Cerebral palsy and aging

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Cerebral palsy (CP), the most common major disabling motor disorder of childhood, is frequently thought of as a condition that affects only children. Deaths in children with CP, never common, have in recent years become very rare, unless the child is very severely and multiply disabled. Thus, virtually all children assigned the diagnosis of CP will survive into adulthood. Attention to the adult with CP has been sparse, and the evolution of the motor disorder as the individual moves through adolescence, young adulthood, middle age, and old age is not well understood. Nor do we know what happens to other functional domains, such as communication and eating behavior, in adults with CP. Although the brain injury that initially causes CP by definition does not progressively worsen through the lifetime, the effects of CP manifest differently throughout the lifespan. The aging process must inevitably interact with the motor disorder, but we lack systematic, large-scale follow-up studies of children with CP into adulthood and through adulthood with thorough assessments performed over time.

In this paper we summarize what is known of the epidemiology of CP throughout the lifespan, beginning with mortality and life expectancy, then survey what is known of functioning, ability, and quality of life of adults with CP. We conclude by describing a framework for future research on CP and aging that is built around the World Health Organization's International Classification of Functioning, Disability, and Health (ICF) and suggest specific tools and approaches for conducting that research in a sound manner.

LIST OF ABBREVIATIONS

AAC	Augmentative and alternative communication
HRQoL	Health-related quality of life
ICF	International Classification of Functioning, Disability, and Health
MACS	Manual Ability Classification System for children with CP
NHP	Nottingham Health Profile
OAEs	Otoacoustic emissions
QoL	Quality of life

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It is encouraging to note that two recent workshops in the USA supported by the Cerebral Palsy International Research and Education Foundation (formerly the United Cerebral Palsy Research and Education Foundation) have addressed issues of prevention and treatment of functional abilities in adults with CP, including the Arnold Werner* Workshop in May 2008 at Michigan State University and the meeting in conjunction with the American Academy for Cerebral Palsy and Developmental Medicine in September 2008 in Atlanta. Continued recognition of the research and medical needs of adults with CP will hopefully spur new and rigorous longitudinal studies of functional abilities that can inform preventive and management approaches to aging with CP.

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CONFLICTS OF INTEREST

The authors declare no conflicts of interest.

*Arnold Werner MD (1939–2007), an outstanding psychiatrist and a professor at Michigan State University, who had spastic diplegia that worsened considerably in his sixties, did much to raise national consciousness in the US on the importance of addressing CP in adult life.

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Cerebral palsy (CP) is often seen as a disorder involving children only. But children with CP nearly always grow up to become adults with CP, and with continuing improvements in survival, it has become increasingly important to plan appropriate service provision for such adults. Our current knowledge base for such planning is woefully thin. Critical questions that need answering include the following: (1) What do we know about the evolution of motor disorders in CP in adult life? (2) What happens to associated dysfunctions, such as problems in communication and in eating, as children with CP become adults? (3) What is quality of life for adults with CP? (4) What are the key research needs in this area, and how should they be addressed?

In this paper, we address these questions on the basis of the existing literature and recommend research approaches to fill in the many gaps.

PREVALENCE OF CP

CP has recently been defined as 'a group of permanent disorders of movement and posture causing activity limitation that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication and behavior, by epilepsy, and by secondary musculoskeletal disorders.'[1] Understanding the population of adults currently living with CP must begin with an examination of CP prevalence over the past decades.

A comprehensive 2006 survey of CP registries across the world revealed a fairly steady, if slightly increasing, prevalence rate of approximately two cases of CP per 1000 live births from the 1950s to present.[2] Until recently, the only population-based CP registry data in the USA were from the metropolitan Atlanta region, and they confirmed that the prevalence rate, which ranged from 1.7 per 1000 1-year survivors in 1975 to 2.0 in 1991, was in the same range as the national survey.[3] More recently, however, a higher prevalence of CP – three to four cases per 1000 school-age children – has been reported from three populations in the USA.[4] Additional research will be needed to see whether this finding represents a trend, an unusual outlier finding, or the use of a denominator that might create a prevalence rate influenced by the movement of the families of children with CP to areas with more sophisticated medical care facilities. It is clear, however, that the worldwide prevalence of CP in developed countries is not substantially declining, notwithstanding the many changes in the management of pregnancy and labor of recent decades. Health-care providers will therefore need to address the needs of adults with CP as today's children age into adulthood.[5]

CP MORTALITY AND LIFE EXPECTANCY

Two periods of the life span need consideration when describing mortality among adults with CP. The first period is survival through infancy and childhood into adulthood; the second is survival in adulthood. Mortality in CP is highly concentrated in infancy, but such deaths are difficult to attribute firmly to CP because they generally occur in infants with severe brain injury who are too young to be diagnosed with CP but are presumed to be at very high risk for it.

Mortality patterns from childhood on have been well described in several studies. Hutton[6] reported that the number and severity of CP-associated disabilities (relating to ambulation, manual dexterity, intellect, and vision) have a marked impact on survival to adulthood. Among children with CP studied in the UK, 99% with CP and no severe impairment survived to age 30, and 95% with just one severe disability survived. Survival was substantially lower in children with additional disabilities. Only 78% of children with two disabilities were likely to reach adulthood, and just 59% of those with three disabilities survived. Among the few children with four disabilities, only 33% survived to age 30.

California data are somewhat more positive. Data collected from persons with CP in that state showed that if CP was not severe, 98.2% of children aged 4 to 14 survived 20 years (1983–2002); if the CP was described as severe, 85% survived that period of time.[7] There was improvement in survival over time; in that same data set, mortality among the most impaired children improved at an average rate of 3.4% per year. With relatively high survival rates for those with minor impairment and increasingly better survival for the most impaired, one can conclude that the population of adults with CP will grow to include a larger number of more severely handicapped persons in the coming decades.

Survival in adults with CP is also quite good, though still lower than in the general population. Among a cohort of individuals with CP born between 1940 and 1960 in Bristol, UK, Hemming et al.[8] reported that 86% of those alive at age 20 survived to age 50. This compares to a survival rate of 96% during that same 20-year period in the general UK population. The relative risk of death was higher at all ages for adults with CP compared with age- and birth cohort–matched adults, but it decreased steadily with age. After age 50, women with CP experienced a slight excess of risk of dying compared to the general population, but men with CP did not. Men with CP born between 1940 and 1950 experienced higher mortality than women with CP, but no sex gap was observed for individuals born from 1950 to 1960. The factor most associated with increased mortality was intellectual disability; nonetheless, adults

with CP who had no intellectual impairment were still at somewhat greater risk of death than the general population. Causes of death also differed: adults with CP were more likely to die of respiratory diseases, but less likely to die of injuries and accidents, than the general population. Sex was not related to CP mortality in the California database described above.

FUNCTIONAL CHANGES IN ADULTS WITH CP

Changes in motor function

Anecdotal reports from adults with CP often include symptoms perceived as premature aging, in some cases starting in the twenties. The New York State Developmental Disabilities Planning Council identified issues in health and in physical, social, and psychological functioning in adults with CP.[9] They noted that early onset of musculoskeletal complaints was particularly prominent in persons with CP. Possible causes of decreased function and mobility mentioned were changes in muscle flexibility, strength, and endurance; increased spasticity; arthritis; falls and fractures; pain; and fatigue. Limited weight bearing, medications (especially for epilepsy), inadequate nutrition, and other causes were considered potential risks for the early development of osteoporosis in these individuals. The authors suggested that early interventions, surgery, exercise, and assistive devices might stem declines in function.

Declines in function may actually start earlier than adulthood. Gross motor function abilities in children with CP have been measured with the Gross Motor Function Measure (GMFM) to show changes over time. Rosenbaum et al.[10] created longitudinal curves from sequential GMFM measurements in 657 children ages 1 to 13, representing the spectrum of CP severity levels categorized by the Gross Motor Function Classification System (GMFCS). These curves showed that children in all severity levels achieved most of their potential function early; by age 7, function generally began to level off.

The only large study of motor functioning that is truly longitudinal comes from the McMaster group that developed the GMFM.[11] A subset of 229 of the children in the Rosenbaum et al. study described above participated in additional longitudinal GMFM assessments up to age 21. After combining measurements from all ages, the researchers analyzed the data by contrasting models for each severity level that incorporated no functional loss or a peak and decline in function. Among children in the most severe levels of disability (as estimated by the GMFCS), these curves estimated declines in function into adulthood. Possible explanations proposed for the motor decline were increased body size, decreased activity, and changes in spinal alignment.

All other studies of this subject have been cross-sectional, with retrospective assessment of prior functional ability. Murphy et al.[12] examined a convenience sample of 101 adults with CP (ages 19–74) living in the community in California. The participants had a variety of CP subtypes and a range of disability. Of 67 participants using a wheelchair at the time of the survey, 26 (40%) had previously been ambulatory and the majority stopped walking in young adulthood: 12 of them before age 21; six between ages 21 and 28; and eight between ages 38 and 68. Andersson and Mattsson[13] surveyed 363 Swedish adults with CP of varying subtypes, but excluding individuals with 'learning disabilities' (presumably thus excluding participants with cognitive impairment). Among 221 respondents (age 20–58), 75% had ever walked and 9% had once walked but were no longer able to. More than a third of the sample (35%) reported decreased walking ability over time, citing knee and balance problems, increased spasticity, and lack of physical training. Half of study participants who had stopped walking (10 of 20) were under the age of 14, and another eight were under the age of 35 when they stopped. Noteworthy is that 19% of ambulatory CP participants reported *improved* walking ability. The authors note the absence of published studies on exercise and outcome measures in adults with CP, resulting in a lack of guidelines about whether physical training would be worthwhile in preventing decline in ambulation.

Sandstrom et al.[14] in another Swedish study, conducted functional assessments of 48 adults (mean age 32) with CP, nine of whom had cognitive impairment. One-third of these participants had declined in motor function, a conclusion based on comparing GMFCS levels from adolescence to adulthood. Authors of a Norwegian survey[15] of 406 adults with CP 18 years and older without cognitive disability reported that 75% walked at the time of the survey, but that nearly 50% reported declines in walking ability, most often before their mid-thirties, while 10% had stopped walking entirely. On the other hand, 27% reported improvement in walking, usually before the mid-twenties.

Bottos et al.[16] assessed 72 adults ages 19 to 65 (86% under age 40) followed through pediatric rehabilitation units in Italy. They observed that the effects of CP evolve in adulthood, with some loss in ambulation and deterioration in walking distances. Among the 72% of the sample who had ever walked, 24% had stopped walking, mainly before age 40. The California Developmental Disabilities database also provides evidence of declines in ambulation, as well as loss in dressing skills, especially in older adults.[17] Ando and Uedo[18] found functional deterioration in 35% of 686 Japanese participants with CP surveyed while working in community workshops. The authors cite both

intrinsic and environmental factors as possible explanations. A subsequent survey found an additional 7% experiencing further declines over a 5-year follow-up period.

Communication functioning

Little is known about speech, language, and hearing in the adult with CP. However, children with CP can have speech, language, and hearing impairments that cannot be remediated and thus persist into adulthood. In addition, one or more of these areas (e.g. hearing) may decline as part of the aging process or because of comorbidities such as acquired neurological conditions. Speech sound errors with the primary disorder of dysarthria[19–21] are the most common communication disorders in CP. Dysarthria is an umbrella term for speech disorders caused by damaged central and/or peripheral motor-sensory loops that interfere with one or more component of the speech-production system (i.e. respiration, phonation, resonance, articulation). Language skills can be described on the basis of what the adult with CP understands (i.e. receptive language skills) and of what the adult says (i.e. expressive language skills). Hearing assessment using pure tone audiometry or physiological tools[22] such as otoacoustic emissions (OAEs) or auditory brainstem response are the most commonly reported communication measures in CP studies. However, several CP surveillance systems use 40 or 70dB hearing loss cut points to describe hearing loss, instead of 20dB, as is more commonly used in audiological research and practice, which results in underreporting of milder hearing difficulties in individuals with CP.

Eating and swallowing

The act of eating can be thought of as a timeline of motor actions: getting and keeping food and drink into the mouth, oral preparation, oral transport, pharyngeal transport, and/or esophageal transport.[23] People with CP may have trouble with one or more of these motor actions, which may make getting adequate nutrition and hydration difficult. Some people with CP use assistive technology, including tube feeding, to eat. Eating and swallowing impairments may develop or worsen as the adult with CP ages. Some clinical anecdotes suggest that changes in eating and swallowing can happen rapidly, which may mean that adults, caregivers, and professionals should monitor for early symptoms of eating difficulties (MS Workinger, personal communication).

QUALITY OF LIFE IN ADULTS WITH CP

Quality of life (QoL) and health-related quality of life (HRQoL) among children and adolescents with CP is often assessed; however, research by Svien et al.[24] and an updated literature search by the authors of this paper found no similar measurements for older adults with CP. To better manage CP symptoms and preventive care, and plan for social and work roles, adults need reliable information on age-related changes in both functioning and quality of life. An important part of the systematic study of the natural history of CP, addressed below, is developing measures to assess overall quality of life in older adults with CP.

Bjornson et al.[25] note that terms such as *health status*, *functional status*, *well-being*, *quality of life* and *health-related quality of life* are used often interchangeably in the QoL literature, often under umbrella term 'quality of life.'

Measuring quality of life

Two types of quality of life measurements can be found in the literature: QoL and HRQoL. Although QoL is sometimes used as an overall term for both QoL and HRQoL, it is also used as a measure distinct from HRQoL. QoL refers to the 'notion of holistic well-being,'[26] and it 'includes aspects of the social and physical environment that may or may not be affected by health or a treatment.'[25] HRQoL emphasizes 'health-related components judged to be associated with life satisfaction' [26] and focuses on the aspects of life and activity that are influenced by health conditions or services.[25]

Physical, mental, and social well-being and life satisfaction are the most common domains in QoL and HRQoL, but the measures differ conceptually and operationally. Both measures have three versions: a general version, a specific version, and a version that integrates the general and the specific. General versions are applied to the general population to capture physical, psychological, and social well-being, and to specific demographic groups such as those characterized by race, sex, and age. Specific versions of QoL and HRQoL are designed for populations with specific conditions, whether these are socioeconomic circumstances or diseases. A special version of HRQoL, targeted to the clinical condition under study, is particularly helpful to evaluate clinical trials and medical treatments. Integration of generic and specific versions in measures of QoL and HRQoL has become the norm in studies of people with disabilities, including children and adolescents with CP. [27,28]

Table I summarizes domains included in measurements of QoL and HRQoL or health status in several studies on children and adolescents with CP. Domains in the general versions of QoL instruments include emotional reactions, energy, pain, physical mobility, vitality, general and mental health, physical and social functioning, and emotional and physical roles. Specific versions have also included mobility, self-care, anxiety and depression, and usual activities.

Table 1: Measures of quality of life (QoL) and health related [HRQoL] in studies of children and adolescents with cerebral palsy

Studies	QoL	HRQoL or Health Status
Spilker[40]	1. Physical status and functional abilities	
	2. Psychological status and well being	
	3. Social interactions	
	4. Economic and/or vocational status	
	5. Spiritual/religious status	
Schipper et al.[41]		1. Physical and occupational function
		2. Psychological function
		3. Social interaction
		4. Somatic sensation
Rosenbaum et al.[26]	1. Physical well-being	1. Self-care
	2. Mental well-being	2. Mobility
	3. Social well-being	3. Communication
Bjornson et al.[25]	1. Social environment	
	2. Physical environment	
CP QOL-Child[42]	1. Physical well-being	
	2. Social well-being	
	3. Emotional well-being	
	4. School	
	5. Access to services	
	6. Acceptance by others	

QoL in Adults with CP

Little is known about how the experience of aging, perception of health, and QoL for people with CP differs from the aging process in individuals without disabilities and in adults with other disabilities. In a convenience sample of 20 adults with CP, loneliness as measured by the UCLA Loneliness Scale was higher for people with CP than for people without known disabilities.[29] In follow-up qualitative interviews with seven of CP adults in this study, the importance of communication and social networks in lessening loneliness emerged. While these themes are also found in the general aging literature, some adults with CP may face additional barriers, with communication disorders and community non-acceptance limiting their participation in the community. [30]

Recent developments on measurements of QoL and HRQoL for children and adolescents with CP have emphasized the distinctiveness of QoL and HRQoL measures and their distinct applications.[25,26,31] Although we found no studies with measures of either QoL or HRQoL for people aging with CP, instruments for assessing the QoL in an older population could provide guidelines for creating such measures. A review article on QoL in older people by Haywood et al.[32] has listed 15 QoL instruments categorized in two versions; the generic and specific. Eleven of them are health profiles (generic version) and four are utility measures (specific version). Among these measures, the Short Form 36-Item Health Survey Questionnaire (SF-36), the EuroQol (EQ-5D), and the Nottingham Health Profile (NHP) tested well for reliability, validity, and responsiveness to the construct compared with other instruments.[32]

APPROACHES TO FUTURE RESEARCH

Conceptualizing disability as a common human experience

The International Classification of Functioning, Disability and Health (ICF) from the World Health Organization[33] provides those interested in CP a paradigm shift from a purely medical to a broader biopsychosocial view of health and disabilities.[34] The ICF's biopyschosocial framework describes health and disability from the interactions among: (1) Anatomy and physiology (termed 'body structure and function'); (2) Daily activities; (3) Participation in home, school, work, and community activities; (4) Environmental factors, including physical, attitudinal, policy, and familial issues; and (5) Personal factors (e.g. age, sex, motivation, and preferences).

These interactions suggest that prevention, assessment, and intervention efforts in any of these five areas are likely to affect the other areas and contribute to a person's health. Disability occurs when one or more of these areas are limited. QoL sums up many of these dimensions but the ICF adds the additional subjective assessment of the individual.

CP in adults as related to the ICF domains of activity and function

Activity and participation levels

Although describing body structure and function impairments in CP is useful, exploring other factors, such as activity limitations and participation restrictions, may be more relevant to the daily lives of adults with CP. The ICF also distinguishes between capacity (i.e. what an individual can do, often in an ideal situation) and performance (i.e. what an individual does do in real-life situations). Mobility, handling objects, communicating, eating/drinking are a few ICF activity and participation categories which may be challenging for some individuals with CP.[1]

Mobility describes how people move within their multiple environments of home, school, work, and community. Adults with CP may require or choose to use assistive technology for mobility, such as walkers and powered wheelchairs, for a variety of reasons, such as a desire to conserve energy for other activities or to increase efficiency or speed.

Handling objects describes how people use their hands to accomplish many daily tasks. Many daily activities, including dressing, eating, writing, driving, are accomplished by using one or both hands. Assistive technology can help a person perform these tasks as well as reduce energy consumption.

Communication is the process where people send and receive messages, often by rapidly alternating sending and receiving. Communication relies on the underlying speech, language, and hearing skills of individuals. Communication problems in individuals with CP can limit social interactions, educational attainment, and employment opportunities.[23] Assistive technology including the use of augmentative and alternative communication (AAC) and hearing aids may improve communication performance.

Eating and drinking: Little has been written about activity and participation limitations that could result from difficulties with eating. For example, some individuals with uncoordinated eating skills may not wish to have familiar and/or unfamiliar dining partners watch them eat or drink. Some adults with CP may need assistance in eating from another person. Thus eating patterns may limit an important daily activity that is often shared with other people. As dining together is an important social ritual in family, school, employment, and community settings, a person with eating difficulties may be socially isolated.

Participation patterns and preferences of adults with CP need further research and should consider multiple life situations including home, school, work, and other community settings. A study of 101 adults with CP found that 53% were competitively employed and 67% lived independently.[35] An interaction was noted where nearly all who were competitively employed lived in their own home. In an on-line focus group of eight adults with CP who used power wheelchairs and AAC, the adults identified six benefits, nine barriers, and six supports to participation in leisure activities.[36] The benefits, barriers, and supports could be captured in one or more of the ICF levels: body structure/function (e.g. improved physical health), activity (e.g. communication), participation (e.g. restaurant dining), environment (e.g. societal acceptance, transportation) and personal factors (e.g. fear to try). Researchers from multiple disciplines need to partner with adults who have CP to understand the complex interactions among body structure and function, activity, participation, and contextual environmental and personal factors.

Tools available to describe status within these domains

Describing the extent of any problems at a body structure/function, activity, and/or participation level would be

Table II: Classification systems for functioning in cerebral palsy

Level	GMFCS (Mobility)	MACS (Handling objects)
I	Walks without limitations.	Handles objects easily and successfully.
II	Walks with limitations.	Handles most objects but with somewhat reduced quality and/or spread of achievement.
III	Walks using a hand-held mobility device.	Handles objects with difficulty; needs help to prepare and/or modify activities.
IV	Self-mobility with limitations; May use powered mobility.	Handles a limited selection of easily managed objects in adapted situations.
V	Transported in a manual wheelchair.	Does not handle objects and has severely limited ability to perform even simple actions.

GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System.

an important first step in better understanding and intervening in communication and eating problems of individuals with CP. Many tools that measure body structure and function are currently in use in the field of CP. Studies are needed to use these tools to measure these areas throughout the life span.

Classifying functional abilities and limitations in the domains critical to the daily needs of people with CP can assist in needs assessment for the adult and, in the process, improve our understanding of individuals with CP throughout their lives. With the creation of the Gross Motor Function Classification System (GMFCS),[37] the Manual Ability Classification System (MACS),[38] and the Communication Function Classification System (currently in development), the important areas of mobility, handling objects, and communication respectively are classified at an ICF activity and participation level.[39] (See Table II for a description of classification levels). The three systems are designed to classify performance (i.e. what a person does in daily life) and provide a valid, reliable tool for clinical and research activities. Eating does not yet have a similar classification system. Tools suitable to capture the wide range of adult participants are also needed.

Quality of Life

Our literature review suggests the value of both the QoL and HRQoL measures for people aging with CP. We need to select domains that are common to both the CP and the aging literature and to conceptualize measures of QoL and HRQoL for people aging with CP. Svien et al.'s [24] work indicated the usefulness of using the World Health Organization's ICF checklist for constructing measurements of QoL for adults with CP. In addition, we recommend special attention to the following domains particularly relevant to aging with CP: pain, fatigue, mobility issues, and comorbidities. These domains should be included in any newly developed measures, which should then be tested for reliability, validity, and responsiveness.

Future research should be colored by ICF thinking

Adults with CP have many questions about how the aging process applies to their lives. Does any aspect of CP make them more at risk for secondary or tertiary conditions or to develop them earlier in life? Can professionals offer guidance to minimize risks? How can research provide better recommendations and suggestions that will enable the adult with CP to enjoy a long and healthy life? Besides body structure/function, activities, and participation, what environmental and personal factors facilitate or hinder adults with CP striving to achieve their life goals? Answering these questions can be facilitated by adoption of the biopsychosocial ICF framework, from the involvement of multidisciplinary research teams that include broad expertise and by including input from adults with CP and their significant others.

CONCLUSION

The high prevalence of CP, coupled with the high percentage of people with CP now surviving into adulthood, warrants further research into how CP changes over the lifetime. For this research to be fully informative to practitioners, families, and persons with CP, it should be conducted in an epidemiologically rigorous manner. The lack of evidence-based therapy in CP across the life span complicates attempts to assist both children and adults with CP.

Preserving health and mobility in adults with CP is of great significance in employment, independence, and both health-related and general quality of life. Research is needed in which adult CP is categorized by a standard typology of CP and GMFCS severity levels, with independent, longitudinal assessments of standardized outcome measures from childhood through adulthood. Careful assessment of exposures such as early interventions and treatment, exercise, physiotherapy, and environmental factors may help us to learn how to forestall or prevent the functional declines evident in these studies that often start in the late teens and twenties, just as adulthood is beginning.

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