Reviewer's report

Title: SIDS Pathogenesis. A review of hypotheses: Plausibility and evidence

Version: 3 Date: 4 February 2011

Reviewer: Peter S Blair

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My main field of interest is SIDS epidemiology rather than pathology or infant physiology so hope the other reviewers can cover these areas.

This is a provocative and interesting piece and I wonder whether this is actually a review or more of a vehicle to argue one plausible theory (held be the author) over the others. Certainly reading through some of the views and arguments given I think some of the groups mentioned might want a right of reply. This might fit better as part of a series of articles debating the causal mechanisms of SIDS rather than a review.

Major Compulsory Revisions

i) In the opening salvos (and throughout the document) the author refers to his group in the third person. As one of the main proponents of a major causal theory surely this undermines (rightly or wrongly) the independence a reviewer must have in weighing up all the evidence available. Perhaps declaring this conflict of interest head on and slightly rewriting this as a perspective rather than a review would reflect better what is actually being argued.

ii) An impression one gets reading the background section is that the different groups are competing for the one true cause of SIDS. I think it is important to establish (to the readers at least) that in all probability there are a myriad of possible causes. SIDS is a diagnosis of exclusion therefore pathologists only establish what it is not. The 7 salient pathological findings listed on page 3 might be associated with some SIDS deaths but are not used as a prerequisite to define SIDS. SIDS is simply defined as “not knowing why the baby died” and to suggest that “Without these salient findings, in cases where the history and death scene was not congruent with SIDS, experienced pathologists would consider alternative diagnoses.” stretches the idea that we have some sort of understanding of what a SIDS death actually is. I hope that experienced pathologists who can’t find a complete explanation of death would classify the case as a SIDS death.

iii) Linked to this, if SIDS is a collection of deaths due to disparate causes, then the initial premise that each SIDS death has to meet all (or most) of the different criteria set down by the author is a fallacy. Just taking the key epidemiological risk factors for instance; prone sleeping, bed-sharing and head covering are strongly associated with SIDS but very rarely would we find any SIDS death exposed to all 3 risk factors and in quite a number these factors would not be present at all. (Sleeping supine is now the most common position to find a SIDS
infant so does that mean we should be recommending parents to not sleep babies on their back too!

iv) Pathologists are divided on the importance of pathological findings. The ideas put forward by Byard & Krous (San Diego definition of SIDS) have not been endorsed by the wider SIDS community and this, in part, is due to taking associated pathological characteristics or recognised risk factors and assuming these are causal features of “classic” SIDS cases. Mixing up association with causation and assuming we are dealing with a single cause of death are mistaken views that are certainly not held by the epidemiologists working in the SIDS field. The author needs to delineate a bit more between what is a consensus view in SIDS research and what is currently up for debate.

v) Some of the rhetoric used is perhaps a little too provocative “remains obstinate” “illogical” “no justification” “disingenuous” “misguided” and should be toned down

vi) I would strongly challenge the concluding statement the author describes as misguided research “focused on usually a single phenomenon (such as the risk factor – prone sleep position) and which has been undertaken at considerable expense to the public purse.” SIDS research is not just about finding causal mechanisms, indeed, more importantly about reducing such tragedies and there is now a considerable wealth of evidence that shows the change in sleeping position has saved tens of thousand of lives across the world (in the UK SIDS deaths have fallen by 80% since the "Back to Sleep campaign" in 1991, the fall in rates from 1500 deaths a year to 300 suggesting more than 20,000 children are now playing on our streets than would have been if the rates had remained the same!) We may not understand why the prone position poses such a risk but funded epidemiological, observational research has paid back the public purse manifold.

vii) A Table 3 was mentioned but I did not seem to have access to it

Quality of written English: Acceptable

Statistical review: No, the manuscript does not need to be seen by a statistician.

Declaration of competing interests:

I declare that I have no competing interests