Longitudinal Assessment of Motor Development

in Epidemiologic Research

for the National Children's Study

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Fall 2004

Prepared for the National Children's Study
by Battelle Memorial Institute
Under Contract No. 282-98-0019

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PART I: INTRODUCTION AND BACKGROUND

Assumptions and Questions that Underlie the Proposed Approach to the Assessment of Motor Development in Children

In developing an approach to the assessment of motor development for the National Children's Study (Study), several questions were considered.

What developmental impairments are most prevalent, and therefore most important to identify, in the Study?

Drawing on epidemiological evidence concerning prevalent disorders of motor development in childhood (Missiuna et al., 2001), particular attention has been given in this paper to two types of developmental motor disorder. These are:

- Cerebral palsy (CP), with an incidence ~2.5/1000 (thus potentially detecting 250 children in 100,000)
- Developmental coordination disorder (DCD), affecting 5–6 percent of children (50–60/1000).

Thus the Study might expect to find, and want to assess in detail, 5000–6000 children over the course of the project. Other conditions that affect the motor function or development of children include:

- Duchenne muscular dystrophy, with an incidence of ~0.24/1000, will involve only about 10–15 boys among the ~50,000. This is a sex-linked condition affecting only boys (hence the ~50,000) and very rarely affecting girls.
- Juvenile arthritis (JRA), at ~1/1000, should yield 100 children with the condition.
- Spina muscular atrophy (SMA), rare at ~0.1/1000, will yield about 10 children.
- Spina bifida, at ~0.6-1/1000, should yield about 60–100 children.

Note that, with the exception of JRA, all these conditions have, or are thought to have, a primary disorder in some aspect of neurological development.

What are the important developmental ages and stages in children's motor development to which particular attention should be paid in the assessment protocol?

The proposed simple assessment scheme and the recommended choice of instruments address that question. (See Tables 1 and 2.)

What tools can be proposed that cover (relatively) broad age bands?

This question is addressed in the context of what aspects of development are being evaluated at different ages. Emphasis in this white paper is on a combination of the functions assessed by the proposed instruments, the measurement properties of those tools, and the accepted use of those measures, especially for the special populations that have been targeted.

Are there approaches to the surveillance and assessment of 100,000 children that can combine accuracy with efficiency?

The approach proposed in this paper tries to accommodate the competing pressures of detailed clinical assessments of all children with the realities of the costs associated with data gathering, the level of expertise required, and possible respondent burden. A tiered approach is proposed that will involve (1) parent-report surveillance of all children, (2) detailed expert motor assessment of those children identified in the surveillance step, and (3) clinical-diagnostic evaluations of those ascertained in Step 2.

Background Perspectives

In typically developing children, gross motor development—the acquisition of basic gross motor skills for postural control and vertical mobility—is essentially well established by about age 5 years. There are many theories concerning the mechanisms and processes by which this development occurs, well summarized by Piper and Darrah (1994). These issues are judged to be beyond the scope of this paper and probably of the Study. Subsequent changes in motor abilities, which obviously usually continue well into adult life, are conceptualized as reflecting a combination of personal interests, training, and practice—in the context of opportunity (the availability of appropriate resources to enable such skill acquisition to occur). These latter factors are seen to be a function of family and social environment and, while important to children and youth and their families, do not in themselves reflect the biological underpinnings that make motor development possible. For this reason, no assessments of advanced motor skills are proposed.

The first emphasis in this white paper is on what can be thought of as basic gross and fine motor development, and the ways to detect variations in motor development that do, or might, signal evidence of important deviations from typical progress. In other words, when is variation in the course of motor development large enough, in terms of the quantity or quality of performance, to warrant attention and further assessment? To address this first step, the authors propose to use validated, standardized, parent-completed screening tools as a cost-effective initial method to identify children who require more detailed (and expensive) assessment, as well as those who appear to be developing normally in terms of the standards of the screening tool, and who can continue in the screening stream.

The second emphasis in this report is on the development of functionally important motor activities, rather than on a detailed concern with component parts or the purported underlying mechanisms of motor development. It is assumed that the Study is concerned with the identification of differences in child development (for example, identifying children outside the norms) on whom more detailed assessment should be done. The detection of such variations will enable researchers to explore possible correlates of those variations as a basis for understanding etiological mechanisms that may have biological and clinical (including preventive) importance. In other words, in the context of the World Health Organization's International Classification of Functioning, Health and Disability (WHO, 2001) the recommended focus is on the activity component of the WHO framework rather than on the details of body structure and function (what were previously referred to as impairments). The rationale for this focus is an interest in

detecting deviations in motor development that have a functional impact on the day-to-day lives of the children and youth who experience these variations.

While from a measurement point of view it is possible to describe motor function on a continuum of scores on tests, this way of thinking about motor function does not lend itself to assessment and description as clearly as do other aspects of growth and development. For example, physical growth or cognitive performance can be categorized using centiles or other norm-related statistical perspectives that make it possible to explore correlates of variation in aspects of growth or performance in relation to other (perhaps causative) variables. Even here, however, one usually groups findings into predefined categories. Although from a numerical scoring perspective this is possible with motor development, for the purposes of the Study, the direction proposed here is primarily toward a dimensional approach, categorizing children's motor develop into typical, suspect, and abnormal, with subsequent more detailed assessment of the status of children who are suspect or abnormal.

What is proposed is an evaluation of motor development of the children in the Study as a three-step process:

- Step 1 is the screening phase, using parent-completed questionnaires to detect children with, or at risk of, difficulties in motor development, based on parental reports of quantity and quality of motor performance.
- In Step 2, children who are screened positive (that is, children who are caught by the screening step) should be assessed with a more detailed developmental evaluation to ascertain whether in fact the screening assessment was accurate (true positive), or whether the child can pass the second-order developmental assessment and return to the screening stream (false positive).
- In Step 3, based on the dictum that "tests don't diagnose, people do" (Folio and Fewell, 2000), it is recommended that for the clinical assessment phase (that is, for those children who are ascertained to present with quantitative or qualitative differences in motor function), there be protocol-based assessments carried out by regional or national experts engaged to assess the children. There is great value in using expertise for these protocol-based evaluations of children who appear to have difficulties, to ensure standardized approaches to both assessment and diagnosis and to ensure developmental/clinical follow-up of those children.

The authors assume that the purpose of the Study is not to develop norms for motor or other aspects of development, but rather to understand the correlates of impaired or deviant development as detected with existing screening and clinical tools.

A word about fine motor assessment is in order. Manipulative skills (such as those needed for many activities of daily living) are often explored in the course of a variety of neurocognitive evaluations (for example, perceptual-motor evaluations). These assessments are presumably addressed by neurophysiologic evaluations and have not been discussed here. On the other hand, many assessment tools (for example, the Peabody Developmental Motor Scales, the Movement Assessment Battery for Children, and the Bruininks-Oseretsky Test of Motor Proficiency) include some type of object manipulation subscales that involve postural control or gross motor

abilities for their accomplishment (for example, throwing or kicking a ball), and are included.

In is important to outline briefly two further contextual perspectives that guide the thinking behind this report reflecting the senior author's experience as a developmental pediatrician and teacher.

- The first concerns the distinction between detection of variations in quality or quantity of motor development (some of which may in fact be abnormalities), and the interpretation of the basis for the variation (diagnosis). To accomplish the first step, one needs tools that can validly discriminate typical from atypical development. To accomplish the second, one requires a combination of appropriate clinical measures and thoughtful assessment by people with experience in infant and child development. This is important in light of the fact that many, if not most, abnormalities of motor development in children (such as CP or DCD) are described and defined phenomenologically rather than on the basis of diagnostic biomedical (or for that matter, clinical) tests. These conditions can be identified according to agreed definitions and schemas, but they rely on clinical training and expertise for accurate ascertainment.
- The second perspective concerns issues in measurement. Measures may be used to undertake one or more of three functions: to describe or discriminate, to predict concurrent or future status, or to evaluate change over time (Guyatt et al., 1992). The primary focus in this white paper is on measures that have been validated to discriminate, because these are the types of tools required to detect meaningful variations in motor function as the basis for more detailed assessment and follow-up. The evaluation of change and the capacity to prognosticate are important for clinical evaluation and research purposes but are assumed by the authors to be less relevant to the focus of the Study.

One final general thought concerns whether, and how, children ascertained to have abnormal motor (or any other developmental) difficulties will be managed and followed clinically. No doubt this important ethical issue has been considered by the leaders of the Study and is addressed elsewhere.

It will, of course, be possible in the proposed Study to identify children with gross motor difficulties prospectively at a young age (or within specific age windows) and then to follow them forward to chart their development systematically in a way that is rarely possible in clinical settings. As this Study gets underway, there will be an important opportunity for people with special interests (for example, in CP or DCD) to present protocols for the assessment and follow-up of these special populations. These ideas are outlined briefly later in this paper.

PART II: OVERALL GOALS AND RECOMMENDED APPROACH

What follows is a roadmap regarding a strategy for the evaluation of motor development in 100,000 children. Details about possible implementation of this roadmap will presumably be developed by those responsible for the actual mechanics of the Study. This discussion is summarized in Tables 1 and 2.

Develop Assessment Protocols

Phase I: Screening for Motor Function Difficulties in the Preschool Years

Developmentally appropriate screening tools are used to detect preliminary evidence of failure to meet motor milestones at any age—what might be considered manifestations of quantitative differences in motor development appearing as delays or deviations in motor development, as seen for example, in the cerebral palsy syndromes, developmental retardation syndromes, myopathies, environmental deprivation, and others.

The first pass at finding children with problems in gross motor development will be to seek parents' reports of the achievement or non-achievement of a number of common milestones using norm-referenced, standardized, parent-completed screening tools. From the screening perspective, to look broadly at children's motor function, the authors would recommend using the Ages and Stages Questionnaires (Squires et al., 1995), a standardized parent-completion tool, at 6, 12, 18, 24, 36, and 48 months of age. Table 2 provides information on the parent-completion time required. In addition to use of this well-validated, parent-completed, screening test, the authors would recommend adding to this screening assessment process three questions:

- "Is there anything your baby is doing with his/her arms, legs, or body movements that concerns you?"
- "Is there anything your baby is not doing with his/her arms, legs, and body movements that concerns you?"
- "Is there anything that you have tried to teach your child to do, involving his/her hands, or whole body movement, that has taken longer to learn than you think it should?"

These simple questions may alert researchers to ascertain children who are passing the motor milestones but are still causing their family some worry. This can happen because of problems with qualitative aspects of motor behavior that are not detected with quantitative milestone assessments, which children may be able to achieve while still presenting difficulties in the integration of motor development for functional purposes. This recommendation is based on the idea that any time the parents identify something about which they are concerned with respect to arms, legs, or body movements (something their child is not doing that the parent feels they should be able to so, or is doing that raises concerns, or is not learning as described by the wording of the questions) these children should move to Phase II and be assessed as if they had failed the quantitative aspects of the screening.

The addition of the open-ended questions will enable parents to report aspects of their child's motor development that may not be captured by the milestone elements of screening assessment, but which may reflect qualitative observations that can be very important. Responses to these questions should be coded as positive if one or more are answered "Yes." Such positive responses should lead directly to a Phase II assessment, just as is proposed for children who are screened positive based on the quantitative scores obtained with the Ages and Stages Questionnaires.

Developmental motor disorders associated particularly with qualitative impairments in functional development (including, for example, cerebral palsy syndromes which may present as asymmetries in motor development, and DCD) include:

- Children under 18 months—Motor Assessment of the Developing Infant (20–30 minutes by people who are appropriately trained)
- Children 18–48 months—Peabody Developmental Motor Scales (45–60 minutes for full assessment)
- At age ~3–4 years one can begin to screen particularly for the appearance/onset of problems in the development of integrated motor skills in the face of what appear to be adequate gross motor abilities (as happens, for example, in children who begin to manifest evidence of DCD at this age). Here one would use the Peabody below age 4 years and the Movement ABC (30–45 minutes) thereafter.

At age ~3 years one can usually detect the appearance/onset of loss of previously acquired motor abilities (as happens in boys with Duchenne muscular dystrophy). This group of children would also include (rare) conditions associated with loss of gross and fine motor function (for example, neurodegenerative disorders). A simple question for parents, such as: "Is there anything that your child used to be able to do with his/her arms, legs, or whole body movements that is now harder for him/her than before?" may be a useful screening approach to detect these rare conditions.

Phase II: Proposed Second-Order Assessment of Children Who Fail a Screening Phase

Developmentally appropriate measures of motor function can be used to evaluate whether children screened positive have measurable difficulties in gross motor function. This would include a positive parental answer to any of the open-ended questions asked of the parents. Such measures address specifically the motor component of children's development, and provide a level of detail and perspective (for example, centile ranks, qualitative aspects of motor function) about a child's motor development not available with screening tools. They require the skills of trained observers/assessors.

Phase III: Protocol for Specialist (Clinical) Assessment to Detect the Onset/Offset of Specific Developmental Motor Disorders (Dimensional Measure[s] of Motor Difficulty)

Any child confirmed to have problems in motor development in the second-order assessment phase of this proposed schema should be assessed by trained, experienced, developmental clinicians (including, for example, developmental pediatricians, developmental therapists such as occupational therapists and physical therapists, and pediatric neurologists). Their role will be to evaluate and interpret the findings generated by the motor assessments carried out in Phase II of the evaluation, in order to arrive at an interpretation of the findings and a formulation about diagnosis. While standardized tests and measures may be helpful to identify the nature and/or extent of the problems, expert clinical evaluation at this stage requires a detailed understanding of motor (and other) development, as most of the conditions likely to be detected are not diagnosable with biomedical tests. In any case, even when a specific diagnosis is possible, characterization of issues such as severity of the disorder and a description of co-morbidities is important.

It is important to emphasize one other point that may be self-evident, but should be underscored: namely, whenever a child is confirmed to have any definable condition affecting his/her motor development, that child must have a careful assessment of all other aspects of neurodevelopmental status. This is because the large majority of the disorders of motor development that will be detected in the course of the Study have a neurological basis, for example, CP, DCD, Duchenne muscular dystrophy, and Down syndrome. Hence, other aspects of a child's neurological development (intellect, sensory function [specifically hearing and vision], neuropsychological status) may also be affected by the underlying biomedical disorder(s) that have caused the motor disorder.

In cerebral palsies (the group of disorders of development of motor control and posture due to a nonprogressive impairment of the developing central nervous system—mix of quantitative and qualitative impairments—what needs to be assessed are the:

- Severity of motor function difficulties
- Distribution of motor impairments
- Type of motor impairment
- Additional functional impairments.

Developmental coordination disorder (DCD) usually begins to be recognized by parents at age 3 and is assessable from age 4. Assessments should include:

- Presence/absence
- Severity
- Co-existing impairments
- Functional consequences.

An assessment is necessary for a diagnosis of Duchenne muscular dystrophy. A clinical evaluation (including a detailed history and assessment to define a specific diagnosis) is necessary for children who:

- Lose motor skills
- Have a non-specific motor delay or impairment.

Assessment by Developmental Stage

There are a number of indicators of potential developmental difficulties that should be sought in the child and family histories that is assumed will be collected on every child as a basic standardized component of the Study, that will sensitize assessors to the possibility of impaired motor development. These include:

- Prenatal
  - Family history of motor disabilities
  - History of impaired fetal movement in this infant
  - History of previous reproductive disorders
  - Parental substance abuse
- Infancy (1st year of life)
  - Feeding problems/failure to thrive
  - Delayed acquisition of motor milestones
  - Abnormalities of quality of motor control (for example, hypotonia or hypertonicity)

  - Discontinuities in motor development (for example, excellent prone function with impaired supine development)
  - Lateral preferences
- Toddler time (2nd year of life)
  - Delayed acquisition of motor milestones
  - Abnormalities of quality of motor control (for example, hypo/hypertonia)
  - Early strong laterality
- Preschoolers (3rd–4th years of life)
  - Loss of motor skills previously demonstrated
  - Emergence of clumsiness, especially discrepancies between (apparent) understanding of the task and executive function (that is, ability to do it)
- School age (after age 4)
  - Loss of motor skills previously demonstrated
  - Emergence of clumsiness, especially discrepancies between (apparent) understanding of the task and executive function (that is, ability to do it).

Continuity/Organization of Assessments

It is important to use parent questionnaires at all stages as the first approach to evaluation of children's motor (and other) development. The Study should also screen for differences in motor development at every stage, with broad inclusion criteria for who will have more detailed (Phase II) assessments. These criteria can of course be varied from stringent to liberal, depending on the tolerance of the Study organizers and the detailed goals of the Study. A hierarchical model of assessment includes:

- Parent report of concerns and reports of achievement/non-achievement of specific motor milestones, using standard screening tools, as a screening step.
- Clinical assessments to ascertain whether being identified by the screen is a true positive or a false positive.
- Detailed clinical evaluation to assess for specified syndromes, including:
  - Cerebral palsies
  - Myopathies
  - Duchenne muscular dystrophy
  - Developmental coordination disorder
  - Neurodegenerative disorders.

"System" Independent Diagnostic Capabilities

It is important to have a systematic account of dimensions of children's function in addition to the gross motor difficulties that lead to the child being ascertained for a Phase II assessment. In other words, children who are screened positive for motor difficulties, using the tools and approaches proposed in this white paper, should have a comprehensive assessment of other aspects of their physical, intellectual, emotional, and social development (as should happen for children ascertained to have functional difficulties in any other dimension of their development). The point to emphasize is that functional problems in one aspect of development are often associated with functional difficulties in other aspects of a child's development, so any child ascertained to have a problem in any area should be assessed carefully.

It is probably useful to obtain descriptive accounts of any diagnostic labels applied to children who are screened positive in Phase I. The point here is that one need not force every child into a pigeonhole when a clear diagnosis is not apparent. There is, however, an opportunity in the Study to learn how children presenting with developmental motor difficulties are thought about and diagnosed in the community, and to compare these community-based formulations with those arrived at by Study expert assessors. Furthermore, having this type of information on record in some form may enable researchers to detect variations within the normal range as a result of, for example, certain environmental toxicants.

There will likely be children who are screened positive in Phase I, are assessed in the Phase II component of the Study, and are judged not to have a problem—at least not a diagnosable one. These children could potentially be used as a comparison group (contrasted to typical children on one side and children with problems on the other side) thus possibly creating the opportunity for groups of children with varying abilities whose exposures could be explored.

Treatment Variables

It is not clear whether treatment variables need to be captured, or in what ways—though a descriptive account of what diagnoses are being applied and what treatments are being offered (traditional as well as alternate and complementary) would be useful, at least descriptively. This could provide an opportunity to explore how motor development disabilities are formulated in the community, in comparison to what the experts associated with this Study say, based on their use of protocol-driven evaluations of children's motor development.

Protective Factors

- Family/social factors
- Demographic factors
- Intellect.

PART III: PROPOSED SPECIAL ASSESSMENTS OF CHILDREN WHO ARE SCREENED POSITIVE AT ANY STAGE

The measures cited in this section are described briefly in Appendix 1.

Phase II Assessment: To "Rule In/Rule Out" Motor Difficulties

The evaluations proposed at this stage are standardized assessments that should be undertaken by people with both expertise in child development and training in the use of the measures recommended. Unlike screening tests, which are designed to be quick and (relatively) simple, evaluation of children with possible motor development difficulties requires the use of people and instruments appropriate to the task.

Note that, for virtually all the assessments done at this stage, the cut points (or sensitivity) of the ascertainments can be adjusted according to the resources and research goals of the Study. In other words, depending on the expectations set by the planners of the Study, one will be able to identify the lowest 5 percent or 10 percent of children (or whatever other proportion is deemed appropriate), assessed with these norm-referenced Phase II assessments, who are judged to need a Phase III-level clinical evaluation.

- For children up to 18 months (adjusting for prematurity), the Motor Assessment of the Developing Infant (also known as the Alberta Infant Motor Scale or AIMS) (Piper and Darrah, 1994) is recommended.
- For children from ages 18–48 months, the gross motor scale of the revised Peabody Developmental Motor Scales (PDMS-2) (Folio and Fewell, 2000) should be used. The PDMS-2 can be used with children from birth to age 6.11 years. It provides a variety of scores (developmental motor quotient, age equivalents, scaled scores). There are 10 items available for each of 17 age bands in the 0–83 month range covered by the PDMS-2; the recommendation for the Study is that children 18–48 months requiring assessment beyond the screening phase be evaluated with the PDMS-2.
- Children aged 48 months and beyond who are identified for the first time as having motor function problems should be assessed with the Movement Assessment Battery for Children (Movement ABC) (Henderson and Sugden, 1992). This instrument is especially useful in exploring issues in the functional integration of motor control—problems that often appear for the first time in the late preschool and early school years.

  An alternative assessment for children of this age is the Bruininks-Oseretsky Test of Motor Proficiency (described briefly in Appendix 1). The Bruininks is an older test battery, and is both conceptually and psychometrically less strong that the Movement ABC. For these reasons the authors' preference is for the Movement ABC for children from age 4–15 years old.

Infants and children who are ascertained to have difficulties at this second level assessment need then to be evaluated by clinical experts. The most prevalent disorders that may be identified are CP (mainly in children in the first 24–30 months of age) and DCD, often recognized in its nascent form by parents of children as young as 30 months, and certainly by preschool and early school years teachers.

Phase III Assessments: Children Who Might Have Cerebral Palsy

The term cerebral palsy (sometimes called the cerebral palsies) refers to a group of conditions of disordered motor development due to some form of non-progressive impairment of the developing central nervous system. The recognized incidence of CP across the western world has been steady at 2–2.5/1000 for many years (Stanley et al., 2000). Thus, in a study with 100,000 children, there may be up to 250 children with CP, with a slight preponderance of males. These conditions usually present with delay in the acquisition of, or failure to reach, motor milestones:

- Qualitative differences in motor development (for example, early persisting asymmetries of function)
- Marked discrepancies in prone (better) vs. supine (poorer) motor function

- Apparent strengths (such as appearing to want to stand at a very young age) that are in fact evidence of spasticity.

These conditions are heterogeneous. Although a variety of risk factors have been identified in epidemiological studies, cerebral palsies are not definable by any specific biomedical marker. They develop (become apparent) over the first 12–18 months of a child's life, when children fail to achieve motor skills (milestones) in a typical way or at the usual times.

Children who have failed the second-level assessment should be assessed by examiners with expertise in child development, most probably developmental pediatricians or pediatric neurologists, who are trained in the use of the assessment tools recommended for this Study. Adding a random age- and SES-matched sample of children who pass the screening assessments (as a control group) would help in further validation of the screening tools, if there is an interest to do this.

The assessments for evidence and description of CP should utilize the Surveillance of Cerebral Palsy in Europe (SCPE) algorithms (Cans, 2000) as the standard approach to assessment. There is currently in progress (mid-2004)—and soon to be completed—an excellent CD ROM-based Reference and Training Manual, created by the SCPE group, that can be used to train assessors regarding CP. The use of this approach will ensure consistent and standard criteria are applied to the ascertainment of CP. This should prevent the ascertainment of children who have signs without having CP and who therefore appear to outgrow it (Nelson and Ellenberg, 1982).

For children ascertained (diagnosed) to have CP:
- Functional level should be rated and classified with the Gross Motor Function Classification System (GMFCS) (Palisano et al., 1997).
- Clinical status should be assessed with the Gross Motor Function Measure (GMFM) (Russell et al., 2002).

Both tools have been developed with, and standardized for, children and youth with cerebral palsy; both are used in the field internationally (Morris and Bartlett, 2004); and both can be learned and applied reliably with relatively little effort.

There is a valuable opportunity to follow the progress of children with CP prospectively and longitudinally (see for example, Rosenbaum et al., 2002), using the GMFM annually and classifying functional status with the GMFCS every 2 years. Other clinical aspects of the CP picture should be assessed every 2 or 3 years with a standardized protocol, possibly the one that will be recommended by the end of 2004 by an international cerebral palsy working group, of which the author is a member. Alternatives include the use of parent-completed multi-attribute health status (functional) evaluations that provide a profile of abilities across several dimensions of function (Kennes et al., 2002).

Phase III Assessment: Children Who Might Have Developmental Coordination Disorder

Developmental coordination disorder (DCD) is a chronic health condition that can be thought of colloquially as a form of motor learning disability. The DSM-IV definition is: "A marked impairment in the development of motor coordination [that]… interferes with academic achievement or activities of daily living" (APA, 1994). In this condition, there are important and functionally significant difficulties in motor coordination, motor planning, and motor learning. The estimated prevalence of DCD is 5–6 percent, meaning that there may be as many as 5000 to 6000 children with DCD ascertained in the Study. Based on emerging experience and research, the impacts of DCD on self-care, social function, and mental health may be considerable. The Study provides a unique opportunity to look prospectively at children who eventually are recognized to have DCD.

Currently, DCD is thought to be recognizable at age 4, with tools such as the Movement ABC (Henderson and Sugden, 1992). As DCD is presently conceptualized, one also needs to know that a child's verbal IQ (as a marker of intellectual capacity) is >80; it is thought that children with lower IQ and problems in motor planning might have other forms of disability that could explain their functional problems.

At present, much of what is known about the evolution of the DCD picture has been gleaned from retrospective accounts of parents' observations of their child's struggles, or their own (for example, trying to teach their child things they believe the child understands but cannot master). By looking prospectively at a very large inception cohort of infants, charting parents' reports and observations over time, and ascertaining a substantial population of school-aged children with DCD, it will be possible to look systematically at the early presentation of this condition, and possibly to identify important markers or correlates of DCD that are present well before the age of 4 or 5 years, when children with DCD typically begin to come to the attention of the health and educational systems.

Early correlates of DCD may include variations in children's language development. On one hand some parents report advanced verbal development (perhaps associated with the activities such children choose to do when they are not demonstrating the usual physical busyness of preschoolers). On the other hand, many children who later are recognized to have DCD present at age 2 or 3 years with language difficulties that do not improve easily. Whether these are early markers/correlates of DCD, whether they are co-morbid conditions, or whether in fact this is an ascertainment bias from selective experience, remains to be studied prospectively.

In clinical studies, DCD appears to be associated with social function difficulties (bullying, social isolation, teasing) especially in the latency years (age 8 and up). DCD is also strongly associated with internalizing mental health problems such as anxiety and depression, particularly in the adolescent years. As well, there are questions about whether, and to what extent, children with DCD are inactive and, therefore, prone to obesity and poor cardiorespiratory fitness. Follow-up through the adolescent years might allow this question to be addressed systematically.

It may be appropriate to add questions about DCD to the inventories that are used in the mental health dimensions of the Study, to ascertain the proportion of children who are identified with mental health concerns and also have evidence of DCD.

There is some experience with intervention strategies that suggest that children with this condition, and their families, can learn compensatory strategies to manage the disability associated with motor coordination and motor planning problems. This is yet another reason to pursue studies that would make early identification of the DCD syndrome possible.

The importance of a prospective study of children with DCD therefore includes:

- Opportunities to understand the early presentation and natural history of DCD
- Opportunities for earlier intervention with strategies to help parents raising children who are struggling with motor coordination problems
- Opportunities to recognize, define and characterize the co-morbidities often associated with DCD
- Opportunities to explore the possibilities of primary prevention of the mental health and social impacts often found in people with DCD, before these become apparent and dominate children's and adolescents' lives.

Phase III Assessment: Children with Other Motor Impairment Conditions

There are a number of relatively rare disorders of child development in which gross motor function is a more or less prominent feature of the condition. These include, for example, boys (and very rarely girls) with Duchenne muscular dystrophy (DMD), whose development often presents with either overall developmental delay or stalled motor progress, usually before age 3 years, followed by loss of previously acquired skills. The prevalence of DMD is relatively low (ranging from 0.01 to 0.24/1000), meaning that the Study will probably see very few children with this condition. Other neurodegenerative conditions are also very rare, but may first be detected on screening in the preschool years because of failure to make motor progress.

Conditions like Juvenile Arthritis are also relatively rare (prevalence less that 1/1000), and will likely be ascertained as part of an evaluation of general health of children, rather than being detected through screening of motor function.

Other childhood conditions associated with impaired or delayed motor development include Down syndrome (1–1.5/1000 births) and spina bifida (about 1/1000), both of which are expected to be recognized at birth.

Developmental delay is a non-specific term that refers to children whose development is progressing slowly in cognitive and adaptive behaviors. This relatively ill-defined group of children may be as prevalent as 4–12/1000. Many will be detected in the course of the proposed screening and Phase II assessments proposed in this paper. It is expected that at the Phase III evaluation stage many children with developmental delay will be diagnosed more accurately and specifically, and followed according to their developmental and family needs.

POSTSCRIPT

This white paper has been prepared on the basis of epidemiological considerations regarding the relative prevalence of disorders of motor development; an effort to propose sensible and efficient ascertainment plans for a 100,000-child longitudinal study; and a clinical perspective on what aspects of motor development and function are relatively more and less important. The senior author will of course be happy to continue to discuss these ideas as the Study moves forward, and as the planners work to integrate the various perspectives that have been offered regarding the Study.

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Appendix 1: Brief Overview Descriptions of Recommended Measures

Ages and Stages Questionnaire (Squires et al., 1995) (4–48 months)

The Ages and Stages Questionnaire is a parent-completed screening tool that appears to have appropriate measurement properties for the task required. The measure has undergone extensive development over the past 20 years, and has several advantages (outlined below) that lead to this recommendation.

There are in total 11 age-specific components to the screening tool, of which the 6, 12, 18, 24, 36 and 48-month stages should be considered for use in the Study. It is recommended that, in addition to the several questionnaires suggested, for screening purposes parents also be asked three additional questions that are specific to their experience with their child's motor development:

- "Is there anything your child is doing with his/her arms, legs, and whole body movements that concerns you?"
- "Is there anything your child is not doing with his/her hands and arms, legs, and whole body movements that concerns you?"
- "Is there anything that you have tried to teach your child to do, involving his/her hands, or whole body movement, that has taken longer to learn than you think it should?"

These questions complement the questionnaire items and allow parents to identify concerns they may have that are not explicitly captured with the relatively brief screening questions at each age and stage.

The rationale for recommending this parent-completed tool as a screening evaluation of motor (and other) development of infants and children includes the following considerations:

- The importance of obtaining parental perspectives on their child's (early) development, including both "how much" (in terms of milestones) and "how well" (in terms of the quality of the performance).
- The efficiency of parent-completed screening instruments (which can be done by mail-out questionnaire, face-to-face interview, or telephone interview).
- The capacity to set the cut-point for screening positive to allow for varied sensitivity, specificity, true and false positive rates, and rates of under- and over-referral. It is recommended that a cut-point of one standard deviation be used, in order to minimize the rate of under-referral (even at the cost of a relatively high rate of false positives) and over-referral (compared to the use of a more stringent cut-point), because the purpose of the Study is presumably to be inclusive in order to detect children with real as well as possible difficulties with development.

Motor Assessment of the Developing Infant (also known as the Alberta Infant Motor Scale or AIMS) (Piper and Darrah, 1994) (0–18 months)

This norm-referenced tool has been developed and validated expressly to look in detail at gross motor function in infants to 18 months of age. Insofar as it is important to assess carefully those

infants who screen positive with the Ages and Stages Questionnaire, this instrument has an advantage over conventional broadly-based infant-toddler assessments like the Bayley Scales of Infant Development, by addressing gross motor development specifically, at a level of detail not found with broader developmental assessment tools.

The measure is an observational tool to be used by any health professional who has a background in infant motor development and an understanding of the essential components of movement as described for each AIMS item. The measure is designed to capture the infant's spontaneously-observed skills, with little or no handling of the infant. Scoring is simple, and both a table of raw scores by age and a graph with centiles are available by which to interpret scores.

It is assumed that infants found to score below the 10^{th} centile will be referred to a specialist at a developmental center for further diagnostic evaluation.

Peabody Developmental Motor Scales (PDMS-2) (Folio and Fewell, 2000)

This norm- and criterion-referenced measure comprises six motor subtests:

- Reflexes (for infants below 12 months)
- Assessment of stationary positions
- Locomotion
- Object manipulation (involving whole-body postural control)
- Grasping
- Visual-motor integration.

Two composite quotients can be derived: a Gross Motor Quotient made up of parts of the first four subtests (depending on a child's age), and a Fine Motor Quotient comprised of the last two subtests. Finally, a Total Motor Quotient can be computed by combining the two quotients.

A variety of scores can be reported with the PDMS. These include raw scores, age equivalents, centile ranks, and standard scores. The composite scores reflect broader perspectives on gross and fine motor function, while the subtests provide more detailed information about specific activities.

The Gross Motor Function Measure (GMFM) (Russell et al., 2002) (to assess children 0–15 years with cerebral palsy)

The GMFM was developed and validated as a discriminative and evaluative measure of the gross motor function of children with cerebral palsy. It is known to be a unidimensional measure of gross motor function, and does not assess other aspects of a child's development. The GMFM is widely used to provide both a descriptive account of a child's current function, and as an evaluative assessment of change in gross motor abilities over time. The measure is specific to children with cerebral palsy, insofar as the meaning of the scores can only be applied to children like those on whom the measure was developed and validated.

GMFM scores are known to be highly correlated with the level of motor disability as described by the observational Gross Motor Function Classification System (Palisano et al., 1997; Rosenbaum et al., 2002).
The GMFM should only be administered by people who have been trained to use and interpret the measure (Russell et al., 1994). A self-instruction CD ROM program is available for potential users.

Gross Motor Function Classification System for Cerebral Palsy (GMFCS) (Palisano et al., 1997) (to classify motor function limitations in children with cerebral palsy)

The GMFCS was developed and validated as a classification system that would both discriminate levels of gross motor function in children with cerebral palsy and predict later gross motor function from earlier status. Evidence has been produced that shows a close correlation between a child's level on the GMFCS and his/her GMFM scores (Palisano et al., 2000; Rosenbaum et al., 2002).

The GMFCS is easy to apply simply by judging which age-specific word picture best corresponds to a child's observed or reported current gross motor function. The GMFCS is not a test, requires no handling of the child, and can be completed on the basis of written descriptions of a child's function or by parent report (Morris et al., 2004).

The usefulness of the GMFCS for children with cerebral palsy is the ability to provide a validated functional stratification factor that can be used in the analyses of potential etiological or co-morbid factors judged by the Study to be relevant.

Movement Assessment Battery for Children (Movement ABC) (Henderson and Sugden, 1992)

The Movement ABC has been evolving over the past three decades, beginning as the Test of Motor Impairment (TOMI) and eventually being refined to become the measure published in 1992 by Henderson and Sugden. The purpose of the measure is to identify and describe motor impairments in children's everyday activities. In addition to the assessment component of the Movement ABC, an observer or examiner (using a checklist tool) can assess children in static and dynamic activities, and in stable and changing environments. There is also an opportunity to report on the behavioral traits that are observed as children attempt the items. The focus is on motor skills as distinct from motor milestones; in other words, on the integration of movements into successful completion of common activities. The developers of the measure have built two components into the instrument; thus it comprises both a test component and a checklist to be completed by an adult able to observe a child's function in everyday situations in which the child has to function.

The Movement ABC comprises four 2-year age bands: 4 to 6 years, 7 and 8 years, 9 and 10 years, and 11 and 12 years. Four sets of eight tasks are included in each band—addressing manual skills, ball skills, and static and dynamic balance. Higher scores on the measure reflect greater degrees of impairment.

The developers report that the measure can be used to screen children who have difficulties with motor coordination and planning. It can also be applied as an assessment of the extent to which a child falls below age-related norms, and can assess changes in function over time or related to intervention.

Bruininks-Oseretsky Test of Motor Proficiency (B-O). (Bruininks, 1978)

The B-O Test has been available for many years. Its current version was developed in the early 1970s when Dr. Bruininks adapted and expanded the Oseretsky Tests developed in the 1940s. The B-O Test is designed to be used by educators, researchers, and clinicians to assess the motor skills of children, particularly those with motor dysfunctions. The test has normative data available, and test scores can be reported in a variety of ways (standard scores, stanines, centiles, and age equivalents).

The test is considered useful with children ranging from 4½ to 14½ years of age. There are eight subtests in the B-O Test:

- Running speed and agility (1 item)
- Balance (8)
- Bilateral coordination (8)
- Strength (3)
- Upper-limb coordination (9)
- Response speed (1)
- Visual-motor control (8)
- Upper-limb speed and dexterity (8).

The results can be aggregated into a Gross Motor Composite, a Fine Motor Composite, and (with the addition of one subtest—upper-limb coordination) an overall Battery Composite.

Table 1: Recommended Motor Development Screening and Clinical Tests/Assessments

Domain: Assessment of motor development					
Test/Assessment/Classification System	**Infancy 0–2.5 yrs**	**Preschool 2.5–5 yrs**	**Grade school 6–11 yrs**	**Adolescent 12–17 yrs**	**Adult 18+ yrs**
Ages and Stages Questionnaire (parent screening assessment)	x	x			
Motor Assessment of the Developing Infant (AIMS)	x 0–18 months				
Peabody Developmental Motor Scales (PDMS)	x 18–48	x 18–48			
Gross Motor Function Measure	x	x	x	x	
Gross Motor Function Classification System for Cerebral Palsy	x	x	x	x	
Movement Assessment Battery for Children (Movement ABC)		x 4–6	x 7–8, 9–10, 11–12		
Bruininks-Oseretsky Test of Motor Proficiency (B-O Test)		x 4 ½ and up	x	x to 14 ½ years	

Table 2: Selected Motor Development Tests

	Age Range	Outcome Scores	Normative Comparison Groups	Reliability	Validity	Administration Time	Cost
Motor Assessment of the Developing Infant	1–18 months	Total number of items passed. Scores are plotted on a graph to provide centiles raltive to normative data.	2,202 age- and sex-stratified Alberta infants born March 1990 to June 1992	One-time inter-rater reliabilities and inter-rater reliabilities over time all >.824	Overall concurrent validity against Bayley Motor Scale (0.93) and Peabody gross motor scale (0.95)	20–30 minutes	Textbook/manual is $38.00. 50 score sheets cost $19.95 each
Ages and Stages Questionnaires	4–48 months	Screening assessments with variable cut-points for referral (1.0, 1.5, or 2 SD) based on ROC curve	Normative sample (Oregon) 2,008 children (1,620 “risk” and 300 “nonrisk”)	Internal consistency (motor scale) 0.53–-0.87; test-retest by parents (2 weeks) 94% agreement; Inter-observer (2 weeks) 94% agreement	Screening cutpoints determined using sensitivity, specificity and ROC curves. Using 1 SD below mean: sensitivity= 0.97, specificity=0.59, true positive=0.97, false positive=0.41, overreferral=0.36, underreferral=0.003. Concurrent validity done against Gesell and Bayley Scales: overall sensitivity=74.6%; specificity=86.2%; false positive=13.8%; false negative=23.4%; underreferral=0.04%;overreferral=11.9%	10–30 minutes. Available in English and Spanish versions	Complete system: $135.00 in each language
Gross Motor Function Measure (for children with cerebral palsy)	0–15 years	GMFM-66 scores range from 0 to 100	N/A The measure has been developed and validated with children and youth with cerebral palsy	Test-retest (ICC)=0.99; Inter-rater reliability (total scores)=0.99	Construct validity shown by gradients of GMFM-66 scores by GMFCS level, and also by diagnostic type of CP (children with hemiplegic CP score highest, those with quadriplegic CP score lowest.	45–60 minutes	$80; GMFM Self-Instruction Training CD $55.
Gross Motor Function Classification System	0–15 years	Five discrete functional levels, based on pattern matching	N/A The measure was developed and tested with children with	Inter-rater reliability (weighted kappa)=0.55 for children <2 yrs,	Correlation between GMFM scores and GMFCS=-0.91. Prognostic validity demonstrated in longitudinal study of 657 children over 4 years (Rosenbaum et al., 2002)	1–2 minutes	No charge. Freely available on the Web (www.canchild.ca)

	Age Range	Outcome Scores	Normative Comparison Groups	Reliability	Validity	Administration Time	Cost
		between age-stratified 'word pictures' and a child's motor function	cerebral palsy	0.75 for children >2yrs. Parent-therapist inter-rater reliability (ICC)=0.94.	et al., 2002)		
Movement Assessment Battery for Children (Movement ABC)	4–12 years in four age bands	A: Total impairment scores (sum of 8 items assessed), %ile equivalents (with separate values for 4–5 and 6–12+ years). B: Movement ABC Checklist (teacher or therapist observations) to produce and overall score.	1,234 U.S. children, stratified by age, sex, region. Much previous work done in U.K. with the TOMI (precursor to Movement ABC)	Median % agreement for Impairment scores=0.80–0.94. Retest reliability of classification (Impaired)=97% for age 5, 91% for age 7, 73% for age 9.	Concurrent validity with Bruininks-Oseretsky test=–0.53; scores of children with known risk or problems significantly higher (more impaired) than norms ($p<0.001$)	30–45 minutes, depending on child's ability and attention	$585
Peabody Developmental Motor Scales (Second Edition) (PDMS-2)	0–83 months	Gross motor, fine motor and composite scores, expresses in various ways (raw scores, age equivalents, centile ranks, and standard scores)	2,003 children reported to be representative of the U.S. population in the late 1990s	Coefficient alphas for six subscales over six ages range from 0.71 to 0.98.	Items selected using Item Response Theory.Content based on research with typical motor development.Concurrent validity demonstrated (correlations range from 0.73-0.86 against another motor measure). Scores correlate with age (ranging from 0.80-0.93). Scores vary by known subgroups	45–60 minutes, with subtests taking 20–30 minutes each	$413 full kit
Bruininks-Oseretsky Test	4.5–14.5 years	There are 8 subtests of 1	765 children (sexes equal)	Test-retest mean reliability	Test scores vary by chronological age (values range from 0.57 to 0.86)	45–60 minutes for the full	$478 U.S.

	Age Range	Outcome Scores	Normative Comparison Groups	Reliability	Validity	Administration Time	Cost
Oseretsky Test of Motor Proficiency	years	subtests of 1–9 items, with a Gross Motor Composite (subtests 1–4), a Fine Motor Composite (subtests 6–8) and an overall Battery Composite aggregating all the data. There is also a Short Form of the B-O Test.	(sexes equal) age 4.5–14.5 years, selected to reflect the 1970 U.S. census (with a small number of demographically matched Canadian children).	mean reliability values for the Composite scores range from 0.80 to 0.87 (with somewhat more variable values for subtests).	(values range from 0.57 to 0.86). Several studies show discriminative validity with scores varying by whether children were developing typically or had varying grades of developmental difficulty.	for the full assessment or 15–20 minutes for the Short Form	