Evidence-Based Medicine and Shaken Baby Syndrome

Part I: Literature Review, 1966–1998

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In recent years, there has been a clear move toward basing medical practice and opinions on the best available medical and scientific evidence. This process has been termed *evidence-based medicine* (EBM) and involves a review of the quality of evidence that is available in various diseases and fields of inquiry within medicine.

This is the first of 2 articles that attempts to formally rank the available medical scientific evidence by internationally accepted methods, to determine the degree of confidence that can be held on various claims about the condition termed *shaken baby syndrome* (SBS). Areas with good scientific evidence are identified, and shortcomings in the research and publications on the subject are addressed.

Approximately half of all indexed medical publications on the subjects of SBS and shaken-impact syndrome were published before 1999 and half since that time. Given that 1998/1999 is regarded as the turning point in acceptance of the tenets and practice of EBM, it seemed reasonable to assess the quality of evidence before 1999 and compare it with the quality of evidence on the same subject matter since that time. At the conclusion of Part II, the 2 periods are compared to determine the extent to which EBM has affected the field of SBS in terms of quality of available evidence.

The aim of this review is to be neutral on the subject of SBS. Neutrality is difficult to define in this field, in part because of the polarization of opinions on the highly emotional subject of infant injury and death and in part because of clear data deficiencies arising from difficulties in performing experiments. It is clearly unethical to intentionally shake infants to induce trauma, and there is an obvious problem with studies and reports that rely on either indirect or disputed evidence of the occurrence, severity, or type of trauma.

Many studies lacking these critical data make the obvious logical error of selecting cases by the presence of the very clinical findings and test results they seek to validate as diagnostic. Not surprisingly, such studies tend to find their own case selection criteria pathognomonic of SBS.

Neutrality in this review simply means that there is no selective quotation of the available literature, and literature is not chosen to support any particular view. The assessment is of the methods and quality of the actual research, and until this assessment is complete, the content, findings, and recommendations are irrelevant. At the end of the ranking, those studies that achieve the highest QER scores are reviewed for their content, findings, and recommendations. Their outcomes are collated, and the entire published data set is then reviewed as a whole to determine the summarized recommendations, noting areas of agreement, conflict, or controversy. From this, the problems with the published evidence are noted, and data gaps are identified. Recommendations can then be made according to the summarized data.

In assessment of the quality of the available scientific evidence, the author has taken an approach recently defined worldwide as an appropriate scale for review of quality of evidence. This approach has been described recently in context of setting Australian clinical guidelines.

Genuine hypothesis testing requires use of appropriate research methodologies, including collection of relevant control data, and suitable statistical analysis. The interpretation of individual study findings may be constrained by factors such as whether the cohort examined was adequately representative of the patient population in general. Replication across studies and in independent research centers is a key factor in the reliability of evidence.

Compelling evidence comes from consistent findings in 2 or more well-constructed, controlled trials or population-based epidemiologic studies (i.e., level I or level II evidence). By contrast, clinical practice guidelines with level IV evidence represent consensus statements of the expert panel according to clinical experience and limited scientific data. Although these statements may influence current practice,

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they are likely to be modified in response to further research findings. Data from a single case series without control subjects provide little more than a stimulus for subsequent hypothesis testing.

Quality of Evidence Ratings

I: Consistent evidence obtained from more than 2 independent, randomized, and controlled studies or from 2 independent, population-based epidemiologic studies. Studies included here are characterized by sufficient statistical power, rigorous methodologies, and inclusion of representative patient samples. Meta-analysis of smaller, well-characterized studies may support key findings.

II: Consistent evidence from 2 randomized controlled studies from independent centers, a single multicenter randomized controlled study, or a population-based epidemiologic study. Data included here have sufficient statistical power, rigorous methodologies, and the inclusion of representative patient samples.

III-1: Consistent evidence obtained from 2 or more well-designed and controlled studies performed by a single research group.

III-2: Consistent evidence obtained from more than 1 study but in which such studies have methodologic constraints, such as limited statistical power, or the inclusion of patient samples that may be nonrepresentative.

III-3: Evidence obtained from a single case study or a selected cohort study.

III-4: Conflicting evidence obtained from 2 or more well-designed and controlled studies.

IV: Consensus opinions of authorities according to clinical experience or descriptive reports.

SBS: Literature Review (1966–1998)

Overview and Methods

The entire Biomednet Medline database (http://www.biomednet.com/db/medline) was searched by using the search term *shaken baby syndrome* and Internet Explorer in late November 1998. Other articles identified that had not yet been indexed on MEDLINE but had been published were also included.

The entire set of retrieved articles was reviewed, and those in which SBS was only peripherally mentioned or in which SBS was unrelated to the original article were omitted. Letters and brief correspondence were also discarded, unless they added new information or data on SBS. Articles in non-English journals that lacked an English abstract were also generally excluded from assessment.

These exclusions reduced the initial list of 71 articles to 54, which were reviewed, categorized, and ranked according to the QER above. To these was added the important study by Jayawant et al. from *BMJ* of December 5, 1998. The editorial was omitted because it added nothing to the original article.

It was impossible to review the full original article in many cases, although all of the major articles were reviewed in full. The remainder was assessed for categorization by using the authors' abstracts.

Each article was assigned to 1 of 4 categories: (1) randomized controlled trial; (2) case series with or without controls (with date, series size recorded); (3) single case reports; and (4) other, including review articles, opinion pieces, and articles on social implications.

Results of Quality of Evidence Ratings

Fifty-four articles or abstracts were reviewed. One was a randomized controlled trial.[34] This trial was not relevant to the general topic of SBS because it assessed a diagnostic technique (electroretinograph) that proved unsuccessful in diagnosis. Twenty-six were case series.[1,2,7,9,11,13–15,18,19,26,28–30,33,36,39,42,45–50,52,53] Twenty-five were retrospective studies, and 1 was prospective.

In total, 307 SBS cases were claimed to have been assessed among the 23 articles in which numbers of SBS patients were provided, with a mean study size of 13 cases and a median of 7 cases per series.

Selection criteria for SBS cases were unstated in 12 articles, based on presumption or suspicion in 10, and confirmed in 4 by confession or conviction. Two studies had appropriate control groups, 3 had inappropriate control groups, and 21 were case series without control groups. Twelve studies were case reports:[3–5,8,10,17,22,24,31,32,51,55]

Retinal pathology in suspected SBS, 5 cases

Blunt head injury at autopsy, 1

Subdural hemorrhage (SDH) and retinal hemorrhage (RH) as a result of fall and chest compression, 1 case

Shaking causing traumatic aneurism, 1 case

Arteriovenous malformation, not SBS, as cause, 1 case

Intentional asphyxia and shaking of 15-week-old baby, 1 case

Magnetic resonance imaging value in diagnosis, 1 case

Raised intracranial pressure as cause of RH, 1 case

Fifteen were "other" articles[6,12,16,20,21,23,25,27,37,38,40,41,43,44,54]

Historical reviews of SBS,10 articles

Opinion papers without original material, 3 articles

Social issues and SBS, 1 article

Review of imaging in SBS (computed tomography versus magnetic resonance imaging), 1 article

RESULTS

The randomized controlled trial was unrelated in any aspect of interest in this review and addressed a method of assessment of the retinae that proved to be unsuccessful.

Of the case series, the flaws noted above are relevant. All but 1 was retrospective, and all but 5 had no control population to compare cases with. Three of those that did

include controls chose insufficient or inappropriate controls (head impact trauma, without healthy controls or other illness unrelated to head injury). This shortcoming would normally have excluded these studies from the literature review (because they do not fulfill criteria for inclusion). Given the difficulties inherent in assessment of SBS and of identifying appropriate control groups, however, these articles have been included as QER III-2.

In studies with confirmed (i.e., admitted or observed) trauma and SBS, there were few common findings, apart from the presence of SDH accompanied by RH in 80% of examined cases. Some articles attempt to measure other risk factor, and the study by Jayawant et al.[42] is noteworthy in this respect.

Finally, the "other" articles do little but summarize opinions and summarize past data. Such articles do not add to the quality of understanding of the condition, nor are they necessarily accurate in what has become a rather emotionally charged area of research and polarized opinion.

In this article, the quality of evidence, rather than the predominance of findings, is being assessed. The issue of the evidence for SBS appears analogous to an inverted pyramid, with a small database (most of it poor-quality original research, retrospective in nature, and without appropriate control groups) spreading to a broad body of somewhat divergent opinions. One may need reminding that repeated opinions based on poor-quality data cannot improve the quality of evidence.

Data Gaps Identified

There exist major data gaps in the medical literature about SBS. There is a very obvious lack of clear definition of cases. For valid studies, some method of determining cases of actual proven shaking must be found, and appropriate control groups (trauma without shaking, other illness, healthy controls) must be defined and assessed blindly. This gold standard has yet to be achieved in even a single study in the field of SBS. There is a lack of useful and specific laboratory or other markers proven to identify SBS. There is poor definition and quantification of the social and family risk factors to provide guidance on likelihood of abuse for a given set of circumstances. Last, there is a strong need for a checklist or other diagnostic or management tool to assess cases and to quantify index of suspicion of shaking.

CONCLUSIONS

There was no evidence on the subject of SBS that exceeded QER III-2 by the end of 1998, which means that there was inadequate scientific evidence to come to a firm conclusion on most aspects of causation, diagnosis, treatment, or any other matters pertaining to SBS.

The majority of evidence achieved only a level of QER IV, opinions that shed no new light upon SBS and did not add to knowledge about SBS. Many of the authors repeated the logical flaw that if RH and SDH are nearly always seen in SBS, the presence of RH and SDH "prove" that a baby was shaken intentionally. Many other studies assumed that the presence of RH and SDH was sufficient to make the diagnosis of SBS in terms of case selection.

The remainder of articles are QER III-3, and as noted above, the inclusion of case series without controls would normally not occur. Thus, the data available in the medical literature by the end of 1998 were inadequate to support *any* standard case definitions, or *any* standards for diagnostic assessment.

Before 1999, there existed serious data gaps, flaws of logic, inconsistency of case definition, and a serious lack of tests capable of discriminating NAI cases from natural injuries. By 1999, there was an urgent need for properly controlled, prospective trials into SBS, using a variety of controls. Without published and replicated studies of that type, the commonly held opinion that the finding of SDH and RH in an infant was strong evidence of SBS was unsustainable, at least from the medical literature.

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