Proposal for an International Classification of SUDI: A response to Blair, Byard and Fleming

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In 2009, Blair, Byard and Fleming, drawing on discussions between SIDS (sudden infant death syndrome) researchers and practitioners, proposed an international classification scheme for SUDI (sudden unexpected death in infancy) (Blair et al., 2009). As indicated in their proposal, the term SUDI incorporates a range of causes of death, both apparent and hidden. Whilst the proportion of SUDI for which a cause is found has risen, at least 50% remain unexplained in all published studies. Blair, Byard and Fleming point out that, although an internationally agreed definition of SIDS exists, use of the term is inconsistent, and both practitioners and researchers often resort to alternative labels for those deaths that remain unexplained. Furthermore, approaches to the investigation of SUDI vary both between and within countries. These inconsistencies complicate research in the field, and potentially hamper our search for understanding of these deaths. Nevertheless they are a reality which will not go away.

Blair et al’s proposal is timely. It is essential, as overall numbers of unexpected infant deaths fall, for there to be collaboration within and between countries in continued investigation of the underlying causes and contributory factors for these deaths. Most researchers and practitioners recognise that SIDS is unlikely to be a single entity for which a simple explanation or cure can be found, rather it is likely that those SUDI that currently remain unexplained include deaths from undiagnosed infections, other underlying medical conditions, including various genetic, cardiac or metabolic conditions, and accidents or non-accidental deaths which we are currently unable to identify. The proposed classification allows for this, through the combination of the “Avon” grading through Ia to III, the addition of category 0 to account for incomplete investigations, and the subgroups of explained deaths within the categories of SUDI.

To be useful, any classification system needs to be simple, yet comprehensive, and to be acceptable to both practitioners and researchers. It needs to accommodate differences in practice between individuals and on a wider basis. The proposed system of Blair et al goes a long way to encompassing these elements. Experience in the UK however has shown that the Avon classification can be difficult for practitioners to understand and work with, and a simplified version has now been widely adopted by child death overview panels and teams responding to unexplained child deaths (Sidebotham et al., 2008). This dispenses with the (a) and (b) subclassification and simply uses a 0 – III classification (0 – information not available; I – no factors or non-contributory factors identified; II – factors possibly contributing to death; III – fully explained). Individuals using this system may choose to subdivide categories I and II, but overall it leads to greater uniformity and is more practical for front-line practitioners.

There have previous attempts to classify SIDS and SUDI, including the San Diego, GeSID and ESPID classifications (Krous et al., 2004, Kerbl et al., 2003, Findeisen et al., 2004). These classifications acknowledge the variations in presentation of SUDI and the differences in approaches to investigation. Although they do not directly conform to the current proposal, there is sufficient overlap to propose a merger of these systems (Table). It will prove difficult to achieve full consensus on any international system of classification, even amongst researchers, let alone practitioners; nevertheless, this proposal should prompt further debate in the right direction.

Thus far, the lack of a generally accepted classification system makes it difficult to reliably compare SIDS or SUDI figures worldwide, and these figures usually contain incomplete information concerning autopsy rate and the incidence of factors potentially contributing to death in the recorded so-called SIDS cases. A starting point for all epidemiological research should be a description of all infant deaths; the next stage would be to define what is included within any categorisation of SUDI; and finally a clear description of those finally labelled as SIDS, conforming to the internationally agreed definitions (Krous et al., 2004, Willinger et al., 1991), but making clear what parameters have been used within any particular research project. This in itself is not necessarily straightforward, as different areas will include different groups of deaths within their definition of SUDI. Nevertheless, by including all SUDI, within the framework suggested, it is possible to separate out those for which a cause is ultimately found from those that remain unexplained, regardless of the term used to describe this group. In order to fully understand these deaths, we need a system which will allow us to go beyond a simple diagnosis of cause of death, to incorporate the various shades of grey inherent in our approach to investigation and the findings thereof. The proposed system by Blair et al goes some way to achieving this, and we believe, with some modification, could be adopted by researchers and practitioners alike.

The proposed classification system could potentially be a starting point for further research and clarification. There remain difficulties in relation to which deaths should be included: for example, in the UK and New Zealand, all sudden unexpected deaths in infancy are referred to the coroner, but this is not necessarily the case elsewhere, and in many places, sudden deaths for which a cause is immediately apparent, including some accidental deaths, or those with an apparent medical cause
may not be included. It is likely that within the spectrum of SUDI, different patterns may emerge at different ages, and some sort of developmental trajectory could be developed to further explore how different infant and environmental factors may be associated with different developmental stages, and different categories of both explained and unexplained sudden infant deaths – an extension of the widely accepted triple risk model of SIDS (Guntheroth and Spiers, 2002). Further work would be needed also to try to achieve some consensus over which associated factors would be included in category II. There may, for example, be reasonable consensus now to include

<table>
<thead>
<tr>
<th>Simplified Blair et al Proposed classification</th>
<th>Categories of SUDI</th>
<th>San Diego Classification</th>
<th>GeSID classification</th>
<th>ESPID classification</th>
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<tbody>
<tr>
<td>0</td>
<td>Unexplained, incomplete investigation. This category may be applied in conjunction with other categories where any part of the investigation is incomplete. This category expands on other classifications by incorporating cases with missing elements of the investigation, not just non-autopsied cases.</td>
<td>Unclassified Sudden Infant Death: a) deaths that do not meet the criteria for category I or II SIDS, but for which alternative diagnoses of natural or unnatural conditions are equivocal (these may be better classified as category II SUDI, with or without a category 0 to indicate inadequate investigation) b) cases for which autopsies were not performed</td>
<td>The GeSID classification was based on pathology findings; being a research based classification, only cases in which a full autopsy had been performed were included.</td>
<td>Non-autopsied SUDI Includes any SUDI for which an autopsy has not been performed; does not account for other missing aspects of an investigation</td>
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<td>I</td>
<td>Unexplained; no contributory factors identified. This classification does not take account of the positive features of a classic SIDS case suggested in the San Diego classification</td>
<td>Category I SIDS a) classic features of SIDS present and completely documented b) classic features of SIDS present but incompletely documented (there is some overlap between this classification and SUDI category 0) There may be some overlap between category I SIDS and SUDI category II, depending on which factors are deemed to be potentially contributory.</td>
<td>Category 1: Without pathological findings from autopsy and additional investigations Category 2: With minor pathological findings in autopsy and investigations</td>
<td>Classical SUDI</td>
</tr>
<tr>
<td>II</td>
<td>Unexplained; possible contributory factors identified</td>
<td>Category II SIDS Meet category I criteria, except for some features which raise possibilities of an alternative explanation (e.g. outside the typical age range; close family history of SUDI; possibility of mechanical asphyxia; abnormal autopsy findings insufficient to determine a cause of death)</td>
<td>Category 3: Severe findings, but not sufficient to fully explain the death</td>
<td>Borderline SUDI</td>
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<td>III</td>
<td>Explained: Rapid infection Rapid onset of acute medical condition Unrecognised pre-existing medical condition Accidental death Non-accidental death</td>
<td>Category 4: SUDI with clear cause of death found at autopsy. Excludes known unnatural deaths</td>
<td>Explained SUDI</td>
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prone sleep position, cigarette smoking in pregnancy and necropsy findings of minor infection; whilst other factors, such as the role of co-sleeping may prove more controversial. Finally, there is a continued need to move towards greater standardisation of the processes by which a diagnosis and categorisation of SUDI are achieved. The criteria required for a diagnosis of SIDS have previously been agreed within the international research community (Willinger et al., 1991) and include a complete autopsy and review of the circumstances of death and the clinical history; what specific elements should be incorporated within that, and how the review should be carried out, for example through a multi-disciplinary case review, remain open to interpretation, and will always be dependent on local resources and working practices, but it is hoped that over time greater consistency can be achieved, leading in turn to more robust descriptions of the causes and circumstances of sudden unexpected deaths in infancy.

REFERENCES

FIRST ANNOUNCEMENT:
SORIA MORIA MEETING 2011
MAY 19-21

THEMES:
● Brain stem research and SIDS
● Genetic risk factors for SIDS
● Domestic violence and effect on early brain development
● Impact of death scene investigation in sudden deaths in infants and small children

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