2. Health Services Research for Children with Disabilities

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GROWING NUMBERS OF CHILDREN AND ADOLESCENTS have long-term disabilities, but they often are excluded from health services research on people with disabilities because of concerns about risks and the special problems in studying this population. As a result, we have little information about the epidemiology and characteristics of children and adolescents; the use, organization, and financing of services for them; the best practices; methods of assessing and improving care; ways of keeping them safe and protecting them from medical errors; and basic issues of long-term clinical management.

By deciding on specific criteria for their inclusion, recent research has helped clarify the definition of children's chronic health conditions (McPherson, Arango, Fox, et al. 1998; Perrin, Newacheck, Pless, et al. 1993; Stein, Bauman, Westbrook, et al. 1993). Further work has improved the systematic specification and identification of this population and indicated how different purposes (e.g., epidemiologic research or program eligibility) may determine who is included (Newacheck, Strickland, Shonkoff, et al. 1998; Stein, Westbrook, and Bauman, 1997). There has been less focus, however, on defining the subset of disabilities that chronic health conditions cause. In this article, I define *disability* as the inability to carry out age-appropriate daily activities as a result of a health condition or impairment; *chronic conditions* are health conditions

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(e.g., leukemia, diabetes) that at the time of diagnosis are predicted to last longer than three months. Much of the research reviewed here does not distinguish between chronic condition and disability, although most of the factors discussed pertain to both groups. Several factors distinguish children with disabilities from adults with disabilities: their epidemiology, the dynamic nature of children's development, children's dependence on adults for care, and the health care and financing systems supporting children. These factors form the outline of a health services research agenda (Forrest, Simpson, and Clancy 1997; Kuhlthau, Walker, Perrin, et al. 1998; Perrin, Shayne, and Bloom, 1993).

The first factor on the agenda is the epidemiology of childhood disability, which differs greatly from that of adults. Adults relatively frequently have chronic conditions—such as hypertension, arthritis, coronary artery and other cardiovascular diseases, various cancers, and renal or pulmonary failure—that can lead to disability. Children and adolescents, however, have only a few chronic conditions in frequencies greater than 1 in 1,000 (asthma, developmental disabilities, obesity, certain mental health conditions), and these conditions cause disability even less often (Gortmaker and Sappenfield 1984). Other conditions, individually rare, account for many childhood disabilities. Examples of these rare conditions are unusual chromosome deletions or metabolic disorders (e.g., maple syrup urine disease), many of which have moderate mortality during childhood.

Second, childhood disabilities must be considered in the context of the child's stage of development, because the onset, manifestations, impact, treatment, and relevant outcomes of disabling conditions vary accordingly (Perrin and Gerrity 1984). For instance, the loss of a limb affects newborns and their adjustment differently than it does 15-year-olds. Speech and occupational therapies also focus on different tasks at different ages. And the medications for a 4-year-old child with arthritis are different from those for a teenager with arthritis.

Third, children's dependence on adults means that research on care must take into account a child's family and schools. Because families provide much of children's care, the parents' health and mental health status and sociodemographic characteristics determine their access to care, use of services, and adherence to clinical regimens. In addition, teachers and other providers affect children's access to and the quality of services they receive in school.

Fourth, the great variance in financing systems for the care of children with disabilities creates special problems in studying and improving the

care of young people and their families. Support for children's health care comes from a varied (and variable) mix of public and private insurance, state health programs (e.g., Maternal and Child Health Programs for Children with Special Health Care Needs, state hemophilia programs), special education services, other agencies dependent on philanthropic or government support, and the Supplemental Security Income (SSI) program (Perrin 1999). (The SSI program provides cash benefits for low-income people with severe physical, developmental, or mental disabilities. In most states, SSI recipients also receive Medicaid coverage even when the household income may be higher than the state's usual eligibility levels for Medicaid.) This mix of sources of support is made even more complex because of the various places and service systems in which children receive care.

In this article, I describe some of the current health services research on child and adolescent disability in the areas of epidemiology, organization and financing of care, quality and outcomes, and information bases for childhood disability. I also identify key issues for developing new and important health services investigations in these areas.

Children and Adolescents with Disabilities: Epidemiology and Characteristics

More and more children and adolescents are found to have chronic health conditions, many of which cause disabilities. In 1960, approximately 2 percent of American children and adolescents were identified by their parents as having to limit their activities because of a health condition lasting longer than three months; by the mid-1990s, this number had risen to more than 7 percent (Newacheck and Halfon 1998). Ironically, much of this rise in child and adolescent disability reflects the markedly improved survival rates for many health conditions that had much higher rates of mortality half a century ago. These improvements in health care also include better trauma services, which increase the survival rate of children and adolescents but also result in about 10 percent of their long-term disabilities (MacKenzie 2000).

Despite their better chance of survival, children and adolescents now face a number of potentially disabling health conditions that occur at much higher rates than was evident in earlier years. Most important are major new epidemics of several conditions, including obesity (Fredriks,

van Buuren, Wit, et al. 2000), asthma (Volmer, Osborne, and Buist 1998; Weitzman, Gortmaker, Sobol, et al. 1992), attention deficit hyperactivity disorder (ADHD), type II diabetes (Rosenbloom, Joe, Young, et al. 1999), and depression. Laplante and Kaye (1998) note that in the past several years, children and adolescents have had the highest growth rates of disability of any population cohort.

The primary diagnoses by which low-income children and adolescents with the most severe disabilities have gained eligibility to the SSI program are mental retardation (29%), chronic physical disorders (38%), and other mental health conditions (33%) (Social Security Administration 2000). Although the 29 percent of SSI recipients with mental retardation represent a fairly homogenous group functionally (despite a relatively wide variety of etiologies), the conditions causing disability among the other 71 percent are quite diverse. Consequently, we probably cannot generalize the standards or quality of care for any single condition to the overall quality of care and outcomes for children with other disabling conditions.

Most childhood chronic health conditions do not cause disability. The Ontario Child Health Study indicated that 15 percent of children aged 4 to 11 years old have chronic health conditions, but only about 2.5 percent of all children have disabling conditions (Cadman, Boyle, Szatmari, et al. 1987). Newacheck and Taylor's 1992 study of the chronic conditions listed in the 1988 National Health Interview Study found that about 31 percent of children under 18 years of age have a chronic condition. Of these, about 8 to 9 percent have relatively minor limitations of activity because of their condition, and about 1.5 to 2 percent are significantly limited. Thus, a relatively small number of children and adolescents are actually disabled, although several conditions can cause disability (Gortmaker and Sappenfield 1984). Although these numbers vary partly because of differences in the definitions used in the studies, they help illustrate the different rates of disability and chronic health conditions among children and adolescents.

The disabilities of young people also have different trajectories from those of adults. Childhood and adolescence are characterized by more change and fewer lasting disabilities than are older age groups (Gortmaker, Perrin, Weitzman, et al. 1993; Pless and Wadsworth 1989). The conditions of working-age and elderly populations are commonly either stable or decline over time, although they, too, may go into remission or become worse, as children's conditions do. In contrast, children

may have a disability or a chronic condition at one time in their lives but appear much less ill or disabled five years later, and vice versa.

Healing and habilitation processes proceed differently in younger populations as well. For example, recent unpublished analyses of the children in the National Longitudinal Survey of Labor Market Experience, Youth Cohort (NLSY), show slowly increasing rates of significant chronic illness with age, although about 40 percent of the children identified in 1988 no longer appeared to have a chronic condition in 1994, indicating also a substantial number of children with new chronic conditions during this period.

Poor children have particularly high rates of disability, with at least some evidence that not only does poverty increase the incidence and severity of disability but also that a child's disability may impoverish the family even more (Mashaw, Perrin, and Reno 1996). In addition, many conditions are more common in poor populations. Controlling for potential confounders, poor children are 80 percent more likely to be limited in activity because of a chronic condition (Newacheck and Starfield 1988). A child's disability has a major impact on the family's time and money, because of both their greater needs and the less time their parents can spend at work (Kuhlthau and Perrin 2001). A study by Aday, Aitken, and Wegener (1988) found that 23 percent of primary caregivers in families caring for children assisted by ventilators were employed full time, and 15 percent worked only part time. Fifty-four percent of primary caregivers of children assisted by ventilators reported having to stop working because of the child's disability. In contrast, 82 percent of the secondary caregivers worked full time, and none had to stop working because of the child's disability. In a study of mothers' employment after a first hospitalization, parents of a disabled child newly dependent on technology were more likely to stop working than were parents of children who were hospitalized for acute conditions. Furthermore, mothers who had quit their jobs had little family support and a poor quality of life (Thyen, Kuhlthau, and Perrin 1999).

Organization of Care for Children with Disabilities

Children and adolescents with disabilities typically need many primary and subspecialty services, including various specialized therapies to

maintain or improve their functioning (Hobbs, Perrin, and Ireys 1985). Problems that children with disabilities face include lack of adequate primary and preventive services, barriers to subspecialty care and specialized therapies, limited coordination of services, and uneven access to schools and other direct-service providers.

Children's access to primary care varies with respect to both having a dependable source of primary care services and receiving appropriate care services. Children with spina bifida, for example, receive fewer immunizations than do comparable populations, despite having similar access to primary care (Raddish, Goldmann, Kaplan, et al. 1993). In regard to more complex disabilities, primary care physicians may not have adequate knowledge of the condition, its treatment, and the best monitoring practices. Only a few disabling conditions (e.g., motor disabilities originating in the central nervous system) are common enough that primary care clinicians have much experience with them.

Centralizing the subspecialty treatment of children with relatively rare disabilities would help ensure at least a coordinated system of care for larger numbers of children, with the prospect of having stronger evidence to support care management recommendations and of being able to identify rare complications. For example, a few children with diabetes develop problems with joint mobility, but the infrequency of this complication makes it unlikely that clinicians treating just a few children will ever see it (or recognize it). Those centers that attract enough children with specific disabilities gain substantial experience with relatively unusual complications and provide access to newer treatments as they become available, although the children seen in these centers may not receive preventive services. Such centers exist almost entirely in academic health centers, although occasionally large health plans have centers for more common conditions. Earlier studies showed that most children with fairly uncommon disabling conditions receive their specialized health care (especially medical and surgical care) in centralized units, usually academic health centers (Weitzman 1985). Recent studies of children insured by Medicaid (Kuhlthau, Ferris, Beal, et al. 2001) indicate, however, that subspecialty care is not always available.

The complexity of a child's disabilities—for example, a child requiring mobility assistance, specialized respiratory care, and nutritional supplementation—also means that a single health clinician cannot arrange or deliver all of these services. Indeed, coordinating primary and subspecialty care is difficult, indicating a need to study ways to enhance

communication and coordination among clinicians. Ideally, children and their families should receive most services in their home community, even if it is far away from a tertiary, highly specialized health center. They also should have access to such a health center for complications, updates on new treatments, and review of the child's treatment plan, especially as the child grows older. Coordinating care probably helps minimize the duplication of services and increase families' access to needed services, although its effects have not had careful assessment (American Academy of Pediatrics 1999).

Many children with developmental disabilities receive care in specialized centers of excellence (University Centers for Excellence in Developmental Disabilities Education, Research, and Services), which provide a multidisciplinary team approach to evaluation and treatment. Supported by several sources, these centers can be found in every state, all associated with major universities and academic health centers. Although these units may emphasize diagnosis and evaluation, in many cases they also provide longitudinal care, based on a model that recognizes the multiple facets of a child's care in the context of family and community.

The idea of a multidisciplinary team may have important implications for children with other kinds of disabilities. The subspecialty programs of many academic health centers have physicians, nurses, social workers, nutritionists, and other therapists collaborating together. For example, these teams may help a child disabled from cancer treatments or recovering from a brain injury to reenter school and community programs. Users of comprehensive hemophilia and sickle cell disease treatment centers have had better outcomes (mortality, emergency department, and hospital use) than nonusers, although the studies did not control for selection biases (Soucie, Nuss, Evatt, et al. 2000; Yang, Shah, Watson, et al. 1995).

Children with disabilities may also receive services from providers outside the usual health services. For instance, public special education programs offer many services to children with disabilities, mainly through programs authorized by the Individuals with Disabilities Education Act (IDEA). This act requires these programs to provide certain health-related services to ensure that children with disabilities receive appropriate educational services. The act also provides for early intervention programs for children from birth to 3 years of age, also often including certain specialized therapies.

We have no reports detailing the scope of services actually offered in educational settings. School systems vary greatly in how they provide (and pay for) these services. Some hire therapists and offer services directly, and others contract with outside agencies to provide services. States and school systems also finance school-based services differently, particularly in how much the schools can use Medicaid to pay for their services. Clearly, coordinating school services more closely with other health services could both help families and lighten the administrative burden and redundant services.

The federal and state Title V Maternal and Child Health programs also offer additional direct services to children with chronic conditions and developmental disabilities, again varying across the states (Ireys, Hauck, and Perrin 1985). Some states provide both outpatient and inpatient health care services directly, whereas others offer only some subspecialty medical and surgical care programs. Still others support a variety of services not usually covered by public or private insurance, and some use their resources mainly to develop systems of care.

Financing Care for Children with Disabilities

Care for children with disabilities is financed by a variety of insurance programs, both private and public, as well as the public and private direct service programs noted earlier. Most American children, including those with disabilities, are insured by their parents' employment benefits (Fox and Newacheck 1990). Medicaid pays for the care of a moderate percentage of children, particularly those with disabilities, mainly because the SSI program in almost all states provides access to Medicaid insurance (Perrin and Stein 1991).

Data from the early 1980s show that children with chronic health conditions had lower rates of insurance coverage than did children without such chronic conditions (Butler, Budetti, McManus, et al. 1985). Since then, children with disabling conditions have had better access to public insurance, especially because more are enrolled in SSI. Using the 1988 Child Health Supplement to the National Health Interview Study, Holl and colleagues (1995) found no differences in rates of insurance coverage among normal children, as opposed to children whose activities were limited due to a chronic condition, although the measure of chronic condition here was relatively crude.

Children with disabilities have higher rates of utilization and expenditures than other children do (Lewit and Monheit 1992; Newacheck and McManus 1988). Sixty percent of Medicaid's expenditures for children are spent by about 10 percent of Medicaid recipients (Adams, Ellwood, and Pine 1989; Kuhlthau, Perrin, Ettner, et al. 1998). The amount spent on children covered by SSI is 2.9 to 9.4 times higher than that spent on children insured by Medicaid but not enrolled in SSI. But if one exclude the approximately 10 percent of children with high expenditures (more than \$10,000 per year), the average SSI expenditure is only 1.5 to 2.7 times higher than the non-SSI average. Those children on whom the most is spent are likely to use hospitals and long-term care, which account for more than half this amount. It is important to note that many non-SSI children also use these services, indicating that many children with disabilities may not receive SSI benefits (Ireys, Anderson, Shaffer, et al. 1997). Generally, pharmaceuticals account for a smaller proportion of expenditures for children than for adults, although here, too, the patterns are uneven, with children with cystic fibrosis and diabetes, for example, having relatively large pharmaceutical expenses (Ireys et al. 1997).

In general, compared with most private health insurance plans, Medicaid offers relatively generous benefits for people with disabilities, including much better long-term care benefits. Nonetheless, the payment and support for such items as assistive devices can be very difficult to obtain, even for Medicaid users. Many children who need specialized devices (such as wheelchairs) that will fit their growing bodies find that Medicaid allows replacements only infrequently, and they often outgrow their equipment well before the authorized replacement time.

Different financing arrangements can either enhance or hinder care for children and adolescents with disabilities (Perrin, Kuhlthau, Walker, et al. 1997), although the research literature in this area is sparse. Organized multispecialty programs with integrated information systems that coordinate care and communication more efficiently can ease families' access to needed services. The different financing arrangements offer choices of whether copayments should be required and what should be included in the benefit package. Without having to adjust their payments to account for greater needs and use, plans have an incentive to minimize the use of services, so people with high health costs may face both direct and indirect barriers to enrollment. In this case, risk-adjusted payments can help with access to needed services.

Changes in health plans may also disrupt previous arrangements with both primary care and subspecialists. Some plans assign patients to primary care providers and then refuse to refer children with disabilities to subspecialists (Ferris, Chang, Perrin, et al., 2002). Although enrollments in private plans may have peaked, the states now increasingly include their Medicaid populations in managed care plans but differ greatly in the degree to which they include children with disabilities (Kaye, Curtis, and Booth 2000).

Medicaid contracts and managed care plans may ration the provision of specialized therapies for people with disabilities (e.g., physical and occupational therapy, respiratory therapy, speech and language services), given the limited evidence for their efficacy. In Medicaid and private insurance contracts, “medical necessity”—the basis for determining whether to pay for a particular service—also may limit access to certain specialized therapies, especially when no trials of their efficacy have been conducted (Fox and McManus 2001; Wehr 2001). A child who has an ambiguous diagnosis but a clear need for specialized therapies is one example of this problem. Accordingly, it is important to clarify the strength of the evidence for the use of specialized therapies, in order to fill in the gaps in evidence and to consider alternatives to determining medical necessity while awaiting better evidence. The remarkably small amount of evidence regarding the use of specialized therapies for children and adolescents makes determining medical necessity particularly important for this age group.

Quality and Outcomes

The field of quality assessment in general lacks an adequate evidence base. Most evidence reports and practice guidelines have dealt with relatively brief episodes of care or illness, paying less attention to long-term clinical management. A recent report by the Institute of Medicine evaluating the quality of long-term care concentrated on nursing homes, emphasizing standards, staffing, and measurement, but admitted that there was little information about quality assessment and improvement in other long-term care settings (Institute of Medicine 2001a). Although the report documented the growing numbers of children and adolescents with disabilities needing care over the long term, it offered little advice about assessing quality for this population. Thus, it has been difficult to

measure the quality of long-term clinical management and care for all people with disabilities, especially for younger populations who receive most of their care in noninstitutional sites.

Until recently, the published practice parameters and clinical guidelines of the American Academy of Pediatrics (AAP) have addressed only acute situations. For example, the AAP published guidelines for the management of acute exacerbations of asthma, but none for the long-term management of asthma (although it did endorse the NHLBI guidelines). The AAP recently published its first guidelines for a chronic condition, the treatment and long-term management of children with ADHD, although the publication notes the limited evidence for key aspects of long-term care (American Academy of Pediatrics 2001).

Disease management programs have improved the care of adult populations with several chronic conditions. Their applicability to children's health has been limited mainly to asthma, and the epidemiology of childhood disability (with large numbers of individually rare conditions) makes the widespread use of these programs less promising than for adults. Nonetheless, the care guidelines of some specialized centers could supplement the disability management programs for certain childhood populations, even when the condition is relatively rare—for example, the care of children hospitalized with sickle cell disease or leukemia.

Few studies or controlled trials have examined the efficacy of specialized therapies for children, and those few offer only limited support for their use (Palmer, Shapiro, Wachtel, et al. 1988; Piper and Pless 1980). Several systematic reviews of therapies note this lack of evidence. For example, the authors of a study of the effectiveness of speech and language therapy noted, "We wanted to take a systematic approach to reviewing the research, but there are not enough controlled studies for us to confine ourselves to this approach. We therefore extended our review to studies displaying the state of knowledge and the main therapeutic challenges" (Enderby and Emerson 1996, 1655). Similar issues emerged in a review of chest physiotherapy in mechanically ventilated children, which concluded, "Despite its widespread use, almost no literature dealing with this treatment modality in pediatric patients exists" (Krause and Hoehn 2000, 1648). Because of the very limited database and because practitioners have legitimate disagreements about the appropriate management of different conditions, most guidelines can only attempt to represent a consensus. It is important, therefore, that the utility and

value of a variety of commonly used specialized treatments for disabling conditions be examined.

Measuring the outcomes and health status of children is more challenging than it is for adults. Several measures are available for specific areas of functioning (e.g., mobility, emotional, developmental) and a few for general child functioning, including that developed by Stein and Jessop (1990; FSII-R, applicable to children aged 0 to 16 years); the Child Health Questionnaire (applicable to children older than 5) developed by Landgraf, Abetz, and Ware (1996); and the Child Health and Illness Profile—Adolescent Edition (Starfield, Riley, Green, et al. 1995). Varni, with a particular interest in children with chronic conditions, has developed health-related quality of life measures for children (Varni, Seid, and Kurtin 2001; Varni, Seid, and Rode 1999).

In addition, the 1980 World Health Organization (WHO) International Classification of Impairments, Disability, and Handicaps (ICIDH) categorized several areas of disability and functioning and associated clinical conditions with their likely outcomes in these areas. Over the last five years, the WHO has revised and updated this classification for the new International Classification of Functioning (ICF), now including more direct attention to disabilities in children and adolescents and to the measurement of their functioning (World Health Organization 2001).

But despite these studies, there are few good measures of outcome and health status for children, especially covering multiple aspects of child functioning, largely because of the difficulties of measuring functioning and ability at different ages and stages. The functioning of a 3-year-old child with cerebral palsy requires a very different assessment from that of a 15-year-old adolescent with cerebral palsy. Given the dependence of young children on the adults around them, usual measures of activities of daily living (ADLs) have little applicability, and the development of measurements has been hampered by the difficulties in determining significant deviations from normal early development and behavior (Stein 2001).

Few studies have addressed the errors in the long-term health care of young people with disabilities. The complex communications among family, child, primary care clinician, and subspecialty and specialized therapy providers offer many opportunities for errors of omission, with each party thinking that another is responsible for certain parts of the treatment program (Kanthor, Pless, Satterwhite, et al. 1974). Problems

in communication may also mean that a treatment plan has not had adequate attention or definition or that a recommendation from one provider has never found its way to the family. A second error-related problem has been in the area of access to the appropriate preventive services needed by all young people. As noted earlier, often most of the treatment plan and services provided are directed to the specific disabling condition rather than to the general health-related issues of the person with disabilities (Kuhlthau, Walker, Perrin, et al. 1998). Other sources of errors are those in prescribing, dispensing, and adhering to both medications and other aspects of treatment plans. Errors are most likely to occur during times of transition (e.g., from home to hospital, hospital to school, into adolescence, home to community). Transitions are stressful for children and parents and often require transferring direct care responsibilities, monitoring, and supervision from one provider to another or to the family.

Information Bases for Child and Adolescent Disability Research

The available databases for child and adolescent disability research include both administrative and survey data. In most cases, administrative data are limited to Medicaid claims, with relatively little information available about children insured by the more prevalent employment-based health insurance plans. Medicaid provides substantial information about the changing patterns of disability among children and adolescents, their utilization of services, access to specialized services, and expenditures.

The growth of Medicaid's managed care arrangements has made claims data less useful, insofar as providers have little incentive to use reliable diagnostic or procedure codes for patients. Furthermore, schools or early intervention programs provide many services for children with disabilities (e.g., occupational therapy, speech and language services), which may not appear in any claims files. Analyses of Medicaid claims data require substantial technical expertise concerning eligibility, coding, provider identification, and the merging of complex files (Cooper and Kuhlthau 2001). Our analyses indicate an underreporting of mental health and developmental retardation in claims data analyses (Perrin, Kuhlthau, McLaughlin, et al. 1999), although others report fewer difficulties in

identifying children with these conditions (personal communications, March 2001, with Elizabeth Shenkman, Ph.D., and John Neff, M.D.).

Several national surveys provide important information about patterns of care of young people with disabilities, especially the National Health Interview Survey (NHIS), the National Survey of Children with Special Health Care Needs (CSHCN), the Medical Expenditure Panel Survey (MEPS), and the National Longitudinal Survey of Youth (NLSY). Only the last of these contains much longitudinal information (MEPS has some two-year longitudinal data), allowing assessment of the clinical and developmental trajectories of young people with disabilities. The surveys have gained much helpful information from dedicated attention to children (e.g., the 1982 and 1988 child supplements to the National Health Interview Survey) or their disabilities (the 1994–95 supplement to the NHIS), and similar special samples in MEPS. These surveys contain good national estimates of rates of chronic conditions and disabilities, although in most surveys, the numbers of children with disabilities in general and with specific conditions are relatively small. The CSHCN includes much larger numbers of children with special health care needs (750 per state) and thus will offer much richer information about children with disabilities. The initial data from this survey should be available in the fall of 2002 (Van Dyck, McPherson, Strickland, et al. 2002).

The broad variations in pathways for young people with disabilities and the importance of understanding the interaction of social characteristics, program participation, and long-term disability make a strong case for obtaining better longitudinal data for this population. U.S. researchers have made little use of the longitudinal data from other countries, especially the several longitudinal cohorts followed with extensive social, health, and utilization data in England and Scandinavia.

Addressing Gaps in Health Services Research for Children and Adolescents with Disabilities

Most striking in my review is what we do *not* know about health services for children and adolescents with disabilities. The following is a partial list of potential research topics, organized according to the sections of my review. The list reflects those areas that appear most promising—or are most needed—to improve the quality of care of young people with

disabilities and their families. The goals for children with special health care needs set by the Maternal and Child Health Bureau for *Healthy People 2010* and the systems change grants awarded by the Centers for Medicare & Medicaid Services provide additional guidance to needed research.

In clinical and health services epidemiology, we need a better description of the changing numbers of young people with disabilities and especially how their functioning has been affected. The changes in young people's disabilities merit careful evaluation of how the patterns of disability change over time, how the different courses for young people can be predicted, and what programs or services appear to influence those courses. We also need to know how to minimize the effects of disability on young people's abilities to engage in age-appropriate activities. What characterizes the successful transition to adulthood by adolescents with a disability, and what services promote a successful transition? Finally, research should focus on the social and structural determinants of new epidemics of chronic conditions among children and adolescents and what determines whether these conditions become disabling.

As noted earlier, the evidence on which most of the long-term clinical interventions and treatment are based is quite weak. Research agencies, especially the National Institutes of Health, should begin extensive and coordinated research on chronic diseases and the treatment and management of disabilities. Given that most of the money spent on health care is for the nation's chronic diseases and disabilities (Fox 1989), this lack of investigation seems remarkable. Future research agendas should address the efficacy of many specialized therapies, explore their usefulness for different disabilities, determine dose effects (if any), and examine mechanisms by which they work. We also need studies of these interventions' effectiveness and cost benefits.

Several organizational and financing questions merit study, including research on the organization of care that takes into account the variations in providers of care and describes the arrangements by which children with disabilities receive services. Such research should look at the current relationship between primary and subspecialty care providers and how variations in these arrangements affect the treatment of children and adolescents with disabilities. Studies of ways to enhance communication and other collaboration should focus on the principal organizational aspects of such arrangements, their effects on patient care processes and outcomes, and costs and efficiencies. These studies should also explore methods

of program collaboration and the tradeoffs (clinical, developmental, and health services) of providing different services in different settings. How have different organizational frameworks—for example, organized multispecialty programs or the use of multidisciplinary teams—succeeded in or failed to obtain needed care for children with disabilities?

Additional research areas include the roles of families and the relationships of these roles to primary and subspecialty services. How can we improve the communication among clinicians and between clinicians and families? We should clarify what services families provide for children and adolescents with disabilities and how much they cost in time and money; how families interact with other aspects of the care delivery system; the out-of-pocket costs of different family care arrangements; and the cost of families' lost opportunities. How do children's disabilities affect the families' structure and participation in the workforce and other community activities?

Comprehensive studies of expenditures and utilization, taking into account the various sources of payment, are clearly needed, as is how different financing arrangements affect the organization of services. How do the different incentives for patients and providers affect access to and use of services for children with disabilities, including methods of adjusting payment for risk? To determine benefit packages, plans need substantial information about costs and pricing. The recent Institute of Medicine report *Crossing the Quality Chasm* (2001b) includes in its recommendations the idea of basing care on continuous healing relationships between clinicians and patients. The report offers only a few recommendations about financing these relationships, however. Research on financing should examine incentives to support the longitudinal view of a person rather than short-term episodic financing. As noted earlier, the databases for assessing children's health are sparse. One improvement would be making the Medicaid claims files consistent, similar to the Medicare claims data systems used for research on the care of elderly populations.

Research on quality and outcomes should focus on developmentally appropriate measures of health status and quality of life. Although recent work by the Foundation for Accountability has led to the creation of a quality measure for children with chronic conditions to be used in the assessment of the National Committee for Quality Assurance health plan (Bethell, Read, Stein, et al. 2002), much more work in this area is needed. Almost no attention has been paid to safety in the care of children and

adolescents with disabilities. Research should determine the steps in care at which errors can occur and examine ways to minimize errors and improve safety.

These are only a few suggestions for the research needed to improve outcomes for young people with disabilities. Attention to these issues can provide a much stronger base on which to determine the necessary services, recognizing the changes in disability among younger populations.

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