Deep brain stimulation in cerebral palsy: an opportunity for collaborative research

Bill Silverman was one of the fathers of modern neonatology. He was also considered by many to be a demanding colleague. He challenged his peers and himself to embrace modern scientific methods to address the clinical problems we face. His poignant and powerful book *Retrolental Fibroplasia: A Modern Parable* describes the years of rancorous, opinion-based debate about whether retrolental fibroplasia, which led to blindness in over 10,000 preterm infants in the 1940s and 1950s, was caused by excess or insufficient supplemental oxygen. The situation was only resolved when a large collaborative randomized clinical trial was undertaken and a definitive answer reached – after which debate essentially ended.

In the modern era of evidence-based medicine, most of us would hope to be guided as much as possible by the best evidence. Sadly, there are still too few available ‘facts’ in our field, and even these can often be viewed through many lenses and variably interpreted. One reason for this reality is that unless one pays careful attention to the details of each report about, for example, a new treatment, one can too easily miss nuances that significantly influenced what was done, and found, and reported. This is in part because there are so many sources of variation that can have an impact on the outcome of a treatment – the age and developmental stage of the children treated; their level of impairment; the nature, timing and details of their treatments; supplemental and adjunctive interventions; the timing and nature of assessment of the effects of the treatment - the list can go on and on.

Unlike the field of childhood cancer, we have not been particularly effective in developmental disability in pooling our intellectual and clinical resources to address new ideas (and of course old problems) patiently, collaboratively, and systematically.

One such opportunity is now at hand, namely the collaborative international exploration of deep brain stimulation (DBS) as an intervention for children with movement disorders associated with cerebral palsy (CP). Koy et al. have published a recent meta-analysis of 20 studies of the management of the dyskinetic type of CP (apparently all in adults) using DBS. The main common outcome of interest was clinical assessment of dystonia using several tools. After noting that these 20 reports involved only 68 participants in total, the authors comment ‘Most articles were case reports reflecting great variability in the score [of the outcome measure] and duration of follow-up.’

The early and uncritical adoption of an exciting idea happens far too easily. Our field is replete with examples of apparently good ideas that have, like fireworks, exploded on the scene, illuminated the ground below, elicited gasps from the viewers, and then quickly disappeared in a puff of smoke. In most cases new ideas are never subjected to proper study to help us find whether there are benefits to be harvested from the new approaches. Even in the few situations in which new ideas have been assessed with robust randomized clinical trials (such as in selective dorsal rhizotomy in CP), the surgical and post-operative management regimens were importantly distinct one from another, making meta-analysis challenging. We may have thrown some babies out with the bathwater!

It is essential that cooperative planning and aggregation of collective experience in CP be undertaken before an apparently ‘good idea’ like DBS simply becomes another treatment of uncertain value. Without such efforts DBS may be offered by well-intentioned practitioners without detailed understanding about whether the intervention does more good than harm, and without knowing for whom it works better and less well.

Of course there must be both academic leadership within the field, and human and financial resources that make it possible for people from several centres (and in this case from several countries) to map out a 5- to 10-year proposal for thoughtful and planned observation, description, data collection, and long-term follow-up of people who are treated and studied in a limited number of academic centres.

The Cerebral Palsy International Research Foundation (Princeton Junction, NJ, USA) is facilitating the establishment of a DBS in CP registry with the intent of an eventual international study of this procedure with uniform selection criteria, operative techniques (including brain localizations), outcome measurement, and appropriate follow-up. An experienced collaborative group from Europe, Israel, and North America has met to determine the data elements for the registry. Once established, any centre will be welcome to contribute cases to the registry. Data from the registry will be essential in establishing the protocol for an adequately powered prospective clinical trial.

Let us hope that this experience sets a new standard for the whole field.

DISCLOSURE

The author is a member of the board, and Chair of the Scientific Advisory Committee, of Cerebral Palsy International Research Foundation (CPIRF). The opinions expressed in this editorial are his alone.

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