Life expectancy in cerebral palsy: an update

David Strauss* PhD FASA;
Jordan Brooks BS,
Life Expectancy Project, San Francisco, CA, USA;
Lewis Rosenbloom FRCPCH,
Royal Liverpool Children's NHS Trust, Liverpool, UK;
Robert Shavelle PhD FAACPDM, Life Expectancy Project,
San Francisco, CA, USA.

*Correspondence to the first author at 1439 – 17th Avenue, San Francisco, CA 94122, USA. E-mail: Strauss@LifeExpectancy.org

DOI: 10.1111/j.1469-8749.2008.03000.x

This paper clarifies and updates some issues of life expectancy in cerebral palsy. These are: (1) the definition of life expectancy and how it is calculated; (2) the secular trends that have occurred since the data for the 1998 paper were collected; (3) revised estimates reflecting improvements of some of the analytical methods and statistics provided in that paper; (4) comparison of life expectancies among countries; (5) issues regarding quality of care; and (6) consideration of prospective life expectations in addition to current life expectancy.

This paper discusses and updates the 1998 paper by Strauss and Shavelle on the life expectancy of adults with cerebral palsy (CP)¹ and the 2007 study by Strauss et al.²

Life expectancy and calculation

The standard scientific definition of life expectancy is the average survival time of the members of a population. The life expectancy of a given individual is thus the average survival time in a large group (real or hypothetical) of similar individuals. It should, therefore, be clear that life expectancy does not refer to the actual time that an individual will live (i.e. the individual's actual survival time), which could be much longer or shorter than the life expectancy.

Life expectancy can be viewed as a convenient summary of the death rates at all ages, and as such can serve as a measure of health. For example, the steady increase in life expectancies that have been observed for several centuries in developed countries reflects improvements in medicine and public health. Further, in the case of those who are injured and will need compensation for their future care, their life expectancy may be an essential input in the assessment of economic damages. (We have used the qualifier 'may be' because a court may award a lifetime stream of 'periodic payments' rather than a lump sum. In that case an estimate of life expectancy would not necessarily be required by the court, though it would be required by the company that provides the payments.)

Life expectancy in CP is of interest for all these reasons. As a result there has been a continuing flow of publications on the longevity of individuals in this group. Some of the more recent are Hemming et al.,³ Hutton and Pharoah,^{4,5} Blair et al.,⁶ and Strauss et al.^{2,7}

There are different general approaches to the estimation of life expectancy. The so-called *top-down* approach starts with the general population figure and then subtracts years for various adverse factors. Although this may be reasonable in cases of near-normal life expectancy, it is unreasonable in the case of medical conditions that dramatically alter the

pattern of morbidity and mortality. As an example of the former, it may be appropriate to make a literature-based reduction from normal life expectancy for smokers. At the other extreme, if a patient is in the permanent vegetative state it would be absurd to start with normal life expectancy and make deductions for the immobility, swallowing problems, etc. This is because normal life expectancy largely reflects the pattern of morbidity and mortality characteristic of old age, most importantly heart disease and cancer. By contrast, major causes of death in the vegetative state are respiratory infections, septicemia, and general organ failure.

An estimate based directly on data on a population of individuals with similar characteristics, without the use of the general population life expectancy as a starting point, has sometimes been termed *bottom-up*. This is in fact the normal scientific method of following a population of individuals with a given condition and recording their actual survival experience. In a given case its application may be criticized on various grounds – for example, the individuals in the study may differ in important respects from the individual of interest – but the *principle* of the method is well established.

It is clear there are some factors whose effect on life expectancy can be numerically quantified, based on population data, and some that cannot. In the case of CP, it is possible to take account of the key factor *severity of disabilities*, as measured by gross motor function, need for gastrostomy feeding, etc. There are also some population data available on the effect of epilepsy, hydrocephalus with shunt dependence, and severe underweight. There are, however, potentially significant factors for which population data are generally lacking. Examples are unsafe swallowing, frequency and severity of respiratory infections, and severe scoliosis.

Given this, our approach to estimating life expectancy in CP is to start by taking account of the factors whose effect can be quantified (using the literature and/or a suitable database). We then consider the other factors that have not been taken into account. If, on balance, the pattern of these other factors is considered more favorable than average among individuals in this group, it is reasonable to argue for an upward adjustment to the life expectancy. The converse applies if the balance is considered less favorable than average. At this stage the views of an experienced clinician may be helpful.

Secular trends: past and future

There seems little doubt that survival in CP is better now than, say, 50 years ago. If, however, attention is restricted to the past 20 to 30 years, the picture is less clear. In their Western Australia study, Blair et al.⁶ reported that they found no secular trend over their study period. Hutton et al.,⁸ in their 1994 study, report that they found no difference in survival between cohorts stratified by time period of birth. Hutton et al.⁴ found no birth cohort effect for children of normal birthweight but some effect for low birthweight. Hemming et al.³ reported 'no evidence of any secular changes in survival'. Further, although only indirectly relevant to CP, recent work on individuals with spinal cord injury⁹ indicates that the improvements in survival are largely confined to the critical first few years after injury.

More recent research in CP, using refined statistical methods and a very large database, suggests that, in fact, there is a

mixed picture.² There has been a marked improvement in survival for two groups with the most severe disabilities, namely children who are largely immobile and fed by others, and adults who are dependent on gastrostomy feeding. In these groups mortality fell by some 3.4% annually. For the other groups, there *may* have been a modest trend, though the effect is only marginally significant statistically. If so, the improvement is roughly comparable to that in the general population, i.e. an annual decline of approximately 1%.

Revised estimates of life expectancy of adults with cerebral palsy

Table III of the 1998 study by Strauss and Shavelle¹ on this topic provided estimates of life expectancy for males and females of ages 15, 30, and 45 with various patterns of abilities and disabilities. These estimates have been widely cited. It now seems appropriate to update the estimates, for a number of reasons:

- (1) In the 10 years since the research was carried out, a great deal of new data have become available, and the methodology for life expectancy estimation of individuals with chronic disabilities has been refined. ^{10,11} For all but the highest-functioning groups, we have now used the method of Proportional Life Expectancy. ^{10,11} In brief, this method assumes that the *proportion* of normal life expectancy for a given medical condition is the same at every age. As a mathematical consequence, this determines the excess death rates for the condition at all ages: they prove to be inversely proportional to the remaining life expectancy at each age. For a complete description of the methodology used for estimates in this article, see Appendix I.
- (2) The life expectancies were previously derived using the assumption of 'linearly declining log-relative-risk'. ¹² We emphasize that this led to overestimates of life expectancy, which are corrected in the present work. Again, for details see Appendix I.
- (3) There has been a trend towards improved survival of adults who are fed by gastrostomy,² and this needs to be taken into account. Further, there has been a modest increase in the life expectancy of the general population, and this too is reflected in the revised estimates.
- (4) In our 2007 study on improved survival in CP,² we commented that in the relevant groups (here, adults who are fed by gastrostomy), the effect of the secular trend was to increase previous estimates by approximately 5 years. This is correct. However, the increase is largely offset by the reductions that result from the technical refinements noted above in (1). Further, because of these refinements the new estimates for individuals who are *not* fed by gastrostomy are in some cases *lower* than before.
- (5) Table III of our 1998 study¹ gave different results for males and females. Subsequent research showed that as would be expected such differences apply mainly to individuals who do not have the most severe disabilities. At the extreme end of the severity spectrum, an individual's sex has no appreciable effect on life expectancy.
- (6) The highest-functioning group considered in the study, namely 'rolls and/or sits, and self-feeds', sometimes appears to have been misinterpreted. Because it was the highest-functioning category it included individuals with a wide range of disabilities. At the lower end of the group were individuals who could roll over and finger feed but, for

example, could not stand unaided and had no useful form of mobility. At the higher end were those who could self-feed with utensils and walk without support. Evidently the life expectancies in these two groups are quite different, and the estimates in Table III were a composite that were too high for the first group and too low for the second. We have now stratified the category according to whether the individual can walk unaided, which proves to be a useful predictive factor.

- (7) In the database we worked with there is a six-level feeding scale, ranging from fed by others (level 1) to finger feeding (levels 2 and 3) up to 'uses fork and spoon without spillage' (level 6). We used the phrase 'at least some self-feeding (SF)' in the earlier article simply to contrast levels 2 to 6 with level 1. To qualify for this the person must take a *significant* proportion of his nutrition by SF. We perhaps did not make it sufficiently clear that *children who take only 10%*, say, of their nutrition by SF would not be considered to have 'at least some SF' for our purposes.
- (8) By contrast with Table III, Figure 2 of the 1998 study¹ gave survival curves for the various cohorts, rather than life expectancies as in Table III. The method for constructing these curves is the standard Kaplan–Meier estimator. The survival curves for the groups that are tube fed (TF) do not capture the secular trend that was subsequently identified, but apart from this, Figure 2 unlike Table III does not require major revision.
- (9) There seems to have been some misunderstanding of a comment in our article¹ (p373): We extracted an additional, bigbest-functioning, subgroup with full sitting, ambulatory, and feeding skills... On average, their life expectancy was reduced by only 5 years compared with the general population. First, highest-functioning on these items includes the requirements that the person could: (a) walk well alone for at least 20 feet and balance well; and (b) use eating utensils consistently without spillage. Second, 'on average' referred to the ages 15, 30, and 45, where normal life expectancy ranged from 65 down to 31 years. A typical reduction from normal was 10%. Third, in some cases young individuals with reasonable ambulation become wheelchair-bound in later life, and the above 10% reduction is generally no longer appropriate for them (see point 10 on the following page). Fourth, as is discussed below this calculation does not refer to projected life expectancy figures.

Table I shows our revised estimates of life expectancy, and supersedes those given in Table III of the earlier study. To facilitate interpretation of this table we make the following comments:

- (1) As in the previous article, feeding skills are stratified into three categories: TF, fed orally by others (FBO), and has some SF ability.
- (2) Regarding motor function, the first three categories (cannot lift head in prone, ¹ lifts head or chest in prone, and rolls and/or sits independently) are as before except that these groups are now restricted to individuals who cannot walk unaided.
- (3) Persons who walk unaided, now the highest-functioning group in the table, are shown in the second column from the right.
- (4) The ages are the same as before, except that estimates are now provided for high-functioning individuals of age 60.
- (5) The missing values in the table correspond to unusual combinations of skills (e.g. TF but ambulatory or unable to lift head but able to self-feed), which must be treated on an individual basis.
- (6) The general population figures in the table are typically 1 to 2 years higher than previously, reflecting improvements in the US general population over the intervening 10 years.
- (7) The estimates of life expectancy for the *individuals* who are TF are higher than those from the earlier study. For example, Figure 2 of that study showed a median survival time of 8 years for the lowest functioning group those who were TF and did not lift their heads in prone. This can be shown to correspond to a life expectancy of 11 years. The estimate in the above table, 13 years, is about 20% higher.
- (8) Some of the estimates in Table I are lower than those reported in the previous study, especially for females. Examples are the estimates for females with the most severe disabilities. The 1998 study reported life expectancies of an additional 21 years for 15-year-old females who could not lift their heads and were FBO and an additional 21 years for those who could lift their head and/or chest using arm support and were TF. The new estimates of 16 additional years in each case are some 5 years lower.
- (9) The reader may also notice that generally in the new table all life expectancy estimates in the rolls/sits category are somewhat lower than previous estimates. The primary

Table I: Life expectancy (additional years) by age and cohort

Sex/age (y)	Cannot lift bead			Lifts bead or chest			Rolls/sits, cannot walk			Walks unaided	General
	TF	FBO	SF	TF	FBO	SF	TF	FBO	SF		population
Female											
15	13	16	-	16	21	_	21	35	49	55	65.8
30	14	20	_	15	26	_	16	34	39	43	51.2
45	12	14	_	13	16	_	14	22	27	31	37.0
60	_	_	_	_	_	_	_	_	16	20	23.8
Male											
15	13	16	_	16	20	_	19	32	45	51	60.6
30	14	19	_	15	24	_	16	31	35	39	46.5
45	12	14	_	13	15	_	14	20	23	27	32.8
60	_	_	_	_	_	_	_	_	13	16	20.4

TF, tube fed; FBO, fed by others, without feeding tube; SF, self-feeds.

reason for these differences is that the rolls/sits category presented here is further restricted to individuals who are unable to walk unaided. As noted previously, this distinction based on ambulation is a useful discriminator among higher-functioning individuals with CP. With the exception of the 'walks unaided' group, all of the groups are restricted to individuals who do not walk unaided.

(10) To estimate the life expectancy of young individuals who can walk without support, one needs to make an assumption about whether this skill will persist through to late adulthood in the present analysis. The assumption is made that the individuals will continue to be able to walk unaided until at least age 60. We recognize that this will overestimate the walking skills in later life of many individuals, but we chose to display these figures as an upper bound on life expectancy for a person who currently has at least some independent walking. The true life expectancy of such individuals must lie between the estimates based on (a) the above upper bound, and (b) the group that rolls/sits and self feeds but does not walk without support. This range is typically some 4 years wide according to Table I, which may be narrow enough to be helpful in practice.

(11) We remind the reader that the categories in Table I are rather broad, and the figures given are averages. Further distinctions within each group could, of course, have been made and the resulting life expectancies may vary by several years from the reported averages. Within the ambulatory group, for example, life expectancies vary considerably according to the extent to which the person can self-feed and otherwise participate in activities of daily living. In particular, if a person's pattern of disabilities is at an extreme end of the range for a given group, their life expectancy may differ substantially from the group average. Further adjustments may be indicated for epilepsy, cognitive function, low weight, and other medical conditions.

Comparisons between countries

There has been speculation about whether life expectancies of individuals with CP are similar in different states or countries. Of course, for a meaningful comparison we must compare children with similar patterns of abilities and disabilities. This is only possible if databases are available with enough information that like children can be compared with like. There are several such databases in the UK, associated with the work of Professor Jane Hutton and her colleagues, a Western Australia database that has been analyzed by Dr Eve Blair and ourselves, and our own California database. To our knowledge, these are currently the only large databases that permit appropriate comparisons.

It appears that when proper comparisons are made, experiences from these databases are similar. A previous comparison of the California and Australian databases shows good agreement in the survival of groups of comparable children. 13 The study also noted comparability of estimates from the California and the UK. Figure 1 compares survival of children with severe disabilities in the California database and the 2005 study of Hemming et al.³

The survival curve from the Hemming et al.³ study is for children with 'four severe disabilities'. These are with respect to ambulation, cognitive function, hand use, and visual function. These criteria can be reproduced reasonably closely in the California database. As evaluations in the UK database are made at the approximate age of 4 years, we worked with a California cohort of children at that age. For the Hemming et al.3 study we plotted the probabilities of survival conditional on being alive at age 4.

Each age-specific conditional survival probability is calculated as the ratio of the probability of surviving to that age to the probability of surviving to age 4. The actual survival probabilities for the Hemming et al. cohort were measured directly from the published curves.3

Thus, both curves start at age 4 with 100% of the cohort alive. As may be seen, survival over the 16-year interval was very similar in the two countries.

Quality of care and life expectancy

The effect of quality of care on life expectancy is frequently discussed, and it is sometimes asserted, without any supporting evidence, that quality of care is a critically important factor. This issue is more complex and less clear than is often assumed and the following brief discussion summarizes some of the reasons for this. Some of these points have been made at greater length in a recent review article on life expectancy after traumatic brain injury by Shavelle et al. 14 Quality

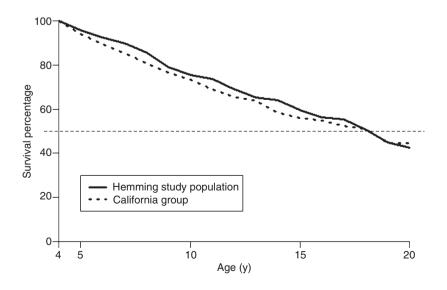


Figure 1: Comparison of UK and California survival rates of children with severe ambulatory, manual, cognitive, and visual disabilities.

of care is a rather vague term that may refer to any or all of the following:

- (1) The expertize of the caregivers, ranging from highly qualified professionals to relatively unskilled (and low paid) staff. A complicating factor is that caregivers are often family members, who generally do not have formal qualifications but in some cases become highly skilled carers.
 - (2) The accessibility of physicians and emergency services.
- (3) The quantity of care and equipment provided, which is often a reflection of the funds available.

Next, the effect of quality of care on life expectancy surely depends on what is being compared. If, for example, it is good care versus grossly inferior care, the difference in life expectancy will doubtless be large. That comparison, however, is generally not of interest. The most relevant comparison is between (1) the reasonable and necessary standard care available in most developed societies, and (2) the care expected given that the patient has a carefully prepared and well-funded life care plan.

It might be argued that the care embodied in (2) represents the best case in practice, as one cannot forecast exactly what care the patient will receive, or will choose to receive, in the coming decades.

It is sometimes asserted that quality of care is the most important determinant of life expectancy. If the comparison is between (1) and (2) above, this assertion is clearly wrong: the most important determinant is undoubtedly the severity of the disabilities. For example, literature from many countries documents that young patients in the permanent vegetative state have mortality rates up to 500 times larger than in the general population. If quality of care is as important a determinant of mortality risk, then death rates under 'standard' care would have to be 500 times higher than they would be under (2). This is surely inconceivable.

Further, some states or countries provide services to individuals with disabilities as an entitlement. For example, California provides annual person-centered individual program plans plus provision of all indicated care. In such cases it is not clear what is the difference, if any, between (1) and (2) above

It may also be noted that researchers at The Dartmouth Atlas Project have found that care beyond what is reasonable and necessary does not significantly prolong the life span of individuals in the general population, ¹⁵ and in some instances it appears to have an adverse effect. ¹⁶ The project's website also provides an annotated list of 387 additional references supporting these conclusions. ¹⁷

Projected life expectancies

The question of changes in mortality rates over previous decades should not be confused with what improvements may be expected in the *future*. Here again, it has been speculated that further gains in CP survival can be expected. An obvious difference, of course, is that we do not have data on the future to confirm or deny this prognosis. It is, therefore, necessary to make some assumptions.

In this context we note the current practice regarding 'normal' life expectancy in UK and Australian courts, though not to our knowledge any other countries. This is to work with *projections of future mortality rates* at each age rather than with current rates. These projections assume a steady decline in mortality rates at all ages. The result is an increase

of some 7 to 9 years in a typical child's life expectancy, by comparison with the standard figure based on current mortality. The current UK female life expectancy at birth is 81 years, ¹⁸ for example, compared with the projected figure of 90 years. ¹⁹ We note that the choice between current and projected rates in litigation is a legal issue rather than an actuarial one: the scientific community is not unanimous on what should be assumed about future mortality rates. ^{20,21}

If projected rates are to be used as the baseline, there are several ways to adjust the life expectancy estimates to reflect this. We illustrate with an example. For a US female of age 15, normal life expectancy is 66 additional years. ²² Consider a female of age 15 whose life expectancy is estimated to be 29 additional years, which is 44% of normal. How should this be applied in the case of a similar child according to, for example, UK projected mortality rates, given that normal UK *projected* life expectancy is 74 years, 8 years higher than the US current figure?¹⁹

One extreme is to argue that the original estimate of 29 years still applies because an increased life expectancy for individuals with CP in another country has not been formally documented. The other extreme is to argue for the full increment (i.e. 8 years), and thus for a life expectancy of 37 years. Intuition suggests that the truth is somewhere between these extremes. For example, if a patient with terminal cancer has a life expectancy of one year in the US, one would not add eight years to this because the patient lived in the UK. On the other hand, if a patient with very mild CP has a near-normal life expectancy, he should be credited for nearly all of the increment.

There is no 'correct' answer because the issue is partly an empirical one. For example, if the improved life expectancy were entirely a reflection of better lifestyle factors, such as weight reduction and smoking cessation, then little of the improvement would apply to individuals with CP. On the other hand, if the gain in life expectancy was solely a reflection of improved medical care or treatment then one would expect the improvement to apply to CP.

The issue is clearly complex but we would suggest that if a simple rule is to be adopted, a *constant percentage* is not an unreasonable choice. In the above example, where life expectancy is 44% of normal, we would thus apply this percentage to the UK general population figure of 74 years. The result is 33 years, an increase of four years over the US figure. Thus, the individual receives 44% of the 8-year increment associated with UK projected rates.

Finally, we note that, as the example illustrates, the percentage-based adjustment can be used when applying the estimates from one country to another, as well as to reflect the use of projected mortality rates.

Accepted for publication 19th December 2007.

References

- Strauss D, Shavelle R. Life expectancy of adults with cerebral palsy. Dev Med Child Neurol 1998; 40: 369–75.
- Strauss D, Shavelle R, Reynolds R, Rosenbloom L, Day S. Survival in cerebral palsy in the last 20 years: signs of improvement? *Dev Med Child Neurol* 2007; 49: 86–92.
- 3. Hemming K, Hutton JL, Colver A, Platt MJ. Regional variation in survival of people with cerebral palsy in the United Kingdom. *Pediatrics* 2005; **116**: 1383–90.

- Hutton JL, Pharoah PO. Effects of cognitive, motor, and sensory disabilities on survival in cerebral palsy. *Arch Dis Child* 2002; 86: 84–9.
- Hutton JL, Pharoah PO. Life expectancy in severe cerebral palsy. Arch Dis Child 2006; 91: 254–8.
- Blair E, Watson L, Badawi N, Stanley FJ. Life expectancy among people with cerebral palsy in Western Australia. *Dev Med Child Neurol* 2001; 43: 508–15.
- Strauss D, Ojdana K, Shavelle R, Rosenbloom L. Decline in function and life expectancy of older persons with cerebral palsy. *NeuroRebabilitation* 2004; 19: 69–78.
- 8. Hutton JL, Cooke T, Pharoah PO. Life expectancy in children with cerebral palsy. *BMJ* 1994; **309**: 431–5.
- Strauss DJ, DeVivo MJ, Paculdo DR, Shavelle RM. Trends in life expectancy after spinal cord injury. Arch Phys Med Rehabil 2006; 87: 1079–85.
- Strauss DJ, Vachon PJ, Shavelle RM. Estimation of future mortality rates and life expectancy in chronic medical conditions. *J Insur Med* 2005; 37: 20–34.
- Anderson TW. Life expectancy in court: a textbook for doctors and lawyers. Vancouver, BC: Teviot Press, 2002.
- 12. Strauss D, Shavelle R. Life expectancy of persons with chronic disabilities. *J Insur Med* 1998; **30**: 96–108.
- Shavelle RM, Strauss DJ, Day SM. Comparison of survival in cerebral palsy between countries. *Dev Med Child Neurol* 2001; 43: 574. (Letter)
- Shavelle RM, Strauss DJ, Day SM, Ojdana KA. Life expectancy. In: Zasler ND, Katz DI, Zafonte RD, editors. Brain injury medicine: principles and practice. New York: Demos Medical Publishing, 2007: 247–61.
- Dartmouth Atlas of Health Care. Center for the Evaluative Clinical Sciences at Dartmouth Medical School. http:// www.dartmouthatlas.org/faq.shtm. (accessed 2 August 2007).
- Fisher ES, Welch HG. Avoiding the unintended consequences of growth in medical care: how might more be worse? *JAMA* 1999; 281: 446–53.
- 17. Dartmouth Atlas of Health Care. Center for the Evaluative Clinical Sciences at Dartmouth Medical School. http://www.dartmouthatlas.org/ismorebetter/is_more_better_1.php. (accessed 2 October 2003). [The list of 387 studies does not currently appear on the project's website, but it may be obtained from the present authors.]
- Government Actuary's Department. Interim life tables for England, 2002–2004. London: The Stationery Office, 2005.
- Government Actuary's Department. Actuarial tables with explanatory notes for use in personal injury and fatal accident cases, 6th edn. London: The Stationery Office, 2007.
- Oeppen J, Vaupel JW. Demography. Broken limits to life expectancy. Science 2002; 296: 1029–31.
- Olshansky SJ, Passaro DJ, Hershow RC, Layden J, Carnes BA, Brody J, et al. A potential decline in life expectancy in the United States in the 21st century. N Engl J Med 2005; 352: 1138–45.
- Arias E. United States Life Tables, 2003. National Vital Statistics Reports, Vol 54, No 14. Hyattsville, Maryland: National Center for Health Statistics, 2006.

List of abbreviations

FBO	Fed by others
SF	Self-feeds
TF	Tube fed

Appendix I

This Appendix provides technical details underlying the life expectancy estimates presented in Table I. The first section describes the improvements in methodology since the 1998 article of Strauss and Shavelle¹ was published. The second section provides further details on the methods used.

1. Improvements in life expectancy methodology

- (a) For individuals of age 15, mortality rates up to age 30 were computed directly from the data. The question is what mortality rates should be assumed for all subsequent ages, as these are needed in the construction of a life table and thus in the computation of a life expectancy.
- (b) The previous study¹ assumed that the excess death rate (EDR) at age 30 stayed constant up to age 40, and then employed the assumption of 'linearly declining log-relative risk'. That is, the logarithm of (the individual's death rate at age x divided by the corresponding death rate in the general population) declines in a straight line fashion after age 40, reaching parity (equal death rates in the two groups) at age 90.
- (c) Subsequent research showed that both of these assumptions led to a moderate degree of overestimation of life expectancy. Specifically, death rates in fact increase appreciably between ages 30 and 40, both for the general population and for persons with cerebral palsy, and the parity age of 90 proves to be too low, 100 being a more appropriate figure.
- (d) On average, the overestimation introduced by the above assumptions was approximately 5 years, though this figure varied considerably in different cases.
- (e) In the current research we have worked with the method of proportional life expectancy, which eliminates the above problems. This method was outlined in the body of this article. See Strauss et al. ¹⁰ for further detail.
- (f) As an illustration of the method, suppose that in a given application the EDR at age 30 is 0.01, i.e. 1 death per 100 persons annually. The US male general population life expectancy at age 30 is 47 years, and at age 40 is 37 years. The EDR at age 40 is therefore assumed to be $0.01 \times (47/37) = 0.0127$.
- (g) In addition, the models used in the 1998 study¹ assumed a difference in life expectancy by sex even in the lower-functioning groups. Subsequent research showed, as would be expected, that the sex difference was in fact negligible for individuals with the most severe disabilities. Thus, the results in Table III for such individuals were too high for females and too low for males, other things being equal. In some cases, therefore, estimates for a low-functioning female were too high by more than 5 years.
- (h) As an example of the above, consider the case of a female of age 15 who is tube fed and lifts her head in prone but does not roll, sit, crawl, or feed independently. Table III of the 1998 article indicated a life expectancy of 21 years, and the addition of 5 years for the secular trend suggests a life expectancy of some 26 additional years. But this is too high for three reasons: (1) The female figure in Table III should be reduced and male figure increased as noted in (g) above. This accounts for 2 years. (2) As noted in paragraph (d) above, the methods used in the 1998 article¹ resulted in an overestimation of approximately 5 years. In the present case, the figure is in fact 6 years. (3) In the present case, the effect of the secular trend is an increase of 3 years. As we had noted an 'average' figure of 5 years in previous research², this means

that the increase is 2 years less than might have been anticipated. The sum of these three adjustments is 10 years, which explains why the life expectancy given in Table I here is, appropriately, 16 years rather than 26.

2. Further technical details on the life expectancy methods

- (a) We first identified persons with cerebral palsy who were evaluated at ages 15, 30, 45 and 60.
- (b) Survival times were calculated as the minimum of (i) the time until death or study end date, and (ii) the time until 3 years after the date of the person's final evaluation. Deaths were obtained from the California vital statistics records.
- (c) Cox regression analysis was used to compute the hazard rates over the next 15 years for each group and sex.
- (d) The cohort of 15-year-olds was stratified into nine groups based on the three levels of motor function combined with the three levels of feeding (tube fed/fed orally by others/self-feeds). Data were restricted to those who could not walk without support.
- (e) Two of the above combinations ([1] Does not lift head, self-feeds; and [2] Lifts head but does not roll, self-feeds) were rare and were eliminated from the analyses. One group was added: persons who *do* walk independently at least 10 feet. There are thus eight (= 9–2 + 1) groups in the new Table.
- (f) In the 30-, 45-, and 60-year-old cohorts, survival for those who were tube fed and unable to walk without support was not statistically significantly different by gross motor function. These groups were therefore combined for modeling purposes.
- (g) Our analyses indicated that the proportional hazards assumption implicit in the Cox models could not be rejected at the 5% significance level.
- (h) The table below provides, for each cohort, the total number of people, the number of deaths, the model X^2 (based on the likelihood ratio) and its *p*-value.

- (i) We chose to compute three 5-year average hazard rates for each cohort. As an example, for the 15-year-olds the first 5-year rate was used for ages 15 to 19; the second 5-year rate for ages 20 to 24; and the third for ages 25 to 29.
- (j) Hazard rates in the tube-fed groups were then adjusted to reflect the secular trend of improvement in cerebral palsy mortality rates as described previously. In particular, 5-year hazard rates were multiplied by 0.966^(2002-k), where k is the average calendar year in each 5-year follow-up period. The average values of k in all tube fed groups were 1996, 1998, and 2000, for the follow-up periods 0 to 5, 5 to 10, and 10 to 15 years, respectively.
 - For example, the average year for the first of the three 5-year periods was 1996. This is 6 years earlier than 2002, and the multiplying factor is, therefore, $0.966^6 = 0.81$. That is, the adjusted hazard is 81% of the unadjusted rate.
- (k) For those who could not walk independently, mortality rates over the remainder of the life span were computed under the assumption of the proportional life expectancy (PLE) method.
- (I) For the walks unaided group, we used the hazard rates from the four models at ages 15, 30, 45, and 60 directly, and assumed PLE beyond age 75.
- (m) The resulting mortality rates were used to construct life tables corresponding to each age, sex, and group combination. Life expectancies were obtained from the life tables.

Table I

Age (y)	Persons	Deaths	Model X^2	df	p value	
15	10721	928	1113.601	8	< 0.0001	
30	8571	721	444.116	6	< 0.0001	
45	4694	485	177.119	6	< 0.0001	
60	1180	300	53.781	6	< 0.0001	

df, degrees of freedom.