Letters to the editor

‘Neurodevelopmental treatment for cerebral palsy: is it effective?’

SIR—We appreciate this opportunity to respond to the article by Charlene Butler and Darrah, ‘Effects of neurodevelopmental treatment (NDT) for cerebral palsy: an AACPDM evidence report’. We also acknowledge the time and effort that the Academy has put into compiling research related to NDT, an important area of therapeutic practice. However, we were concerned that the report had methodological limitations as well limitations in its approach to evaluating evidence for the effectiveness of NDT in children with cerebral palsy (CP). We stress that it would be premature and mistaken to conclude that NDT is not beneficial. Absence of evidence of effectiveness in an evidence report cannot be construed as proof that a treatment is not effective. Rather, it may reflect areas in which more meaningful research is needed.

To assess accurately the effectiveness of NDT in CP there needs to be consistent operational definitions of both NDT and CP. It is important to remember that NDT has evolved and changed. For example, the evidence review stated that originally, NDT advocated placing children in reflex inhibiting postures. This is no longer practiced. Yet the evidence report included early articles on such abandoned practices as evidence that NDT is not effective. We point out that the reviewers compared studies published before 1990 with those published between 1990 and 2000 and found the more recent results favored NDT, especially in the areas of motor impairment and motor activity measures. However, this information was not included in their summary.

The reviewers also included articles that may not have used NDT per se. They cited articles that described procedures that specified inhibition of primitive responses and pathological reflexes but were not stated to be NDT. It is unfair to the NDT approach to suggest that an article that did not specify that NDT was utilized constitutes valid evidence that NDT is ineffective.

There are many obstacles that make it difficult, if not impossible, to answer the research question: ‘Is NDT effective for CP?’ First, NDT is not a ‘treatment’ for CP. It is an approach used to assess and to assist children with CP to perform functional tasks sooner and better and with minimal negative effect on future functional abilities. Second, CP is as broad and heterogeneous as its management is challenging. Children with CP include children with spastic diplegia, spastic quadriplegia, ataxia, and athetoid movements. When studying treatment of the disorder, it is hard to recruit a homogeneous population, harder to recruit one of ample size, and nearly impossible to recruit a study population with homogeneous functional goals. Third, identifying and measuring outcomes that can test whether NDT is effective is difficult. Scores on Denver Developmental Screening Tests or the Bayley Motor Scale are examples of measures of function used in the cited studies. These scales are not sensitive to the types of outcomes expected when using an NDT intervention. NDT focuses on discrete and individual functional needs of involved children and not developmental milestones. For example, a child treated with NDT who has feeding problems may benefit from an individualized plan to improve her feeding capability. While the treatment may reduce the danger of aspiration and improve the child’s current nutritional state, it would be inappropriate to expect that such treatment will improve the child’s performance on the Bayley Motor Scale.

Because of these difficulties, we are pleased that the AACPDM is accepting the methodology of single-subject research design as a valid, evidence-based form of research. Other than ‘n-of-1 studies’, we were unaware of levels of efficacy for single subject research design and applaud the AACPDM for seeing a need in the area of rehabilitation research. It would be interesting to open up the discussion of whether the scientific community as a whole accepts categorizing single-subject research design evidence according to the AACPDM’s ranking.

We suspect that trying to evaluate the effectiveness of NDT for CP as was done in this report, is neither useful nor appropriate. We stress that NDT is a method of assessing diverse, neurologically impaired children, identifying their functional state and needs, planning and, yes, treating the child to facilitate meeting those needs. Consequently, focusing on levels of evidence as to NDT’s effectiveness may not be warranted.

Can we design a level 1 or level 2 study to research the efficacy of a complex, multifaceted assessment as well as treatment approach? To ask if such an approach ‘works’ strikes us as analogous to asking, ‘Does the neurologist’s approach work to ameliorate CP?’ The immediate answer is no, because neurologists do not cure CNS dysfunction. A better question might be: ‘Does the neurologist assist children with CP to be more functional?’ This is a more appropriate question, but still too global and multifaceted. However, if one were to define operationally an aspect of the clinicians’ intervention and appropriately define children with specific and well-delineated impairments, as well as have functional outcomes, then research would be possible. It necessitates asking many questions about specific techniques for specific impairments of children with CP working toward specific functional goals.

We have reservations about whether the conclusions reached by the reviewers are justified. For example, the Adams article is identified as one of the studies reviewed, but then it is left out of evidence Table VI and not cited as a study that demonstrated functional gains associated with NDT. Also, articles with results that suggested benefit from NDT that did not reach statistical significance were analyzed as though they were the same as articles where the results favored the control treatments. This is a bias in analysis that fails to recognize the likelihood that such studies favorable to NDT might contain type II errors and thus fail to demonstrate the superiority of NDT because the sample size was too small to achieve statistical significance. For example, the evidence review states under the heading Societal Limitation/Context Factors: ‘Only one of 14 results supports the expectation that NDT would confer a greater benefit to maternal–child interaction’. It would have been more factual to say: ‘Only one of two studies supports the expectation that NDT would confer a greater benefit to maternal–child interaction; the other study did not show a statistically significant benefit’. Using the AACPDM method, it would be as appropriate to say that the Palmer’s study supports NDT in relation to maternal–child interaction (responsiveness and involvement) because their results were positive, but not statistically significant. Yet, these results were listed as part...
of the 14 results that did not support benefit to maternal–child interaction.

We were also surprised that an AACPDM evidence report would state that: ‘Therapists who attended an NDT training course seldom had continuing education in neurodevelopmental treatment although they had joined the NDT Association.’ We certainly do not believe this is true and we are writing on behalf of the NDT Association. We would like to know where the Academy found such data.

We agree with the evidence review that there is too little research assessing the effect of specific treatment techniques (whether they arise from NDT or some other approach) on achievement of functional goals in neurologically impaired children. NDT focuses on discrete functional goals and cannot and should not be viewed as a ‘treatment’ for CP. Therefore, using randomized controlled trial methods to determine if NDT is an effective ‘treatment’ for CP is not appropriate, as the question as posed is unanswerable. However, it is imperative that NDT therapists operationally define treatment techniques for homogeneous populations and look at their effect on functional outcomes.

We appreciate the AACPDM’s efforts to focus on the impact of the NDT approach. The evidence review of the effect of NDT on CP outcomes has furthered the discussion surrounding research of treatment strategies for children with CP. It brings to the forefront many complex and confounding issues which we have highlighted in this letter.

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References

‘AACPDM Treatment Outcomes Committee replies’
SIR–First, we thank the authors of this letter for their thorough review of the tables in this evidence report which identified a typographical error in the evidence table (Table VI) on page 787. The six outcomes from the Adams article are, in fact, present in the table but were incorrectly cited. To correct Table VI, six of the seven results that were cited as having come from the study by Trahan, et al. (51) should be changed to the study by Adams et al. (52) Table V summarizes all results and can be consulted to identify the six outcomes from the Adams et al. study. The article in the AACPDM Database of Evidence Reports at www.aacpdm.org has been corrected.

We would like to remind readers that AACPDM evidence reports simply aggregate and describe research outcomes that have been published. Reviewers and readers may disagree with some of the outcomes measured, or be disappointed about the lack of specificity of the interventions, the sample of individuals studied, and so on. It is the task of the reviewers only to report what has been done and to categorize the results by type of outcome and strength of validity. No attempt was made to ‘compare’ studies. Comparison of studies is not part of the systematic review methodology adopted by the Treatment Outcomes Committee of the AACPDM.

The letter from NDTA restates issues that were identified and discussed at length in the evidence report. Its authors suggest that these issues invalidate the evidence report or render inappropriate any attempt to evaluate evidence about NDT. We disagree.

All the studies included in the evidence report were reported by their investigators to be either NDT, Bobath method or, in two early studies, therapy based on the neurodevelopmental treatment principles advocated by Bobath, Rood, and Ayres. This is shown in Table III: Summary of studies—interventions and participants.

While we agree with the limitations of the NDT research enumerated in the letter, we reach a different conclusion. We do not believe that these limitations lead to the conclusion that NDT, or any other intervention, cannot be evaluated for the extent, types, and strength of evidence currently available to us. Rather, we conclude that recognition of these limitations will allow and stimulate NDTA and other practitioners of neurodevelopmental treatment to move beyond a ‘first generation’ research effort characterized by studies which tried to evaluate an ill-defined intervention on small, heterogeneous groups of children. The NDT evidence report highlights that it is time to implement a ‘second generation’ of research characterized by robust studies that systematically evaluate thoroughly described intervention techniques (including their intensity and setting) in order to determine what works for specific types and ages of children. Many of the same limitations are being revealed by all the evidence reports that are in various stages of completion. It is not a problem unique to NDT but plagues the field of developmental disabilities in general.

The letter from NDTA challenges the grouping of results that are not statistically significant (ns) with results that were found to be unchanged or not different between groups. This has been done because all of these results are indicators that the null hypothesis could not be rejected.

Special caution was given in the evidence report to readers about interpretation of ns results, that is, that they may result from inadequate power in studies to detect differences, if differences actually exist. Only one study stated it had appropriate power. Future studies that include power calculations and adequate numbers of participants are critically needed.

The evolution of NDT and the resultant complication in
evaluating measures of effectiveness prompted careful study by the evidence report authors and review panel. Based on the neurodevelopmental treatment intervention methods described in the articles and the Bobaths’ final publication in 1984, the AACPDM report also described the results of ‘earlier’ versus ‘later’ studies. In their letter, however, the NDTA states that it no longer considers NDT a therapeutic treatment, but an approach. This is new information for the field and is counterintuitive since the name remains ‘neurodevelopmental treatment’. Even so, this raises the issue of terminology. The term, ‘treatment’, in the medical context, does not necessarily infer an attempt at a cure. Nevertheless, the terms ‘intervention’, ‘management’, or ‘approach’ are often used to infer more clearly an aim to aid in the everyday management of the child disabled by cerebral palsy rather than to cure.

Irrespective of the terminology being used, however, if children are being seen, assessed, and provided with interventions for which outcomes and reimbursement are expected, measures of those interventions are required for understanding effectiveness. Systematic attempts to measure interventions should be made and these attempts analyzed and criticized.

In addition, some practitioners may still believe that use of NDT can permanently alter the natural history of some cerebral palsies. We believe that it is important to report literature that might examine that claim.

A benefit of the evidence report may be to act as an impetus for the NDTA and others to clarify what NDT is today and to disseminate this among NDT practitioners, other professionals in the field of developmental disabilities, and consumers. It may be difficult to demonstrate that NDT, as it is described in this letter, can help in the management and care of a child with CP. If, however, there continue to be techniques or activities that are specific to NDT, then NDT may best be studied by measuring these specific techniques or therapeutic activities. They will need to be operationally defined such that the intervention is recognizable as NDT and can be replicated by clinicians and researchers.

The NDTA disputes the appropriateness of using levels of evidence in the review of evidence about NDT. Carl Sagan said, ‘Science is far from a perfect instrument of knowledge. It’s just the best that we have.’ Using levels of evidence to categorize strength of validity of outcomes is a scientific instrument. Though imperfect, it helps us apply greater scientific scrutiny to outcomes research. Use of the concept of a hierarchy of research designs (i.e. levels of evidence) moves us beyond the uncritical acceptance of outcomes as being ‘true’ just because they arise from ‘research’, or beyond the too-critical rejection of research in general as being inconclusive, as even the best studies are never absolutely conclusive. The categorization of studies based on the concept of levels of evidence is particularly helpful in teasing out some of the confounding variables that plague the present literature. More robust studies can then be designed using either group research designs or single subject designs with multiple replications.

The objective of the AACPDM evidence reports is to provide the biomedical research and clinical practice communities with the current state of evidence about various interventions for the management of developmental disabilities. The goal is to stimulate research to fill the gaps in our knowledge base. The first step is this type of dialogue and, speaking on behalf of the Academy, we look forward to continued dialogues in multiple forums about a whole host of interventions and medical conditions.

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Movement Disorders in Children
By Emilio Fernandez-Alvarez, & Jean Aicardi

This book examines movement disorders as applied to individual children, and as distinct from traditional adult neurology. The authors of this comprehensive volume have undertaken a panoramic search of literature and, combined with knowledge of their own experience in decades of clinical work, have managed to collect and sort a vast range of material. Movement Disorders in Children is designed to be used clinically, is especially useful for clinicians with difficult diagnostic cases, and is an essential paediatric neurology text.

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