SECTION 2: MEASUREMENT

Consideration of the Use of Health Status, Functional Outcome, and Quality-of-Life to Monitor Neonatal Intensive Care Practice

Maureen Hack, MB, ChB

ABSTRACT. Measures of health status, functional abilities, and quality-of-life are being used increasingly to evaluate health care practice, and to measure outcomes from the patient’s perspective. There is thus a need to reassess the use of growth and neurodevelopmental status that have traditionally been used as measures of outcome after neonatal intensive care.

The quality of neonatal intensive care constitutes only one factor among many that determine the functional health and quality-of-life of survivors of neonatal intensive care. These include genetic disposition, intrauterine events, the effects of sociodemographic factors on the health and development of the child, and on the parents’ assessment of their child’s functioning. To obtain health status, functional and quality-of-life measures, parents need to act as proxy for the child during infancy and childhood. The parents’ cultural, social, and educational background and the specific experience of the parent with children may influence their responses. Furthermore, their perspective may differ from that of the child.

Measures that have been used or have the potential to measure health status, functioning, and quality-of-life include the National Health Interview Survey, the National Health Insurance Study, the Functional Status II, the Multi-Attribute Health System, the Functional Independence Measure for Children, the Vineland Adaptive Behavior Scales, the Adolescent Child Health and Illness Profile, and the Child Health Questionnaire for children, infants, and toddlers. Knowledge of the validity of the use of these measures among survivors of neonatal intensive care is, however, sparse.

Studies have shown that the collection of a standard core of data from various national sources with specific criteria for defining severe disability at 2 years of age is feasible in Great Britain. However, questionnaires or available national databases provide global and epidemiologic information on outcomes rather than identifying the specific pathogenesis or rates of impairments.

To determine the possible deleterious effects of new therapies, specific diseases or impairments will need to be identified rather than the global effect on functioning or health related quality-of-life. Examination of the proximal neonatal impairments that predispose to later disability, such as rates of periventricular hemorrhage or retinopathy of prematurity, are probably better measures for evaluating quality of neonatal care rather than distal impairments such as cerebral palsy, growth imperfections, or reactive airway disease.

The ultimate goal of neonatal intensive care is to provide survival without impairment. Objective measures of specific impairments and their residual disability are thus better measures of the quality of neonatal intensive care than subjective assessments of children and their families. Pediatrics 1999;103:319–328; childhood, quality-of-life, health status, functional ability.

ABBREVIATIONS. HRQL, health-related quality-of-life; WHO, World Health Organization; NHIS, National Health Insurance Study; FSII, Functional Status II; VLBW, very low birth weight; WeeFIM, Functional Independence Measure for Children; CHQ, Child Health Questionnaires.

Measures of health status, functional ability, and quality-of-life are being increasingly used to evaluate health care practice and research, and to measure outcomes from the patient’s perspective. This necessitates a reevaluation of the traditionally used measures of neurodevelopmental status for examining outcomes of neonatal intensive care.

This article will review the definitions of health status, functional outcome, and quality-of-life in general, and for infants and children in particular, the currently available instruments and experience with these instruments among neonatal intensive care survivors and the feasibility of using this methodology to monitor quality of care and changes in neonatal intensive care practice. Because the reported literature pertains mainly to preterm and low birth weight children, the review will focus on this population.

HISTORICAL OVERVIEW OF THE MEASURE OF NEONATAL OUTCOMES

A review of the reporting of outcomes during the preintensive care era provides perspective to the current methods used to measure outcomes after neonatal intensive care.

During the first half of the 19th century reports of outcomes of low birth weight children were mainly descriptive and, in most instances, lacked formal psychometric measures. They pertained to physical and mental development, behavior, scholastic achievement and, to a certain extent, to resource utilization such as the need for special schools.1,2 Hess, who developed the first premature nursery in Chicago in 1922, also reported on personal, family, and social functioning, including the number of sur-
vivors serving in the armed forces, marriages, and resulting births.\(^3\) In the 1940s and 1950s quantitative measures began to be used to report the iatrogenic disasters that resulted from various neonatal care practices and therapies.\(^4\) The major outcomes reported were mental and neurosensory impairments, such as rates of subnormal cognition, cerebral palsy, blindness, deafness, and physical growth failure.\(^5\)\(^-\)\(^7\) These traditional objective measures of growth and neurodevelopmental status continued to be used in the late 1960s after the introduction of neonatal intensive care practices and remain, with few exceptions, the predominant measures of long-term neonatal outcome in general,\(^8\)\(^-\)\(^11\) as well as for randomized, controlled trials.\(^12\)\(^,\)\(^13\)

Suggestions for the design, analysis, and reporting of neonatal outcomes have been published,\(^14\)\(^-\)\(^16\) but there has been an ongoing criticism of the methodology. This includes poor conceptualization in designing studies, incomplete description of the populations at risk, lack of control populations, the measures selected, and lack of uniformity in their use.\(^17\)\(^-\)\(^21\) Assessments of cognitive function are strongly influenced by sociodemographic factors and considered to be culturally biased, and the diagnosis of cerebral palsy is not exact. The use of neurologic and cognitive status has, however, proven useful in comparing outcomes between the preintensive and early postintensive care eras,\(^23\)\(^-\)\(^27\) and in classifying levels of impairment for cost analysis and assessment of quality-adjusted lives saved by neonatal intensive care.\(^28\)\(^-\)\(^30\) Trends in the rates of cerebral palsy have also proven useful in tracking the overall impact of changes in perinatal care on brain injury.\(^31\)\(^-\)\(^33\)

The increased survival since the 1970s has been associated with the appearance of new morbidities, such as bronchopulmonary dysplasia and necrotizing enterocolitis, together with a resurgence of the preintensive care morbidities, retrolental fibroplasia, and neonatal infections among the least mature survivors.\(^34\)\(^,\)\(^35\) These medical problems have resulted in increased health care utilization after discharge, and the need for better measures of the effects of nonneurologic medical conditions on the growth, health status, and functioning of the survivors. More attention is also being paid to subtle attentional, behavioral, and learning difficulties in children who have no major neurodevelopmental impairments but which affect their school functioning and behavior, and may potentially have an enormous impact on ultimate psychologic health, educational attainment, careers, and economic well-being.\(^8\) Table 1 presents a list of medical problems and therapies commonly used as measures of health and health care utilization among survivors of neonatal intensive care.

McCormick\(^21\)\(^,\)\(^36\) has been a pioneering proponent for changing how neonatal outcomes are measured and has stressed the importance of using multidimensional measures of health status. She, together with the perinatal research group from Oxford, England, have also suggested simplifying the whole process of neonatal follow-up by the use of questionnaires and existing databases.\(^37\)\(^,\)\(^38\) These could potentially be less expensive than the traditional examination of the neurosensory and developmental status of each child.

### DEFINITIONS OF HEALTH, FUNCTIONING, AND QUALITY-OF-LIFE

Health has been defined by the World Health Organization as “a state of complete physical, mental and social well being and not just the absence of disease or infirmity.”\(^39\) This all-encompassing definition is very similar to that of quality-of-life, which is generally defined as total well-being encompassing physical and psychosocial determinants. The term health-related quality-of-life (HRQL), which is used when examining the impact of disease on quality-of-life, is increasingly used in adult populations as an outcome measure in clinical trials, effectiveness research, and research on quality of care.\(^40\)\(^-\)\(^43\) There is much debate in the literature and very little consensus as to what constitutes HRQL, because nonmedical aspects of life, which include economic, educational, and environmental factors, may impinge on the health of an individual. HRQL is often used interchangeably with health status or functional status and may be considered globally, with the various domains of health combined to give an overall rating, or be considered specific to a disease (for example, asthma).\(^44\)

In 1980 the World Health Organization (WHO) proposed an international classification for classifying the consequences of disease, including impairment, disability, and handicap, to be used as an extension to the international classification of diseases coding system.\(^45\) Impairment was defined as any loss or abnormality of physiologic or anatomic structure; disability was defined as any restriction or loss of ability (attributable to an impairment) in performing an activity in a manner and range considered normal for a human being. A handicap was
defined as a disadvantage for a given individual, resulting from a disability or impairment, that limits or prevents the fulfillment of a role that is normal for that individual (depending on age, sex and social, and cultural factors). An alternative conceptual framework proposed by Nagy has excluded the category of handicap and includes four components: pathology, impairment, functional limitation, and disability, with disability defined as “the inability or limitation in performing socially defined activities and roles expected of individuals within a social and physical environment.” The National Center for Rehabilitation Research has further expanded these models to include societal limitations on persons with disability including disadvantage and discrimination.

Figure 1 presents Wilson and Cleary’s conceptual model of quality-of-life that links clinical variables with functional status and overall quality-of-life, together with the many intervening variables that may mediate these effects. The outcomes are divided into five levels: biologic and physiologic factors, symptoms, functioning, general health perceptions, and overall quality-of-life. Moving left to right in the model, one moves from cellular pathology and physiology, to the individual, and then to the interaction of the individual as a member of society. At each level there are increasing numbers of inputs that may influence the overall quality-of-life. Factors that may potentially affect the progression from pathology, disease, and impairment to disability, include quality of health care, cultural and sociodemographic factors, educational enrichment and vocational training, individual personality characteristics, and the ability to compensate by using aids to prevent disability or taking advantage of alternate abilities.
parents' responses include their educational level, whether they speak a foreign language, the mother's mental health status, and her overall concerns or perception of health problems in the child. Information on health varies depending on whether it is collected by interview, self-administered, or via health diaries. Both recall of past events and recording of daily events related to health and clinic visits has been found to be inconsistent.

Considerations in using health status, functioning, and quality-of-life measures in childhood include the fact that children's abilities change over time, as does their expected role in society. There are large variations and ranges for the normative growth and development of young children. Sociodemographic factors, specifically poverty, may play a much larger role in childhood compared with adulthood, because it may permanently impede the growth and intellectual development of the child. Parental understanding and interpretations of questions might differ as to what is considered a functional difficulty or disability, or there may be denial or undue concern with underreporting or overreporting of problems. Furthermore, mother's, father's, and teacher's assessment of a child's health and functioning may differ. Children's perception of life may differ according to their developmental stage, and their reporting of health and well-being may differ from that of the parents. Children who have subnormal cognitive development may not understand the questions. They may also be unaware to what extent they differ from other children.

Specific considerations with regard to preterm children include the fact that discrepancies may arise if the parent uses the postnatal, rather than postconceptual, age of the child when answering questions involving functional level or age-appropriate activities of daily living below 2 years of age. Bronchopulmonary dysplasia is considered a chronic disease in infancy; however, unlike other chronic illnesses, the respiratory symptoms gradually subside and disappear in the majority of infants by 1 to 2 years of age. In contrast, some disabilities, such as learning problems, may only become evident when the child has to comply with demands at school. Thus, HRQL and its impact on the family may, over time, improve in some respects, but deteriorate in others.

Table 2 presents a description of selected health status and quality-of-life measures that have been used, or have the potential for use, among survivors of neonatal intensive care.

The National Health Interview Survey includes an overall assessment of health, questions concerning specific diseases and health problems, whether a problem is chronic (defined as more than 3 months' duration), whether it affects activities of daily living, and the degree to which it limits a major activity.

The National Health Insurance Study (NHIS) includes questions concerning physical and mental functioning, social relations, a rating of general health, and the child's resistance or susceptibility to illness, as well as the parents' satisfaction with the development of the child.

The Functional Status II (FSII), which measures the parents' perception of the child's physical, psychologic and social functioning, and the impact of disease on these functions, was originally developed by Stein for infants and children with chronic disease, but has been used to examine functional outcomes of very low birth weight (VLBW) populations, including children without chronic disease. It is currently being used by the National Institute of Child Health and Human Development Neonatal Research Network to assess the parents' perspective of the functional status of <1000-g birth weight children at 18 months of age.

The Multi-Attribute Health System was developed at McMaster University to provide a means to classify health status in terms of the ability to function on each of a set of attributes or dimensions of health status. The relative value attached to health states was also tested through utility measurements that rate the desirability or undesirability of a specific health state. In the 1970s, this system was used for an economic evaluation of neonatal intensive care, and included, at that time, four attributes of health (physical function, role function, social, and emotional function). The Multi-Attribute Health System has been used for children with asthma and after pediatric intensive care. The six health attributes currently used for children include: sensation, mobility, emotion, cognition, self-care, and pain, with ratings of function varying from poor to good or normal. Based on assessments by health professionals, Saigal et al initially applied the multi-attribute approach to describe the health status and health related quality-of-life of <1000-g birth weight children at 8 years of age. Saigal and her collaborators later administered the questionnaires to a cohort of <1000-g birth weight and term 12- to 16-year-old adolescents and to their parents to obtain self-perceived health status. In addition, direct measurement of abilities was obtained using the Standard Gamble Technique to provide a globalized HRQL. Similar techniques were used to obtain the perspective of parents on the health status and HRQL of their children. Although teenagers rated their health problems and functional status worse than that of normal birth weight controls, the majority, even those with neurosensory impairments considered that they had a fairly good HRQL. Saigal et al have more recently developed a multi-attribute preschool health status classification to be used with parents to describe the functional health status of 3-year-olds. This comprises 12 dimensions (vision, hearing, speech, mobility, dexterity, self-care, emotion, learning and remembering, thinking and problem-solving, pain, general health, and behavior), with between three to five levels of functional limitation for each dimension.

The Functional Independence Measure for Children (WeekFIM) based on the measure for adults was developed by Msall and has been used to report preterm outcomes. The child's level of independence on six functions is rated on levels ranging from total dependence to independence (see Table 2).

The Vineland Adaptive Behavior Scales are administered via interview of the parents by a trained profes-
sional. Questions asked included various aspects of daily living, socialization, and motor skills of the child.

Msall examined the functional status as measured by the WeeFIM compared with the Vineland in a study of 148 4½-year-old children born at less than 28 weeks' gestation between 1983 through 1986. Seventeen percent of the population had WeeFIM scores consistent with the need for caretaker assistance, whereas 36% had low functional scores on the Vineland. This discrepancy might be attributable to methodological differences between the two measures.

The Adolescent Child Health Illness Profile, developed by Starfield is a self-administered questionnaire for teens (age 11–17 years) which includes six domains (activity, comfort, satisfaction, disorder, achievement, resilience). This instrument, which takes about 30 minutes to complete, is very user-friendly and allows the adolescent to rate the various health and life experiences on a Likert Scale. It has been used for children with chronic conditions. We are currently using the Adolescent Child Health Illness Profile to examine the various domains of functioning as well as current health, recreational activities, and health risk behaviors compared with normal birth weight controls in a study of 750-g birth weight teens born in 1982 through 1986 and in a study of 1.5 kg birth weight young adults born in 1977 through 1979.

### TABLE 2. Health Status, Functional, and Quality-of-Life Measures for Children [Modified from Pal, Jenney, and Vohr]

<table>
<thead>
<tr>
<th>Measure</th>
<th>Respondent</th>
<th>Age Group (Years)</th>
<th>Domains</th>
<th>Studies of Low Birth Weight Children</th>
</tr>
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<tbody>
<tr>
<td>National Health Interview Survey&lt;sup&gt;64&lt;/sup&gt;</td>
<td>Parent</td>
<td>&lt;5, 5–17</td>
<td>Overall assessment of health. Questions concerning specific diseases and problems. Whether a problem is chronic (&gt;3 months), whether it prevents activities of daily living, and the degree to which it limits a major activity. Bed days attributable to illness.</td>
<td>Overpeck&lt;sup&gt;68&lt;/sup&gt; McCormick&lt;sup&gt;11&lt;/sup&gt; McGaughey&lt;sup&gt;69&lt;/sup&gt;</td>
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<tr>
<td>National Health Insurance Study&lt;sup&gt;65&lt;/sup&gt;</td>
<td>Parent</td>
<td>0–4, 5–13</td>
<td>Physical (activity, role functioning, self-care, mobility), mental (anxiety, depression, positive well-being, mental health index), social relations, general health ratings (also current and previous health, resistance/susceptibility to illness), parent satisfaction with development</td>
<td>McCormick&lt;sup&gt;11,67,71&lt;/sup&gt; IHDP&lt;sup&gt;70&lt;/sup&gt;</td>
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<tr>
<td>Functional Status II&lt;sup&gt;72&lt;/sup&gt;</td>
<td>Parent</td>
<td>0–16</td>
<td>Communicating, mobility, mood, energy, sleeping, eating, toileting, and impact of chronic illness on these functions.</td>
<td>Cronin&lt;sup&gt;73&lt;/sup&gt; McCormick&lt;sup&gt;71&lt;/sup&gt; IHDP&lt;sup&gt;70&lt;/sup&gt;</td>
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<tr>
<td>Multiattribute Health System&lt;sup&gt;74&lt;/sup&gt;</td>
<td>Child/Parent</td>
<td>2–16</td>
<td>Sensation, mobility, emotion, cognition, self-care, pain.</td>
<td>Boyle&lt;sup&gt;75&lt;/sup&gt; Saigal&lt;sup&gt;78–81&lt;/sup&gt;</td>
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<tr>
<td>Functional Independence Measure for Children WeeFIM&lt;sup&gt;83&lt;/sup&gt;</td>
<td>Professional assessment</td>
<td>6 months–8 years</td>
<td>Self-care, sphincter control, transfer, locomotion, communication, social cognition.</td>
<td>Cryotherapy and Surfactant trials&lt;sup&gt;49&lt;/sup&gt; Extreme prematurity&lt;sup&gt;44&lt;/sup&gt;</td>
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<tr>
<td>Vineland Adaptive Behavior Scales&lt;sup&gt;85&lt;/sup&gt;</td>
<td>Professional interviews</td>
<td>0–18</td>
<td>Communication, daily living, socialization, and motor skills.</td>
<td>Ventrilolostomy&lt;sup&gt;86&lt;/sup&gt; Hack&lt;sup&gt;87&lt;/sup&gt; Saigal&lt;sup&gt;89&lt;/sup&gt;</td>
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<tr>
<td>The Adolescent Child Health and Illness Profile&lt;sup&gt;90&lt;/sup&gt;</td>
<td>Child</td>
<td>11–17</td>
<td>Activity, comfort, satisfaction with health, disorders, achievement, resilience.</td>
<td>Boyle&lt;sup&gt;75&lt;/sup&gt; Saigal&lt;sup&gt;78–81&lt;/sup&gt;</td>
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<tr>
<td>Child Health Questionnaire-PF50 (CHQ PF 50)&lt;sup&gt;92&lt;/sup&gt;</td>
<td>Parent</td>
<td>5–15</td>
<td>Global health, physical activities, growth and development, bodily pain/discomfort, temperament and moods, behavior, general health perceptions, change in health, mental health.</td>
<td>Ventrilolostomy&lt;sup&gt;86&lt;/sup&gt; Hack&lt;sup&gt;87&lt;/sup&gt; Saigal&lt;sup&gt;89&lt;/sup&gt;</td>
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<tr>
<td>Child Health Questionnaire (CHQ87)</td>
<td>Child</td>
<td>5–15</td>
<td>Parent impact: emotional/time.</td>
<td>Ventrilolostomy&lt;sup&gt;86&lt;/sup&gt; Hack&lt;sup&gt;87&lt;/sup&gt; Saigal&lt;sup&gt;89&lt;/sup&gt;</td>
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<tr>
<td>Infant and Toddler Health Questionnaire*</td>
<td>Parent</td>
<td>2 months–5 years</td>
<td>Family activities/cohesion.</td>
<td>Ventrilolostomy&lt;sup&gt;86&lt;/sup&gt; Hack&lt;sup&gt;87&lt;/sup&gt; Saigal&lt;sup&gt;89&lt;/sup&gt;</td>
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* Landgraf JM, unpublished.
Bjerager interviewed 85 young VLBW adults born 1971 through 1974 and normal birth weight controls by telephone to estimate both objective and subjective quality-of-life subscores in four areas: elementary biologic needs, warm human relations, meaningful occupations, and diverse and exciting experiences. Additional questions on alcohol and drug abuse, mental health, school performance, and self-esteem were also included. Overall the group of VLBW subjects tended to have lower scores in all areas of objective and subjective aspects of quality-of-life. However, the only significant differences pertained to the 19 VLBW subjects who had neurosensory or physical handicaps who scored significantly lower on the elementary and biologic needs component that included questions regarding the absence/presence of pain or disease, need and quality of sleep, sex life, and physical shape.

The Dutch Collaborative Project on Preterm and Small For Gestational Age Infants born in 1983 has been the only study to apply the WHO classification of impairment, disability, and handicap to the description of 5-year outcomes.\cite{95-97} Impairments, disabilities, and handicaps were classified as none, minor or major and considered separately by system and then globally. The specific systems or domains considered were congenital malformation, neuromotor development, mental development, hearing, visual function, speech and language development, musculoskeletal system, ear, nose and throat, and respiratory problems. Problems with growth and behavior were excluded from the ratings as the investigators did not feel that they had a good assessment of behavior, and subnormal growth had not been defined in the WHO classification. At 5 years of age, 50\% of the <32 week gestation and <1.5-kg birth weight children were classified as “impaired” with impairments including conditions such as syndactyly, hemangiomata, slightly abnormal posture or movement, nystagmus, more than 10 colds a year, or recurrent wheezing.\cite{98} The majority of these children had no associated functional disability. This study did not have a term born control population, however the authors estimated that the rate of impairment without disability would be similar in a normal population. The rate of disability was, however, considered to be higher than expected with 13\% of the total population classified as disabled and 14\% as handicapped. Handicaps were mainly attributable to abnormalities of neuromotor function, mental development, or language and speech development.

An overall examination of the specific psychometric properties of the instruments described above is beyond the scope of the present review. The only published study of the validity of their use among low birth weight children is that of Scholle,\cite{99} who used the 2- and 3-year data on morbidity, functioning, and health care utilization collected by the multicenter Infant Health and Development Project of Low Birth Weight Children\cite{100} to examine which measures of health and functioning the NHIS and the FSII questionnaires\cite{101,102} tapped and how useful they were in predicting health problems and health care utilization 1 year later. Neither the NHIS, the FSII, nor a global rating of health were related to growth, health utilization, or functional measures after controlling for sociodemographic variables and study site. The mother’s global perception of her child’s health was related to outpatient utilization and behavior 1 year later and the NHIS was weakly related to morbidity 1 year later but did not predict hospitalizations or outpatient visits; neither did the FSII. The poor concurrent and predictive validity of the NHIS and the FSII in this low birth weight population is in contrast to that found in an older group of children with chronic illness, mainly asthma.\cite{103} This may be related to the fact that the health problems associated with bronchopulmonary dysplasia, the predominant residual medical illness among VLBW children that requires doctor visits or hospitalizations, gradually resolve over the first few years of life, in contrast to chronic conditions such as asthma which rarely show resolution over time.

**IMPACT ON THE FAMILY**

The total dependence and intimate relationships between a young child and parents makes it imperative to measure the impact of the child’s health status on the life of the family, and to examine the reciprocal relationships between the two. A good example of this reciprocity is maternal depression, which is prevalent among mothers giving birth to VLBW infants. Maternal depression will have a deleterious effect on the child’s well-being and care, and also influence the mother’s responses to questions on child behavior; however, the health and behavior of a chronically ill or impaired child may further exacerbate maternal mental health and family stress. Specific questions concerning impact on the family include whether the child’s preterm birth or health has previously or is currently affecting family activities, routines or relationships, effects on siblings, economic hardship, or decisions to have more children.

The Impact on Family Scale of Stein and Reisman, which was developed to assess the financial impact, personal strain, familial social impact and coping, and mastery abilities of parents of children with chronic illness, has been used for low birth weight populations including those without chronic illness.\cite{71,73,105} Impact on the family of VLBW children is dependent on the child’s age, the presence of a major neurosensory handicap, and the educational and social background of the family.\cite{71,73,94,101,102} McCormick examined the impact on the family 1 to 4 years after the birth of <1750-g birth weight children and found higher impact scores for parents of children requiring greater health care services and for parents with fewer resources.\cite{71} Cronin compared the Impact on Family scores of 96 parents of VLBW children, 1 to 4 years after the birth of the child, to matched parents of normal birth weight children. Parents of VLBW children had higher scores on the familial burden, financial/social impact, personal strain, and mastery scales.\cite{71} Within the VLBW cohort parents experienced more impact when children had more functional handicap and when parents were of low education and family income. Cronin, similar to Rivers who interviewed parents of neurologically-impaired
VLBW children, found that the families were less likely to have given birth again. However, neither author examined whether this was attributable to an obstetric problem that might also have caused the initial preterm birth or to the stress and impact associated with having a preterm child.

SUGGESTIONS FOR A CORE DATA SET IN BRITAIN

The National Perinatal Epidemiology Unit in England and the Oxford Regional Authority have suggested that a standard core of data be obtained from various national sources in Britain to provide an ongoing assessment of the effects of neonatal intensive care and the need to plan for community health and educational services. This minimum data set includes maternal and infant identifiers, maternal social and demographic measures, and perinatal data such as birth weight, gestational age, multiple birth, and death before discharge. Information at 2 years of age includes health and functional status, severity of functional loss within various domains as well as documentation of the underlying disease or impairment. Documentation up to school age to identify lesser learning difficulties has also been suggested.

Suggested criteria for defining severe disability at age 2 years are listed in Table 3. According to these criteria, any one item may define severe disability of function of a specific organ system (for example, respiratory or cognitive disability). Many children have multiple disabilities. For example, a child with cerebral palsy who cannot walk and has a neuromotor disability very often has a disability in cognitive function and communication, and may also have subnormal growth. Similarly, a child who requires oxygen or assisted ventilation at 2 years of age may also have a cognitive disability, hearing loss, or a disability in communication.

Feasibility studies of collecting this minimum data set indicates that the required 2-year follow-up data are being collected, but there is a need for improvement of linkage of records and standardization of clinical findings. Information obtained via postal questionnaires of parents’ knowledge of their child’s development, in a severely impaired population of 30-month-old children with posthemorrhagic hydrocephalus, was in fairly good agreement with a detailed examination by a developmental pediatrician. Parents, however, identified mainly impairments associated with functional loss. In a later cohort with posthemorrhagic hydrocephalus, the children’s general practitioners were asked to assess the developmental status from their medical records rather than via a detailed examination, and similarly identified only impairments associated with functional loss. In a study of the feasibility of using a postal survey in the region of Oxford, 95% of a 7-year-old birth cohort was traced. Parental questionnaires were completed in 65% of the birth cohort alive at 7 years, with teachers’ questionnaires in 56%. The rates of health and educational problems identified via these questionnaires at 7 years of age were similar to those reported in the literature. The investigators thus concluded that this method could potentially be used to estimate regional changes in health and educational needs.

FUNCTIONAL HEALTH AND QUALITY-OF-LIFE AS A MEASURE OF THE QUALITY OF NEONATAL INTENSIVE CARE

There are serious concerns when considering using functional status or quality-of-life as measures of the quality of neonatal intensive care. Stewart et al have questioned the use of questionnaires and maintain that ascertainment of impairment by trained personnel according to specific protocols and standards needs to be the primary outcome measure for assessing outcomes of neonatal intensive care. Although information on severe functional impairment or disability is ascertainable via questionnaires, such information, for example, that a child is not walking at 18 months of age or that a child is blind, gives no information about lesser, nondisabling impairments, or in the case where a disability is present, of the type of impairment that underlies the disability. Assessment of quality-of-life using multi-attribute scales without concurrent medical records and objective assessments similarly gives little indication of the primary disease or impairment that may predispose to a poorer quality-of-life. Ens-Dokkum has

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<tr>
<th>TABLE 3</th>
<th>Suggested Criteria for “Severe Disability” at Age 2 Years</th>
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<tbody>
<tr>
<td><strong>Criteria for Severe Disability</strong></td>
<td></td>
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<tr>
<td>Malformation</td>
<td>Any malformation that, despite physical assistance, impairs the performance of daily activities.</td>
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<tr>
<td>Neuromotor Function</td>
<td>Unable to sit</td>
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<td></td>
<td>Unable to use hands to feed self</td>
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<td></td>
<td>Unable to control head movement without support or no head control</td>
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<tr>
<td>Seizures</td>
<td>More than 1 per month despite treatment</td>
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<tr>
<td>Auditory Function</td>
<td>Hearing impaired, uncorrected even with aids</td>
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<td>Communication</td>
<td>Unable to comprehend word/sign in cued situation</td>
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<td></td>
<td>Unable to produce more than 5 recognizable sounds or no vocalization</td>
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<tr>
<td>Visual Function</td>
<td>Blind or sees light only</td>
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<tr>
<td>Cognitive Function</td>
<td>About 12 months behind at 2 years or more</td>
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<td></td>
<td>More than 3 standard deviations below mean or standardized assessment</td>
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<tr>
<td>Other Physical Disability</td>
<td>Respiratory</td>
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<td></td>
<td>Requires continual oxygen therapy</td>
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<td></td>
<td>Requires mechanical ventilation</td>
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<td>Gastrointestinal Function</td>
<td>Requires tube feeding</td>
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<td>Requires parenteral nutrition</td>
</tr>
<tr>
<td>Renal Function</td>
<td>Requires dialysis</td>
</tr>
<tr>
<td>Growth</td>
<td>Height or weight more than 3 standard deviations below mean for age</td>
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From: Disability and Perinatal Care: Measurement of Health Status at Two Years. A report of two working groups convened by the National Perinatal Epidemiology Unit and the former Oxford Regional Health Authority, March 1994. Oxford, England: National Perinatal Epidemiology Unit.
stressed that what we evaluate in using quality-of-life outcomes is not only perinatal treatment, but the sum of genetic disposition, intraterine events, and postnatal life. Davies has emphasized that demographic factors influence not only the obstetrical situation and the condition at birth, but also the infant’s progress after discharge from the neonatal unit, so that the quality of neonatal interventions may play only a part in determining subsequent status or outcome. The effects of cultural and educational differences on parents’ assessments of their children’s health functioning and behavior may be no less than the effects of sociodemographic factors on measured cognitive skills. Attempts to compare functional and quality-of-life outcomes between neonatal intensive care units that differ in the sociodemographic descriptors of their population is thus fraught with error.

The state of the art and science of neonatal intensive care practice is still evolving. During the last 10 years new therapies have included antenatal steroid therapy, surfactant and indomethacin therapy, and high-frequency ventilator strategies. Nitric oxide therapy, liquid ventilation, and other treatment strategies are currently under investigation. To determine the possible deleterious effects of new therapies, specific diseases or impairments will need to be identified rather than their global effect on functioning or HRQL. Examination of the proximal neonatal impairments that predispose to later disability, such as the rates of periventricular hemorrhage, periventricular leukomalacia, retinopathy of prematurity, prolonged oxygen dependence, or abnormal base of brain-evoked responses are probably the best measures for evaluating the quality of neonatal care at the present time. Assessment of the distal impairments such as cerebral palsy, growth impairments, or reactive airway disease and their long-term implications should be restricted to academic/research programs that would have the resources and expertise necessary to measure outcomes via standard protocols and validated methodology.

There is a need for research to continue to validate and test the available measures of childhood health status, functioning, and quality-of-life among VLBW survivors. Studies need to be performed to test the available measures of childhood health and functional status with these more recent comprehensive health and functional status measures. They should also be used concurrently with in-depth analyses of the pathophysiology of impairment and its relationships to later disability and handicap. Evaluation of outcomes via questionnaires or available national databases are important for the purpose of providing global and epidemiologic information on outcomes, and for the planning of services for children with disabilities rather than for identifying the specific pathogenesis of the impairments. There is furthermore a need for ongoing clinical assessment of the health, development, and growth of all survivors of neonatal intensive care to identify health and potential educational problems, and appropriately intervene when necessary.

CONCLUSION

The ultimate goal of neonatal intensive care should be to provide survival without impairment. Disabled persons might rate their quality-of-life as good or acceptable; however, their impairment and its resulting disability is a poor outcome for neonatal intensive care. Objective measurement of specific impairments and their residual disability should thus be the measure of quality of neonatal care, rather than the subjective assessment of children and their families.

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Maureen Hack

*Pediatrics* 1999;103;319

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