Response to David Strauss and Dr. Richard Katz, November 14, 2005

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In February 2004, David Strauss and his colleagues posted, on their website (www.lifeexpectancy.com), a commentary about my previously published study concerning survival rates of children with severe neurologic disabilities[5]. This commentary appeared exactly 6 years after the publication of my study, and exactly 6 years after receiving congratulatory phone calls from David Strauss. I wrote a response to this commentary[9], and it was posted for two months on the website until it was removed without explanation.

Recently, the text of the commentary has been modified. Thus, an updated response is necessary.

In Paragraph 1, David Strauss contends that my published data contradicts “a large scientific literature from around the world.” This statement is totally incorrect. My data confirms the findings of many investigators that clinical factors decrease survival of disabled individuals (including immobility, tube feeding, epilepsy, and mental retardation; see references 5-8). Direct comparisons of my results to published reports outside of the US are not possible to do (although David Strauss suggests that this can be done). The reason is that the definition of “severe” cerebral palsy in international studies is so broad that all six groups of disability fit into the definition of “severe.” The most disabled group 1 has a high mortality rate, whereas the less disabled groups 3 through 6 have low mortality rates. Thus, without knowing the exact number of individuals from groups 1 through 6 in a “severe” category, direct comparisons cannot be made. Furthermore, the more recently published studies by David Strauss and his colleagues, when compared to my published results, give very similar, and at times nearly identical survival rates for individuals who are less disabled[6-8]. These more recent Strauss results are almost an independent confirmation of the validity of my previously published results. The divergence between my results and those of David Strauss are in the survival rates of the more disabled groups.

It should be noted that the articles published by David Strauss and his colleagues repeatedly report that the presence of epilepsy or any other serious medical illnesses have no impact on survival rates[1, 2, 15]. These claims are totally contrary to the published literature, to clinical experience, and to common sense. A disabled child, who is having 6 or 7 pneumonias per year, cannot have the same prognosis as a disabled child who is having no pneumonias. David Strauss and his colleagues themselves have, in fact, contradicted “a large scientific literature from around the world.”

Paragraph 2 gives a hypothetical example in which after two years of life, the survival rate would be 5% (using my methods; graphic representation can be found in ref. 9). David Strauss claims that my method would result in a survival rate of 9.5%. David Strauss misrepresents my statistical methods, builds a straw man, and then is able to deftly knock it over.

Paragraph 3 is a discussion about using “age-specific mortality rates”. This suggested approach is a perfectly valid one, but not the method that I used. In all of the graphs in my study are labelled “% Surviving”, namely survival rates. This is a different approach, in no way less valuable—just different.

Further, in Paragraph 3, David Strauss again misrepresents my data. All of the patients in my study had their neurologic disability since birth. There were no cases of acquired encephalopathy at a later age. Thus, anyone of a given age was “exposed” to the risk of dying on every previous day of their lives. No one was immune from the possibility of death.

In Paragraph 4, the logic is unfathomable. In 4.1, David Strauss notes that in group 4 of my study, deaths took place at the ages of 4 and 20. That is correct. In Paragraph 4.2 he writes that I had “assumed that all 11 people were alive both at age 4 and at age 20.” How is it possible that I could assume that all were alive at the points in time when my results clearly indicate that two had died (at the ages of 4 and 20)? This comment is incomprehensible.

This unfathomable logic continues in Paragraph 4.3. In a further analysis of group 4 results, David Strauss writes, “6 of the 11 persons were initially age 15 or over.” Then, two sentences later, he concludes: “Thus, at most 5 of the 11 were alive at age 4.” How is it possible for 6 individuals to be over the age of 15, and not have been alive at the age of 4? Perhaps they were all born on their 15$^{th}$ birthday? Again, the comments are incomprehensible.

The problem with the entire line of commentary is that David Strauss did not evaluate my data as presented and published, rather looked at it using totally different (not more valid, just different) statistical approaches. This led to Straussian results that at best are straw men, and at worst absurd.

In analyzing the California data base, I am sure that the statistical methodologies used are fine. However, the major problems lie in the data base itself. These problems have been thoroughly reviewed[6-8]. The four most important issues are:

(1) The California data base starts with data from the beginning of 1980. Access to medical care for severely disabled children only became a reality in the mid 1980’s thanks to US Federal government intervention. Prior to 1985 the death rates of disabled children were very high[12]. That is why we started our data collecting in 1985 when medical care had been standardized. Including data from 1980-85 in the data base, and using it, produces artificially high death rates.

(2) Despite US Federal regulations, many physicians still practice with an attitude that severely disabled children should receive minimal care, and should be allowed to die quickly (I do not subscribe to this abhorrent philosophy). This attitude, which cannot be weeded out of the California data base, significantly decreases survival rates.

(3) The California data base does not account for the issues of access to acute medical care, or to on-going optimal medical care. David Strauss has reported that home and group home settings, where access to acute medical care is difficult, result in much higher mortality rates[13]. In my study, acute medical care was available 24-7, and the quality of medical care provided was the best that we could provide (optimal). Lack of access to acute medical care, and lack of on-going optimal medical care, both decrease survival rates.

(4) All of the data is collected by individuals with no medical training or background, thus making data base extremely deficient in medical information. Dr. Grossman, one of the authors of the early California studies, wrote that the data was "notoriously unreliable[3]". If one starts with "notoriously unreliable" information, how can one expect to get reliable results? Several examples can be given. It is entirely unknown whether tube-feeding is by gastrostomy tube or by nasogastric tube (which is associated with much higher death rates[5]). Whether a person has epilepsy, or not, was totally unknown in 36% of cases. In three different studies, no basic neurologic diagnosis existed in 39.7%, 78% and 82% of cases. "Notoriously unreliable" appears to have been an accurate evaluation. (For a more detailed analysis see references 6-8.)

As far as the validity of my published data is concerned, it should be noted that I have more clinical experience in providing medical care, 24-7, to severely disabled children and young adults, than probably any physician in North America. The data that we published was collected and finalized in 1996, almost ten years ago. Since that time we have made considerable progress in improving the medical care delivered to the most disabled individuals in our society[10]. My clinical experience since the data was collected, corroborates the accuracy of our original publication. My suspicion is that if the study were repeated today, given the improvements in medical care that we have implemented, the survival rate results would be even better.

In comparison, how much hands-on clinical experience does David Strauss and his colleagues have? They are statisticians, and have no clinical experience. Lacking clinical experience, they cannot vouch for the accuracy of any of the medical information in their computers.

The Strauss commentaries, and their revisions, provide erroneous information about my work, and suggest a total inability to do medical research. I have 126 medical research publications to my credit. Most of the publications required statistical analysis—which was done adequately, and underwent peer review. I have received 10 major medical research grants, for a total of $2.8 million in funding. Research grants are not provided if statistical methodology is flawed.

Was it not possible for David Strauss and his colleagues to get it right the first time? The revision was more flawed than the first attempt.

What are we to expect next? More straw men?

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I recently became aware of a review article about survival rates of disabled children written by Dr. Richard Katz[4]. In this article he made several comments about my previous publication[5]. But first, it should be noted that in the extensive bibliography that Dr. Katz cites (a total of 61 references) there is not one survival rates study that he himself has undertaken and published. Indeed, a Medline search revealed that he has never published any original study on this topic.

The only substantial criticism that Dr. Katz made is that in one place the number 367 appears, and in another it is 371. This is clearly a typographical error, and this should have been obvious to Dr. Katz.

With typographical errors out of the way, other issues can be addressed:

(1) Good medical outcomes in children with tracheostomy have also been noted in a report by David Strauss[15]—thus my result is in keeping with the published literature.

(2) Dr. Katz wrote that the Southern Medical Journal is "outside the literary arena of most persons interested in this topic area." This journal is indexed by Medline shortly after publication. Indeed, within two weeks of publication, David Strauss telephoned me to congratulate me. Thus, this journal clearly is inside the mainstream. In the category of general medical/surgical journals, the Southern Medical Journal had the third largest circulation in the US.

(3) Dr. Katz claims that the statistical methods are not clear, yet in the methods section is found: "For statistical analysis chi-square testing was used. Data are presented as the mean $\pm$ 1 standard deviation of the mean." The statistical methods were clearly stated.

(4) Dr. Katz criticized the comparison of my data to that of Eyman[2]. In fact, that was exactly the stated purpose of my study, and the purpose was accomplished. Subsequently, I did compare my results to those found in the general medical literature[6-8].

(5) Dr. Katz notes that my study had small numbers—that is correct. However, when one does a detailed analysis and a careful comparison to the report by Eyman[2], the stunning result is that my numbers are actually 2.2 times larger than Eyman's[11].

(6) Dr. Katz comments that all of the children, who I cared for, resided in skilled nursing facilities—that is correct. Under these circumstances we could provide optimal acute and optimal chronic medical care. Is this undesirable? Isn't providing optimal medical care something that all physicians and nurses strive to do? Shouldn't optimal medical care outcomes be considered a "gold standard" according to which home-based outcomes could be compared?

Thus, in summary, the only substantive flaw that Dr. Katz found in my study is one typographical error. For that correction, I am grateful.

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