The Participation of Young Adults with Mild Intellectual Disabilities and Cerebral Palsy in the Decision to Receive Botox Treatment for Muscle Spasticity

A DISSERTATION
SUBMITTED TO THE FACULTY OF THE GRADUATE SCHOOL
OF THE UNIVERSITY OF MINNESOTA
BY

John Leland Belew

IN PARTIAL FULFILLMENT OF THE REQUIREMENTS
FOR THE DEGREE OF
DOCTOR OF PHILOSOPHY

Barbara Leonard, PhD, RN

April 2010

Acknowledgements

I first must recognize that without the enthusiastic involvement of the study participants, and the trust they placed in me as an investigator, there would be no dissertation. Barbara Leonard has provided valuable guidance and encouragement for this project over the years since I first presented this topic as the focus of my dissertation in 2004. Ann Garwick offered me many opportunities to learn about qualitative methods and has been a valued mentor. Diane Rose's audit of the conceptual framework was critically important. Conversations with Rick Cardenas at key junctures were invaluable. Gillette Lifetime Clinic staff were instrumental in the successful recruitment of the participants. Chris Milbrath and Karen Brill were generous in their support of my academic work. Thanks to Nancy for supporting my evolution into a nurse researcher, and to Jasper and EmmaLee, for learning to ask at an early age, "What's a dissertation?" Barbara and Helen have had ongoing enthusiasm about the progress of this project. And Margaret provided preliminary feedback on the conceptual framework at a key time.

Research expenses were paid by a Special Olympics Healthy Athletes Health Professions Student grant. The grant review committee and staff made valuable comments that contributed to the project. This project was also supported in part by the Center for Children with Special Health Care Needs grant number T80-MC00010, from the Maternal and Child Health Bureau (MCHB), Health Resources and Services Administration, Department of Health and Human Services.

Abstract

Background: While a large proportion of youth with intellectual disabilities (ID) live with chronic health conditions, it is unlikely that these individuals will be prepared to participate in health-related decisions. **Purpose**: This study examined the phenomenon of health-related decision-making among young adults with cerebral palsy and mild or borderline ID. The specific clinical context for the study was the decision to receive intramuscular injections of botulinum toxin type A (Botox) for skeletal muscle spasticity. **Methods**: A multicase methodology was used, based on a narrative approach. Data were gathered during semi-structured interviews with young adults and their parent guardians. Participants described their experiences related to the decision-making process. **Results**: Preliminary findings included a conceptual framework of the decision-making process based on case reports and case narratives. The four main concepts of the conceptual framework were *goals*, *information*, *relationships*, and *deliberation*. Thematic analysis was conducted using cross-case comparisons; data from field notes were also integrated. The main themes were *agency*; *communication and cognition*; *information gathered independently*; and *relationships*. The themes were further developed by synthesizing information from current literature. **Conclusions**: Participants described varying degrees of explicit participation of the young adult making the decision. Communication patterns were a significant factor in making decisions. Youth were supported in various ways with communication and deliberation. Participants considered peers as an important source of information about Botox. The young adults, their parents and their physicians weren't the only people involved in the decision-making process.

Table of Contents

List of Tables

List of Figures

Introduction

As adolescents make the transition to adulthood it is expected that they will become autonomous in all aspects of life. Young adults with mild intellectual disabilities (ID) face myriad challenges when they seek self-determination in their lives. While a large proportion of youth with ID live with chronic health conditions, it is unlikely that these individuals will be prepared to negotiate health-related decisions. This study examined the phenomenon of health-related decision-making among young adults with mild ID and cerebral palsy. The specific clinical context for the study is the decision to receive intramuscular injections of botulinum toxin type A (Botox) for skeletal muscle spasticity. Data were gathered during interviews with young adults and their parent guardians (and in one case, only a legally autonomous young adult).

Research Problem

Individuals with ID constitute a small but significant portion of the population. These individuals are at greater risk for health impairments. Yet they rarely participate in decisions about their own healthcare. Some of these individuals are legally autonomous, while many have guardians who are responsible for making health-related decisions on their behalf. Parents are often their guardians during early adulthood. As these individuals grow older, a sibling or other relative might assume guardianship, but it is increasingly likely that they will have guardians who aren't familiar with them (e.g. public or private guardians).

Individuals with ID and health.

Using data gathered by the Social Security Administration in 1993, it has been estimated that the prevalence of intellectual disabilities (ID) in the United States is 0.76% (Nehring & Poindexter, 2005), though other estimates range as high as 2% (Larson et al., 2001; Oliver et al., 2003).

Compared with their peers in the general population, individuals with ID are more likely to have diminished health and they are more likely to face barriers in accessing health services (USDHHS, 2002; Walsh, 2008). Compromised health can be a burden for any person. However, for individuals with ID, diminished health status can also contribute to the need for a more restrictive living situation, due to the need for more assistance with health care needs (Marks & Heller, 2003). These individuals are more likely than those without ID to experience compromised health as a result of one or more of the following conditions: seizure disorders, mental health disorders, obesity, thyroid disease, oral health issues, skin disorders, sensory impairments, constipation and fracture risk (Bohmer, Taminiau, Klinkenberg-Knol, & Meuwissen, 2001; K. Fisher, 2004; Jansen, Krol, Groothoff, & Post, 2004; Kozma & Mason, 2003; Patja, Molsa, & Iivanainen, 2001; Sullivan et al., 2006; Sutherland, Couch, & Iacono, 2002; Turk, Geremski, Rosenbaum, & Weber, 1997). Sometimes these risks are directly related to the underlying condition that causes the ID. For example, the original insult to the brain that caused an individual's cerebral palsy can also cause a seizure disorder.

This study targets young adults with ID who have cerebral palsy (CP). CP is the most common cause of physical disability among children, with a prevalence of 3.3 per 1,000 (Yeargin-Allsopp et al., 2008).

> Cerebral palsy describes a group of permanent disorders of the development 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 cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems (Bax et al., 2005).

The longevity of adults with CP has steadily increased in recent decades (Strauss, Shavelle, Reynolds, Rosenbloom, & Day, 2007), and these adults are at increased risk for musculoskeletal deformities, dysphagia and gastrointestinal disease (Liptak, 2008; Reilly & Morgan, 2008).

Guardianship trends.

Even though there are no estimates of the proportion of adults with mild ID who are legally autonomous as compared with those who have proxy decision-makers, some related trends shed light on this topic. Recent legislation in the UK (Mental Capacity Act, 2005) requires that individuals must be presumed competent to make legal decisions unless the person is demonstrated incapable, *after* making necessary accommodations for particular information needs (e.g. through the use of visual aids; (Keywood & Flynn, 2006). In the United States, when an individual with ID reaches the age of 18, she becomes legally autonomous, unless a court has adjudicated that she is incompetent; in this case, the court names a guardian.

Two trends coincide to pose a potential threat to the well-being and self-determination of adults with ID. First, it is quite common for the appointed guardians of adults with ID to be one or both of the young adult's parents. And adults with ID are likely to be living in the family home. The most recent reports estimate that 60% of adults with ID live in the family home (Braddock, Emerson, Felce, & Stancliffe, 2001; Fujiura, 1998). Australian researchers concluded that parents continued in their roles as primary caregivers for their adult children with ID due to a perceived shortage of satisfactory residential housing (Eley, Boyes, Young, Hegney, & Hegney, 2009). In a population-based study of family caregivers of adults with ID conducted in Ireland, researchers found that the average age for moving from the family home was 35 (Barron, McConkey, & Mulvany, 2006).

The second trend is that the life expectancy of individuals with ID has steadily increased in recent decades (McCallion & McCarron, 2004). Together these two trends illustrate a growing concern: as adults outlive their parents or move away from the family home, their parents will be replaced by guardians who are not well-acquainted with the communication patterns, personal histories, preferences and values of these adults with ID. While many adults with ID continue to live in the family home, adults with ID are increasingly likely to be living in group homes or assisted living settings (Braddock et al., 2001).

Perhaps this loss of the parent's involvement can be ameliorated if young adults with ID actively participate in health-related decisions. The young adult's participation can be supported and promoted beginning in early adolescence. Yet there is very little evidence that young adults with ID are being encouraged and educated in the abilities needed to make meaningful health-related decisions. Seltzer and Krause (2001) comment that "particularly glaring is the absence of research literature on the degree of choice, personal autonomy, and self-determination of… adults with MR/DD who live outside the formal residential system (p. 112).

Young adults and transition.

There is growing attention to the myriad issues associated with the transition of young adults from child-centered to adult-centered health care services (Betz, 2004; Kelly, Kratz, Bielski, & Rhinehart, 2002; Scal & Ireland, 2005). All adolescents and their parents face uncertainty as the youth moves from child-centered to adult-centered care. Youth who have substantial ongoing need for health services face a significantly more stressful transition. Individuals with ID and their families often face yet another level of uncertainty related to transition to adult health services. Health care professionals generally lack expertise in the health concerns common to the population of adults with ID, and they rarely know how to offer anticipatory guidance for these individuals (Betz & Redcay, 2003; B. Hudson, 2003). "Adult services appear to be fragmented and unprepared for young adults with developmental disabilities who are living longer lives and expecting to live as full citizens in society" (Rosenbaum & Stewart, 2007, p. 1081).

Study Aims

The aims of this multicase study were:

- To describe the decision-making process of young adults with cerebral palsy and mild or borderline intellectual disabilities considering Botox therapy for relief from muscle spasticity.
- To describe the roles of the young adult and their guardians in this decision.

Research Design Overview

This study uses multicase qualitative methods, drawing from methods developed by Stake (1995; 2006) and Yin (2003). The investigator adopted a narrative approach to the collection and analysis of data (McCance, McKenna, & Boore, 2001).

The investigator sought as much homogeneity as possible with regards to the clinical context of health-related decision-making. So the context selected for the study is the decision regarding the use of Botox for the treatment of muscle spasticity. Young adults with cerebral palsy (and their family guardians) were targeted because the investigator sought to explore the experiences of young adults who have had need of medical interventions through their entire lives. These extensive life experiences with healthcare and health-related decisions provide them with a perspective that was invaluable in providing insights during the interviews.

Joint interviews were conducted with the young adults and their family guardians. This strategy was chosen as part of the narrative approach. The investigator's premise was that decision-making processes are a joint construction of the involved individuals. Since the young adults and their family guardians were both key participants in this process, they were interviewed together so that they could jointly reconstruct the story of how the decision was made.

Investigator's Background and Assumptions

The investigator's interest in this topic originated in his experiences as a nurse working with children, adolescents and adults in a variety of settings. Over the past 16 years he has worked in hospitals, at a residential school for special education, and in residential facilities for adults with intellectual disabilities.

When undertaking a research project the investigator must be vigilant in attending to underlying assumptions that he brings to the study. At each stage in the development and conduct of the study, one's preconceptions influence the decisions made during an investigation. For this reason qualitative researchers often state whatever assumptions of which they are cognizant. This attempt to explicitly outline assumptions alerts readers to specific opinions of the investigator that might have influenced key decisions.

Self-determination.

The investigator has adopted the definition of self-determination as developed by Abery and Stancliffe (2003) in the Tripartite Ecological Model (TEM) of self-determination. The TEM is composed of three elements that, in combination, indicate the level of an individual's self-determination:

- Importance: How important is it to the individual that she has control over a specific aspect of her life?
- Desired control: How much control over that aspect of life does the person want to have?
- Exercised control: How much control over that aspect does the person actually have?

What is most striking about the TEM is that it describes that self-determination is optimal when a person controls those things that she finds important for her to control, at the degree of control that she prefers. This definition is distinct from other discussions of self-determination and autonomy which imply that *more control* indicates *more self-determination*. A concrete example of an application of the TEM model would be the manner in which a person controls her personal finances. While this individual may want to have access to a limited amount of discretionary funds, she may prefer to have minimal involvement in how the details of her overall finances are managed. An application of the TEM would reveal that to pressure her into being more involved in all aspects of controlling her personal finances would actually *reduce* her self-determination.

The investigator conducted this study while holding the opinion that it is desirable for young adults with ID to be self-determined. This assumption is significant because study participants might not share this premise, and these contrasting perspectives might influence interactions during the interviews. For example, if parents sensed that the investigator expected them to promote the youth's self-determination they could be tempted exaggerate or fabricate statements in the interest of social desirability, rather than come into conflict with the investigator's strong opinion that youth autonomy should be a priority.

Decision-making and risk.

The investigator assumed that health-related decisions are often sensitive and personal events, and that there is no simple or direct way to access an individual's experiences related to these decisions. The optimal context for this study would involve a decision where the risks involved are not extraordinary. If participants were asked to describe their process for deliberating on a decision that involved grave risks, they may be inclined to offer responses they consider to be socially acceptable. If the interviews are about a decision with relatively minimal risks, the participants may feel freer to be more candid about the process.

Defining health.

The investigator assumed that health-related decisions are similar to but distinct from other decisions. (The investigator's definition of *health* is stated below.) The distinction is based on a conceptualization of health as a dimension of life that permeates all aspects of life.

Significance

Current estimates are that between 1% and 2% of the population have intellectual disabilities (Larson et al., 2001; 2872 Nehring & Poindexter, 2005; Oliver et al., 2003). This means that in the US there are about three million people with ID. Implementing effective strategies for routine health promotion and disease prevention activities for these individuals can be particularly challenging, considering that information and interventions must often be modified to meet individuals' unique physical and cognitive needs. As has been described above, individuals with ID also have higher rates of morbidity than the general population. The complexities of health management for individuals with ID can be particularly daunting, yet these challenges can be ameliorated if the specific self-care abilities of these individuals are tapped. What is lacking is the expertise for recognizing and cultivating individuals' abilities.

Although self-determination is widely acknowledged as being of fundamental importance in health care, for those with ID self-determination is commonly quite restricted. In the present study the investigator explored the ways that adults with ID were involved a key aspect of self-determination: health-related decision-making.

Twenty-eight years ago a Presidential commission charged with making recommendations on the informed consent process in health care stated that clinicians are obligated to specifically determine individuals' capacity for medical decisions, rather than categorically discounting abilities, e.g. based on having ID:

> If people have been able to form their own values and goals, are free from manipulation, and are aware of information relevant to the decision at hand, the final aspect of self-determination is simply the awareness that the choice is their own to make. Although the reasons for a choice cannot always be defined, decisions are still autonomous if they reflect someone's own purposes rather than external causes unrelated to the person's "self." Consequently, the Commission's

> concept of health care decision-making includes informing patients of alternative courses of treatment and of the reasoning behind all recommendations. Self-determination involves more than choice; it also requires knowledge. (President's Commission, 1982, pp.46-47).

Twenty years later, a report of the Surgeon General stated that one goal (of six) for improving the health of individuals with ID was to "increase knowledge and understanding of health and mental retardation, ensuring that knowledge is made practical and easy to use" (USDHHS, 2002, p. 5). This report draws attention to the increasing importance of health care providers specifically addressing the needs and preferences of individuals with ID. In the ensuing years there has been very little research-based evidence guiding the implementation of these recommendations for adults with ID, especially in the US. Editors at the medical journal the Lancet (2009) recently stated that health care providers can pose a significant *barrier* to the independence of individuals with ID by not recognizing the strengths and capacities of these individuals.

The Health Issues Special Interest Group of the International Association for the Scientific Study of Intellectual Disabilities recently recommended, "persons with ID are capable of assuming greater control over their lives, and they deserve the opportunity to do so" (Scheepers et al., 2005, p. 253). "Self-determination has been identified both as a means of enabling people with intellectual disabilities to achieve valued life outcomes…and as a valued life outcome in and of itself" (Shogren, Wehmeyer, Reese, & O'Hara, 2006, p. 107). The current study contributes to the knowledge of how to support young adults with ID in becoming more self-determined with regards to health-related decisions.

Individuals with ID are categorically excluded from virtually all health sciences research (USDHHS, 2002) so very is little is known how these individuals are similar or different from their peers. Researchers have offered different rationales for this exclusion, although frequently this exclusion criterion is stated without further explanation. The most common reason presented for excluding individuals with any type of cognitive impairments is that the self-reports of these individuals are likely to be suspect. In other words, inclusion of individuals with likely impairments of reasoning would decrease the reliability and validity of self-reported data. But it must be pointed out that individuals with ID are also routinely excluded from studies that don't involve self-report. One is left to assume that the presence of a diagnosed cognitive impairment somehow adds unwanted variability in data and additional complexity to analysis.

Key Concepts and Definitions

Definitions of key concepts used in this dissertation are stated here.

Intellectual disabilities.

The term intellectual disability (ID) is generally equivalent to and currently preferred over *mental retardation* (MR). The term ID aligns with recent efforts to more carefully specify various disabilities, and the term MR has gained an increasingly derogatory association (Hahn & Marks, 2003). In 2007 the American Association on Mental Retardation changed its name to the Intellectual and Developmental Disabilities

Association, and other national organizations have made similar changes in their names (Prabhala, 2007). (The term MR will occasionally be used here in order to accurately report research findings from other studies that used this term.)

> The American Association on Mental Retardation has defined MR (and ID) as a substantial [limitation] in present functioning…characterized by significantly subaverage intellectual functioning, existing concurrently with related limitations in two or more of the following applicable adaptive skill areas: communication, self-care, home living, social skills, community use, self-direction, health and safety, functional academics, leisure, and work. Mental retardation manifests before age 18 (Leonard & Wen, 2002, p.119).

In the U.S., the term developmental disability (DD) is used to determine individuals' eligibility for public resources (K. Fisher, 2004). While most individuals with ID are considered to have DD, it is estimated that about half of those individuals with DD do not have ID (Larson et al., 2001). For example, many individuals with autism (and hence with DD) have normal or higher intelligence quotients (IQs). Literature regarding individuals with DD has been included in this study when it is applicable to individuals with ID. Another point of potential confusion is the term learning disability (LD). In the UK the term LD is frequently used as an equivalent to ID and MR. However, in the U.S. learning disability refers to individuals with average or higher IQ who have specific learning difficulties in specific functional areas, such as reading or math.

The population of individuals with ID is extremely heterogeneous. While cerebral palsy, Down syndrome, or metabolic syndromes can cause intellectual disabilities, these conditions have little else in common. In fact, no etiology for an intellectual disability will be identified for most individuals with ID (Arvio & Sillanpaa, 2003). While the etiologies of ID are diverse, individuals with ID as a group are likely to face similar challenges in society. An international workgroup has stated that individuals with ID deserve to be recognized as a distinct group within the greater population of individuals with disabilities: "Persons with ID should not be subsumed into a broad 'disability population definition'" (Scheepers et al., 2005, p. 250).

Supports.

We typically consider *independence* to mean the degree with which an individual is capable of functioning without any assistance. In recent years there has been growing recognition that, for individuals with a variety of disabilities, the concept of independence should be reframed. When accommodations are available, an individual's abilities to be independent can broaden. For example, a person who cannot walk independently can nevertheless be considered "independently mobile" if a power wheelchair is considered as an accommodation that is an integral component of the individual's functioning. These ideas regarding independence and accommodation are often conceptualized as *supports*. "Supports are resources and strategies that enhance human functioning" (Thompson 2009 p. 136). Examples of supports are a wheelchair and an electronic communication device. When a personal care attendant reads prices to an individual with ID while he is shopping, this activity is also a support.

Health.

A range of conceptual approaches to the definition of *health* have been offered in the literature (Fawcett, 1999; McDowell, 2006). The choice of definitions has profound implications on the scope of investigations characterized as being *health*-related. The following definition is used in this dissertation:

- *Health* refers to those aspects of physical or mental well-being that are amenable to services offered by health care professionals.

This is an admittedly constrained and arbitrary definition, and neglects many aspects of life that could easily be considered as being health-related. However, this definition serves to limit the scope of this investigation, including issues closely related to a medical context.

Terminology.

Several specialized terms and abbreviations are used in this dissertation:

- **ACD:** Augmentative communication device. Any type of technology used to facilitate verbal communication.
- **Baclofen pump**: This implanted device delivers the muscle relaxing medication baclofen (Lioresal) through a catheter to the spinal cord (intrathecally), providing relief from muscle spasticity.
- **Meaningful decision**: An individual comprehends aspects of the decision beyond simply declaring a choice (see Literature Review chapter).
- **PCA:** Personal care attendant. PCAs are professional staff that provide a variety of services for individuals, usually in the home.

- **Supports**: “Supports are resources and strategies that enhance human functioning” (Thompson 2009 p. 136). This term is widely used to describe a variety of ways assistance can provided for individuals with disabilities, minimizing obstacles to independent functioning.
- **Young adult; youth**. The term *youth* has been identified by advocacy organizations as a preferred term to describe adolescents and young adults (National Collaborative on Workforce and Disability, 2009). The term has value in that it doesn’t introduce artificial and arbitrary limits on age when grouping individuals who share many developmental goals. The terms *young adult* and *youth* are used interchangeably in the text to reduce awkward phrases.

Chapter Summary

There is a high prevalence of a variety of chronic health conditions among individuals with ID. Many young adults with mild ID have their parents as guardians. Individuals with ID are increasingly likely to survive their parents. These facts indicate a looming trend, where adults with ID who aren’t legally autonomous will have guardians who aren’t familiar with their individual values, preferences, and life history. It has been recommended that parents and professionals promote and support self-determination of adults with ID.

In this study the investigator focuses on a specific-health related decision, which is a consideration of Botox injections for the treatment of muscle spasticity. Using a multiple case methodology the investigator will describe the decision-making process and the roles of these young adults and their parents in this decision.

Literature Review

There has been scant research addressing the topic of the participation of adults with ID in health-related decisions. In this chapter the investigator has drawn from related literature to identify and evaluate current approaches to this topic.

Search Strategies

The investigator has found that the literature related to health issues of individuals with ID is scattered across a wide variety of journals. Inconsistencies in terminology related to this population pose a challenge to any exhaustive searches in this area of interest. For instance, the term *intellectual disability* has only recently been indexed as an equivalent term with the medical subject heading *mental retardation* in the Medline database; in 2009 the use of *intellectual disability* would not link to any keyword in Medline. And the term *learning disability*, used in the UK as an equivalent for *intellectual disability*, refers to a wholly distinct diagnosis in the US (i.e. those with average or higher than average intelligence and a specific deficit in reading, writing or mathematics).

The investigator used several strategies to identify literature pertinent to this study. In addition to a conventional keyword search of literature databases, the investigator searched journals and citation lists by hand, and newer electronic technologies were utilized.

The investigator first conducted an extensive database search seeking literature that addressed the key phenomena related to the research topic of this study. The investigator searched the MEDLINE, CINAHL, PsychInfo, ERIC, and Google Scholar literature databases. The following keywords were used in various combinations:

- Intellectual disability.
- Mental retardation.
- Learning disability (for literature from the UK).
- Developmental disability.
- Cerebral palsy.
- Down syndrome.
- Health; illness; chronic condition (added when searching the non-medical databases: PsycInfo, ERIC and Google Scholar).

The investigator used the Science Citation Index to identify literature that cited seminal articles. Reference lists of all relevant articles were hand searched for additional references. To remain current with emerging literature on any of these related topics, the investigator employed electronic tools. With RSS feeds and auto-alerts the investigator receives regular notification of literature as it is published or indexed in the literature databases.

Topics Addressed in Literature Review

The next section is a critical appraisal and synthesis of literature. The topics addressed in this review of the literature include:

- Conceptual approaches to an understanding the decision-making process (in the general population).

- The capacities of individuals with ID to participate in decision-making.
- The involvement of these individuals in decision-making.
- The role of health care providers in supporting IWID in decision-making.

Health-related decisions.

The investigator conducted an extensive review of the literature for conceptual analyses of the health-related decision-making processes. There is a surprisingly small body of literature addressing this topic. The main focus of this body of literature is the *locus of control* of the decision. That is, the literature primarily addresses the degree of patient autonomy exercised by patients during health-related decisions, as contrasted with the influence of the physician (Bodenheimer, Lorig, Holman, & Grumbach, 2002; Degner & Sloan, 1992; Degner, Sloan, & Venkatesh, 1997; Emanuel & Emanuel, 1992; Entwistle & Watt, 2006; Gore, Johnson, Caress, Woodcock, & Custovic, 2005; Graham & O'Connor, 2006; Kiesler & Auerbach, 2006; Makoul & Clayman, 2006; Mead & Bower, 2000; Murray, Charles, & Gafni, 2006; Murray, Pollack, White, & Lo, 2007; Say, Murtagh, & Thomson, 2006; Sheridan, Harris, Woolf, & Shared Decision-Making Workgroup of the U.S. Preventive Services Task Force, 2004; Swanson, Bastani, Rubenstein, Meredith, & Ford, 2007; Tyler & Horner, 2008; Young, Moffett, Jackson, & McNulty, 2006; Zoffmann, Harder, & Kirkevold, 2008).

Emanuel and Emanuel (1992) developed one of the most widely cited conceptual approaches to locus of control in health-related decision-making. These physicians identified four idealized models of the physician-patient relationship as it pertains to decision-making:

- The paternalistic model. The physician determines the course of medical care unilaterally, or attempts to persuade the patient to assent to this course of care.
- The informative model. Also known as the consumer model, the physician provides the patient with pertinent information, and the patient determines the course of care.
- The interpretive model. The physician counsels the patient to reflect on her values in light of the information surrounding the decision at hand.
- The deliberative model. The physician engages with the patient to better appreciate his values, and then the physician advocates a decision that she feels best represent these values.

Degner and colleagues have developed a method for identifying a patient's preferred degree of control in health-related decision-making (Degner et al., 1997). The Control Preference Scale has been used in both a clinical and a research context (Degner et al., 1997; O'Donnell & Hunskaar, 2007; Pyke-Grimm, Degner, Small, & Mueller, 1999). To plumb a patient's control preference, he is presented with five cards; on each card is a description and illustration representing a different level of preferred degree of patient control (relative to the degree of a physician's control) during health-related decision-making. Four of the levels closely match Emanuel and Emanuel's (1992) four models, with the addition of a collaborative role where the patient and doctor "share responsibility" (Degner, p. 23).

While the approaches of Degner and the Emanuels provide a framework for exploring patients' desired degree of autonomy within the context of the patient-physician relationship, other key elements of decisions are not accounted for in these approaches (Entwistle & Watt, 2006). For example, how do patients (or their legal proxies) negotiate the information about the decision? How do patients involve other important people in the deliberative process (e.g. spouses, parents, children)? These issues are developed in the current study and presented in the Results chapter.

Cognitive capacities of adults with ID.

This section is a discussion of the literature addressing the cognitive capacities of individuals with ID to participate in decisions. Wong, Gunn & Holland (1999) explored the issue of competency with regards to providing consent in a research context. They incorporated previous standards for competency and developed a schema of five "abilities," which are the abilities to:

1. Declare a choice.
2. Understand factual information.
3. Remember factual information.
4. Use good judgment in manipulating information.
5. Appreciate the context and consequences of the decision.

Building on this conceptual approach to competency, the investigator will use the term *meaningful decision*. A meaningful decision will refer to a choice was based on the second, third, fourth or fifth of Wong, Gunn & Holland's standards. In other words, a simple declaration of choice, unsubstantiated by additional validation of competency, will not be considered a meaningful decision.

Capacities for participation in decision-making and consent.

In investigations researchers have reported on the demonstrated abilities of adults with ID to make decisions, or to provide (research or medical treatment) consent. (Consent for medical treatment is closely related to consent for participation in research, as both situations involve an appraisal of personal risk and benefit. Consent will be treated as a special case of decision-making.)

Researchers in the US studied the capacity of adults with ID to understand and provide meaningful consent for three hypothetical health-related treatments (Cea & Fisher, 2003). Adults with mild ID (n=30) and moderate ID (n=30) were compared with adults without ID (n=30). Information was provided in sections. These researchers developed an instrument based on standards for determining capacity for consent similar to those developed by Wong et al. (1999; Applebaum & Roth, 1982). Approximately 30% of the individuals with mild ID were able to provide meaningful consent for any of the three scenarios. A noteworthy finding was that for one of the scenarios, on one of the test items, the adults *without* ID had a lower mean score than the adults with mild ID. This finding perhaps highlights the complexities of providing information and assessing abilities related to consent.

Members of this research team also studied capacity for meaningful consent for participation in research by comparing adults with mild ID (n=50), moderate ID (n=50) and freshman college students without ID (n=50; Fisher, Cea, Davidson, & Fried, 2006). The researchers developed an instrument to quantify individuals' abilities in appraising consent issues related to a hypothetical randomized clinical trial (the instrument's basic features are similar to that used in Cea & Fisher, 2003). Nearly half of the subjects with mild ID demonstrated an understanding of the concept of human subject protection in research. Most of the subjects with ID had scores similar to the control group with regards to an understanding of *research procedures*, an *appreciation of the medical disorder* represented in scenario, and an *appreciation of consequences* of research participation.

In a comparative study conducted in the UK, the decision-making capacities of adults from the general population were compared with adults with mild ID, schizophrenia, or dementia (Wong, Clare, Holland, Watson, & Gunn, 2000; n=82). The assessments for decision-making capacity were made in the context of routine phlebotomy procedures that the individuals were undergoing regardless of study participation. Capacity was determined using an ad hoc measure consisting of five elements testing subjects' comprehension of information. Subjects' responses were classified as to the method they used in communicating comprehension: some subjects paraphrased details of the phlebotomy procedures completely, some paraphrased sections (with prompting) and some demonstrated the procedure without using words. While the adults with ID (n = 20) demonstrated poorer decision-making abilities than the general population group (n = 20), 65% of the adults with mild ID (n=20) demonstrated a capacity for meaningful decision-making.

Researchers in the UK examined the capacity of individuals with mild or moderate ID to make a meaningful decision whether or not to consent for medical treatment (Arscott, Dagnan, & Kroese, 1999). In this study subjects were presented with three hypothetical vignettes involving medical treatment. The information was presented twice; the second time it was presented in smaller sections. Simple line drawings accompanied the textual information. Using an adapted instrument (the Ability to Consent Questionnaire: ACQ) that scores responses according to appropriateness to the given situations, the researchers found that 26 of the 40 subjects (65%) were capable of providing consent for at least one of three hypothetical scenarios. However, only five subjects (12.5%) were found to be capable of providing consent for all three scenarios. The study included an estimate of subjects' relative cognitive abilities (based on assessment of receptive vocabulary); no correlations between capacity for consent and cognitive testing scores were reported. Subjects' verbal and memory abilities were tested, and deficits in these areas were associated with reduced ability to participate meaningfully in the decision. Most of the subjects could participate to some degree in the decision, though very few met the challenges of meaningful decision-making across multiple scenarios.

Another research team in the UK conducted a randomized, controlled trial using a modified version of the ACQ (Dye, Hare, & Hendy, 2007). These researchers studied the benefits of two innovative strategies for aiding individuals with mild or moderate ID in comprehending consent for participation in research as subjects. For the control group consent information was read through twice. In one of the experimental groups subjects had consent information presented to them in smaller sections, with consent for each described portion solicited at the completion of each section. A second experimental group was presented with photographs intended to aid understanding of the consent information. There were no differences in subjects' capacity to make meaningful decisions regarding consent based on group assignment. Of 85 subjects five (8.5%) were assessed as being competent when asked to respond all seven of the questions in the ACQ. However, 53% of the subjects were assessed as able to comprehend the impact of the decision as well as the options available.

In these studies, individuals with mild ID did not perform as well as those without ID in studies involving testing of discrete skills associated with meaningful decision-making. Yet these studies help to highlight the complex nature of the idea of "capacity for decision-making." When competency is considered as either "absent" or "present," the evidence for competent participation in decision-making by individuals with mild ID is quite limited. Competency for meaningful participation in decision-making can be considered as a continuum; each individual has strengths and limitations that have bearing this ability (Harris, 2006).

Involvement in self-care activities.

If a broader array of abilities is considered, there is compelling evidence that many or most individuals with ID have the capacity to meaningfully participate in some aspects of decisions. What does the literature tell us about the actual participation of adults with ID in decisions? This literature is presented here, following a brief discussion of the participation of adults with ID in other self-care activities.

Participation in self-care.

It is expected that young adults with chronic health needs develop *self-care* abilities in anticipation of the emerging independence that typically comes with adulthood (Rosen et al., 2003). Self-care encompasses those activities an individual performs on behalf of herself with limited or no assistance. (Horan, Doran, & Timmins [2004a; 2004b] evaluated Orem's self-care deficit nursing theory for application to improved understanding of health management of individuals with ID and concluded there was a poor fit between this theory and the complexities of care needs of many individuals with ID.)

Participation in health-related decisions certainly is an aspect of self-care (Geenen, Powers, & Sells, 2003), yet the literature addressing self-care in individuals rarely explores decision-making as a component of self-care. Instead this literature focuses on specific self-care *skills*. (The term *skill* is often used to designate a technical ability that is acquired following intensive training, as contrasted with higher level, cognitive abilities.) Lunsky, Straiko & Armstrong (2003) found that following an eight-week health curriculum, US women with mild and moderate ID demonstrated significant improvements in health knowledge and coping strategies. In a qualitative study in the US researchers found that 11 of 12 of the adult participants with mild ID were able to describe "routine preventative procedures…e.g. weight, blood pressure" (Horrell, MacLean, & Conley, 2006, p. 243). Eight participants demonstrated basic knowledge about their own medications.

Decision-making is a key aspect of self-care, perhaps especially for those individuals with physical limitations who can't participate in many other self-care activities due to motor limitations. In effect, making decisions is the one aspect of health-related self-care that *anyone* with sufficient cognitive and communicative capacity can participate in, regardless of physical disabilities.

Participation in decision-making.

Three studies explored the participation of individuals with ID in decision-making. A team of Dutch researchers studied the phenomenon of decision-making in a group of individuals with all levels of ID residing in a residential facility (n=15: Vallenga, Grypdonck, Tan, Lendemeijer, & Boon, 2006). Using a multiple embedded case study design the researchers focused on the decision-making process involved in managing the individuals' risks for injuries related to seizures. For these individuals *protective measures* had to be balanced against a sense of an *acceptable risk*. For example, extremely restrictive equipment (protective measures) might protect against injuries from falls during seizures, yet using this equipment could dramatically affect the individual's quality of life. The researchers found that parents of younger residents were more involved in decisions than parents of older residents; they surmised that these differences were based on different generational assumptions about the degree that family involvement in decisions is appropriate. The researchers found that the residents were not provided with adequate information to aid them in determining a preference about injury prevention measures. The facility had not conducted assessments of residents' decision-making capacities.

In a large national study in the US (n = 2,398), Lakin et al. (2008) investigated choice making among adults with ID who resided in settings other than the family home. The investigators were primarily interested in whether differences in residential settings were associated with involvement in choices. Choice making was measured using an instrument that categorized choices as *everyday* (e.g. choices about clothing or bedtime) or *support-related* (e.g. choices about residential setting). Individuals receiving home and community-based services had more choice than those in larger residential facilities. Higher functioning individuals had more choice.

Prader-Willi syndrome (PWS) is a rare genetic syndrome where individuals are at high risk for life-threatening obesity. These individuals do not experience satiety and will tend to eat constantly to suppress a constant sense of hunger. Most individuals with PWS also have moderate to borderline ID, and behavior problems such as tantrums and skin picking are common. Cognitive limitations mean that issues of autonomy and control of access to food are of grave importance. Dutch researchers investigated the issue of strategies for handling the autonomy of adults with PWS (van Hooren, Widdershoven, Candel, van den Borne, & Curfs, 2006). The investigators presented parents and professional caregivers of individuals with PWS with hypothetical scenarios involving diet and behavioral issues. These subjects then selected interventional strategies that were conceptualized according to four styles drawn from Emanuel's and Emanuel's (1992) four models (described earlier in this review). Subjects were more likely to intervene according to a paternalistic approach in scenarios involving food as compared to scenarios involving behavior. Parents also tended toward a paternalistic approach more than did the professionals. The researchers noted their surprise at the apparent reluctance of all caregivers to consider a more collaborative approach to handling the admittedly formidable issue of eating behavior.

While there is evidence to indicate that individuals with ID might have the capacity to participate in at least some aspects of decisions, these three studies indicate that decision-making is not widely supported or promoted for these individuals.

Role of health-care professionals.

There is growing recognition of the need for specialized expertise in managing health needs of adults with ID (Barber, Garnham, Lovell, Camus, & Persaud, 2008; Betz, 2007; Burge, Ouellette-Kuntz, Isaacs, Lunsky, & Undergraduate Medical Education in Intellectual Disabilities Group at Queen's University, 2008; Llewellyn & Northway, 2007; Martin, Philip, Bates, & Warwick, 2004; Phillips, Morrison, & Davis, 2004; Sheerin & McConkey, 2008). "Inadequate acknowledgement and accommodation of [the health care needs of adults with ID]…has contributed to poorer health outcomes in this group, as evidenced by the high numbers of missed diagnoses and untreated health conditions, and the lower life expectancy for these individuals" (Wallace & Beange, 2008, p. 358). Salvador-Carulla (2009) commented that it is typical for physicians to receive minimal or no training of in the care of individuals with ID. And in 2006 the Commission on Dental Accreditation in the US called for dental schools to provide students with education and training to better care for individuals with developmental and other disabilities (Waldman & Perlman, 2006).

The literature indicates that progress on addressing this lack of expertise among health care professionals has been slow. In a qualitative study of adults with ID and professional care providers, participants reported that they faced challenges in finding physicians who were familiar with specialized health concerns of individuals with ID (Jones, McLafferty, Walley, Toland, & Melson, 2008). Canadian researchers found routine health screens and health promotion activities were more likely to be overlooked for adults with ID (Ouellette-Kuntz, 2005). In a survey of 227 general practitioners providing care for individuals with ID in Australia, most physicians indicated that they had significant concerns about completing basic elements of clinical services, such as obtaining a complete medical history and verifying instructions (Iacono, Davis, Humphreys, & Chandler, 2003). In the same survey family and professional care providers of individuals with ID reported concerns regarding physicians' and nurses' knowledge of and attitudes toward individuals with ID. Iacono and Davis (2003), in a survey of adults with developmental disabilities (n=328), found similar concerns about health professionals' expertise in working with this population.

This literature addresses health care providers' familiarity with health issues common to individuals with ID. Beyond the provision of health services, are these providers prepared to support and promote the participation of individuals with ID in decision-making? Recently a group of physicians and other professionals developed guidelines regarding optimal health management of individuals with ID. The guidelines grew out of a colloquium held on this topic, and drew from current literature as well as the experience of the authors (Sullivan et al., 2006). These guidelines state "adults with

DD should have the opportunity and support needed to participate in making informed health care decisions” (Sullivan et al., p. 1410).

Geenen (2003) studied how physicians are doing in supporting adolescents with a variety of disabilities in the transition from child-centered care to adult-centered care. The investigators surveyed parents (n = 753) and physicians (n = 141). Subjects prioritized 13 transition activities. Five of the items pertained to helping the youth develop self-care skills (the remaining items pertained to services provided to the young adults). But none of the items included mention of decision-making participation or skills.

Chapter Summary

A critical review of the current literature reveals that:

- No models of the decision-making process have addressed issues beyond the locus of control in the patient-physician relationship.
- There is evidence indicating that individuals with mild ID have the capacity to participate in some aspects of health-related decisions.
- The literature describes a need for expanded expertise among health care professionals in the specialized health issues of individuals with ID, yet evidence indicates this need has not been ameliorated.
- There is limited recognition of the role of adults with ID in the process of making health-related decisions.

Methods

A study of decision-making faces several hurdles. As has been noted, the regarding health-related decision-making has a quite narrow scope. In a qualitative study of this process the investigator must encourage participants to divulge details about decisions that can be sensitive and troubling. In this chapter the methodology and methods used to achieve the study's aims are presented. Here the philosophical premise of data collection and data analysis are presented. The steps used in data analysis are described in detail. Issues of trustworthiness are addressed.

Study Aims

As stated previously, the aims of this study were:

- To describe the decision-making process of young adults with cerebral palsy and mild or borderline intellectual disabilities considering Botox therapy for relief from muscle spasticity.
- To describe the roles of the young adult and their guardians in this decision.

Participants

Inclusion criteria.

Young adult participants were included who:

- Have mild or borderline ID.
- Are ages 18 to 30.
- Have a legal guardian who is a family member.

- Are considering Botox injections for treatment of muscle spasticity.
- Have a diagnosis of cerebral palsy.

The rationales for these criteria are presented here.

Mild and borderline intellectual disabilities.

Young adults were included who had mild or borderline ID. The primary rationale for this criterion was so that young adult participants with ID would have cognitive capacities allowing them to participate in a reflective discussion of issues related to the decision-making process. Individuals with mild ID have intelligence quotient scores ranging from 50 to 69, with an estimated age-equivalent cognitive capacity from age six to under age nine (Harris, 2006; Oliver et al., 2003). Individuals with mild ID also represent the largest proportion of individuals with ID. It has been estimated that approximately 75% of individuals with ID have mild ID (Murphy, Yeargin-Allsopp, Decoufle, & Drews, 1995).

In addition to individuals with mild ID, this study includes subjects with borderline ID. (Borderline intellectual impairment generally includes individuals who fall more than one standard deviation below the mean for intelligence, i.e. with IQ scores in the 70 to 85 range). This study targets young adults with CP who face decision-making challenges due to cognitive vulnerabilities. Young adults with borderline intellectual impairment are a key segment of this targeted population. Further explanation for this rationale is found in this citations from Tymchuk, Lakin, & Luckasson (2001), who coined the term *mild cognitive limitation* as an umbrella term that includes both mild and borderline ID:

> The term *mild cognitive limitations*... is being used to describe people who fell outside existing diagnostic criteria for mental retardation during school but who nonetheless displayed similar learning characteristics, living circumstances, and communities as did people who filled those diagnostic criteria. Labels such as learning disabled, mildly disabled, and emotionally disturbed may have been applied to these individuals. *Mild cognitive limitation also is applied to individuals whose measured IQ score is greater than the traditional ceiling of 70-75 but for whom education and/or support along continua* (i.e. from more to less; continuous to periodic) *may be essential to their success in more complex situations* (e.g. planning for and making major decisions, such as *consenting to health care* or other forms of treatment, participation in research, or entrance into contractual relationships; for self and child care; protecting self and family from harm; responding to accusations of behavioral or criminal misconduct; defending self; initiating political self-activism) and in roles that require more complex learning, judgment, and other cognitive functions across the life span. (Tymchuk et al., p. xxv; italics added).

While most individuals with mild cognitive limitations are legally autonomous, these individuals are likely to face a challenge in negotiating complex health-related decisions independently (Tymchuk et al.). Medical decisions are often complex and involve consideration of several abstract considerations.

Transition age.

Individuals in young adulthood (ages 18 to 30) were targeted in this study, as this is typically the time when young adults develop autonomy. They have also started the transition to adult health services. By focusing on young adults the investigator also was able to explore the decision-making process in the context of the parent-child relationship. The upper age limit was set at age 30 because many adults with ID live in the family home well into their 30's, and for many the transition to adult centered health services is an extended process that occurs throughout their 20's.

Guardianship.

It is very common for adults with ID aged 18 to 30 to have their parents or other family members named as their guardians. The investigator sought to engage participants in a discussion of the decision-making process in the context of the youth's life, so individuals with private or public guardians who might not have rich knowledge of the young adult were excluded.

Young adults who are legally autonomous (or *self-guardians*) were also included. Many young adults with mild ID, and most of those with borderline ID, are legally autonomous. These individuals were included to provide a diverse perspective on how issues of guardianship pertain to decision-making.

Botox treatment for relief of muscle spasticity.

The context for this study is the decision-making process regarding initiation or continuation of intramuscular injections of botulinum toxin type A (Botox) for skeletal muscle spasticity. The rationale for the choice of this context is that this treatment decision is relatively low-risk, and the experience is relatively homogenous across individuals. (Further discussion of this rationale is presented below.) The mechanism of action of Botox involves the interruption of the actions of individual nerve fibers that stimulate muscle contractions; the paralyzing effects are gradually reversed as new neural pathways are established (Pidcock, 2004). Hence the spasticity relieving effects of Botox are temporary, lasting from 12 to 16 weeks (Ramachandran & Eastwood, 2006). This treatment has been demonstrated to be effective for improving function and reducing pain by relieving acute muscle spasticity associated with conditions such as cerebral palsy, stroke and dystonia (Koussoulakos, 2009). The most common risks associated with Botox treatments are pain during injections and excessive weakness, beyond the desired relief of muscle spasticity (Ramachandran & Eastwood). (There have been recent reports of very serious but rare risks associated with Botox treatments; this information will be addressed in the Discussion chapter.)

While Botox has a low risk-profile, the experience of receiving the treatment can be quite distressing. Each treatment session involves from five to 25 intramuscular injections. Many patients become increasingly distressed with subsequent treatments as they anticipate the pain and distress (Symons, Rivard, Nugent, & Tervo, 2006). Interventions that can reduce pain and distress during Botox injections include pharmacologic interventions (sedatives, analgesics, nitrous oxide) and non-pharmacologic interventions (aromatherapy, distraction; Zier, Rivard, Krach & Wendorf, 2008).

Cerebral palsy.

Young adult participants were limited to those with cerebral palsy primarily because this is by far the most common diagnosis among young adults receiving Botox treatments at the recruitment site. (Other conditions treated with Botox for muscle spasticity include spinal cord injuries and genetic conditions.) The investigator narrowed the sample to this single diagnosis in the interest of recruiting a homogenous sample.

The population of adults with CP is also desirable in an exploration of health-related decision-making as these individuals – and their parents – commonly have extensive experiences with health services and decisions. This background provides a good context for a discussion of a current decision-making experience.

Clinical Context

The investigator sought to gather data from individuals who had recently made a health-related decision. Lacking any established theory or instruments for investigating this topic in the target population, a qualitative approach to addressing the study's aims was selected. The investigator wanted to give the participants the opportunity to freely describe their experiences related to a specific decision-making event. After determining that a qualitative multiple case approach was appropriate, the investigator next selected an optimal clinical context for addressing the study's aims.

Determining an appropriate clinical context for the study of health-related decision-making is particularly challenging. The investigator determined that, optimally, the clinical context for this study would have the attributes of low-risk and homogeneity across cases.

- The decision should involve a relatively low-risk treatment. Decisions that have the potential for extremely adverse or unexpected outcomes (e.g. major surgeries) can be extremely sensitive topics for interviews. If participants were asked to describe the process they used for deliberating on a decision that involved grave risks, they may be inclined to offer responses they consider to be socially acceptable. If the interviews are about a decision with relatively minimal risks, the participants may feel freer to be more candid about the process. Many other health-related decisions are often considered too trivial to even be considered as decision-making events (e.g. changes in medications). This posed a challenge for the investigator to identify a health-related decision that was neither too sensitive nor too inconsequential.
- The decision should be as homogenous as possible across cases with regards to risk and decision-making complexity. The decision-making in each case should be comparable to facilitate cross-case comparisons. The investigator wanted to avoid making comparisons between dissimilar situations, such as a decision about having spinal fusion surgery in one case, and a decision to get a hearing aid in another case.

The investigator settled on the clinical context for the study to be *the decision whether or not to undergo Botox injections for treatment for muscle spasticity*. This clinical context met the criteria for being a treatment with relatively low risks as well as the criteria for homogeneity. The Botox treatments provide a comparable experience for across-case comparisons. While individuals get the injections in different sites of their bodies (e.g. legs or arms), the overall experience (pain, related distress, trajectory of temporary relief of muscle spasticity) is comparable between individuals.

Sampling and recruitment

Participants were recruited at a clinic that specializes in the care of young adults with a variety of disabilities, including cerebral palsy. For eight years the Gillette Children's Specialty Healthcare' Lifetime Clinic has offered specialty services for adults who would likely otherwise seek medical care from pediatric providers. This unique clinic was established to specifically address the needs of youth making the transition from child-centered to adult-centered health services. Approximately 300 adults with CP attend the clinic each year. At the Lifetime Clinic physicians provide Botox injections as an ambulatory procedure. Clinicians offer a variety of interventions intended to relieve the pain and distress associated with Botox injections.

The investigator used purposive sampling methods in recruitment. Clinic staff reviewed clinic schedules, identified clinic patients were eligible for participation in the study, and then mailed recruitment letters to these patients and (in most instances) to their guardians. Clinic physicians also invited eligible patients to consider participating in the study during clinic appointments.

Philosophical Approach

Narrative.

This study employs a narrative approach to empirical data (McCance et al., 2001). Narrative methodology derives from the assumption that individuals use stories to make sense and find meaning in their lives (Bamberg, 2006; Poirier & Ayres, 1997; Sandelowski, 1991). The use of narrative in research is related to a phenomenological approach in that data is drawn from the participants' subjective experiences. However, while a phenomenological approach typically explores participants' unmediated experiences, a narrative approach asks participants to make sense of and interpret these experiences as stories.

A narrative approach serves as the epistemological and ontological foundation for the study. In other words, the investigator sought to elicit participants' stories as vehicles for learning about what happened during the decision-making process. The investigator also accepted that these narratives are actually very much a part of what actually happened. Ricouer (as quoted by Mishler, 1991) stated that from narratives "we are able to extract a configuration from a succession" (p. 148). We generally think of stories as having a specific chronology, such as a beginning, middle and ending. But the term narrative is also applied to non-chronological representations of events (Charon, 2006; Riessman, 2007). The essence of the narrative is the creation of an interpretation of past events, assembled in the same way that one tells a story.

Narrative methodology offered the investigator an opportunity to integrate any of the information provided by participants as they described the central phenomenon (the decision-making process regarding Botox treatments). The story that the young adult and parent established together was the basis for the investigator's understanding of the phenomenon in the context of the participants' experiences.

Research Design

The investigator used a multicase study design for several reasons. This design is appropriate for topics that have been little studied, where key contextual elements have not yet been identified (Stake, 2005; Stake, 2006; Yin, 2003). Although there are many adults with cerebral palsy who have mild ID, it can be particularly difficult to identify and recruit these individuals in sufficient numbers to conduct measurement studies (Oliver et. Al., 2003).

Case studies and case series are a familiar approach for presenting information in clinical settings. A case approach provides clinicians with key elements of actual situations; data are presented with contextualized descriptions, and are outlined to encourage reflection on how the case is representative, or not representative, of similar cases.

A narrative ontological and epistemological perspective is an appropriate approach to a topic that has received little attention in the literature. Concepts are developed through the integration of participants' descriptions with those found in the literature.

This qualitative approach, where participants' stories are the data in a series of comparable cases, also creates a link with the ways that these findings might ultimately be used. And there is growing recognition that stories can serve a valuable role in health-related decisions. "Researchers may be more effective in translating evidence into practice and policy if they recognize the power of stories to affect decisions, and use stories to lessen the tensions between words and numbers, and between the perspectives of the individual and the group" (Steiner 2007, p. 1603). It is noteworthy that, just as in the current study a narrative approach guided the inquiry, researchers have begun to explore how clinicians might use stories to facilitate the involvement of patients in decision-making (Mazor et al., 2007; Winterbottom, Bekker, Conner & Mooney, 2008).

Multicase, narrative design.

To integrate the rich detail specific to each young adult's circumstances into an understanding of his or her participation in health-related decision-making, a qualitative, multicase study design was selected. As described above, the investigator sought to elicit participants' narratives of the decision-making process. These narratives were the primary data.

Data-Collection Methods

The primary source of data was semi-structured interviews with participants. In addition to the interview data, the investigator collected demographic data from participants and recorded field notes.

Interviews.

The investigator specified semi-structured interviews as the primary source of data. This strategy was consistent with a narrative approach. For the cases where the parents were guardians, the investigator conducted joint interviews. The rationale for the joint interviews was the assumption that the narrative about the decision to undergo Botox treatments was a shared narrative.

The investigator conducted interviews about one week following a Botox treatment appointment at the Gillette clinic. This interval was chosen as a time interval when the memory of the decision to receive Botox could be recalled in some detail.

For each case the interview recording was transcribed word for word by a professional transcriptionist. The investigator listened to each recording from beginning to end to make corrections in the transcription document where the transcriptionist didn't accurately capture words that were clear to the investigator during the interview. During this process the document was de-identified: all names were replaced with pseudonyms, and any other identifiers were removed.

Interview guide.

The interview guide for this study used three levels of organization as described by Rubin and Rubin (1995): main questions, probes and follow-up questions. (The actual questions that guided the semi-structured interview are found in Appendix A.) The investigator initially asked *main questions*, which are open-ended questions broad enough to encourage participants to respond freely and at length. Riessman (1993) recommends using five to seven main questions in narrative inquiry. However, "when responses lack sufficient detail, depth, or clarity, the interviewer asks a *probe* to complete or clarify the answer or to request further examples and evidence" (Rubin & Rubin, p. 146). *Follow-up questions* help the investigator to pursue information related to participants' responses, such as learning about the consequences of a participant's reported actions. Mishler (1986) cautions that, when using a narrative approach to participants' understandings, the investigator should be cautious so that the prepared main questions don’t interrupt storylines.

Demographic information.

In addition to the interview data the investigator gathered the following information from participants:

- Young adult’s age.
- Young adult’s current residence (e.g. shares parent’s home, lives in a group home) and length of time in that residential setting.
- How long ago did the young adult begin Botox treatments, and approximately how many treatments has he or she received so far?
- Parent assessment of young adult’s level of physical functioning (using the Gross Motor Function Classification System).

This information aided the investigator in a description of the individual cases in the final report.

Field notes.

The investigator recorded field notes during and immediately following the interviews. The purpose of the field notes was to gather descriptive information about the participants, such as descriptions of the young adult’s communication patterns or physical functioning. These notes were not intended to challenge the veracity of the participants’ narratives. These observations were gathered in an effort to create a richer description of the context of each of the cases. The field also notes helped the interviewer to recollect the context for key statements made during the interviews, and aided in making corrections to the typed transcripts of the recorded interviews.

Data Analysis

In qualitative data analysis the investigator endeavors to generate meaningful knowledge from subjective data. The narrative methodology used in this study is fueled by purely unverifiable subjective data: the participants' narratives of the decision-making process. Therefore the soundness of resulting findings relies primarily on a consistent and coherent analytic strategy.

The specific methods of analysis used in this study are described in some detail below. The process is complex and nuanced. The detailed description of the analysis process provides the reader with an opportunity to evaluate the appropriateness of the derivation of findings and conclusions from the data.

Stake (2006) recommends that during the analysis of data in a multicase study the investigator should strive to sustain a tension between the *within-case* perspective and the *across-case* perspective. On the one hand, the investigator maintains an appreciation of the contextualized data within each case. Yet the investigator also attends to the research questions, concepts and themes that are recognized when cases are compared.

There were three stages in data analysis (see Figure 1):

- Data reduction to generate preliminary findings.
- Thematic development using cross-case comparisons.
- Interpretation, synthesis and reflection on findings.

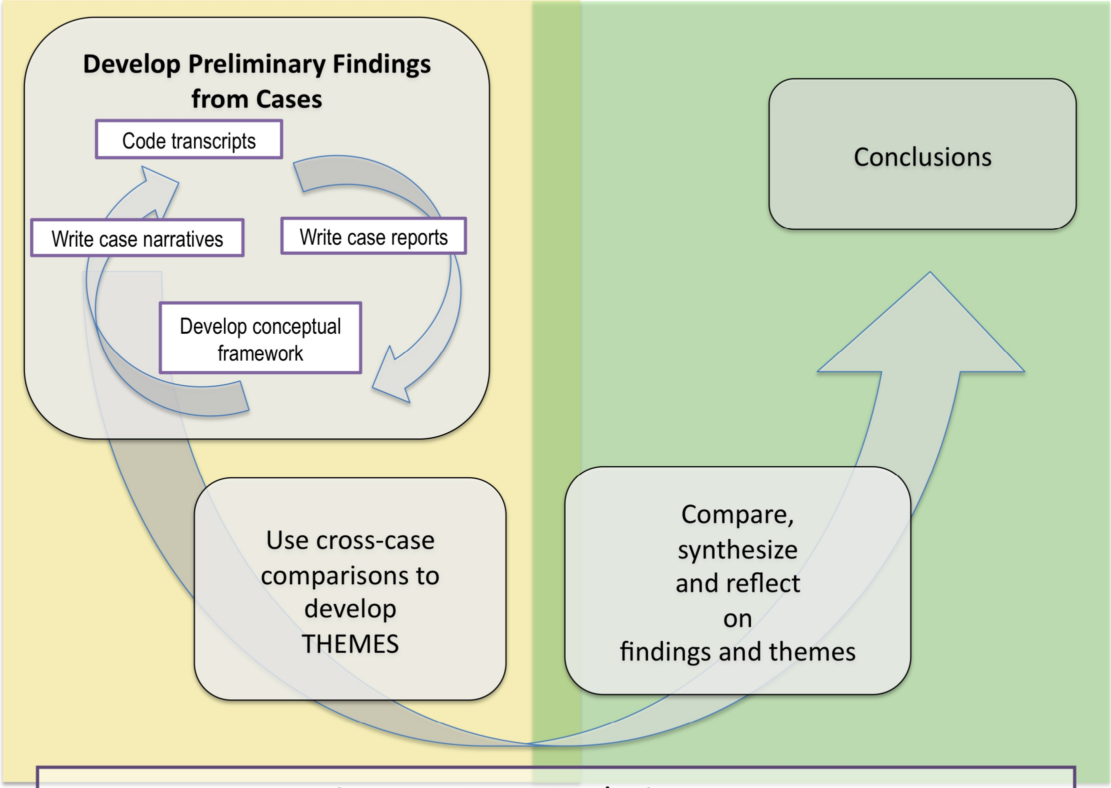

Figure 1: Data Analysis Process

Preliminary findings from cases.

Data were first analyzed at the case level using narrative analysis. Narrative analysis has been defined as "the systematic study of stories" (Bailey, 1996, p. 187) and "the contextual analysis of stories" (Bailey, p. 192). "The basic work of narrative analysis is to produce an integrated story with a plot for each of the participants in the study. Studies using narrative analysis usually produce a case study of each participant and then, a commentary addressing the differences and similarities among the case studies" (Polkinghorne, quoted in McCance, McKenna, & Boore, 2001, p. 355).

The first stage in data analysis involved using data reduction and analysis to generate preliminary findings. Stake (2006) described data reduction as the process of distilling from raw qualitative data salient information that will help to address the study's aims. The investigator performed four distinct steps in the analysis data to generate preliminary findings.

- Coding of interview transcripts.
- Creating case reports.
- Creating case narratives.
- Development of a conceptual framework of the decision-making process.

These steps in the preliminary analysis were not always chronologically sequential, at times the steps overlapped, and the analysis process was cyclical (see Figure 1).

The investigator sought to keep the process of preliminary data analysis closely linked to the narratives of the participants. For this reason the investigator listened to each of the interview recordings from beginning to end at three stages in the data analysis:

1. When verifying that the transcripts were accurate.
2. While coding each transcript.
3. When drafting each of the case reports.

Coding of transcripts.

Coding provides a system for recognizing key ideas as they might appear in slightly different forms throughout the data. (For example, if a mother mentioned that she talked with her son about his symptoms that included pain, this statement would be assigned the codes "communication" as well as "symptoms.")

Each transcript was imported into the qualitative data management program NVivo 8.0 (QSR, 2008). NVivo allows the researcher to code sections of transcripts in a variety of ways that facilitate qualitative analysis. Sections of text can be assigned any number of codes. Codes can be organized into a hierarchical structure. For example, the category “People” contains a sub-category “Healthcare professionals,” with “Physicians” as a more specific sub-sub-category.

The investigator began coding interviews with a preliminary outline that matched the Main Questions outlined in the interview guide (see Appendix A). As the investigator coded interview transcripts he identified new elements that did not match the preliminary outline. Moving back and forth from outline to transcript, the outline gradually evolved into a conceptual framework of the decision-making process. The transcript coding was subsequently modified to match the final conceptual framework.

Case reports.

A case report provides a summary of the analysis within a case (Stake, 2006; Yin, 2003), allowing the investigator ready access to narrative representations of the decision-making process in each case. The investigator drafted a case report for the decision-making process identified in each of the cases. The case reports were structured according to the conceptual framework of the decision-making process (see below) and recorded in a matrix format. (See Appendix B for a sample case report.)

Case narratives of decision-making process

Drawing from case reports, transcripts, and interview recordings, the investigator drafted a narrative for each of the cases. These narratives are the participants' stories of the decision-making process. (See Appendix C for a sample case narrative.)

Conceptual framework of the decision-making process

The investigator created a conceptual framework of the decision-making process. Using this framework the investigator accounted for key elements of the decision-making process identified in each of the cases. The framework also facilitated cross-case comparisons of decision-making processes.

The investigator chose three key criteria to guide the development of the conceptual framework: salience, parsimony and coherence (Fawcett, 1999).

- The criterion of *salience* was met by accounting for all issues that the investigator found to be compelling in each case.
- The criterion of *parsimoniousness* was met by including only elements that contributed to a coherent outline of the decision-making process as the investigator observed it in the cases.
- The criterion of *coherence* was met by organizing the elements into a relational structure that is not difficult to understand, building links between similar concepts.

The investigator coded salient sections of each transcript in NVivo while simultaneously listening to the recording of the interview. Thus the conceptual framework was developed in a cyclical, reiterative process, moving from an immersion in contextualized data to an abstracted representation of key ideas identified in the interviews (the evolving conceptual framework).

Once the preliminary analysis was completed, the investigator engaged in the next level of analysis: thematic analysis. This process was built upon the preliminary findings.

Cross-case analysis: Thematic analysis

Thematic analysis involved a comparison of codes and key concepts as they appeared across the cases. During the process of preliminary analysis described above the investigator developed the narratives of the decision-making process as represented by the participants. Cross-case analysis involved matching and contrasting the themes of the stories reported in each case (Ayres, Kavanaugh & Knafl, 2003). Ayres (2000a and 2000b) used a similar analytic strategy for a multi-case study of family caregivers.

The investigator identified specific themes by being immersed in the case reports and case narratives. Cross-case comparisons were made using a matrix representation of the conceptual framework. While identifying the themes, the investigator maintained a narrative perspective, i.e. he did not draw from his field notes, demographic data, or any other information except that described by the participants in their stories.

The purpose of the next stage in analysis was to identify and develop the ideas represented by the themes. The investigator reflected on the cases, in themselves and in comparison, building upon the preliminary findings and themes through interpretation, synthesis and reflection.

Comparisons, synthesis and reflections.

The investigator compared the themes and preliminary findings with the assumptions identified at the beginning of the study (as stated in the Introduction chapter). These assumptions are based on the investigator's professional experiences and serve as a contrasting information source, helping to illuminate and to contribute meaning to the study findings.

The investigator also made comparisons between the study findings and any applicable findings in the literature. With this synthesis important links were established between various sources of evidence. Finally, the investigator reflected on the themes and preliminary findings, comparing the contextualized data (case reports and case narratives) with field notes and demographic information. The resulting conclusions from the analysis are presented in the Discussion chapter.

Trustworthiness

Guba and Lincoln (2004) have proposed that in qualitative methodologies *trustworthiness* is an acceptable approach to the issues usually conceptualized as validity and reliability in quantitative research. Trustworthiness refers to a demonstration that the investigator's arguments and assertions are sound (Richards, 2004).

Throughout data collection and analysis the investigator maintained an *audit trail* that included documentation of ideas and decisions made during data analysis (Ayres, 2007). This method allowed the investigator to keep track of the development of concepts and themes during data analysis.

Once the investigator had developed a complete version of the conceptual framework of the decision-making process, a colleague audited the resulting the conceptual framework. She specifically targeted the coherence and parsimoniousness of the conceptual framework. (The colleague is a doctorally prepared nurse who has experience with analyzing qualitative data, who is also familiar with issues faced by children with complex health needs and their families.) First the investigator provided the colleague with a thorough description of the elements and organization of the conceptual framework. Then the investigator provided the colleague with selections from the interview transcripts that exemplified the elements of the conceptual framework. The results of the colleague's audit were that she found the conceptual framework coherent and parsimonious.

Ethical Considerations

The investigator received approval from the University of Minnesota's Institutional Review Board (IRB) prior to initiating any recruitment activities. (This IRB also oversees ethical conduct of human subject research for Gillette.)

Potential participants first learned about the study from one of the clinic physicians during a clinic appointment. To minimize coercion the investigator had no contact with clinic patients until they expressed interest in speaking with the investigator about the study. During the consent process the young adult were consented separately from their parents. The consent forms also included a separate consent for the use of participants' direct quotes in publications and presentations (Mishler, 1986).

Chapter Summary

The investigator used a multicase methodology to study a specific health-related decision. Semi-structured interviews were conducted with young adults with cerebral palsy and ID (and their guardians). Participants described their experiences in the decision-making process as stories, in accord with a narrative perspective. Case narratives were analyzed to generate preliminary findings, which included case reports, case narratives, and a conceptual framework of the decision-making process. Thematic analysis was made using cross-case comparisons. Themes were developed by synthesizing information from current literature.

Results

The primary aims of this study were:

- To describe the decision-making process of young adults with cerebral palsy and mild or borderline intellectual disabilities considering Botox therapy for relief from muscle spasticity.
- To describe the roles of the young adult and their guardians in this decision.

To address these aims, the researcher analyzed the data to develop preliminary (case-based) findings and thematic (cross-case) findings.

Description of Participants

Seven cases were included in the study. The investigator had set a target for 12 cases. After 11 months of recruitment activities he was successful in enrolling only 14 participants in the seven cases. The first phase of recruitment lasted five months and involved the mailing of recruitment letters to Gillette's Lifetime clinic patients and guardians who met study sample criteria; eligibility was identified by staff review of patients' records. However, only one guardian contacted the investigator after receiving the recruitment letter (the young adult was not eligible for enrollment).

The second phase of recruitment was successful in recruiting the participants for the seven included cases. This phase lasted six months. Clinic staff provided the investigator with the times of clinic appointments each week when potential participants would be attending clinic. The investigator came to the clinic at these times, to remind the physicians of the study's inclusion criteria, and to be available to speak with patients and guardians who expressed interest in learning more about the study. (It is noteworthy that none of the enrolled subjects ever mentioned receiving an introductory letter.) Two patients declined participation after being introduced to the study in person by the physician.

Table 1 provides descriptive information of the sample subjects. (All names used are pseudonyms.) These descriptive elements include:

- Guardianship status.
- Current living situation.
- The number of Botox treatment sessions the young adult has undergone.
- The particular sites on the body where the young adult has received Botox injections.
- Other health-related decisions that were brought up during the interviews.
- Gross Motor Function Classification System level. GMFCS scores provide a description of gross motor functioning for individuals with CP, based on a description of the individual's abilities with regards to sitting, transfers and mobility. Level I indicates the ability to walk independently; Level V indicates that mobility is only with use of a wheelchair, with limited trunk control (McCormick et al., 2007).

Table 1. Attributes of case participants.

Gender	Age	Name	Guardianship status	Living situation (parent study participant)	# of Botox sessions	Botox injection sites	Other health related decisions discussed	GMFCS level
m	28	**Bill**	self-guardian	with parents (mother)	3	wrists	(none)	**III**
m	24	**Mike**	parents	group home (father)	4	arms & legs	urinary bypass surgery	V
m	24	**Joe**	parents	with parents (mother)	5	wrists	baclofen pump	IV
m	27	**Tom**	self-guardian	independent	3	legs	baclofen pump	**III**
m	19	**Scott**	parents	with parents (father)	(5 years)	legs	baclofen pump; spinal fusion	V
f	18	**Cathy**	parents	with parents (mother)	2	legs	shunt revisions	**II**
m	21	**Mark**	parents	with parents (mother and father)	(9 years)	arms & legs	baclofen pump; orthopedic surgery	IV

It is important to note that all of the young adult participants had received Botox treatments more than once. As a result, there were two different types of decisions discussed during the interviews: the decision to try Botox the first time, and the decision(s) to continue to return for subsequent Botox treatments.

The inclusion criteria allowed for young adults who had a non-parent family member (e.g. sibling, aunt, uncle, grandparent). But only parent guardians were subsequently recruited.

Preliminary Findings

Preliminary findings are drawn from the participants' experiences and meanings as presented in their narratives. As described in the previous chapter (also outlined in **Figure 1**), the first stage in data analysis was to generate preliminary findings, consisting of *case reports*, *case narratives*, and a *conceptual framework*. The interviews lasted between 30 and 60 minutes (mean length = 48 minutes). Interview transcripts were between 13 and 24 pages in length. The case reports and case narratives are not reported here. (A sample case report is found in Appendix B, and a sample case narrative is found in Appendix C.) These documents served as tools used by the investigator to facilitate cross-case comparisons, analysis and synthesis.

Conceptual framework of decision-making process.

The conceptual framework presents key elements of the decision-making process identified in the cases. Key elements have been organized into a nested hierarchy where similar ideas are brought together under the four major concepts: goals, information, relationships and deliberation. The major concepts are presented in an order that corresponds to a chronological sense of the decision-making process. No relational claims are stated, beyond the presentation of ideas in an outline format. (See **Figure 2** for a graphic representation of the conceptual framework. The complete conceptual framework of the decision-making process, with illustrating examples from the case narratives, is found in Appendix D.)

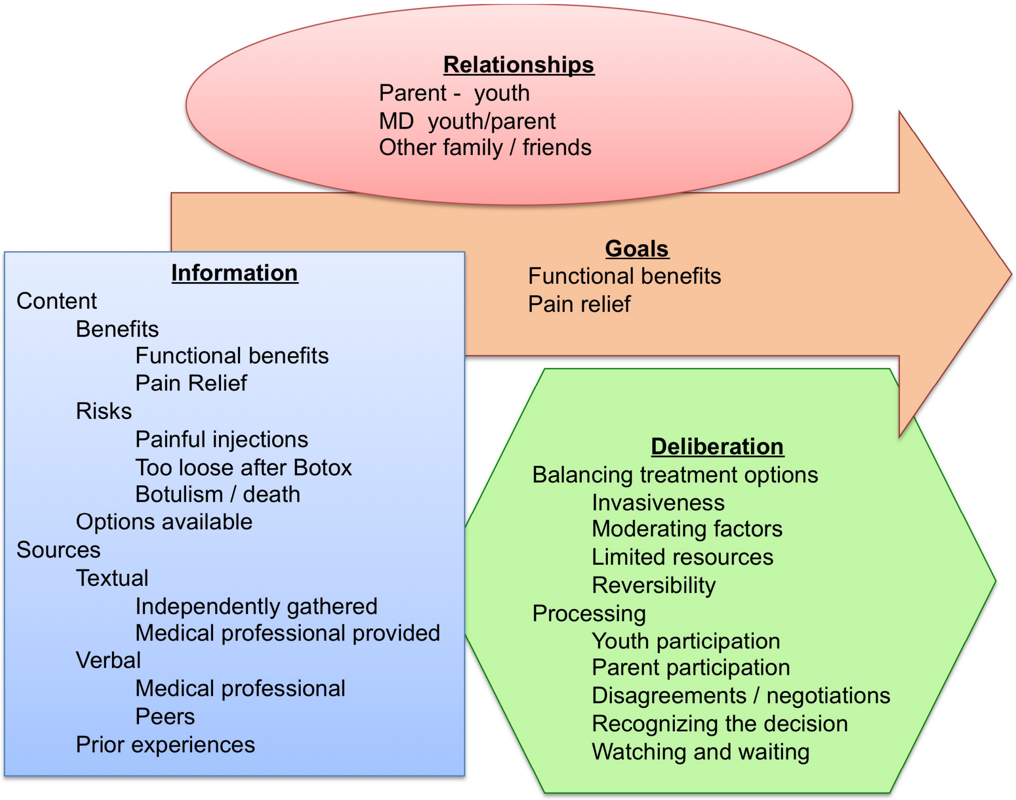

Figure 2: Conceptual Framework of Decision Making Process

Goals.

Goals are the desired state of being that the participants were seeking for the young adult with regards to managing muscle spasticity.

- *Functional benefit* refers to goals with regards to relieving muscle spasticity in order to improve or maintain function.
- *Pain relief* refers to goals with regards to relieving muscle spasticity in order to reduce or eliminate pain?

Information.

Information includes the information content used in the decision-making process as well as the sources of this information.

- *Information content*: What was the information about?

    - Potential benefits of Botox.
        - Functional benefits of Botox.
        - Pain relieving benefits of Botox.
    - Potential risks of Botox.
        - Pain and distress associated with Botox injections.
        - Botox injections can result in the muscles becoming too weak.
        - Over the past few years there have been alerts regarding a very small number of deaths associated with Botox treatments for muscle spasticity.
    - In addition to Botox, what are alternative treatment options for addressing treatment goals?
- *Information sources*: Where did the information come from?
    - Textual information.
        - Information sought by youth or guardian, independent of that provide by medical professionals.
        - Information provided by medical professionals.
    - Verbal information based on other peoples' experience or expertise.
        - Verbal information from medical professionals.
        - Information from peers and others who have experience with Botox.
    - Information from prior experiences with spasticity, Botox and other treatment options.

Relationships.

These are the relationships with people who are brought into the decision-making process. This means that the opinions and recommendations of these individuals were part of the decision.

- The relationship between the youth and the parent.

- How was the physician involved in the decision?
- Beyond the parent, who else was involved in the decision?

Deliberation.

Deliberation refers to the process of developing the decision.

- *Balancing treatment options* refers to the appraising of the relative merits and liabilities associated with different elements of the decision.
    - *Invasiveness*: Different treatment options for managing muscle spasticity involve different levels of invasiveness, ranging from surgery to massage.
    - *Moderating factors* are methods of ameliorating any of the risks associated with Botox, specifically the pain of the injections. Examples are nitrous oxide and EMLA anesthetic cream.
    - *Limited resources*: Different treatment options involve a commitment of limited resources, such as financial cost, or the time or energy involved in different treatment options.
    - *Reversibility*: Some treatment options are essentially reversible over time, e.g. Botox and oral medications. Surgery is an example of an irreversible treatment option.
- *Processing* includes actions and events that took place during deliberation.
    - How did the youth participate in the decision?
    - How did the parent participate in the decision? (For the youth who are legally autonomous, how is the parent involved in the decision?)
    - Disagreements or negotiations between the young adult, guardian, parents, or physician.

  - Was there a particular moment when the decision was made, or was it an extended process with no clear decision-making event?
  - Once various options are being considered, did the youth and guardians "watch and wait" before committing to any decision?

Themes

Thematic analysis of preliminary findings involved cross-case comparisons of the preliminary documents (the case reports, case narratives, and conceptual framework). The investigator also integrated information from field notes. The investigator identified four main themes and 10 sub-themes:

- **Agency**
  - **Explicit agency**
  - **Implicit agency**
- **Communication and cognition**
  - **Capacity for decision-making**
  - **Complexities of communication**
  - **Use of supports with cognitive tasks**
  - **Distinguishing communication from cognition**
- **Information gathered independently**
  - **Information from peers**
  - **Information from the Internet**
- **Relationships**
  - **Non-family members**
  - **Relationships with physicians**

The themes and sub-themes are described here, and supporting examples from the cases are provided.

Agency

The main theme *agency* was perhaps at the heart of this study's purpose; Stake calls this a "foreshadowed issue" (2006; p. 11). Agency refers to the ability and means to act; in this study it refers specifically to the young adults' volition with regards to the decision whether or not to receive Botox treatment. Young adults' agency was not clearly stated in all of the narratives. The investigator found a clear distinction between varying degrees of the youth's role in the Botox decision: In some cases participants described the youth's explicit, active involvement in the Botox decision. In some cases the narratives implied the youth's involvement. And in other cases parents did not distinguish the youth's role from the parents' role in the decision.

Explicit agency.

Explicit agency refers to instances in the unambiguous identification of the youth's role in decision. Mike's father described Mike's role in the decision:

> My wife and I talked it over with Mike. And he said, well, his words were — "Anything that gives me more ability to do things and have better control, let's give it a try." … But we've always talked about it, and he's indicated his wanting to continue. So this is what we've done…We've always tried to involve Mike [in decisions].

Mark's parents described that they have always been cognizant of their role as proxy decision-makers. Mark's role in the decision was described as being a type of veto power. His father said, "Mark always participates [in decisions]…Mark always has the final, 'No, we're not going to do it.'" His mother described a more active role for Mark in decisions: “Pretty much Mark gets fed all the ideas, and I’ll give him my opinion, and then he generally makes a decision…We’re here to do the research on the computer, and talk to people, and then give our input.” But she also described that for decisions that involved more significant risk, such as surgery, the parents would ultimately make the decision: When deciding about extensive orthopedic surgery, “We were protecting him. He could not have handled that.”

In some cases there was also a description of how the parents or school had worked to develop the young adult’s skills in making decisions. Mike’s parents have always encouraged his self-determination, most explicitly since he moved from the family home to the group home. His father stated, “All the while he was at home and all the opportunity that we get we involve him in all of the decisions. I mean, he made the decisions at home, what he wanted to do. We’ve always wanted him to be an independent thinker.” Joe’s mother reported that Joe had had training in decision-making during his junior year in high school: “They even did little exercises and had games about—where they talked about how to— ‘If you do this, let’s think about what the consequences might be…’” She was disappointed that he had not received any further development of these skills during his subsequent four years in the high school transition program. She was pleased that his current day training program was helping him to develop additional decision-making skills.

Implicit agency

Implicit agency refers to instances where the youth’s role in the decision was implied in the narrative. In two cases parents described that they interpreted the *absence* *of the youth's resistance* to undergoing medical treatments as representing the youth's *implied agreement* with this decision. Scott's parents take control of the big decisions but are very sensitive to his strong feelings. Scott "doesn't like surprises," and he will go along with the parents' decision as long as it is clear and makes sense to him. Scott's dad described how Scott can really get upset, and he can in effect veto a treatment option by getting very upset, or at least force the parents to reconsider the decision. Scott rarely gets upset while at medical appointments or during hospitalizations. But he has become very upset and angry about other types of issues, so his parents trust that he will let them know whenever he feels strongly about the outcome of a decision.

Joe's mother has recognized how passive Joe often is: "Joe doesn't feel that his body is his own. Because all his life, he's had things done for him and to him." He has limited experience with agency, and his mother has identified that she needs to develop Joe's skills, especially with regards to "negotiable" decisions like Botox. Joe's mother interpreted his willingness to return for the second Botox treatment session as his indication that he wanted to continue:

> If he had been adamant about not wanting to do the Botox, we would not have repeated it. But he wasn't. We don't talk about it anymore. The last time we went in, we didn't discuss whether or not to do it again. I just said, 'You've got an appointment with Dr. Smith. We're going to do the Botox injections.' And that was fine.

Mark's father described how he and his wife strive to empathize with Mark's situation while the family deliberates:

> So, you know, it's an education process for the patient and the people that live with the patient to try and understand how do you—what is this disease, and how does it affect him? It's hard to put one of us three into Mark's body and think of what that is, cause we aren't there… It's not us that the needles are going in. I'd be a lot more scared than you [Mark] are! [father laughed, Mark laughed as well].

Cathy's mother stated that she has always placed a priority on being respectful and considerate of Cathy when considering decisions affecting Cathy:

> Nothing was done behind her back…. I mean even before she was 18. The age thing didn't matter to me. I guess I tried to put myself in her place, and most people would feel different. I mean I would want to know—I work in health care, so you have to understand that too, but I would want to know what's going to be done to me, and I didn't think that it was fair, it wouldn't have been very fair to her, to not let her be able to expect to know what's coming. Whether it's going to hurt or not, at least you know it's coming, and you know what to expect.

Communication and cognition.

The investigator had anticipated that communication and cognition would be key issues, i.e. this was a foreshadowed issue. Communication impairments are common in individuals with cerebral palsy. The label *intellectual disability* is predicated on diagnosed cognitive impairments. These two concerns can easily become entangled.

Capacity for decision-making.

Parents who were guardians described that some decisions were too difficult for the young adult to handle, and for these decisions the parents would take full control. As previously described, Joe's mother characterized decisions as *negotiable* or *non-negotiable*; she considers the decision about Botox treatments as an example of a negotiable decision, because while it can certainly affect Joe's comfort and function, the outcome of the decision is not critical. She brought up the distinction as part of a growing awareness of how little control Joe has over his life and even his body. She stated, "There are certain things that *have* to be done;" with this statement she seemed to imply that critical decisions were beyond Joe's capacity.

Cathy's mother made a similar statement regarding the severity of the consequences of different decisions: "I mean with the shunt decision, that's do or die, really. And the Botox, you know, you either do it or you don't do it." Similar to Joe's case, the context of the statement seemed to imply that some decisions were beyond Cathy's capacity. And Mark's mother's statements were also consistent with these examples. With regards to a decision about a major orthopedic surgical procedure she said, "We were protecting him. He could not have handled that."

In a sense these parents were describing some sort of threshold for seriousness or complexity of decision-making. The critical and difficult decisions were the parents' domain, and the young adult would be encouraged to participate in negotiable or perhaps easier to understand decisions.

Complexities of communication.

Many individuals with CP have sensory and motor impairments that result in dysarthria, which is the inability to clearly articulate when speaking. About 60% of individuals with CP have some degree of dysarthria (Rapp 2000). Dysarthric speech patterns can pose tremendous obstacles in daily functioning, especially when attempting to communicate with someone who is unfamiliar with an individual's unique speech pattern. With regard to dysarthria, the young adults in the study covered a spectrum, with one adult whose speech was mildly dysarthric on one extreme, and a youth who had no capacity for speech on the other extreme.

Augmentative communication devices (ACDs) are any form of technology used to assist an individual in verbal expression. Electronic ACDs can facilitate extremely detailed and extended verbal communication. But the process of using an electronic ACD is often very slow if the user has poor motor control, as is often the case in individuals with spastic CP. Two of the young adults used electronic ACDs during the interviews. They both demonstrated extensive facility in generating very precise statements using their ACDs. However, it was striking that both of these young men seemed to only use their ACD's as a last resort, when verbal expression or gestures failed. It took 74 seconds for Bill to express the word "inward," while describing that muscle spasticity was forcing his hands *inward*, which was the issue that brought him to finally decide get the Botox injections. He once tried to speak the word, and when neither his mother or the investigator understood, he began to spell the word using his ACD. When using his ACD throughout the interview, most of his messages took longer than 60 seconds to compose.

Scott has severe dysarthria, with essentially no verbal speech. Yet it was uncanny how Scott could bring his father to understand him with only a subtle gesture or gaze. The investigator would restate the father's words to solicit Scott's endorsement of the statement. Each time Scott responded strongly to either confirm or reject his father's words as being an accurate representation of his own. (In nearly every instance Scott endorsed his father's words.) His father offered some insight to this phenomenon: "Sometimes I think I'm like reading his mind now, but it's just so many years worth of different things that he's doing, that it's repetitive then. I just know what he's trying to bring out."

Even with ACD's, and parental involvement, communication can be a significant obstacle. Scott's father described a recent situation where Scott was very agitated when he didn't understand the parents' plan for a weekend vacation. Despite the use of Scott's ACD, and the involvement of both parents and his long-time PCA, it took what seemed like a very long time to help figure out why Scott was so upset.

Mike has severely dysarthric speech and doesn't use an ACD. He uses a method of consonant substitution in his speech. For example, he can't close his lips to make the "b" sound, so he substitutes a hard "g" sound. His speech is not hard to understand once the listener has had an opportunity to acquire an ear for this speech pattern.

In addition to using ACDs, these young adults rely on the assistance of those who know them well to facilitate their communication. Bill's mother was very active during the interview in helping the interviewer to understand Bill's words.

Joe's mother described how she sometimes acts as an interpreter for Joe, aiding both his receptive and expressive communication: "

> It's just people that don't know Joe. It's difficult. And I hesitate—like when you were asking Joe questions — I hesitate to interrupt, and re-phrase what that person said, but I know that I can re-phrase it in a way that something clicks for Joe….I guess I kind of serve as an interpreter a lot of times, both ways. If Joe will say something that might seem out of context or not to make any sense, I know him well enough to know where that came from. And so I'll kind of re-phrase and interpret what—and I'll ask him, I'll say, "Is that what you were talking about, or is that what you meant?" And he'll say "Yes"… It's a challenge, to make sure that he understands, and that he is being understood. So that's why I go to all the doctor's appointments with him.

Use of supports with cognitive tasks.

The participants described different processes for providing the young adult with support for the cognitive aspects of decision-making. Mike (whose parents are his guardians) said, "I like to take control as much as possible...*but*...I have to have somebody go through the information with me..." His father confirmed this: "He doesn't like to make decisions unless we've talked about them."

As described above, Joe's mother aids in Joe's comprehension of information while she is also facilitating communication, or "translating." She also carefully sets up decisions, presenting him with smaller elements of information and decisions.

> I try to come up with things that he can relate to. Either things that have happened in the past, or experiences that we have all the time, and say, "You know how—so and so and so..." Yeah, it's usually one thing at a time. And I try not to overwhelm him with too much information at once. And sometimes I even have to talk to him a little bit one day, and let him kind of process it. And then we'll talk about it again the next day.

Mark's mother described how she and her husband support his decision making (in an excerpt also cited above): "Pretty much Mark gets fed all the ideas, and I'll give him my opinion, and then he generally makes a decision…We're here to do the research on the computer, and talk to people, and then give our input."

Distinguishing communication from cognition.

The issue of communication is distinct from cognition, yet the issues are not easily distinguished; when communication is not effective, there is no easy way to determine whether the failure in communication is a result of expression or cognition. In other words, is the individual unable to articulate the desired words, or is he perhaps misunderstanding the topic at hand? And because dysarthric speech patterns of individuals with CP often involve long or unpredictable pauses during conversations, others can interpret the speech pattern as an indication of lack of comprehension.

Joe's mother described how his communication pattern poses a significant obstacle in being recognized as an individual with volition. It's not just that his words are hard to understand; people tend to assume he can't think clearly because he can't speak clearly: “Joe’s not always easy to understand. And a lot of people, if they don’t understand what he says, dismiss it.”

Another dramatic example of the confusion of communication and cognition arose from the recruitment process. During study recruitment at the Lifetime Clinic, clinic staff reported to the investigator that they estimated Scott as having a moderate or even severe level of ID, because they had never witnessed him communicating. This was because Scott had never come to the clinic with his electronic ACD. Once the investigator met with Scott and his father at their home, Scott quickly demonstrated his dexterity with the ACD. (Using only a head switch as an interface device, Scott uses Facebook to keep in contact with his family and friends, and navigates the Major League Baseball website to watch several ballgames simultaneously.) Based on the fact that they had never witnessed any direct communication from Scott, the assumption of the clinic staff was that his extreme communication impairment was matched with a similarly impaired cognitive capacity.

Information gathered independently.

It is not surprising for an investigation of health-related decision-making to include an exploration of the issue of *information*. What does a patient learn about the decision and how did she receive this information? Discussions of health information and patient education often address the type of information provided by health-care professionals. What is often overlooked is the fact that individuals are also likely to seek information on their own, either from peers or, increasingly, via the Internet.

Information from peers.

Individuals with CP are very likely to become acquainted with one another throughout their lives, through clinical and educational settings that provide specialized services for this population. Parents of children with CP often meet one another in the same ways. In five of the cases, participants described how they had gathered information about Botox treatment for muscle spasticity from the young adults' peers.

Participants described that information from peers was as important as any other source of information. Joe's parents declined Botox for more than a year because a peer of Joe's had found the injections to be terribly painful. Scott has a good friend with CP ("Sarah"), and his parents consult with Sarah's parents on all health related issues because her experiences are invaluable as they consider what might be ahead for Scott. His father stated, "She's a year older, and she literally was just like one year ahead of him, of having all these different things done, just like Scott. They could be twins."

Tom found that the information he received from his friends was most helpful. In fact the information about Botox that he received from his friends was really the only significant source of information he considered while considering Botox treatments. Although Tom received written information from Dr. Smith, he considered that this information was not as relevant as the testimonies of his friends who had experience with Botox treatments for muscle spasticity:

> [Dr. Smith provided] a handout to read about Botox. But I didn't have to read about it, cause my friends already told me about it. So I have it filed with all of my medical appointments. Why read about it when your friends already told you

> about it? And your friends are not going to lie to you. I trust my friend before I'd read something, you know?

Information from the Internet

Individuals frequently turn to the Internet as a source of health-related information (Keselman, Logan, Smith, Leroy, & Zeng-Treitler, 2008). In two cases participants mentioned getting information from online sources. Scott's family decided to stop his Botox treatments when they found news stories on the Internet reporting several deaths associated with the use of Botox for muscle spasticity. They waited to see if new reports of serious complications associated with Botox would continue to appear. They continued the Botox treatments when they were satisfied that risk of complications was relatively small. This deliberation was made by Scott's parents, without Scott's involvement. Mark's mother also mentioned in passing that they search the Internet when seeking to learn about a health issue.

Relationships.

It is noteworthy that in the decision-making literature (described in the literature review chapter above), the only individuals considered as participants in the decision are the patient and the physician. No mention is made of other individuals, such as family members, with whom the patient might confer. In the investigator's personal and professional experience, family members and friends are often consulted when significant medical decisions are being considered. In fact, extended care planning meetings, involving family members and others, are becoming increasingly common across healthcare settings (P. Hudson, Quinn, O'Hanlon, & Aranda, 2008).

Non-family members.

In addition to the young adult, the parent and physician, participants described that they involved other key individuals in the decision-making process. Joe's mother stated that she highly values his personal care attendants' (PCAs') opinions while making important decisions.

> Well, his PCAs are really good sounding-boards for me. Because I think out loud [laughs]. And I'll ask them—well, they've been with him so long now, they just come to me and say, "You know, I'm noticing this, and this seems to be a little bit tighter," or "This seems to be a little harder," or "Joe seems to be not quite himself." … I know I talked with them about the baclofen, and explained what it was and why we were doing it. But I think, now, today, yeah, I probably would ask for their input and their opinions, even more than I did two years ago. Because they know Joe.

Scott's PCA Sally has been a part of Scott's life for 13 years. Scott's father described that, "She's Scott's big sister, second mom." When it comes to making decisions on behalf of Scott, his father stated, "It's probably the four of us. It's Sally, Scott, me and mom." Scott's brother Barry, who also lives in the family home, does *not* want to be involved in the details of Scott's health issues. Scott's father said that while Barry is very much involved with Scott's life,

> [Barry] is a little squirmish when it comes to making medical decisions on his brother... cause he feels the pain that [Scott] goes through— So we'll tell brother what's going on…But if we tell him everything that's involved in that then he kind of gets a little worried of things that could go wrong…So we kind of—not leave him out of the loop, we keep him informed.

Relationships with physicians.

Participants described how they had included the physician's recommendations while considering Botox as a treatment option. Participants also described many positive interactions with physicians. Bill admitted that he felt that his physician trusted him, as a legally autonomous adult, to make his own decision, without trying to sway him. In several cases parents described long-lasting relationships with physicians that involved many difficult decisions and experiences over the years; the relationships with the physicians were described in very positive ways.

Mark and his mother both described how they were favorably impressed when a physician disclosed to them that he wasn't sure what the best decision for them would be. Mark said, "I went to the doctor... that does [the baclofen pump], and it's like, he said, 'I don't know!,' which was actually pretty good!" His mother reinforced this opinion in her statement that it is frustrating when health professionals express only certainty about treatment options and rarely disclose uncertainty about the optimal treatment option: "We're guardians to protect him from—like different doctors have different opinions, and they all are so sure of them."

In each case participants described at least one occasion where a physician had recommended a significant medical or surgical intervention that the family had declined. With regards to Botox treatments for muscle spasticity, in four cases the families passed up offers to try Botox before eventually agreeing.

Chapter Summary

Preliminary findings were based solely on participants' stories. A conceptual framework was developed to coherently represent key issues identified in participants' narratives of the decision-making process. A thematic analysis identified four main themes and 10 subthemes. Participants described varying degrees of explicit participation of the young adult in the decision-making process. Communication patterns were a significant factor in making decisions. Youth were supported in various ways with communication and deliberation. Participants considered peers as an important source of information about Botox. The young adults, their parents and their physicians weren't the only people involved in the decision-making process.

Discussion

The conceptual framework and themes are compared with related findings from current literature. Conclusions are outlined, and recommendations for professional practice and further research are stated. First a discussion of the strengths and weaknesses of the research methods are presented.

Strengths & Limitations

Features of the dissertation that strengthen or limit the significance of findings are addressed here. Generalizability is considered within the discussion of the study's limitations.

Strengths.

A specific strength of this study is that it examines a specific health-related decision that is easily compared across cases. Even at a clinic that provides services to a large number of patients who fit the inclusion criteria, there were ultimately only a handful of patients who met the criteria who were also able to manage the logistics of recruitment and interviewing. This pursuit of homogeneity of clinical context very likely contributed to the small number of participants recruited. There is no doubt that additional cases would have provided a better understanding of the key phenomena. But the price of a small number of cases was accepted because of the value of having those cases be as closely comparable as possible.

The investigator was quite familiar with the clinical issues discussed by the participants (i.e. management of muscle spasticity associated with cerebral palsy). It is likely that this familiarity contributed to the extensive descriptions participants offered during the interviews. As has been mentioned, several of the participants characterized the issue of the Botox decision as making a mountain of a molehill. Cathy's mother commented dryly, "We've made bigger decisions." But the investigator was able to catch subtle references to the various concerns that arise related to spasticity and cerebral palsy. This attentiveness perhaps encouraged participants to go on at some length about a topic that they did not find particularly significant or interesting. At the end of the interview, Mike's PCA observed, "He usually doesn't talk this much." It wouldn't have been surprising if some participants were reticent with regard to the extremely narrow focus of the interviews. One participant – Joe - excused himself early on in the interview; all of the other participants (including Joe's mother) provided rich and insightful descriptions. The interviews ranged from 30 to 60 minutes, averaging 48 minutes.

The narrative methods used in this multicase study provided an opportunity for the participants to freely offer their reflections on the decision-making process. These methods also provided a chance for the young adults with unique communication patterns to contribute to the data.

Limitations.

An obvious limitation of the narrative approach is that the investigator has no means of determining to what degree these narratives are grounded in actual events. In the context of the interviews parents might have felt pressure to exaggerate or fabricate the "story" about the youth's involvement in the decision. It is also unlikely that the participants would report during the interviews any significant conflicts between the parents and the young adult.

The investigator had assumed that the unique communication patterns of the young adults would not pose a significant obstacle during the interviews. It is understandable if readers are skeptical and suspect that perhaps these interviews are the result of ventriloquism on behalf of either the parents or the investigator. The investigator made certain to confirm with the young adult every restatement or clarification made by the parent. Yet it may be hard for others to consider that the young adults unembellished statements survived this process that one parent described as “translation.”

The investigator made no attempt to establish the competency of the young adult participants for making meaningful decisions. This issue was beyond the scope of the study, but it is certainly an important consideration. For example, it would be important to know how parents’ estimations of the young adults decision-making capacities would compare with systematic assessment of these abilities, e.g. with the use of the Ability to Consent Questionnaire used by Arscott et al. (1999).

It is important to consider how an investigation of a health-related decision with a relatively low risk profile (e.g. Botox) is distinct from a decision involving significant risk. A consideration that the investigator might have been *making a mountain out of a molehill* raises both potential strengths and limitations of this study. It is likely that the low risk profile offered participants an opportunity to discuss their decision-making experiences more freely than they might when reflecting on a “high stakes” decision. On the other hand, it is also likely that a conceptual model of the decision-making process regarding higher risk treatments might include different elements than the one developed in this study.

Generalizability.

The investigator's intention was to offer new knowledge on a little studied topic, in the form of insights drawn from conversations that were conducted in a systematic way. Clearly the results of seven cases cannot provide a basis for extremely specific generalizations about a population. Therefore the conclusions stated here are limited assertions.

The participants are not considered to be representative of all young adults with CP and mild cognitive limitations. It is very likely that the investigator recruited parent participants who are more likely to be closely involved in the lives and healthcare of their adult children than is typical. It is also likely that these parents represent a higher degree of commitment to their children's self-determination. And there was no representation of young adults who have private or public guardians, or even those who have non-parent family guardians (e.g. siblings or grandparents).

Since the investigator did not have the resources to incorporate translators into the data collection process, only English speaking participants were included. This meant that at the Lifetime clinic many patients and families who spoke Spanish, Hmong, Somali, or American Sign Language were excluded.

The young adult participants were also predominately male. Studies of individuals with CP commonly report a disproportionate number of males as compared with females, though this imbalance has not been addressed in the literature.

Comparisons with Literature

Findings from the study will be compared with current research literature. These comparisons provide perspective on the study findings and will also shed new light on other researchers' evidence. (For some of the sub-themes there was no literature available for comparison.)

Conceptual framework.

Three studies of decision-making processes provide contrast with the conceptual framework developed in this study. Two of these studies address the issue of locus of control. As discussed above, an extensive search for conceptual approaches the process of health-related decision-making resulted in literature that was surprisingly narrow in scope, i.e. a focus on the locus of control in the decision.

In a qualitative study that addressed the issue of locus of control, Young (2006) reported on children's, parents' and therapists' views as to who made decisions about physical therapy programs. (The children all had cerebral palsy.) While some parents expressed frustration at not being more involved in therapy related decisions, other parents seemed to defer to the therapists' expertise. Several parents "showed a sense of surprise" at being asked about their role in the decision-making process (Young, p. 119). These parents had assumed that the "right" decision would be made unilaterally by the therapist, based on her professional expertise. Other parents described a balance or tension between their own specific knowledge of their children and their confidence in the therapists' expertise. Parents and therapists described that the decision-making process involved substantial negotiations: The therapists might develop the therapy plan unilaterally, but the parents' played a critical role in their decisions as to whether or not to implement the therapy plan at home. In contrast, parents in the current study reported that they were confident in making healthcare decisions independently of the recommendations of healthcare providers.

Neely-Barnes, Graff, Marcenko & Weber (2008) explored patterns of family involvement in planning and service decisions. Specifically, the roles of family members were contrasted with the roles of case managers. These US researchers surveyed family members (90% parents, 5% siblings) caring for adults with developmental and intellectual disabilities in the family home (n = 547). Siblings did not have a different level of involvement in decision-making involvement from parents, though younger parents were more involved than older parents in decision-making. Family members of individuals with more severe disabilities were more likely to be active in decisions. (There was no mention of the participation of the individuals with ID in decisions about their own services.) With such a large sample this study provides some insight into what might motivate family members of adults with ID to be more actively involved in decisions. These findings also pose an interesting question in light of the findings of the current study: Why would family members of the higher functioning adults with ID be more likely to defer to case managers with regards to service and planning decisions? The current study's participants very likely represent some of the most involved families of adults with ID; nevertheless, none of the parents expressed anything close to deference to anyone else with regards to decisions on behalf of the adult children with ID. The locus of control with regards to the Botox decision (and other decisions they described) was held closely by the participants.

In a qualitative study Canadian researchers explored factors influencing elderly patients' decisions whether or not to proceed with total joint replacement surgery upon their doctors' recommendation (Clark et al., 2004). To explore the decision-making process, these investigators conducted semi-structured interviews with patients who were considered as candidates for total knee or total hip replacement surgery (n = 17). Based on their findings these investigators developed a conceptual approach to the decision-making process. Several of the elements of their model were similar to those in the conceptual framework developed in the present study, including:

- Prior medical/surgical experiences.
- Information sources (physicians and other health care professionals; family; friends; Internet).
- Balancing treatment options based on risk and benefit.
- Watching and waiting.

It is interesting to note that in their conceptual approach to the decision-making process based on semi-structured interviews, Clark et al. did *not* find the patient-physician locus of control to be a central factor in the patients' decision.

Themes.

Agency.

Dutch researchers studied the process of decision-making regarding the care of individuals with various levels of ID residing in a residential facility (Vallenga et al., 2006). These individuals had intractable epilepsy, and the context of the decision was the prevention of injury from falls during seizures. These researchers found that the individuals were essentially excluded from the decision-making process. While 80% (12) of the subjects had mild or moderate ID, none of the residents were explicitly involved in the decision-making process, beyond "being given the opportunity to react to" a fait accompli (Vallenga et al., p. 608). In the current study the narratives were not always clear with regards to the youth's agency. In some cases, it seemed possible that the young adults were indeed responding to faits accomplis: Scott's father and Cathy's mother may have been describing this approach when they stated that they were forthright with these young about impending health-related decisions. There is a clear distinction between encouraging participation in a decision and providing a notice of the results of that decision. However in four of the seven cases the narratives clearly identified youth's explicit participation in the Botox decision. While no conclusions are merited, there is evidence to support a claim that individuals who live in the family home are given more opportunity to exercise decision-making agency than those who live residential settings (Lakin et al., 2008).

How prepared are the parents of young adults with mild ID for their adult children to become autonomous? Researchers in the UK conducted a grounded-theory qualitative study exploring the experiences of adults with mild ID who belonged to a self-advocacy group (n = 8; Beart, Hardy, & Buchan, 2004). Some participants reported that involvement in the self-advocacy group was somehow threatening to family members and professional care providers. Several participants did not disclose their involvement in self-advocacy activities due concerns that parents would disapprove.

Geenen, Powers, & Sells (2003) surveyed parents to explore their expectations of health care providers (HCPs) with regards to preparing youth (ages 13 to 21) with various disabilities during the transition from child-centered to adult-centered health care. Parents of youth with developmental disabilities (DD; n = 165) were compared with a larger group of parents of children with a variety of disabilities (n = 753). These researchers found that the parents of the youth with DD ranked health-related self-care activities as a lower priority than did the other parents. However, there was no description of the level of developmental disabilities of these youth, which makes it difficult to interpret these findings. For example, parents of young adults severe cognitive limitations might be less likely to have expectations of the young adult's participation in health-related self-care.

A recent mixed-methods study of older parents (mean age 63) of adults with ID living in the family home was conducted on Prince Edward Island (Weeks, Nilsson, Bryanton, & Kozma, 2009). Thirty-three mothers participated in semi-structured interviews to explore their concerns regarding their adult children with ID. The mothers described that they hoped that their adult children would receive good quality services and access to employment and activities. There was no mention that mothers identified any goals for their children associated with self-determination or agency. There was limited description of the levels of ID of the adult children; the degree of cognitive impairment might have influenced the mothers' aspirations. It is also possible that a cohort effect related to the mothers' generation (or another demographic variable) resulted in lower expectations for youth's agency. For example, these mothers may not have been exposed to values of normalization and mainstreaming that have become well established in special education programming in recent decades. (Vallenga, Grypdonck, Tan, Lendemeijer, & Boon [2006] speculated that a similar dynamic was responsible for generational differences identified in their study.)

Communication and cognition.

Capacity for decision-making

Parents in the current study described that they determined that some decisions were too critically important or perhaps too complex for the young adults to participate in. Vallenga, Grypdonck, Tan, Lendemeijer, & Boon (2006) also found that, in a Dutch residential facility, decision-making options available to adults with ID were extremely limited when the decision outcome was potentially critical (i.e. head injuries resulting from falls during seizures). VanHooren (2006) described that caregivers would override the choice of individuals with Prader-Willi syndrome when they indulged in overeating behaviors (which are associated with grave consequences in this population).

Complexities of communication.

Researchers recently investigated issues related to conducting focus groups with adults with CP who use augmentative communication devices (ACDs; Hemsley, Balandin, & Togher, 2008). In this qualitative study researchers found that participants made strategic use of non-verbal communication in the focus groups. These researchers described that individuals used non-verbal communication “to gain the attention of the group, to indicate agreement or disagreement, and to add emphasis to a spoken message” (Hemsley, p. 116). The two participants in the current study who used ACDs made even more extensive use of non-verbal communication, though this was also in the presence of parents who were familiar with their extensive non-verbal “vocabularies.”

Ziviani, Lennox, Allison, Lyons & Del Mar (2004) conducted a qualitative study to explore the factors influencing the success of communication between physicians and adults with ID. They conducted semi-structured interviews with three adults with ID, five physicians who care for patients with ID, and nine caregivers or advocates. The physicians emphasized that it is important for an individual with ID to have access to appropriate supports during clinic consultations. It is particularly important for the individual to be accompanied by someone who is familiar with the individual’s unique communication pattern. Individuals with ID, and their advocates, reported that they found it common for physicians to not be willing to take the time and effort to communicate directly with the individual.

Use of supports with cognitive tasks.

Parents in the current study described that they used a strategy of breaking up larger pieces of information into more manageable bits when supporting the young adults in understanding and processing health-related decisions. Investigators studying the capacity of individuals with mild and moderate ID to provide consent for participation in research have used a similar strategy (Arscott et al., 1999; Cea & Fisher, 2003; Dye et al., 2007). Grisso and Appelbaum (1991) have named this strategy *single-unit disclosure*, where each "unit" consists of a single idea or paragraph in the information being reviewed. The parents' strategies differed from the single-unit disclosure method in that the parents would extend the process over the course of days, giving the young adult additional time to consider different aspects of a decision. This is a different process than asking an individual to integrate all of the components of a decision at one sitting. There is no way of determining the effectiveness of these parents' strategy, aside from their convictions that it helps the young adult to better engage with complicated issues.

Information gathered independently.

Clark et al. (2004) described that peers were an important source of information used by adults considering total joint replacement surgery. This phenomenon was also described by participants in the current study, though this is not addressed in other health-related decision-making literature.

Relationships.

As has been previously noted, the literature addressing health-related decisions has emphasized the issue of whether the locus of control rests with the physician, the patient, or is shared. What is overlooked in this discussion is the role of other individuals in supporting the patient's deliberation. While this idea was clearly presented in the case narratives, there is no description of this issue in current research literature. With regard to the issue of informed consent with individuals with ID, Sullivan et al. (2006) recommend that clinicians should "involve family or social support network members to facilitate informed decisions" (p. 1413). In the current study, participants described that they had confidence that the youths' physicians respected their decision-making roles. In a qualitative study, parent guardians described that they had distinctly supportive and trusting relationships with the physicians caring for their adult children with ID (Horrell et al., 2006).

Conclusions

The investigator developed conclusions that are founded on preliminary and thematic findings. The conclusions are also considered in light of related findings from the literature. The investigator's conclusions are stated below (in bullets).

Conceptual framework.

- The conceptual framework of the decision-making process identifies key elements of the process.

The investigator has explored the decision-making process of young adults with cerebral palsy and mild or borderline intellectual disabilities considering Botox therapy for relief from muscle spasticity. Conceptual approaches to health-related decision-making described in the literature have focused on the issue of locus of control. The predominance of paternalism found in many studies of health-related decision-making does not resonate with the investigator's experiences with this population. In the investigator's experience, parents of children with lifelong disabilities are generally quite empowered with regards to their opinions about decisions affecting their children. Only one study has addressed the role of parents of children with disabilities in health-related decision-making (Young 2006). The elements of the decision-making process presented in this conceptual framework help to expand an understanding of the details of this process, beyond the aspect of locus of control. Ideas represented in this conceptual understanding are reinforced by current evidence and recommendations (Clark et al., 2004; Sullivan et al., 2006).

Themes.

The investigator identified themes and sub-themes that convey salient ideas found during analysis of findings in the current study. These themes describe several aspects of decision-making not previously reported in the literature.

Agency.

Anyone who spends time with individuals with cognitive disabilities can observe that the agency of these individuals is constantly threatened. It is not uncommon for the label of intellectual disability to be considered indistinguishable from delimited agency; we often are quick to ask, "Is he his own guardian?," and this type of query implies that absence of legal autonomy is equivalent to absence of agency.

The current literature does not offer a firm foundation for generalization about the attitudes of parents toward the self-determination of their adult children with ID. It is noteworthy that in the current study there are scant examples of parents actively seeking opportunities to expand the agency of their adult children with mild ID. In his widely cited book *Families, Illness, & Disability*, Rolland (1994) proposes that in families of youth with chronic health needs, the often intense family cohesion can be threatened as the youth's autonomy emerges. Parents can be particularly averse to risks that might accompany youth's autonomy if they have faced life-threatening events in the child's past. The family has well-established patterns for managing a variety of health issues and crises that they have navigated through the child's life. It is understandable that parents are distressed at the prospect of helping this adult child to embark on a new, self-determined life.

- There is a spectrum of agency of young adults with mild ID and CP.

A consideration of the decisions described in the case narratives reveals a spectrum of the young adults' agency. This spectrum ranges from legally autonomous and unsupported decision-making (e.g. Tom), to decisions where the parents decided unilaterally on behalf of the young adult. The degree of agency of a young adult with mild ID and CP can be described in four levels:

1. The young adult has legal autonomy without need for support from parents.
2. The parents facilitate the decision, and the youth's role in the decision is explicit. The parents provide supports for the youth in the decision-making process.
3. The parents facilitate the decision, and they value importance of the youth's values and preferences, but the youth has no explicit or active role in the decision-making process. The decision-making event is often described in terms such as, "We decided…."
4. The parents take full control of a decision.

This depiction of an array is not intended to indicate as a generalization that one level is more appropriate than another. Like the Tripartite Ecological Model of self-determination (Abery and Stancliffe, 2003), what is important is for youth to be at a level of his or her preference.

Explicit agency

- Parents of young adults with mild ID and CP may find it challenging to identify and support the young adult’s role in health-related decisions, distinct from the parents’ opinions.

During the interviews the investigator sensed tension in the parent participants when he asked them to specifically describe the young adult’s role in the decision to receive Botox. The investigator carried the assumption that it is desirable for young adults with mild ID to be active in decisions about their own health, which parents perhaps perceived. Yet not all of the parents were prepared to offer clear descriptions or examples of the youth’s participation in the decision.

Implicit agency

- Parents of young adults with mild ID and CP value the young adult’s opinions even when they have not promoted a role for the young adult that is distinct from the parents’ opinions.

In each of these cases it appeared that the parents were well attuned to the young adult’s preferences. The parents and youth described being in accord with regards to decisions: The parents honor the children’s values and preferences when making decisions on their behalf. Yet one can detect an impending conflict within these narratives.

Consider how Bill, who is legally autonomous and lives at home, asked for his mother to participate in the interview. She was quite active in the interview, facilitating his communication and comprehension, and spoke more than Bill did during the interview. The descriptions provided by Bill and his mother made clear that they find Bill to be independent with regards to decision-making. Yet how would Bill negotiate decisions or medical appointments without the fairly intense involvement of his parents? Implicit agency appears vulnerable if the family was no longer intimately involved with day-to-day affairs of young adult.

Scott's parents apparently have not yet come to focus on Scott's role in decision-making. The parents and PCA are so attuned to Scott that they can successfully anticipate his preferences. They know he can raise a vigorous protest if he is opposed to a decision. But there doesn't seem to be an expectation that Scott's agency might diverge in unexpected ways from his parents.

Communication and cognition.

Communication and cognition are key issues related to the youth's involvement in decision-making. The investigator anticipated that these would be important issues in the case narratives. Yet these issues were not extensively described by participants in their narratives. It is reasonable to speculate that the participants didn't elaborate extensively on issues of cognition limitations or impaired communication due to a sense of frustration or embarrassment. Individuals with ID face unique issues of stigma and acceptance in society (Smith, 2008).

Capacity for decision-making

- Parents of young adults with mild ID and CP are hesitant to involve the young adults in decisions considered too risky or complicated.

In the case narratives participants provided two examples of instances where parents estimated the capacity of the young adults' for participation in health-related decisions. Two of the parents described that, in their estimation, some decisions were too consequential or too complicated for the young adult to participate in. The parents did not describe how they had come these conclusions.

This strategy of the parent unilaterally making the most serious decisions without consulting the young adults echoes the way that parents handle decisions of every kind on behalf of a young child. During adolescence, most children are gradually granted a more significant role in decisions, until the time that they are functionally and legally autonomous adults. For example, children with childhood onset diabetes take on increasing responsibilities for managing this condition (Alderson, Sutcliffe, & Curtis, 2006). For youth with ID, this process of becoming autonomous can plateau and stall; they are granted participation in some decisions, but there can be a threshold, beyond which the more serious or complex decisions are made unilaterally by the guardian.

Complexities of communication.

- Communication is commonly a factor that complicates the decision-making process for individuals with CP and mild ID.

Individuals with CP are likely to have some degree of dysarthria (Rapp & Torres, 2000). Many use augmentative communication devices. It can take far more time for them to express themselves than it might take another person. Their communication patterns are often unique, and optimal communication is facilitated by the assistance of someone who knows them. In a health care context these communication issues can have undesirable consequences. Clinicians typically have limited time to spend in consultations. Many health issues are complicated and sensitive in nature, and often decisions must be made expeditiously.

Use of supports with cognitive tasks.

- Individuals with mild ID may benefit from alternate methods of presentation of information to support their participation in decisions.

Participants described that the young adults benefited from various types of support from their parents while developing their decisions. This meant helping the young adult to understand fragments of the decisions, or giving the young adult time to slowly consider the decision. The literature does not provide unequivocal evidence that these methods improve individuals' performance in decision-making. However, parents in the current study described that they would allow the youth to consider a decision over the course of days, rather than soliciting a decision in one sitting, as is typical in clinical and research settings.

Distinguishing communication from cognition.

- Cognitive limitations are not easily distinguished from communication impairments of individuals with ID and CP.

It is often difficult to distinguish communication issues from cognitive issues (Tuffrey-Wijne & McEnhill, 2008). We can never be certain when an individual comprehends what we tell him; we rely on reciprocal communication to confirm an understanding between the two individuals. For instance, if a young adult responds to a query with a statement that does not seem appropriate in the context of the interview, is this because he didn't understand the question? Or is his message getting lost in the complexities of his communication process? When an individual's communication difficulties are confused with limited cognitive abilities, there is a great risk for inappropriate abridgement of the person's autonomy (Wagner, 2003).

Information gathered independently.

- While considering a health-related decision, young adults with mild ID and CP, or their guardians, are likely to seek information on their own, supplementing information provided by health care professionals.

Young adults with CP, along with their families, typically have extensive experiences with involving medical treatment. They also have considerable interaction with other individuals and families with similar life experiences. Subsequently many of these families are inclined to explore various sources of information to aid them in deliberation. These sources of information may be as important or more important that the information or opinions provided by the youth's physician.

(As mentioned above, Scott's parents learned of recent alerts regarding cases of serious adverse effects, including death, resulting from Botox treatments for muscle spasticity in individuals with CP. The use of Botox for this indication is "off-label," meaning that the Food and Drug Administration has not approved the medication for this use. The likelihood of patients developing significant adverse effects from Botox injections are considered quite small, comparable to rare events associated with risk profiles of other treatments (Hurley, 2008; Partikian & Mitchell, 2007).

Relationships.

- While considering a health-related decision, young adults with mild ID and CP, or their guardians, are likely to seek involvement of other people whom they trust.

These individuals are likely to have been through many serious decisions in the course of the young adult's life, and they have come to rely on their own judgment, rather than deferring key decisions to be made by solely upon the recommendation of the physician. Individuals with ID often are supported by several key people. In addition to parents, it may be siblings, extended family members, residential staff, a case manager, school nurse, teacher, or another physician, who might be consulted for a health-related decision.

Recommendations

Choice, *autonomy*, *decision-making*, *independence*, *self-determination*….When considering these closely related concepts, the spirit of *individual agency* can be easily lost in a struggle over semantics. *Agency* can be hard to define, but we recognize it when we witness it. In this study the investigator has attempted to focus on the spirit of agency: How can an individual's preferences be meaningfully solicited when choosing among options that influence the person's well-being? It is common for adults with ID to participate in mundane decisions while being excluded from decisions that will have pervasive impact on their lives. Choosing between two meal options is a far cry from helping to decide whether or not to proceed with a potentially life-altering surgery.

Here the investigator presents recommendations for improved clinical practices that can promote opportunities for increasing participation in health-related decision-making of young adults with mild ID. Below are also recommendations for further research needed to support these practices. Specific recommendations are presented with bullets.

Professional practice.

- Increase expertise among health care professionals in the care of adults with ID.

There has been growing attention in the literature regarding the need for health care professionals who have expertise in the health care issues of individuals with ID (Camus, 2008; Jones et al., 2008; Sullivan et al., 2006; van Schrojenstein Lantman-de Valk, HMJ, 2009; Wallace & Beange, 2008). This literature has primarily focused on knowledge of the particular health issues that are common in the population. It would serve this population well if these experts would also develop skills in promoting the participation in decision-making of individuals with ID.

- Support individuals with ID in the development of decision-making skills.

In developing skills in decision-making skills individuals with ID can expand their self-determination. Heller & Miller (2000) conducted a quasi-experimental study of an intervention intended to promote choice-making and self-advocacy for adults with mild and moderate ID. Compared with a control group (n = 22), the intervention group (n = 38) showed a greater increase in the exercise of choice. Shogren et al. (2006) posit that "encouraging self-determination in health care may well be a strategy for reducing health disparities experienced by people with intellectual disabilities" (p. 109). Abery has developed instruments measuring health-related self-determination (personal communication, 2007).

- Support guardians in recognizing the role of young adults with mild ID in decisions.

Guardians might be reluctant to allow the young adult to voice an opinion that conflicts with their own. Professionals can help guardians to recognize when the young adult's voice is not distinct in a decision. Professionals can encourage guardians to avoid dismissing the youth's ability to be involved in decisions involving serious risk and consequence.

- Assess cognitive capacity of patients with ID.

Sullivan et al. (2006) have recommended that primary care providers learn to assess capacity for consent of their patients with ID. Healthcare professionals have a responsibility to determine whether their patients comprehend information they provide (Wagner, 2003). These professionals can help guardians to explore the capacity for the young adults' participation in decisions of more serious consequence. We often find that health-related decisions only get harder as we get older, e.g. deliberations regarding advance directives or cardiac surgery. When guardians learn to hear the voice of the adult with ID they can feel more confident that they are honoring the individual's wishes when they ultimately make a decision.

- Anticipate the need for adaptations for unique communication patterns.

Sullivan et al. (2006) have recommended that primary care providers provide appropriate adaptations to accommodate the communication patterns of individuals with ID. Particularly challenging is the need for extra time that is often necessary when communicating with these individuals during clinical consultations. Innovative approaches might be appropriate in order to establish effective communication, such as communication aids that target decision-making subjects (Cameron & Murphy, 2007). At Gillette's Lifetime clinic patients with need for communication adaptations are scheduled for a longer appointment time, even though third-party reimbursement does not account for this impact on clinician time and revenue.

- Adapt methods of the presentation of information to accommodate cognitive limitations.

Clinicians can develop alternative ways of presenting decision-making and consent information. Parents in the current study described how they would present information to the youth in ways that helped them to comprehend issues. Researchers have developed an innovative “nominal technique” to elicit views of individuals with mild and moderate ID in focus groups (Tuffrey-Wijne, Bernal, Butler, Hollins, & Curfs, 2007). This technique for exploring decisions on sensitive topics could perhaps be adapted as a method of eliciting an individual’s preferences in a decision-making context. A recent compilation of literature includes a wide variety of ideas and resources for promoting participation in decision-making among individuals with ID (Warner, Mariathasan, Lawton-Smith, & Samele, 2006).

- Develop familiarity with different manifestations of the locus of control in decisions.

The literature regarding the locus of control in health-related decision-making has been addressed extensively. Primary care providers are gaining abilities in handling the nuances of collaborative relationships with their patients with chronic conditions (Bodenheimer et al., 2002). Clinicians can also learn that for individuals with ID, agency is not an all-or-nothing consideration. Even if the individual with ID is not legally autonomous, she may exercise some influence over decisions of which she is the subject.

- Remember to consider important relationships when soliciting a decision.

Patients are likely to include key individuals in the process of deliberation, even though it is usually only the patient and guardian who meet with the health care provider during clinical consultations. It is prudent to be aware the young adult or parent will not always be making the decision in isolation of other people that are important in the young adult's life (Sullivan et al., 2006).

Further research.

- Use care when specifying "intellectual disability".

Currently, it is quite common for individuals with ID to be portrayed in the literature as a monolithic population. In other words, authors apparently assume that these individuals have extensive similarities based on the single fact of a diagnosis of ID (for example, Wullink, Widdershoven, van Schrojenstein Lantman-de Valk, H., Metsemakers, & Dinant, 2009). This approach overlooks the heterogeneity of individuals with the diagnosis *intellectual disability* (Perkins & Small, 2006). It is always important but rarely described that there is found in this population tremendous diversity with regard to:

- Etiology of ID.
- Associated conditions.
- Relative level of ID (i.e. profound, severe, moderate or mild ID).

Meaningful interpretation of data about individuals with ID is difficult when these factors are not described or accounted for. Rather than considering individuals with ID as a population, intellectual disability may be better conceptualized as a variable. The long-demonstrated, often unexamined bias against including individuals with ID in research can perhaps be redressed by better clarifying how the variable of intellectual ability interacts with other variables.

- Specify more carefully the concept of health.

There is a growing body of literature in multiple disciplines addressing health issues of individuals with ID. The utility of this literature is often hindered by the fact that investigators fail to provide a definition of *health* (e.g. Wullink et al., 2009). Too often it is unclear what researchers are investigating when they explore a “health-related” topic, and this confusion is no doubt reflected and compounded by different interpretations of *health* brought to investigations by study subjects. Individuals define *health* differently from one another, and even these personal definitions can vary depending on context (Smith, 1981).

Closing Remarks

Young adults with mild ID and mild cognitive limitations have abilities that should be recognized when health-related decisions are made on their behalf. While they are still closely involved with their parents, these abilities are likely to be subsumed. Yet it is during young adulthood that self-determination abilities, such as decision-making, can be most fruitfully developed. These abilities can serve the individual throughout life, after their parents are no longer central to their lives.

In summarizing the findings of this study and their importance, a key issue remains that has only been mentioned in passing, which is the stigma faced by individuals with intellectual disabilities and mild cognitive limitations. Specific cognitive abilities are increasingly valued and rewarded in today’s society. These abilities are typically brought under a broad conceptualization as *intelligence*. Individuals who have impaired or questionable intelligence are quickly sidelined with a label (e.g. MR, ID, or DD). Individuals with these labels face ridicule, denigration and hostility throughout life.

Professional and academic considerations of intellectual disabilities are not immune from such deprecation. A recent article described an actual hospital ethics committee's consult involving an adult with severe ID with a life threatening cancerous abdominal mass (Baumrucker et al., 2008). The philosophy professor on the committee stated that, because of this patient's "decreased mentation…she has no narrative, no perspective, and no personhood….[She] has inadequate mental capacity to be truly *suffering*….She lacks [the] bare minimum of mental capacity to even suffer or be in pain" (p. 57-58). Neither the co-authors nor the editors made any comment regarding this characterization of the individual as not actually being a person due to the trait of severe intellectual disability.

For individuals with less severe ID the stigma can be particularly bracing, because they can readily perceive when they are being treated differently from their peers. Participants in the current study made little mention of the youths' cognitive limitations, which was likely attributable to self-consciousness. As these young adults knew that they had been recruited for the study due to the attribute of having an intellectual disability, they were no doubt sensitive to the likelihood that the investigator viewed them primarily as members of a marginalized group.

Perhaps a way forward is for professionals, academics and families to consider cognitive abilities as a *difference* rather than as a *disability*. Individuals may have needs of a variety of supports to compensate for cognitive deficits. But everyone requires individualized attention to particular needs at some time or other. We provide better services, and gain more sound knowledge, when we more carefully specify individual differences and avoid unnecessary aggregation of people according to labels.

References

Abery, B. H., & Stancliffe, R. J. (2003). A tripartite-ecological theory of self-determination. In M. L. Wehmeyer, B. J. Abery, D. E. Mithaug & R. J. Stancliffe (Eds.), *Theory in self-determination: Foundations for educational practice* (pp. 43-78). Springfield, IL: Charles C. Thomas.

Alderson, P., Sutcliffe, K., & Curtis, K. (2006). Children as partners with adults in their medical care. *British Medical Journal, 91*(4), 300.

Appelbaum, P. S., & Roth, L. H. (1982). Competency to consent to research: A psychiatric overview. *Archives of General Psychiatry, 39*(8), 951-958.

Arscott, K., Dagnan, D., & Kroese, B. S. (1999). Assessing the ability of people with a learning disability to give informed consent to treatment. *Psychological Medicine, 29*(6), 1367-1375.

Arvio, M., & Sillanpaa, M. (2003). Prevalence, aetiology and comorbidity of severe and profound intellectual disability in Finland. *Journal of Intellectual Disability Research, 47*(Part 2), 108-112.

Ayres, L. (2007). Qualitative research proposals-part V: Rigor in data analysis. *Journal of Wound, Ostomy and Continence Nursing, 34*(5), 489.

Ayres, L. (2000a). Narratives of family caregiving: Four story types. *Research in Nursing & Health, 23*(5), 359-371.

Ayres, L. (2000b). Narratives of family caregiving: The process of making meaning. *Research in Nursing & Health, 23*(6), 424-434.

Ayres, L., Kavanaugh, K., & Knafl, K. A. (2003). Within-case and across-case approaches to qualitative data analysis. *Qualitative Health Research, 13*(6), 871-883.

Bailey, P. H. (1996). Assuring quality in narrative analysis. *Western Journal of Nursing Research, 18*(2), 186-194.

Bamberg, M. (2006). Stories: Big or small: Why do we care? *Narrative Inquiry, 16*(1), 139-147.

Barber, C., Garnham, L., Lovell, S., Camus, H., & Persaud, M. (2008). Galvanising the role of learning disability nursing. *British Journal of Nursing (BJN), 17*(4), S3-S3.

Barron, S., McConkey, R., & Mulvany, F. (2006). Family carers of adult persons with intellectual disabilities on the island of Ireland. *Journal of Policy and Practice in Intellectual Disabilities, 3*(2), 87.

Baumrucker, S. J., Sheldon, J. E., Stolick, M., Morris, G. M., Vandekieft, G., & Harrington, D. (2008). The ethical concept of "best interest". *The American Journal of Hospice & Palliative Care, 25*(1), 56-62.

Bax, M., Goldstein, M., Rosenbaum, P., Leviton, A., Paneth, N., Dan, B., et al. (2005). Proposed definition and classification of cerebral palsy. *Developmental Medicine & Child Neurology, 47*(8), 571-576.

Beart, S., Hardy, G., & Buchan, L. (2004). Changing selves: A grounded theory account of belonging to a self-advocacy group for people with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 17*(2), 91-100.

Betz, C. L. (2004). Transition of adolescents with special health care needs: Review and analysis of the literature. *Issues in Comprehensive Pediatric Nursing, 27*(3), 179-241.

Betz, C. L. (2007). Facilitating the transition of adolescents with developmental disabilities: Nursing practice issues and care. *Journal of Pediatric Nursing: Nursing Care of Children and Families, 22*(2), 103-115.

Betz, C. L., & Redcay, G. (2003). Creating healthy futures: An innovative nurse-managed transition clinic for adolescents and young adults with special health care needs. *Pediatric Nursing, 29*(1), 25-30.

Bodenheimer, T., Lorig, K., Holman, H., & Grumbach, K. (2002). Patient self-management of chronic disease in primary care. *JAMA, 288*(19), 2469.

Bohmer, C. J. M., Taminiau, J. A. J., Klinkenberg-Knol, E. C., & Meuwissen, S. G. M. (2001). The prevalence of constipation in institutionalized people with intellectual disability. *Journal of Intellectual Disability Research, 45*(part 3), 212-218.

Braddock, D., Emerson, E., Felce, D., & Stancliffe, R. J. (2001). Living circumstances of children and adults with mental retardation or developmental disabilities in the United States, Canada, England, and Wales. *Mental Retardation and Developmental Disabilities Research Reviews, 7*(2), 115-121.

Burge, P., Ouellette-Kuntz, H., Isaacs, B., Lunsky, Y., & Undergraduate Medical Education in Intellectual Disabilities Group at Queen's University. (2008). Medical students' views on training in intellectual disabilities. *Canadian Family Physician Medecin De Famille Canadien, 54*(4), 568-9, 569.e1-6.

Cameron, L., & Murphy, J. (2007). Obtaining consent to participate in research: The issues involved in including people with a range of learning and communication disabilities. *British Journal of Learning Disabilities, 35*(2), 113.

Camus, H. (2008). The way forward for learning disability nursing. *British Journal of Nursing (BJN), 17*(4), S18-9.

Cea, C. D., & Fisher, C. B. (2003). Health care decision-making by adults with mental retardation. *Mental Retardation, 41*(2), 78-87.

Charon, R. (2006). *Narrative medicine: Honoring the stories of illness*. New York: Oxford University Press.

Clark, J. P., Hudak, P. L., Hawker, G. A., Coyte, P. C., Mahomed, N. N., Kreder, H. J., et al. (2004). The moving target: A qualitative study of elderly patients' decision-making regarding total joint replacement surgery. *The Journal of Bone and Joint Surgery, 86*(7), 1366.

Degner, L. F., & Sloan, J. A. (1992). Decision making during serious illness: What role do patients really want to play? *Journal of Clinical Epidemiology, 45*(9), 941-950.

Degner, L. F., Sloan, J. A., & Venkatesh, P. (1997). The control preferences scale. *Canadian Journal of Nursing Research, 29*(3), 21-43. (39 ref).

Dye, L., Hare, D. J., & Hendy, S. (2007). Capacity of people with intellectual disabilities to consent to take part in a research study. *Journal of Applied Research in Intellectual Disabilities, 20*(2), 168-174.

Eley, D., Boyes, J., Young, L., Hegney, D., & Hegney, D. E. D. (2009). Accommodation needs for carers of and adults with intellectual disability in regional Australia: Their hopes for and perceptions of the future. *Rural and Remote Health, 9*, 1239.

Emanuel, E. J., & Emanuel, L. L. (1992). Four models of the physician-patient relationship. *Readings in Health Care Ethics, 1*, 40–49.

Entwistle, V. A., & Watt, I. S. (2006). Patient involvement in treatment decision-making: The case for a broader conceptual framework. *Patient Education and Counseling, 63*(3), 268-278.

Fawcett, J. (1999). *The relationship of theory and research* (3rd ed.). Philadelphia: F.A. Davis Company.

Fisher, K. (2004). Health disparities and mental retardation. *Journal of Nursing Scholarship, 36*(1), 48-53.

Fisher, C. B., Cea, C. D., Davidson, P. W., & Fried, A. L. (2006). Capacity of persons with mental retardation to consent to participate in randomized clinical trials. *American Journal of Psychiatry, 163*(10), 1813-1820.

Fujiura, G. T. (1998). Demography of family households. *American Journal of Mental Retardation, 103*, 225-235.

Geenen, S. J., Powers, L. E., & Sells, W. (2003). Understanding the role of health care providers during the transition of adolescents with disabilities and special health care needs. *Journal of Adolescent Health, 32*(3), 225-233.

Gore, C., Johnson, R. J., Caress, A. L., Woodcock, A., & Custovic, A. (2005). The information needs and preferred roles in treatment decision-making of parents caring for infants with atopic dermatitis: A qualitative study. *Allergy, 60*(7), 938-943.

Graham, I. D., & O'Connor, A. M. (2006). Research in shared decision making is growing deeper roots and more branches. *Patient Education and Counseling, 63*(3), 259-261.

Grisso, T., & Appelbaum, P. S. (1991). Mentally ill and non-mentally-ill patients' abilities to understand informed consent disclosures for medication. *Law and Human Behavior, 15*(4), 377-388.

Guba, E., & Lincoln, Y. (2004). Competing paradigms in qualitative research. In S.J. Hesse-Biber and P. Leavy (Eds.), *Approaches to Qualitative Research: A Reader on Theory and Practice* (pp. 17-38). New York: Oxford University Press.

Hahn, J. E., & Marks. (2003). Preface: Intellectual and developmental disabilities. *Nursing Clinics of North America, 38*(2), xi-xiv.

Harris, J. C. (2006). *Intellectual disability: Understanding its development, causes, classification, evaluation, and treatment*. New York: Oxford University Press.

Heller, T., Miller, A. B., Hsieh, K., & Sterns, H. (2000). Later-life planning: Promoting knowledge of options and choice-making. *Mental Retardation, 38*(5), 395-406.

Hemsley, B., Balandin, S., & Togher, L. (2008). 'I've got something to say': Interaction in a focus group of adults with cerebral palsy and complex communication needs. *Augmentative and Alternative Communication, 24*(2), 110-122.

Horan, P., Doran, A., & Timmins, F. (2004a). Exploring Orem's self-care deficit nursing theory in learning disability nursing: Philosophical parity paper: Part 1. *Learning Disability Practice, 7*(4), 28-33.

Horan, P., Doran, A., & Timmins, F. (2004b). Exploring Orem's self-care model in learning disability nursing: Practical application paper: Part 2. *Learning Disability Practice, 7*(4), 33-37.

Horrell, S. C. V., MacLean, W. E. J., & Conley, V. M. (2006). Patient and Parent/Guardian perspectives on the health care of adults with mental retardation. *Mental Retardation, 44*(4), 239-248.

Hudson, B. (2003). From adolescence to young adulthood: The partnership challenge for learning disability services in England. *Disability & Society, 18*(3), 259-76.

Hudson, P., Quinn, K., O'Hanlon, B., & Aranda, S. (2008). Family meetings in palliative care: Multidisciplinary clinical practice guidelines. *BMC Palliative Care, 7*, 12.

Hurley, D. (2008). Neurologists' reaction varies widely to FDA safety alert on botulinum toxin for cerebral palsy. *Neurology Today, 8*(6), 5.

Iacono, T., & Davis, R. (2003). The experiences of people with developmental disability in emergency departments and hospital wards. *Research in Developmental Disabilities, 24*(4), 247-264.

Iacono, T., Davis, R., Humphreys, J., & Chandler, N. (2003). GP and support people's concerns and priorities for meeting the health care needs of individuals with developmental disabilities: A metropolitan and non-metropolitan comparison. *Journal of Intellectual & Developmental Disability, 28*(4), 353-368.

Jansen, D. E., Krol, B., Groothoff, J. W., & Post, D. (2004). People with intellectual disability and their health problems: A review of comparative studies. *Journal of Intellectual Disability Research, 48*(Pt 2), 93-102.

Jones, M. C., McLafferty, E., Walley, R., Toland, J., & Melson, N. (2008). Inclusion in primary care for people with intellectual disabilities: Gaining the perspective of service user and supporting social care staff. *Journal of Intellectual Disabilities, 12*(2), 93-109.

Kelly, A. M., Kratz, B., Bielski, M., & Rhinehart, P. M. (2002). Implementing transitions for youth with complex chronic conditions using the medical home model. *Pediatrics, 110*(6): Suppl), 1322-7.

Keselman, A., Logan, R., Smith, C. A., Leroy, G., & Zeng-Treitler, Q. (2008). Developing informatics tools and strategies for consumer-centered health communication. *Journal of the American Medical Informatics Association, 15*(4), 473-483.

Keywood, K., & Flynn, M. (2006). Healthcare decision-making by adults with learning disabilities: Ongoing agendas, future challenges. *Psychiatry, 5*(10), 360-362.

Kiesler, D. J., & Auerbach, S. M. (2006). Optimal matches of patient preferences for information, decision-making and interpersonal behavior: Evidence, models and interventions. *Patient Education and Counseling, 61*(3), 319-341.

Koussoulakos, S. (2009). Botulinum neurotoxin: The ugly duckling. *European Neurology, 61*(6), 331-342.

Kozma, C., & Mason, S. (2003). Survey of nursing and medical profile prior to deinstitutionalization of a population with profound mental retardation. *Clinical Nursing Research, 12*(1), 8-22.

Lakin, K. C., Doljanac, R., Byun, S. Y., Stancliffe, R., Taub, S., & Chiri, G. (2008). Choice-making among Medicaid HCBS and ICF/MR recipients in six states. *American Journal of Mental Retardation : AJMR, 113*(5), 325-342.

Lancet. (2009). Disability: Beyond the medical model. *The Lancet, 374*(9704), 1793.

Larson, S. A., Lakin, K. C., Anderson, L., Kwak, N., Lee, J. H., & Anderson, D. (2001). Prevalence of mental retardation and developmental disabilities: Estimates from the 1994/1995 national health interview survey disability supplements. *American Journal on Mental Retardation, 106*(3), 231-252.

Leonard, H., & Wen, X. (2002). The epidemiology of mental retardation: Challenges and opportunities in the new millennium. *Mental Retardation & Developmental Disabilities Research Reviews, 8*(3), 117-134.

Liptak, G. S. (2008). Health and well being of adults with cerebral palsy. *Current Opinion in Neurology, 21*(2), 136-142.

Llewellyn, P., & Northway, R. (2007). The views and experiences of learning disability nurses concerning their advocacy education. *Nurse Education Today, 27*(8), 955-963.

Lunsky, Y., Straiko, A., & Armstrong, S. (2003). Women be healthy: Evaluation of a women's health curriculum for women with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 16*(4), 247-253.

Makoul, G., & Clayman, M. L. (2006). An integrative model of shared decision making in medical encounters. *Patient Education and Counseling, 60*(3), 301-312.

Marks, B. A., & Heller, T. (2003). Bridging the equity gap: Health promotion for adults with intellectual and developmental disabilities. *Nursing Clinics of North America, 38*(2), 205-228.

Martin, G., Philip, L., Bates, L., & Warwick, J. (2004). Evaluation of a nurse led annual review of patients with severe intellectual disabilities, needs identified and needs met, in a large group practice. *Journal of Intellectual Disabilities, 8*(3), 235.

Mazor, K.M., Baril, J., Dugan, E., Spencer, F., Burgwinkle, P., & Gurwitz, J.H. (2007). Patient education about anticoagulant medication: Is narrative evidence or statistical evidence more effective? *Patient Education and Counseling, 69*, 145-157.

McCallion, P., & McCarron, M. (2004). Ageing and intellectual disabilities: A review of recent literature. *Current Opinion in Psychiatry, 17*(5), 349.

McCance, T. V., McKenna, H. P., & Boore, J. R. (2001). Exploring caring using narrative methodology: An analysis of the approach. *Journal of Advanced Nursing, 33*(3), 350-356.

McCormick, A., Brien, M., Plourde, J., Wood, E., Rosenbaum, P., & McLean, J. (2007). Stability of the gross motor function classification system in adults with cerebral palsy. *Developmental Medicine and Child Neurology, 49*(4), 265-269.

McDowell, I. (2006). *Measuring health: A guide to rating scales and questionnaires* (3rd edition ed.). New York: Oxford University Press.

Mead, N., & Bower, P. (2000). Patient-centredness: A conceptual framework and review of the empirical literature. *Social Science & Medicine (1982), 51*(7), 1087-1110.

Mishler, E. G. (1991). *Research interviewing: Context and narrative* Harvard University Press.

Murphy, C. C., Yeargin-Allsopp, M., Decoufle, P., & Drews, C. D. (1995). The administrative prevalence of mental retardation in 10-year-old children in metropolitan Atlanta, 1985 through 1987. *American Journal of Public Health, 85*(3), 319-323.

Murray, E., Charles, C., & Gafni, A. (2006). Shared decision-making in primary care: Tailoring the Charles et al. model to fit the context of general practice. *Patient Education and Counseling, 62*(2), 205-211.

Murray, E., Pollack, L., White, M., & Lo, B. (2007). Clinical decision-making: Patients' preferences and experiences. *Patient Education and Counseling, 65*(2), 189-196.

National Collaborative on Workforce and Disability. Retrieved December 29, 2009 from http://www.ncwd-youth.info/ .

Neely-Barnes, S., Carolyn Graff, J., Marcenko, M., & Weber, L. (2008). Family decision making: Benefits to persons with developmental disabilities and their family members. *Intellectual & Developmental Disabilities, 46*(2), 93-105.

Nehring, W. M., & Poindexter, A. R. (2005). Epidemiology of intellectual and developmental disabilities. In W. M. Nehring (Ed.), *Core curriculum for specializing in intellectual and developmental disability: A resource for nurses and other health care professionals* (pp. 47-54). Boston: Jones and Bartlett.

O'Donnell, M., & Hunskaar, S. (2007). Preferences for involvement in treatment decision-making generally and in hormone replacement and urinary incontinence treatment decision-making specifically. *Patient Education and Counseling, 68*(3), 243-251.

Oliver, P. C., Piachaud, J., Done, D. J., Regan, A., Cooray, S. E., & Tyrer, P. J. (2003). Difficulties developing evidence-based approaches in learning disabilities. *Evidence-Based Mental Health, 6*(2), 37-39.

Ouellette-Kuntz, H. (2005). Understanding health disparities and inequities faced by individuals with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 18*(2), 113-121.

Partikian, A., & Mitchell, W. G. (2007). Iatrogenic botulism in a child with spastic quadriparesis. *Journal of Child Neurology, 22*(10), 1235-1237.

Patja, K., Molsa, P., & Iivanainen, M. (2001). Cause-specific mortality of people with intellectual disability in a population-based, 35-year follow-up study. *Journal of Intellectual Disability Research, 45*(part 1), 30-40.

Perkins, E. A., & Small, B. J. (2006). Aspects of cognitive functioning in adults with intellectual disabilities. *Journal of Policy and Practice in Intellectual Disabilities, 3*(3), 181.

Phillips, A., Morrison, J., & Davis, R. W. (2004). General practitioners' educational needs in intellectual disability health. *Journal of Intellectual Disability Research, 48*(Pt 2), 142-149.

Pidcock, F. S. (2004). The emerging role of therapeutic botulinum toxin in the treatment of cerebral palsy. *Journal of Pediatrics, 145*(2): Supplement), S33-5.

Poirier, S., & Ayres, L. (1997). Endings, secrets, and silences: Overreading in narrative inquiry. *Research in Nursing & Health, 20*(6), 551-557.

Prabhala, A. (2007, February). *Mental Retardation* is no more – new name is *Intellectual and Developmental Disabilities*. AAIDD News. Available from http://www.aamr.org (retrieved December 31, 2007).

President's Commission for the Study of Ethical Problems in Medicine and Biomedical and Behavioral Research. (1982). *Making health care decisions: The ethical and legal implications of informed consent in the patient-practitioner relationship*. Retrieved December 27, 2007 from https://idea.iupui.edu/dspace .

Pyke-Grimm, K. A., Degner, L., Small, A., & Mueller, B. (1999). Preferences for participation in treatment decision making and information needs of parents of children with cancer: A pilot study. *Journal of Pediatric Oncology Nursing : Official Journal of the Association of Pediatric Oncology Nurses, 16*(1), 13-24.

Ramachandran, M., & Eastwood, D. M. (2006). Botulinum toxin and its orthopaedic applications. *Journal of Bone & Joint Surgery - British Volume, 88*(8), 981-987.

Rapp, C. E.,Jr, & Torres, M. M. (2000). The adult with cerebral palsy. *Archives of Family Medicine, 9*(5), 466-472.

Reilly, S., & Morgan, A. (2008). Dysphagia is prevalent in children with severe cerebral palsy. *Developmental Medicine and Child Neurology, 50*(8), 567.

Richards, L. (2004). Validity and reliability? Yes! Doing it in software. Paper presentation to the Conference on Strategies in Qualitative Research with QSR Software, University of Durham, Durham, UK. Retrieved November 11, 2008 from http://www.lynrichards.org/Qual_Writg.htm.

Riessman, C. K. (1993). *Narrative analysis* Sage Pubns.

Riessman, C. K. (2007). *Narrative methods for the human sciences* Sage Publications (CA).

Rolland, J. S. (1994). *Families, illness, and disability: An integrative treatment model*. New York, NY: Basic Books.

Rosen, D. S., Blum, R. W., Britto, M., Sawyer, S. M., Siegel, D. M., & Society for Adolescent Medicine. (2003). Transition to adult health care for adolescents and young adults with chronic conditions: Position paper of the society for adolescent medicine. *Journal of Adolescent Health, 33*(4), 309-11. (10 ref).

Rosenbaum, P., & Stewart, D. (2007). Perspectives on transitions: Rethinking services for children and youth with developmental disabilities. *Archives of Physical Medicine & Rehabilitation, 88*(8), 1080-1082.

Rubin, H., & Rubin, I. (1995). *Qualitative interviewing. The art of hearing data*. Thousand Oaks, CA: Sage Publications.

Salvador-Carulla, L., & Saxena, S. (2009). Intellectual disability: Between disability and clinical nosology. *Lancet, 374*(9704), 1798-1799.

Sandelowski, M. (1991). Telling stories: Narrative approaches in qualitative research. *Image--the Journal of Nursing Scholarship, 23*(3), 161-166.

Say, R., Murtagh, M., & Thomson, R. (2006). Patients' preference for involvement in medical decision making: A narrative review. *Patient Education and Counseling, 60*(2), 102-114.

Scal, P., & Ireland, M. (2005). Addressing transition to adult health care for adolescents with special health care needs. *Pediatrics, 115*(6), 1607.

Scheepers, M., Kerr, M., O'Hara, D., Bainbridge, D., Cooper, S. A., Davis, R., et al. (2005). Reducing health disparity in people with intellectual disabilities: A report from health issues special interest research group of the international association for the scientific study of intellectual disabilities. *Journal of Policy and Practice in Intellectual Disabilities, 2*(3-4), 249-255.

Seltzer, M. M., & Krauss, M. W. (2001). Quality of life of adults with mental retardation/developmental disabilities who live with family. *Mental Retardation and Developmental Disabilities Research Reviews, 7*(2), 105-114.

Sheerin, F. K., & McConkey, R. (2008). Frontline care in irish intellectual disability services: The contribution of nurses and non-nurse care staff. *Journal of Intellectual Disabilities : JOID, 12*(2), 127-141.

Sheridan, S. L., Harris, R. P., Woolf, S. H., & Shared Decision-Making Workgroup of the U.S. Preventive Services Task Force. (2004). Shared decision making about screening and chemoprevention: A suggested approach from the U.S. preventive services task force. *American Journal of Preventive Medicine, 26*(1), 56-66.

Shogren, K. A., Wehmeyer, M. L., Reese, R. M., & O'Hara, D. (2006). Promoting self-determination in health and medical care: A critical component of addressing health disparities in people with intellectual disabilities. *Journal of Policy and Practice in Intellectual Disabilities, 3*(2), 105-113.

Smith, J. A. (1981). The idea of health: A philosophical inquiry. *Advances in Nursing Science, 3*(3), 43.

Smith, J. D. (2008). Trading one myth for another?: With apologies to Dr. Brabner. *Intellectual and Developmental Disabilities, 46*(2), 142.

Stake, R. E. (1995). *The art of case study research*. Thousand Oaks, CA: Sage Publications Inc.

Stake, R. E. (2005). Qualitative case studies. In N. K. Denzin, & Y. S. Lincoln (Eds.), *The Sage handbook of qualitative research* (3rd ed., pp. 443-466). Thousand Oaks, CA: SAGE Publications.

Stake, R. E. (2006). *Multiple case study analysis*. New York: Guilford Press.

Steiner, J.F. (2007). Using stories to disseminate research: The attributes of representative stories. *Journal of General Internal Medicine, 2007, 22*, 1603-1607.

Strauss, D., Shavelle, R., Reynolds, R., Rosenbloom, L., & Day, S. (2007). Survival in cerebral palsy in the last 20 years: signs of improvement? *Developmental Medicine and Child Neurology, 49*(2), 86-92.

Sullivan, W. F., Heng, J., Cameron, D., Lunksy, Y., Cheetham, T., Hennen, B., et al. (2006). Consensus guidelines for primary health care of adults with developmental disabilities. *Canadian Family Physician, 52*, 1410-1418.

Sutherland, G., Couch, M. A., & Iacono, T. (2002). Health issues for adults with developmental disability. *Research in Developmental Disabilities, 23*(6), 422-445.

Swanson, K. A., Bastani, R., Rubenstein, L. V., Meredith, L. S., & Ford, D. E. (2007). Effect of mental health care and shared decision making on patient satisfaction in a community sample of patients with depression. *Medical Care Research and Review : MCRR, 64*(4), 416-430.

Symons, F. J., Rivard, P. F., Nugent, A. C., & Tervo, R. C. (2006). Parent evaluation of spasticity treatment in cerebral palsy using botulinum toxin type A. *Archives of Physical Medicine & Rehabilitation, 87*(12), 1658-1660.

Thompson, J. R., Bradley, V. J., Buntinx, W. H. E., Schalock, R. L., Shogren, K. A., Snell, M. E., et al. (2009). Conceptualizing supports and the support needs of people with intellectual disability. *Intellectual and Developmental Disabilities, 47*(2), 135-146.

Tuffrey-Wijne, I., Bernal, J., Butler, G., Hollins, S., & Curfs, L. (2007). Using nominal group technique to investigate the views of people with intellectual disabilities on end-of-life care provision. *Journal of Advanced Nursing, 58*(1), 80-89.

Tuffrey-Wijne, I., & McEnhill, L. (2008). Communication difficulties and intellectual disability in end-of-life care. *International Journal of Palliative Nursing, 14*(4), 189-194.

Turk, Geremski, C. A., Rosenbaum, P. F., & Weber, R. J. (1997). The health status of women with cerebral palsy. *Archives of Physical Medicine and Rehabilitation, 78*(12): Suppl 5), S10-7.

Tyler, D. O., & Horner, S. D. (2008). Family-centered collaborative negotiation: A model for facilitating behavior change in primary care. *Journal of the American Academy of Nurse Practitioners, 20*(4), 194-203.

Tymchuk, A. J., Lakin, K. C., & Luckasson, R. (2001). *The forgotten generation: The status and challenges of adults with mild cognitive limitations* Baltimore, MD: Paul H. Brookes Pub. Co.

U.S. Department of Health and Human Services (2002). *Closing the gap: A national blueprint to improve the health of persons with mental retardation*. Available from http://www.surgeongeneral.gov/ (retrieved December 19, 2007).

U.S. Department of Health and Human Services (2005). *The Surgeon General's call to action to improve the health and wellness of persons with disabilities.* Available from http://www.surgeongeneral.gov/ (retrieved December 19, 2007).

Vallenga, D., Grypdonck, M. H., Tan, F. I., Lendemeijer, B. H., & Boon, P. A. (2006). Decision-making about risk in people with epilepsy and intellectual disability. *Journal of Advanced Nursing, 54*(5), 602-611.

van Hooren, R. H., Widdershoven, G. A., Candel, M. J., van den Borne, B. W., & Curfs, L. M. (2006). Between control and freedom in the care for persons with Prader-Willi syndrome: An analysis of preferred interventions by caregivers. *Patient Education and Counseling, 63*(1-2), 223-231.

van Schrojenstein Lantman-de Valk, HMJ. (2009). Healthy persons with intellectual disabilities in an inclusive society. *Journal of Policy and Practice in Intellectual Disabilities, 6*(2), 77-80.

Wagner, L. C. B. (2003). Clinical ethics in the context of language and cognitive impairment: Rights and protections. *Seminars in Speech and Language, 24*(4), 275-84, 1-8.

Waldman, H. B., & Perlman, S. P. (2006). Mandating education of dental graduates to provide care to individuals with intellectual and developmental disabilities. *Mental Retardation, 44*(3), 184-188.

Wallace, R. A., & Beange, H. (2008). On the need for a specialist service within the generic hospital setting for the adult patient with intellectual disability and physical health problems. *Journal of Intellectual & Developmental Disability, 99999*(1), 1-8.

Walsh, P. N. (2008). Health indicators and intellectual disability. *Current Opinion in Psychiatry, 21*(5), 474-478.

Warner, L., Mariathasan, J., Lawton-Smith, S., & Samele, C. (2006). Choice literature review. *London: Kings Fund/Sainsbury Centre for Mental Health,*

Weeks, L. E., Nilsson, T., Bryanton, O., & Kozma, A. (2009). Current and future concerns of older parents of sons and daughters with intellectual disabilities. *Journal of Policy and Practice in Intellectual Disabilities, 6*(3), 180-188.

Winterbottom., A., Bekker, H.L., Conner, M., & Mooney, A. (2008). Does narrative information bias individual’s decision making? A systematic review. *Social Science & Medicine, 67*, 2079-2088.

Wong, J. G., Clare, C. H., Holland, A. J., Watson, P. C., & Gunn, M. (2000). The capacity of people with a 'mental disability' to make a health care decision. *Psychological Medicine, 30*(2), 295-306.

Wong, J. G., Clare, I. C., Gunn, M. J., & Holland, A. J. (1999). Capacity to make health care decisions: Its importance in clinical practice. *Psychological Medicine, 29*(2), 437-446.

Wullink, M., Widdershoven, G., van Schrojenstein Lantman-de Valk, H., Metsemakers, J., & Dinant, G. (2009). Autonomy in relation to health among people with intellectual disability: A literature review. *Journal of Intellectual Disability Research, 53*(9), 816-826.

Yeargin-Allsopp, M., van Naarden, B.K., Doernberg, N.S., Benedict, R.E., Kirby, R.S., & Durkin, M.S. (2008). Prevalence of cerebral palsy in 8-year-old children in three areas of the United States in 2002: a multisite collaboration. *Pediatrics, 121*(53), 547-554.

Yin, R. K. (2003). *Case study research: Design and methods*. Thousand Oaks, CA: SAGE Publications.

Young, B., Moffett, J. K., Jackson, D., & McNulty, A. (2006). Decision-making in community-based paediatric physiotherapy: A qualitative study of children, parents and practitioners. *Health & Social Care in the Community, 14*(2), 116-124.

Zier, J. L., Rivard, P. F., Krach, L. E., & Wendorf, H. R. (2008). Effectiveness of sedation using nitrous oxide compared with enteral midazolam for botulinum toxin A injections in children. *Developmental Medicine & Child Neurology, 50*(11), 854-858.

Ziviani, J., Lennox, N., Allison, H., Lyons, M., & Del Mar, C. (2004). Meeting in the middle: Improving communication in primary health care consultations with people with an intellectual disability. *Journal of Intellectual & Developmental Disability, 29*(3), 211-225.

Zoffmann, V., Harder, I., & Kirkevold, M. (2008). A person-centered communication and reflection model: Sharing decision-making in chronic care. *Qualitative Health Research, 18*(5), 670.

Appendix A – Interview Guide

Ground Rules

Following completion of the consent process I will describe ground rules for the interviews:

- "This interview will last no more than one hour. You can also ask to stop at any time."
- "When a parent and child are interviewed together, it's only natural to want to interrupt or correct each other. But for this study it will be very helpful for me if you try to take turns. Please communicate one person at a time. This helps me to be clear about what each of you knows or believes."
- "Please don't speak on behalf of one another. But please let me know if you think I am misunderstanding either of you."

Main questions, followed by probes, and follow-up questions

Note: Each question will first be directed to the young adult and then to the parent.

Main question 1

Young adult: "You came to the clinic for Botox treatments last week. Tell me about how it was decided that you would get the Botox treatment."

Parent: "[Young adult] came to the clinic for Botox treatments last week. Tell me about how it was decided that [she or he] would get the Botox treatment."

Potential probe questions for those **with prior Botox** treatments:

- Tell me, if you can remember, how you first heard of the Botox treatment.
- What sources of information were important in making this decision?
- Did you receive any written information?

- Did you try to find information on your own that wasn't given to you by the clinic staff? [E.g. on the internet.]
- Did you find any of the information useful?
- How well did the information you received in the past prepare you for your previous Botox injection(s)?
- Did you feel that the information accurately described (young adult's) experience?
- Tell me how the information was helpful or not helpful in helping you to make the decision.
- How did you use the information? [E.g. read them together, by yourself.]
- Tell me how you knew that this was the "right" treatment for [young adult].

Potential probe questions for those **naïve to Botox**:

- Tell me, if you can remember, how you first heard of the Botox treatment.
- What sources of information were important in making this decision?
- Did you receive any written information?
- Did you try to find information on your own that wasn't given to you by the clinic staff? [E.g. on the internet.]
- Did you find any of the information useful?
- Tell me how the information was helpful or not helpful in helping you to make the decision.
- How did you use the information? [E.g. read them together, by yourself.]
- Did you learn anything from talking with other people who had already received Botox?
- Tell me how you knew that this was the "right" treatment for [young adult].

Main question 2

Young adult: How was the decision to get Botox treatment like other important decisions you have been involved in? How was it different?

Parent: How was the decision to get Botox treatment like other important decisions that you and [young adult] have been involved in? How was it different?

Potential probe questions for **all participants**:

- Please describe how [young adult] might have been involved in making other significant decisions in [his/her] life, for example, choosing where to live, what kind of job to take, etc.
    - Young adult: Tell me if you have been involved in making other significant decisions in your life.
    - Parent: Tell me if [young adult] has been involved in making other significant decisions in [his/her] life.
- Please describe if [young adult] has been involved in making other health-related decisions in [his/her] life.
    - Young adult: Tell me if you have been involved in making other health-related decisions in your life.
    - Parent: Tell me if [young adult] has been involved in making other health-related decisions in [his/her] life.
- How was the decision about Botox similar or different from these other decisions?

Main question 3

Young adult: Tell me how getting Botox helps you feel better?

Parent: Tell me how getting Botox helps [young adult] feel better?

Potential probe questions for those with **prior Botox treatments**:

- Why were you considering getting the Botox injections at this time?
    - young adult: Why were you considering getting the Botox injections at this time?

  - Parent: Why was [young adult] considering getting the Botox injections at this time?
- What are the main physical sensations or symptoms that the young adult experiences related to muscle spasticity? --OR-- What is it about the way you feel that makes you consider Botox at this time?
  - young adult: What are the main physical sensations or symptoms that you experience related to muscle spasticity?
  - Parent: What are the main physical sensations or symptoms that [young adult] experiences related to muscle spasticity?
- Can you describe how you talk about these sensations or symptoms (e.g. only when they are really severe, just before going to the clinic appointment, etc.)?
- [*Quantification*.] How did you figure out *how much* discomfort from muscle spasms there was? (E.g. compared to previous times, using a formal or informal for rating pain like "mild / moderate / severe discomfort.)
- How did you know that the Botox was working, or not working, after the last treatment?
- What were some of the bad or uncomfortable things about getting the Botox injections?
- Was there anything that surprised you about the injections, that you weren't prepared for?
- Did the clinic staff do anything that made you feel that it would be ok to get the injections the next time? [Coping -]

Potential probe questions for those **naïve to Botox**:

- Why were you considering getting the Botox injections at this time?
  - young adult: Why were you considering getting the Botox injections at this time?
  - Parent: Why was [young adult] considering getting the Botox injections at this time?

- What are the main physical sensations or symptoms that the young adult experiences related to muscle spasticity? --OR-- What is it about the way you feel that makes you consider Botox at this time?
    - young adult: What are the main physical sensations or symptoms that you experience related to muscle spasticity?
    - Parent: What are the main physical sensations or symptoms that [young adult] experiences related to muscle spasticity?
- Can you describe how you talk about these sensations or symptoms (e.g. only when they are really severe, just before going to the clinic appointment, etc.)?
- [*Quantification*.] How did you figure out *how much* discomfort from muscle spasms there was? (E.g. compared to previous times, using a formal or informal for rating pain like "mild / moderate / severe discomfort.)

Main question 4

Young adult: What did you think were the good things about getting Botox? What are some of the bad things about getting the Botox?

Parent: What did you think were the good things about getting Botox? What are some of the bad things about getting the Botox?

Potential probe questions for those with **prior Botox treatments**:

- What were the benefits (if any) that [young adult] experienced after the last Botox injections?
    - young adult: What were the benefits, if any, that you experienced after the last Botox injections?
    - Parent: What were the benefits, if any, that [young adult] experienced after the last Botox injections?
- What was bad or hard about receiving the Botox injections?
- Can you describe some of the most important benefits and risks involved in deciding to have the Botox treatment?

Potential probe questions for those **naïve to Botox**:

- What would be the benefits of getting Botox injections?
- What would be hard or risky about getting the Botox injections?
- Can you describe some of the most important benefits and risks involved in deciding to have the Botox treatment?

Main question 5

Young adult: Tell me about how you talked with each other about getting the Botox treatment.

Parent: Tell me about how you talked with each other about getting the Botox treatment.

Potential probe questions for **all participants**:

- How did you talk about how big a problem the muscle spasticity was? [E.g. conversations specifically about the spasticity, scattered comments.]
- How did you prepare for the clinic appointment when you talked about the Botox treatment? [E.g. data gathering, writing down questions.]
- Did (parent) and (young adult) have different responsibilities in preparing for the clinic appointment? What were these different responsibilities?
- How did (parent) and (young adult) collaborate on this decision? In other words, how did you work together to figure out what you were going to decide about the Botox treatment?
- How did you collaborate with the physician on this decision? In other words, how did you work together with the physician to figure out what you were going to decide about the Botox treatment?
- How would you describe the effectiveness of the communication between you two about the decision to get the Botox treatment? In other words, do you feel that it was easy to understand each other when it came to deciding about the Botox treatment?
- In what ways was your conversation with the doctor helpful to making this decision?

Main question 6

Young adult: Who else helped you to decide about getting the Botox?

Parent: Who else helped you to decide about getting the Botox?

Potential probe questions for **all participants**:

- Tell me about how important it might have been to deciding to get the Botox treatment to know the doctor who would be providing the treatment.
- In addition to the doctor, do you remember talking with anyone else about your decision to get the Botox treatment? [E.g. teachers, therapists, nurses, other doctors.]
- How important were these other individuals in the decision-making process?
- How important were your doctor or nurse practitioner's opinions and recommendations about getting the Botox treatment? In other words, was the doctor's opinion one of the most important factors in making the decision about the Botox?

Main question 7

Young adult: How was the decision made about getting the Botox?

Parent: How was the decision made about getting the Botox?

Potential probe questions for **all participants**:

- Tell me why it is **important** to you who makes the decision about the Botox treatment. In other words, why does it matter whether the decision is made by you (parent), you (young adult) or by the doctor?
- Tell me if you were involved in the decision as much as would like to be.
- Did you recognize that you were making a decision to get the Botox treatment?

Appendix B

Case Report Sample: Mark

Theme	Subthemes			Data from narrative
Goals	Functional benefit			Mark answered the opening question by saying that he was tight in his arms and legs.
	Pain relief			*- no data from narrative -*
Information	Information content			*- no data from narrative -*
		Potential benefits of Botox		*- no data from narrative -*
			Functional benefits of Botox	*- no data from narrative -*
			Pain relieving benefits of Botox	*- no data from narrative -*
		Potential risks of Botox		
			Pain & distress	*- no data from narrative -*
			Muscle weakness	43:00 Mother says that she has seen other kids become limp from Botox, so she realizes how limp and weak Mark could become.
			Alerts – serious risk	Father recalls first learning about Botox and being aware that it was a *toxin* that was being injected. 7:00
		Treatment options		• Mark decided to go with Botox as an alternative to the intensive PT. • He is going ahead with a trial of baclofen. 2:45 He wouldn't need to return to clinic every 3 months ("6 months" for ITBP). • MASSAGE: Mother. 4:20 – "If I had money, we would do

Theme	Subthemes			Data from narrative
				massage, every day….[Botox] is totally paid for, and it is effective. But the massage, I think, would probably help just as much, and it would be less invasive. And cheaper for them." "And nicer for Mark." • 4:43. Mom "We never stop looking for options." • 6:15 – RE: ITBP – 19:10. F: Well, I think, it's what's available. What we understand is available. What are people asking and what are they saying that the benefit is going to be. And then it comes down to weighing what is feasible, what is financially possible, what is time-constraint possible, and what will have the largest potential benefit? There's a million things you can do, but not all of them have big benefits. And when they would want us to—for instance—work on speech therapy when he was in second grade, well, there were so many other things to do that had a bigger benefit than that— • 52:20 – Mother commented that without the Botox Mark would very likely have had more surgeries.
	Information sources			*- no data from narrative -*
		Textual information		*- no data from narrative -*
			Independently found	• 48:08 Mother: "We're here to do the research on the computer, and talk to people,

Theme	Subthemes			Data from narrative
				and then give our input.” • Parents state that they haven’t looked into newer information about the risks of Botox. Father, 10:00 “Probably not a good thing, to not keep researching.”
			Provided by MD / others	MD had originally provided written information.
		Verbal information		
			Verbal info from MDs	- *no data from narrative* -
			Verbal info from peers	12:15. Conductive education therapists were opposed to Botox. They wanted the children to learn to use the high muscle tone rather than becoming too loose. 6:00 Didn’t know anyone else who had gotten Botox when Mark started. 32:10 - friend with CP doesn’t have spasticity issues.
		Prior experience		• Mother. Mark is uncomfortable “Botox does help, so he puts up with it.” • 15:00 Parents describe dramatic benefits of Botox. “Put your arms down, put your arms down. “ 15:30. Marks elbows would be fully flexed, with his forearms folded up on his upper arms, with his hands at his shoulders. Mom: “It’s really hard [for Mark] to do stuff.” • 14:35 Mark: I could see it. Cause I was much looser than before the first time... I was much looser after I had Botox. • 15:00. Parents describe how obvious the benefits are one

Theme	Subthemes			Data from narrative
				week after the Botox injections.
Relation-ships	Youth – Parent			*- no data from narrative -*
	Physician			12:35. Mark described that when they consulted with an MD about the Botox injections, and the concerns about loosing the benefits of high muscle tone he stated that he wasn't sure that it was appropriate to start Botox. "I went to the doctor... that does this, and it's like, he said, "I don't know!" , which was actually pretty good!" Mark and his mother appreciated the physician's openness to the concerns raised by conductive education therapists.
	Others			30:40 – Mother talks with her sister who is a chiropractor.
Deliber-ation	Balancing treatment options			19:10. F: Well, I think, it's what's available. What we understand is available. What are people asking and what are they saying that the benefit is going to be. And then it comes down to weighing what is feasible, what is financially possible, what is time-constraint possible, and what will have the largest potential benefit? There's a million things you can do, but not all of them have big benefits. And when they would want us to—for instance—work on speech therapy when he was in second grade, well, there were so many other things to do that had a bigger benefit than that— F: So we left that to the school, and we really didn't work on speech therapy at home, even though they would have liked another hour of that.

Theme	Subthemes			Data from narrative
				There's been occupational therapies that have come up that people say, you know, "you got to do this, da da da, and it's going to really make a big difference, because he'll be able to, you know, do this at work someday." And we'd weigh that against all the other things and make a decision based on that. And so there isn't a "let's sit down and talk about Mark for fifteen minute and make a decision," it's his life. It's a process of just how you live. And every day something new pops up that, "well, got to add that into the equation and think of how we can do that or not do it."
		Invasive-ness		• Mother. 4:20 – "If I had money, we would do massage, every day….[Botox] is totally paid for, and it is effective. But the massage, I think, would probably help just as much, and it would be less invasive. And cheaper for them." • Mom- 4:43 – Botox is "not as invasive as surgery."
		Moderating factors		• 27:12. Now the injections are much easier because of EMLA and cold spray. Also, Dr. G. and the staff are very quick with the process, Mother: "They just cream, spray, boom, it's done." 29:28 – Mother: "This last one was a non-event." "I didn't even see you flinch once." • 29:30 – Mother has to be present [not clear exactly why] – seems to help manage

Theme	Subthemes			Data from narrative
				Mark's startle reflex.
		Limited resources		• MONEY: Mother. 4:20 – "If I had money, we would do massage, every day….[Botox] is totally paid for, and it is effective. But the massage, I think, would probably help just as much, and it would be less invasive. And cheaper for them." Conductive education not paid for by state or insurance. 17:18 • TIME: Mother's first comment, in response to the opening question, was about Mark's almost all-day therapy sessions. "It took up too much of his life." 1:45 Mom- 4:43 – Botox is "the least invasive of his *time*. I mean, if he does physical therapy all the time he can become loose." Father – 8:00 – At one point they had added up all of the time that it would take to complete the therapies recommended for Mark, it added up to **more than 24 hours each day**. Therapists weren't aware of the recommendations of other therapists, and the individual treatments were added incrementally: Mother quoting therapist: "It's just 15 minutes." 8:30. Speech, PT, OT. Father, 8:30: "It's more than one person could possibly do in a day." • ENERGY: Father – 9:00: "He'd get exhausted when he'd do ten hours of it or eight hours. And so you make choices based on the reality of

Theme	Subthemes			Data from narrative
				life, not on necessarily what maybe is best or not best, it's what you can actually achieve." 48:15 Father: "I would say that the one thing we haven't talked about is the benefits of any of the procedures that he gets, if they're working, is it allows him to work on something else. Rather than spending the whole day with "hands down", now he could work on his smaller skills of his fingers or something. You know, he has like a remote control that he runs. Well, if he's really tight, just hitting the one or two buttons, but if he's loose he can mess around with it. And I think that in itself becomes somewhat of a therapy for him, as far as using his fingers and using his muscles of re-learning, and—. F: So I think it's opened a lot of opportunities for him to try things and do things that are good things. He rides his bike, and he….Mark: Yup...I was gonna mention that... F: He does his wheelchair very well now, and all of that's because he's loose enough to control himself… And ten years ago he was just learning kind of how to use a wheelchair. And again, you're fighting all the normal stuff to just get to the basic teaching. Well, if you can get past those through some medication of some sort, then you can start teaching, and then you can have a higher quality of life I guess."
		Revers-ibility		*- no data from narrative -*
	Processing			

Theme	Subthemes			Data from narrative
		Youth's partici-pation		• 24:25 Mother: “Pretty much Mark gets fed all the ideas, and I'll give him my opinion, and then he generally makes a decision.” • 47:46 Mother: “Mark always participates. I guess that's—Mark always has the final, "no, we're not going to do it." Unless it was obviously life-threatening or something like that. We're guardians to protect him from—like different doctors have different opinions, and they all are so sure of them. “ • 48:08 Mother: “We're here to do the research on the computer, and talk to people, and then give our input.”
		Parent partici-pation		• 53:25 Father: “Well, it's Mark's life. It's not our life. We're lucky enough to be part of his life, and to have him in our life, but we aren't—it's not us.” • 23:54. Mother: “We would never make a decision like that for him.”**I:** But something like the surgery like you were describing, it's so clear—Mother: Oh, yeah. We were protecting him. He could not have handled that. • 47:46 Mother: “Mark always participates. I guess that's—Mark always has the final, "no, we're not going to do it." Unless it was obviously life-threatening or something like that. We're guardians to protect him from—like

Theme	Subthemes			Data from narrative
				different doctors have different opinions, and they all are so sure of them. “ • 36:25 – Father: “F:...Now what do we do with it? You don't like Valium. (Father and Mark laughing) It can make you real happy. So, you know, it’s an education process for the patient and the people that live with the patient to try and understand how do you—what is this disease, and how does it affect him. It’s hard to put one of us three into Mark’s body and think of what that is, cause we aren’t there. And if we were there—if all of a sudden we were stricken with what he has, try and think that through. We would be complaining, we would be "oh my god, this is horrible, and I can’t do this, and I hurt, and I’m tight, and I’m..."—that must be just bizarre. But you know, for Mark it’s normal, and look at him. He’s just got the best attitude in the world. So it’s hard to read what - -
		Disagreements / negotiations		*- no data from narrative -*
		Recognizing decision		*- no data from narrative -*
		Watch and wait		*- no data from narrative -*

Appendix C

Case Narrative Sample: Mark

Goals

What did the youth and caregivers hope to accomplish with Botox treatments?

- Mark and his parents sought to maintain or improve the flexibility and function of his hands and arms.
- Mark was getting good benefit from intensive physical and massage therapies. But the success of these therapies came at a considerable cost, with regard to time, energy and expense.
    - Time: The physical therapy sessions, at home and at school, occupied a significant portion of Mark's week.
    - Energy: Beyond the amount of time the therapies took, Mark (and his parents) had little energy left to work on other issues beyond his muscle spasticity. For example, he can work on developing abilities involving small motor coordination.
    - Expense: The parents were covering the costs of therapies out of pocket. Botox treatment costs, however, were reimbursed by insurance.

Information

What information was involved in the decision?

- The family was satisfied with the information they had gotten from the physician.

- When Mark was first considering Botox nine years ago his therapists were opposed to Botox.
- Mark's family has had many treatment options to consider since he was quite young. These have involved techniques that aren't part of the standard medical approach to CP and spasticity management. (They didn't describe how they had learned about the therapy methods.)

Relationships

These are the relationships with people who are brought into the decision-making process. This means that the opinions and recommendations of these individuals were part of the decision.

- The decision was made by Mark and his parents, with some input from Mark's aunt.

Deliberation

Deliberation refers to the ***process*** of developing the decision.

- The family has had many treatment options to consider since Mark was quite young.
- The decision to start Botox came as they weighed the various treatment options, along with an appreciation of how Botox might be less of a drain on time, energy and financial resources.
- Even though Botox was invasive, unlike massage and physical therapies, it was also essentially fully reversible, unlike surgery.
- Clinic staff have come up with interventions to make the Botox injections almost painless.
- Mark's parents see themselves as facilitators; they help to frame up the information and decision elements to help Mark make the decision himself.

- But some decisions are too risky for Mark to make. For these decisions, the parents draw on their familiarity with and empathy for Mark.

Appendix D

Key Elements of Conceptual Framework: *Decision-Making Process*

With supporting examples from case narratives

Goals

- ***Goals*** are the desired state of being that the participants were seeking for the young adult with regards to managing muscle spasticity.

Functional benefit.

- What were the goals with regards to relieving muscle spasticity in order to improve or maintain function?

  EXAMPLE Case 6: Cathy describes how hard it was to walk before the Botox treatment: ""Yeah. It was harder to walk through the halls at the school."

  EXAMPLE Case 7: Mark's father describes how the benefit of the Botox treatment is obvious: "The wheelchair is tangible. The bike is tangible. The fact that he can work his computer, or his remote control."

Pain relief.

- What were the goals with regards to relieving muscle spasticity in order to reduce or eliminate pain?

EXAMPLE Case 5: Scott's first statement in the interview (using his electronic communication device) was, "My wrist hurts" due to severe muscle spasticity.

EXAMPLE Case 2: Mike described how the pain from spasticity in his leg increasingly intensified before he got the Botox treatment: "It was all that I could think of, like every minute."

Information

- ***Information*** includes the information *content* used in the decision-making process as well as the *sources* of this information.

Information content.

What was the information about?

Potential benefits of Botox.

Functional benefits of Botox.

Information about how Botox can allow improved functioning by relieving spasticity.

EXAMPLE Case 4: Tom first learned about Botox from his friends who have cerebral palsy: "I'm like 'What are Botox?' And they just told me it helps with your muscles not to be so tight. And, you know, be loose, and people walk better, and you know."

Pain relieving benefits of Botox.

Information about how Botox can reduce pain by relieving spasticity.

EXAMPLE Case 2: Mike's father described how Botox was considered for managing Mike's pain: "He gets pain in the knee. And we had Dr. Smith…take a look at it. And oh dear, a whole series of things. X-rays, and MRIs and that. And really couldn't find nothing wrong with the knee itself. And Dr. Smith determined that because of the tension, and the hamstrings in pulling on it, it was causing this. And so, we just thought it would make it easier to handle with the Botox, but it's also to relieve the pain."

EXAMPLE Case 5: Scott's wrists were much more relaxed within one week of his first injections to the wrists.

Potential risks of Botox.

Pain and distress associated with Botox injections.

What information did the youth and parent receive to help prepare them for the experience of getting the Botox injections?

EXAMPLE *Case 1*: Bill described that although he was aware the injections would hurt, he wasn't well prepared for the actual experience: "May I be honest? Looking back I think they underestimated how much it would hurt."

EXAMPLE *Case 5*: Scott's father described how he felt unprepared for the first injection: "You hear that they numb it and that, but you're still there holding them and that, and he's still jerking. He still can feel it."

Botox injections can result in the muscles becoming too weak.

Botox injections can result in the muscles becoming too weak.

EXAMPLE *Case 4*: Tom had heard that Botox injections could result in muscles becoming *too* weak: "Dr. Smith, and my friends. They're like, 'You might get too weak.'"

EXAMPLE *Case 2*: Mike's father described how he had heard about other people who had become too weak from Botox: "One of the other clients in the house also had Botox. And after the first set of injections she totally was unable to use her hands for a significant period of time."

Warnings of rare but serious side effects of Botox.

Over the past few years there have been alerts regarding a very small number of deaths associated with Botox treatments for muscle spasticity.

> _EXAMPLE_ *Case 5*: Scott's father brought up the issue of serious complications associated with Botox treatment: "We were a little skeptical. There were some problems with Botox that surfaced here a couple years ago. So we had stopped, for a while."

Other treatment options available.

In addition to Botox, what are alternative treatment options for addressing treatment goals?

> _EXAMPLE_ *Case 7*: Mark's parents described how they have explored and used a wide variety of options for managing Mark's spasticity. Mother: "We never stop looking for options." Father: they considered "what we understand is available. What are people asking and what are they saying that the benefit is going to be? And then it comes down to weighing what is feasible, what is financially possible, what is time-constraint possible, and what will have the largest potential benefit? There's a million things you can do, but not all of them have big benefits."

Information sources.

Where did the information come from?

Textual information.

Media refers to different formats or modes of textual information.

Independently gathered.

Information sought by youth or parent, independent of that provide by medical professionals.

> _EXAMPLE_ *Case 5*: Scott's parents followed up on information they heard in the news about Botox: "The warnings and that were coming just from the internet, and from different cases that were being published, that there was a few deaths in there. But, just like anything else, you don't always get all the information. So, after we decided to kind of pass on it a little bit, then we asked more questions, and got a little more comfortable after. And I think there was more information that came out of maybe some of the other cases. You know, what took place."

Medical professional provided.

Information provided by medical professionals.

> _EXAMPLE_ *Case 3*: Joe's mother: "And I went to talk to Dr. Smith again, and re-read all the literature, and talked to my husband."
>
> _EXAMPLE_ *Case 4*: Tom: "That was a handout to read about Botox… I have it filed with all of my medical appointments."

Verbal information.

Verbal information based on other peoples' experience or expertise.

Medical professionals.

Verbal information from medical professionals.

> _EXAMPLE_ *Case 1*: Bill and his mother attended an informational session on the topic of "Growing older with cerebral palsy," where clinical staff discussed Botox as a treatment option.

Peers.

Information from peers and others who have experience with Botox.

> _EXAMPLE_ *Case 4*: Tom: “My friends already told me about [Botox]... Why read about it when your friends already told you about it? And your friends are not going to lie to you. I trust my friend before I’d read something, you know?”

Prior experiences.

Information from prior experiences with spasticity, Botox and other treatment options.

> _EXAMPLE_ *Case 2*: Mike’s father described how the use of Botox injections has evolved as they have seen the effect of the treatments: “We started first with the right arm, because that was the one—he’s right handed, and that was the one that he had the most potential of using. So we started with that. And after about the second or third series of that, we noticed a marked improvement.”

Relationships

These are the relationships with people who are brought into the decision-making process. This means that the opinions and recommendations of these individuals were part of the decision.

Parent – youth relationship.

This is the central relationship in the decision.

> _EXAMPLE_ *Case 4*: Tom is legally autonomous, but he sought his parents’ opinion on a recent health-related decision: “I wanted [mother’s] opinion. And I couldn’t ask anybody else, because nobody else in my town had a [medical device], so I couldn’t ask anybody. So me and my mom and my dad make these decisions.”

Relationship between physician and youth – parent.

How was the physician involved in the decision?

> *EXAMPLE Case 7*: Mark and his mother described how they appreciated that the doctor expressed his ambivalence about whether to start Botox. Mark: “I went to the doctor... that does this, and it’s like, he said, ‘I don’t know!’, which was actually pretty good.” Mother: “People try to talk us into it for so many years. Every appointment we went to they—‘Well, why aren’t you doing this?’... The one doctor who actually does it said, ‘Oh, maybe not.’"
>
> *EXAMPLE Case 4*: Tom described how he was very careful to not mention Botox to his physician. This was a strategic decision; Tom thought that it would discourage Dr. Smith from recommending Botox if Tom raised the idea first: “I was—like I know he’s like, doctor, I know the pump wasn’t working. And I wanted him to mention Botox, because what if he said ‘no’ and I didn’t want to get my hopes up.

Other family, friends or professional staff.

Beyond the parent, who else was involved in the decision?

> *EXAMPLE Case 3*: Joe’s mother described how Joe’s PCA’s are helpful in making health-related decisions: “His PCAs are really good sounding-boards for me. Because I think out loud. And I’ll ask them—well, they’ve been with him so long now…”

Deliberation

Deliberation refers to the ***process*** of developing the decision.

Balancing treatment options.

Refers to the appraising of the relative merits and liabilities associated with different elements of the decision.

> _EXAMPLE_ *Case 3*: Joe's mother gave an overview of the decision-making process: "We decided we would try it [Botox]…And so we talked to Joe about it. And I went to talk to Dr. Smith again, and re-read all the literature, and talked to my husband. And then went over with Joe what would happen. What the Botox injections would be like. And talked to Dr. Smith again, about my concerns, and that's when he said he would prescribe Valium to relax Joe. And so we decided to try it and see."
>
> _EXAMPLE_ *Case 7*: Mark and his family have selected from a wide variety of treatment options over his whole life. His father stated that they have chosen from "what's available [to help manage spasticity]…what we understand is available. What are people asking and what are they saying that the benefit is going to be? And then it comes down to weighing what is feasible, what is financially possible, what is time-constraint possible, and what will have the largest potential benefit? There's a million things you can do, but not all of them have big benefits.

Invasiveness.

Different treatment options for managing muscle spasticity involve different levels of invasiveness, ranging from surgery to massage.

> _EXAMPLE_ *Case 7*: Mark had received intensive massage to reduce spasticity in his arms and legs. The parents preferred massage because it was less invasive than Botox injections, but they preferred Botox as a less invasive alternative to

surgery. Mother: “But the massage, I think, would probably help just as much, and it would be less invasive…And nicer for Mark.”

Moderating factors.

Moderating factors are methods of ameliorating any of the risks associated with Botox, specifically the pain of the injections. Examples are nitrous oxide and EMLA anesthetic cream.

EXAMPLE *Case 3*: Joe receives oral Valium prior to Botox treatments and has no discomfort with the injections. Mother: “He’s always had Valium that morning before we go in…And the first time we did it, Dr. Smith gave him 10 milligrams of Valium, and he was *out*! So he probably doesn’t remember.”

EXAMPLE *Case 4*: Tom benefits from aromatherapy and music as well as EMLA cream and cold spray to minimize discomfort during injections: “They put lavender on, and they had music on, so it’s not—people think those [injections] are just painful, but it’s not. It’s worth it. And they have cream and they have spray.”

Limited resources.

Different treatment options involve a commitment of limited resources, such as financial cost, or the time or energy involved in different treatment options.

EXAMPLE *Case 7*: Intensive massage provided Mark with excellent relief from muscle spasticity. While the massage was less expensive than Botox, massage was an out of pocket expense, whereas Botox treatments were covered by insurance. The massage sessions also were time intensive, involving many hours out of Mark’s week. Mother: “If I had money, we would do massage, every day….[Botox] is totally paid for, and it is effective. But the massage, I think, would probably help just as much, and it would be less invasive. And cheaper for them."

Reversibility.

Some treatment options are essentially reversible over time, e.g. Botox and oral medications. Surgery is an example of an irreversible treatment option.

EXAMPLE Case 2: Mike started out receiving very low doses of Botox injections. If the muscles became too relaxed, muscle tone would return in a few weeks as the effects of the Botox wore off.

EXAMPLE Case 3: Joe's mother: "We decided we would try it. It wasn't anything that was irreversible, or *that* invasive."

Processing.

Processing includes actions and events that took place during deliberation.

Youth participation in decision.

How is the youth active in the decision?

EXAMPLE Case 2: Father's description of the decision to try Botox: "My wife and I talked it over with Mike. And he said, well, his words were —'Anything that gives me more ability to do things and have better control, let's give it a try.' … But we've always talked about it, and he's indicated his wanting to continue. So this is what we've done…We've always tried to involve Mike..." in decisions.

EXAMPLE Case 7: Mother: "Pretty much Mark gets fed all the ideas, and I'll give him my opinion, and then he generally makes a decision…Mark always participates. I guess that's—Mark always has the final, 'No, we're not going to do it.'"

Parent's participation in decision

How is the parent active in the decision? For the youth who are legally autonomous, how is the parent involved in the decision?

EXAMPLE Case 7: Mark's parents are his guardians. His father described that, while they are responsible for the decisions, they appreciate the obligation to respect Mark: "Well, it's Mark's life. It's not our life. We're lucky enough to be part of his life, and to have him in our life, but we aren't—it's not us."

EXAMPLE Case 1: Although Bill is legally autonomous, he lives at home with his parents. His parents are involved with all aspects of Bill's medical care and decisions, although Bill has ultimate responsibility for decisions. When the doctors first offered Botox treatment, they held off because Bill's mother said she needed more information: "Mom wanted more data."

EXAMPLE Case 3: Joe's mother stated, "It's so hard for a parent. There are certain things that have to be done that are not negotiable."

EXAMPLE Case 6: Cathy's mother stated that the Botox decision was not a "major" decision like the decisions involved with managing Cathy's ventriculoperitoneal shunt: "That's do or die"

EXAMPLE Case 7: Mark's father describes what it's like to make decisions on behalf of his son: "It's an education process for the patient and the people that live with the patient to try and understand how do you—what is this disease, and how does it affect him? It's hard to put one of us three into Mark's body and think of what that is, 'cause we aren't there."

EXAMPLE *Case 5*: Scott's father didn't feel fully prepared for the first Botox injection session. The father was expecting that Scott would have *no* sensation of needles: "Maybe I was more jumpy just thinking what would it feel like to *me*."

\EXAMPLE *Case 6*: Cathy's mother described that it's very important to Cathy that they are honest with her about the impact of medical decisions, like the pain she might experience with the Botox injections. Cathy said that knowing what to expect "matters [to her]…now that…something like that would matter to me."

Disagreements or negotiations.

Disagreements or negotiations between the young adult, parents, or physician.

EXAMPLE *Case 3*: Joe's mother stated, "If he had been adamant about not wanting to do the Botox, we would not have repeated it."

EXAMPLE *Case 4*: Tom knew about Botox from friends with CP. He waited for his doctor to introduce the option of starting Botox, because he was certain that if he brought up Botox first his doctor would not agree to starting Botox: "I knew there was one alternative, but I didn't want to say anything, cause when people say something, and [Dr. Smith would say], 'No, that won't work for you.'"

Recognizing the decision.

Was there a particular moment when the decision was made, or was it an extended process with no clear decision-making event?

> *EXAMPLE Case 6*: Cathy's mother described that after the decision to get Botox the first time, subsequent treatments didn't involve significant deliberation: "I think the very first time it was brought up, way back when, we probably discussed it more than we did this time. This time was more definitely of a no-brainer. Before, you know, I can't remember details, but before we definitely had more questions. You know, I knew Botox as these people injecting it in their face. That is what I knew of Botox. But yeah, we definitely asked more questions the first time than we did this time, because we didn't have any bad effects from the first time."
>
> *EXAMPLE Case 3*: Joe's mother describes how she recognized that returning after 90 days for the next Botox was a decision, although she decided not to explicitly confront her son with this decision: "And if he had been adamant about not wanting to do the Botox, we would not have repeated it. But he wasn't. We don't talk about it anymore. The last time we went in, we didn't discuss whether or not to do it again. I just said, 'You've got an appointment with Dr. Smith. We're going to do the Botox injections.'"

Watching and waiting.

Once various options are being considered, did the youth and parents "watch and wait" before committing to any decision?

EXAMPLE *Case 1*: Bill's doctors first recommended Botox for treatment of his muscle spasticity 16 years ago. The family felt that this option was too new. "You thought about it and decided you didn't want to do it—at that time. Remember? Because it was so new." They only reconsidered Botox as Bill's wrists got too tight to operate his wheelchair and communication device.

EXAMPLE *Case 5*: About two years ago news stories appeared in the mainstream press about a small number of deaths associated with Botox treatments for muscle spasticity. Scott's family heard this news. Father: "Well, it came up to where it was his time to go in again, whatever that month period was, and mom and dad heard about it, and then we talked it over with Scott and Sally, and we said, 'Well, you know, until more information comes out, down the line here, maybe we want to wait. And maybe give him something more to help him relax in the meantime, even though it wouldn't be—it's temporary.' So, we just didn't have a comfortable feeling until we could find more out." They continued with Botox when they were confident that more cases of death weren't being reported.