search has also been amended on page 7 of the revised manuscript.

This broadening of the database searches resulted in two additional studies from Embase and two studies from PsycINFO being retained for full text review. Medline and CINAHL searches were also repeated to capture any studies published since the initial search, and three additional studies were retained for full text review. Ultimately, these seven studies were excluded as they did not meet the specified inclusion criteria. Figure 1: the study selection flowchart, attached as a supplementary file, has been amended to reflect this information.

2. It may have been helpful to look at the articles where the term complex medical conditions was used as this term does often encompass children with ID.

Response:

Studies that referred to children with complex medical conditions were scrutinised to determine if the study participants had disorders that include a diagnosis of ID. Studies that did not explicitly involve children with ID were excluded from our review. The author team debated the merits of broadening the inclusion criteria in the way suggested by the Reviewer, but ultimately decided to retain the original distinction. This is because complex medical condition or disability may encompass many diseases and disorders of childhood that do not cause inherent cognitive impairment. Where this term was used, the study participants were reviewed to determine if children with ID were the focus. As there is little research in this area that is specific to children with ID, it was vital that only directly relevant evidence was included in the narrative synthesis to inform the final conclusions. We have identified this as a potential limitation in the original manuscript and expanded on this point on page 17 of the revised manuscript.

3. Key terms seem to be missing. Can they add some detail as to their thoughts on this and either that they did scoping searches and found they damaged or impacted the search or rerun and look for any missing papers and add if needed?

Response:

The search strategy was determined collaboratively by the author team and a senior university librarian who possesses vast experience in developing systematic search strategies. A decision was reached for the search process to be undertaken in two stages; scoping and then the full search. The explode function was employed for searches in Medline and CINAHL to ensure studies that used less common terms were captured. As noted above in response to an earlier comment by the Reviewer, the search strategies for Embase, PsycINFO, Scopus and Web of Science were repeated after receiving feedback on the first submission of our manuscript. The terms Mental Retardation and Developmental Delay were added, but with no additional records included in the review as a result. As the Reviewer may have been alluding to, the terms used for children with ID vary internationally, even among predominantly English-speaking countries. Our appreciation of this challenge motivated us to use a variety of keywords, determined in consultation with subject experts and senior university librarian to maximise study capture, including: iatrogenic disease, medical errors, patient safety,

patient harm; hospitalization; adolescent, child, infant; intellectual disabilities, cognitive disorders, learning disorders, developmental disability. Examples of the Boolean search terms that were applied include: iatrogenic disease OR medical error OR patient safety OR patient harm; intellectual disability OR cognitive disorders OR learning disorders OR developmental disability; hospital\*; infant OR child\* OR adolescen\* OR teenage\*.

There are many and varied key terms used to describe different aspects of the topic examined in our manuscript. Having initially implemented a comprehensive search strategy, which was then further strengthened by including the Reviewer's suggested additional terms, we are confident that the final search terms and strategy outlined in the revised manuscript were sufficiently rigorous to enabled capture of relevant studies.

### **Response to Reviewer 2:**

We thank Reviewer 1 for taking the time to review our manuscript and the helpful comments. We have answered each of the points below.

1. Extent and severity of ID Intellectual Disability and its impact on specific safety risks and outcome could have been elaborated more.

Response:

Two of the studies included in our review used methods to identify the severity of ID (Graham et al, 2008) or cognitive impairment (Malviya et al, 2001) in participants, but this was for the purpose of selection and allocation of participants into the control or ID cohort of the study arms. Neither of these studies stratified their results according to severity of ID. We agree that this would be an important direction for future study and Seliner, Latal and Spirig (2016) highlight the absence of research in this area to characterise treatment outcomes by severity of ID. For these reasons we have made an amendment to the manuscript, page 16, with citation, to highlight this important point.

2. Majority of the studies included Down Syndrome Children, the characteristics of other children included not described and whether how many having physical disabilities along with ID.

Response:

The inclusion criteria concentrated on studies which stipulated the study population of interest was children with ID, or explicitly focused on a disease cohort whose condition included ID, such as Down Syndrome. As nine of the 16 included studies focused on children with Down Syndrome, we noted on page 18 that this group has been the focus of the majority of studies in this area to date and that little is still known about the experiences of those with other IDs.

All of the studies involving children with Down Syndrome were cohort studies reporting surgical outcomes compared to children without Down Syndrome. One reason for the inclusion of Down Syndrome populations in such studies is that cardiac disease (Desai et al, 2014; Fudge et al, 2010) and Hirschspung's Disease (Morabito et al, 2006) are more common in people with Down Syndrome than those without. Children with Down Syndrome are therefore a well-defined cohort for comparative study (Valkenburg et al, 2012). The presence of physical disability was not a focus of the studies included in this review, and presents different and variable challenges distinct from those for children with ID. We agree with the Reviewer that the cumulative impact of physical and intellectual disability should be a focus for future inquiry in this field.

3. The procedures, in addition to cardiac surgeries in Down Syndrome children, requiring hospitalizations not mentioned.

Response:

This deficiency has now been addressed by adding information regarding procedures to the last paragraph of page 9 and first paragraph of page 10 in the study characteristics section of the revised manuscript.

#### **VERSION 2 – REVIEW**

<b>REVIEWER</b>	Thomas, Megan Blackpool Teaching Hospitals NHS Foundation Trust, UK Competing interests: No competing interests
<b>REVIEW RETURNED</b>	10-Dec-2017

<b>GENERAL COMMENTS</b>	Thank you for re-visiting the search strategy and clarifying the justifications for some of the decisions made.
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