

Our results examined a specific diagnostic test—electrodiagnostic testing—for upper-limb symptoms and found low predictive ability for determining study outcomes using the presence of common upper-limb musculoskeletal disorders.

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### Survival of Persons With Locked-In Syndrome

In a 1987 study, Haig et al<sup>1</sup> reported on 27 patients with locked-in syndrome (LIS). A slightly expanded group of 29 patients was studied by Katz<sup>2</sup> and Doble<sup>3</sup> and colleagues. To our knowledge, these are the largest follow-up studies of persons with LIS, and the only ones to report survival probabilities. Their findings on longevity were summarized by Doble as follows: "Five-, 10-, and 20-year survival were 83%, 83%, and 40% respectively."<sup>3(p438)</sup>

Unfortunately, these probabilities reflect a methodologic problem. Survival time was counted from the first anniversary of the onset of LIS. However, in some instances, follow-up began many years later, and thus subjects were "guaranteed" to survive until the beginning of follow-up: those who died in the interim never entered the study. As an illustration of the problem, figure 1 of Doble<sup>3</sup> shows survival over a 25-year period even though the subjects were followed for only 11 years.

To correct the survival probabilities, we counted each person's survival time only from the time at which they were "exposed" to death (ie, if they died they would still have been included in the study). Then a Kaplan-Meier analysis<sup>4</sup> gave survival probabilities of 84% at 5 years and 56% at 10 years. The 20-year survival probability cannot be computed directly from the observed data, but the use of some standard actuarial assumptions leads to an estimate of 31%.

In addition to the above, 2 additional facts should be noted: (1) the subject population included primarily elective admissions to a world-class facility, and this may have led to an overestimate of survival; and (2) any advances in medical care since the study period would suggest that these figures underestimate survival.

Nevertheless, the above figures are compatible with those derived for other types of neurologic injury and comparable physical

disabilities. Examples are the permanent vegetative state,<sup>5</sup> traumatic brain injury,<sup>6</sup> and cerebral palsy.<sup>7</sup>

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### The authors respond

I am grateful that our work in the 1980s has served to reverse the perception that people with locked-in syndrome (LIS) universally die.<sup>1</sup> At a recent meeting of the Association du Locked-in Syndrome,<sup>2</sup> for instance, I had the pleasure of meeting more people with the syndrome than were reported alive in the entire world literature when we first published our 1987 study. Now this year, the acclaimed movie *The Diving Bell and the Butterfly*, about a locked-in author, further humanizes the lives of these people.<sup>3</sup> While our work provides some estimate of survival, I do not believe that the comments of Shavelle and colleagues are scientifically accurate, and I worry about the implications for people who suffer from the disorder in the 21st century.

The length of survival from severely disabling syndromes such as LIS is a serious issue. Drs. Strauss and Shavelle are directors of the Life Expectancy Project, an organization that consults on life-care planning and survival.<sup>4</sup> Millions of clients' dollars ride in the balance when life care planners are in conflict about survival. It is natural, therefore, for them to take a critical look at the literature on which their opinions are based.