The Problem With Prevention: The Case of Spina Bifida

Ann Neville-Jan

In this paper, I present a viewpoint about prevention and spina bifida that is not usually expressed within the occupational therapy literature. Using an autoethnographic account, I convey my experiences as a person with impairments from spina bifida in order to problematize current preventive efforts undertaken to eradicate this birth defect. This self-reflexive account connects my personal experiences to historical and medical views about spina bifida. The messages inherent in preventive efforts are discussed from a disability rights perspective. Occupational therapists are challenged to examine their attitudes toward disability, act as advocates in their practice, and, in a more informed manner, support or contest policy initiatives.

Neville-Jan, A. (2005). The problem with prevention: The case of spina bifida. *American Journal of Occupational Therapy, 59,* 527–539.

Still leaning against the incubators he gave them, while the pencils scurried illegibly across the pages, a brief description of the modern fertilizing process . . . actually showed them how this liquor was drawn off from the test-tubes; how it was let out drop by drop onto the specially warmed slides of the microscopes; how the eggs which it contained were inspected for abnormalities, counted and transferred to a porous receptacle. (p. 5)

—Excerpted from *Brave New World* by Aldous Huxley, 1932

Luckily, queer ones like her don't happen often. We know how to nip most of them in the bud, early. (p. 60)

—Excerpted from *Fahrenheit 451* by Ray Bradbury, 1950

There were only two occasions of release which were not punishment. Release of the elderly, which was a time of celebration for a life well and fully lived; and release of a newchild, which always brought a sense of what-could-we-have-done. This was especially troubling for the Nurturers, like Father, who felt they had failed somehow. (p. 7)

—Excerpted from *The Giver* by Lois Lowry, 1993

The above quotes from futuristic tales depict life-and-death choices made at both ends of the life cycle. Today, these choices are no longer limited to the realm of science fiction. For example, preimplantation genetic diagnosis or PGD (Harper, 2001), the latest technique for detecting certain genetic diseases, allows parents to select a "healthy" embryo for implantation. Preventing the birth of an infant with a disability would appear to be a good thing not only for the child and parents, but, considering the costs related to health care, a good thing for society. My paper provides a different way of framing prevention. Using autoethnography (Ellis & Bochner, 2000), I will convey my experiences as a person with impairments from spina bifida[1] in order to problematize preventive efforts undertaken to eradicate this birth defect.

Autoethnography refers to both a research method and the self-reflexive texts that link ethnography with autobiography. Autoethnography is all at once about

Ann Neville-Jan, PhD, OTR, FAOTA, is Associate Professor, Department of Occupational Science and Therapy, University of Southern California, 1540 Alcazar Street, CHP-133, Los Angeles, California 90089-9003; aneville@usc.edu

self and culture (Reed-Danahay, 1997). For me, as a researcher studying adults with spina bifida, a group of which I am a member, autoethnography provides a means for me to connect my self-as-researcher and self-as-research participant. In previous articles, I have used my personal experiences of living with spina bifida to question the dominant discourses about pain (Neville-Jan, 2003) and about sexuality (Neville-Jan, 2004). Autoethnography as a means to understand the lived experience of disability fits within Kielhofner's (2004, 2005) recommendations for research in occupational therapy that incorporates disability studies. Autoethnography contributes a perspective beyond the personal disability experience to the social and political contexts in which disability occurs.

For the past 3 years, I have been a coinvestigator on two ethnographic studies funded through a disability supplement from the National Institutes of Health (National Institute of Child Health and Human Development, National Center for Medical Rehabilitation Research, #RO1 HD 3887801A1S). The project that my research supplements is an ongoing ethnographic and longitudinal study of 30 black families who have a child with a disability (#RO1 HD 3887801A1). For this project, I have been a participant observer during clinical encounters, interviewed family members and practitioners, and recruited participant families, 4 of whom have a child with spina bifida. The second project used narrative, ethnographic, and longitudinal methods to study 14 adults with spina bifida. I am one of the 14 participants as well as one of the researchers. Through such intense involvement with children and adults who have spina bifida, I have realized the importance of narrating parts of my story as reflexive texts to the ethnographies described above.

For me, the topic of this paper is profoundly disturbing. What if prenatal testing were available when I was conceived? Would I exist today? Is there value to having spina bifida? What messages, both positive and negative, are embedded within current folic acid campaigns and research geared towards early diagnosis to prevent spina bifida?

Occupational therapists are very much a part of this story. They evaluate and treat infants, children, adolescents, and, much less frequently, adults with spina bifida and their families in a variety of settings, including neonatal units, specialty spina bifida clinics, school-based practice arenas, and outpatient centers. Whereas genetics specialists counsel parents about particular disorders, families and individuals with particular impairments frequently direct their questions to frontline professionals such as occupational therapists who engage with them in day-to-day activities. In their everyday practices occupational therapists champion the rights of persons with disabilities through empowerment and advocacy activities. On an individual level these efforts are notable. Yet, how can occupational therapists become more involved in the political and economic barriers that restrict participation in occupation?

In order to apply the perspective of autoethnography to the discussion about spina bifida, I begin by relating selected experiences of growing up with spina bifida and layer them within their medical and historical contexts. I will go back and forth between my experiences (represented in italics) and the literature. A discussion follows with implications for occupational therapy.

1950

I came into the world on December 13th at around 8:30 a.m. I weighed 7 lbs 2 oz and was the third child born into a family that would include one more sibling. My mother described her experience of being told that her newborn infant had spina bifida. She said:

They told me you had spina bifida. I had never heard those words before. They said that some infants are born with their spine exposed, but yours was closed and you had a hair spot. No one in the family had seen anything like that before. I was very upset and they gave me a sedative to calm me down. The obstetrician said you probably wouldn't walk and that we should watch your head because it might get big. He said that you could be mentally retarded.

My mother further told me that she didn't know why it happened but she did recall that her father, my grandfather (born in Sicily), had a brother who had a "tail on his back." His brother never came to America, so we don't know what happened to him.

The worst case scenario presented to my parents was not realized. I walked at 18 months, although my gait was different. Both feet pointed to the right when I walked, and my right leg was less muscular, slightly shorter, and a little contracted at the knee, resulting in a limp when I walked. My right foot had no sensation. My mother said that after I was born, she had no idea what to expect. She recalled:

I would put you in an infant seat in the yard and you'd jump up and down. I thought that was part of the spina bifida, but they said you were supposed to do that. After you began walking, I took you to the pediatrician who was present at your birth and showed him how your foot pointed inward. He said: "That's not a problem. We never thought she'd walk!"

Because 90% of infants born with spina bifida develop hydrocephalus, my parents were advised to monitor the size of my head. Looking back, I feel lucky that I did not develop hydrocephalus. The shunt now typically used to drain the cerebrospinal fluid that accumulated on the brain was not developed until 1952.

What Is Spina Bifida?

Spina bifida occurs during the very early weeks of pregnancy when the neural tube fails to close properly. The type and severity of impairments result from the extent and location of the spinal cord lesion and can therefore be highly variable (Feuchtbaum et al., 1999; Hunt & Poulton, 1995; Mazur, Shurtleff, Menelaus, & Colliver, 1989; Swank & Dias, 1992). Smith (2001), in a historical account, found evidence of spina bifida in skeletal remains dating as far back as 10,000 B.C. Few reports of the long-term consequences of spina bifida existed until 1652, when Professor Nicholas Tulp, a Dutch anatomist, gave spina bifida its name and described the consequences more fully (Smith).

Today, the estimated incidence of spina bifida occurring within the United States ranges from approximately .5 to .8 per 1,000 live births (Lary & Edmonds, 1996; Singh, 2003). Surveillance data (1983–1990) of prevalence rates at birth in the United States (16 states participated) report significant variations by state, ethnicity (Hispanics rate highest, followed by whites, blacks, and Asians), and gender (rates higher for females) (Lary & Edmonds). Trends suggest a decrease in the incidence of spina bifida since the implementation of prenatal screening, followed by abortion, and folic acid supplementation programs (Davidoff, Petrini, Damus, Russell, & Mattison, 2002; Rekate, 1991). Of those born with spina bifida, rates of survival into adulthood have steadily increased, from 60% in 1963 to an almost 90% survival rate in the 1990s (Kinsman & Doehring, 1996). The Spina Bifida Association of America (SBAA) estimates that today there are 70,000 Americans living with spina bifida (Spina Bifida Association of America, n.d.)

Secondary conditions associated with spina bifida can include physical impairments such as pain, bowel and bladder incontinence, pressure sores, scoliosis, orthopedic malformations, and short stature (Farley, Vines, McCluer, Stephans, & Hunter, 1994; Lollar, 1994b). In addition, psychological impairments such as depression, anxiety, impaired body image, and learning difficulties have been documented (Dennis & Barnes, 2002; Dennis, Fletcher, Rogers, Hetherington, & Francis, 2002; Dennis, Rogers, & Barnes, 2001; Lollar, 1994a; Marge, 1994). These secondary conditions, some of which can occur at any time throughout a person's life, can impact activity and participation in education, employment, community living, and health care (Dunne, Gingher, Olsen, & Shurtleff, 1986; Hunt, 1990; Peterson, Rauen, Brown, & Cole, 1994). It has been suggested that the aging process compounds these secondary conditions (Bergman, 1994; Turk, 1994).

Surveys of children and adolescents report a concern for the future as a prominent theme (Appleton et al., 1994; Holmbeck & Faier-Routman, 1995). Specifically, when asked about their future, children and adolescents with spina bifida have described feelings of apprehension in regard to establishing friendships, finding a life partner, holding a job, and sexuality (Lollar, 1994a).

> *Rose, my mother's best friend, had polio. One day Rose's mother and my mother shared stories while shopping at the grocery store. She told my mother, "Make sure your daughter gets as much education as she can." I heard that comment many times growing up and I guess it made a difference because I did just that. Other than going to school, I was not expected to marry or have children. Although when I was a child I had fantasies about getting married and keeping my incontinence a secret, when I found out about sex, I figured getting married was not a possibility. There were no adult role models for me, either, probably because not many infants born before me survived into adulthood.*

To date, only a few studies have been conducted with adults who have spina bifida. These studies have focused on young adults and medical outcomes related to problems such as hydrocephalus, tethered cord, or incontinence (Andren & Grimby, 2000; Dunne et al., 1986; Mazur et al., 1989; Query, Reichelt, & Christoferson, 1990). Researchers have not explored the everyday life experiences of adults with spina bifida, their experiences growing up, and their concerns for the future. Our current research efforts intend to fill this gap in the literature.

1960–1970s

> *I wasn't aware that I walked differently until one day, as I was crossing the street with my best girlfriend Julie—I think I was about 11 years old—she asked me, "Do you ever notice how people stare at the way you walk?" From that point on, I noticed all of the time. Some people would even turn around after I passed them to continue staring. I was different and I didn't want to be "normal" like my friends. In an effort to achieve this, my mother and I saw an orthopedic surgeon, referred by our general practitioner. He slated me for a "makeover." First, he recommended orthopedic surgery to correct my right foot that turned inward. Second, I was referred to a plastic surgeon to remove the* hairy nevus*, the technical name for the hair spot on my back. It was a large rectangular patch of hair about 4 inches by 6 inches. If someone happened to see it, they'd say, "What's that on your back?" Attempts by my mother to shave it never worked. There was always visible stubble. Third, I was referred to a urologist to see what could be done about my incontinence. I wore diapers and rubber pants that my mother constructed to be absorbent, but not too bulky under my clothes. From 1964 to 1970, I*

had four surgical procedures (three on my foot and one on my back). I was very disappointed in the results. I really thought my body would be made to look "normal."

Treating orthopedic and urinary problems resulting from spina bifida has benefited from several technological advancements. New plastic materials introduced in the '70s improved the splints and braces that assisted children in attempting to walk. Today, children who walk primarily with extensive bracing, once in school, move toward a wheelchair that provides more ability to participate in social activities. Urological procedures followed a similar path. During the 16th and 17th centuries, nonmetallic catheters were first used to drain urine. The results were poor due to infection, smell, and social stigma. Techniques developed in the late 19th century provided an alternative to catheterization through the use of various methods to divert the urine. Transplanting the ureters to a loop of ileum became the recommended method to treat urinary incontinence in the early 20th century. Today, there has been a return to catheterization along with surgically augmenting the bladder with artificial sphincters and increasing the bladder capacity. New "odorless" pads and catheterization provide a more socially acceptable and natural means to manage this problem (Smith, 2001).

In 1961, spina bifida clinics, composed of multidisciplinary teams, were established in several major cities across the United States in an effort to coordinate the many complex secondary conditions related to spina bifida. The largest clinic was located in Sheffield, England, and was directed by neurosurgeon John Lorber. In a highly controversial study of 524 infants born with spina bifida between 1959 and 1969, Lorber (1971) developed prognostic criteria to decide which infants should be treated (closing the spina cord) at birth. Those infants thought to be too severely handicapped were not treated but provided analgesics and normal nursing care. They were left to die.

Bladder incontinence became the most stigmatizing impairment for me. In 1970, I had a surgical procedure called an ileal conduit, which diverted urine from my bladder to an outside appliance on my abdomen. After the surgery I finally achieved independence. The following year, I left home, bound for New York City to study occupational therapy. I had experienced physical therapy (specifically, whirlpool for infections and muscle testing) during my hospital admissions but never occupational therapy. I learned about occupational therapy from a book my sister received at a career fair. I remember that the brochure described occupational therapy as a health profession that brought science together with art. This seemed perfect for me and my interests. I majored in biology in college and seemed to have an artistic flair noted by my high school art teacher. I volunteered in an occupational therapy department at Johns Hopkins Hospital during my last year in college and decided this was what I wanted to do.

While living in New York, I continued my quest to be "normal." For me, this meant doing everything an adult developmentally accomplishes—having a career, getting married, and having children. I read about these goals in my developmental theory books in occupational therapy school. Periodically, I was confronted with the fact that having spina bifida meant that I was not "normal." For example, one of my professors, after reviewing a paper I wrote about growing up, said: "You should read other students' papers to learn what it's like to grow up normally." Another encounter occurred during a week-long fieldwork experience. I had requested a placement in a spina bifida clinic to observe the role of occupational therapy in this area. My supervisor was not happy with having me there. I became acutely aware of this after I attended a conference at which the clinic team was discussing an option of institutionalization with a family whose child had spina bifida. After the meeting, the therapist said to me, "I was really nervous about having you witness this meeting. I thought you might overidentify with the plight of the child and it would upset you." Again, I was confronted with someone else's expectation based on my diagnosis. This was the decade of Lorber's selection criteria, which may have added to her worry about my presence in the clinic. At the time, I wasn't aware that selective treatment occurred.

1980s

The Maternal Alpha Fetal Protein (MAFP) blood test became available in the early 1980s. The test, performed between the 15th and 17th weeks of pregnancy, measured the amount of alpha fetal protein secreted from the fetus into the mother's blood stream. An elevated MAFP meant that there was an increased risk for a neural tube defect. A repeat test, ultrasound, and amniocentesis might then be offered to parents for more definitive results (Powell, 2000).

It was February 1982, I was in my bedroom reading. The television was on in the background. I was drawn away from my book when I heard the opening statement on a news show called "The Terrible Choice." The journalist said:

Imagine this. You are pregnant, or your wife is. The fetus is 4 months old, and you are able to look inside the womb and see your baby. And what you see is deformed, crippled, and will be for life. What would you do? (Utley, 1982)

I was glued to the TV set for the rest of the segment. The show was about the newly developed maternal alpha fetal protein blood test that, according to the news report, "Can now determine within almost 100% accuracy, whether a baby will be born with this fatal or crippling defect." After a positive test, an ultrasound scan was typically recommended to determine if the fetal spine showed "evidence of a defect in its formation." Here's how the announcer described spina bifida:

What it usually means is paralysis below the waist. No control over bladder or bowel functions, and often mental retardation. Joan [not her real name] is lucky. She did not suffer brain damage. But every 3 hours, her mother, who adopted her, must insert a tube to drain her urine. That is Joan's life, and this is why. Look through an electron microscope at the fetus of any creature with a backbone, and this is what you will see. First, a tiny plate appears, and begins to curl upward. This is the neural tube, which will form the brain and the spinal cord. By the 30th day of pregnancy, before the mother even knows she is pregnant, the tube must seal itself perfectly from top to bottom. Usually it does. But once or twice in every thousand babies, it doesn't. There is a gap, a neural tube defect. If it is at the top of the tube, the brain will never develop, the baby will almost certainly die soon after birth. If the defect is near the bottom of the neural tube, the baby can survive, but it will face a life which at very best will be severely handicapped. It will have spina bifida. (Utley, 1982)

After reviewing the program again on a rerun and subsequently purchasing a copy of the tape, I found only one sentence devoted to the possibility of a positive outcome or that it might not be a tragedy. "Spina bifida children can and do triumph over their condition; with specialized training and surgery, many become strong, productive adults" (Utley, 1982). However, this was followed by, "For each child, the cost can run to thousands of dollars a year—financed sometimes by a family's private insurance, sometimes by the taxpayer, through Medicaid" (Utley). The announcer interviewed parents who opted for abortion after finding out their growing fetus had spina bifida. It was stated that most of the doctors consulted for the news segment agreed with the parents' decision to not have a child with spina bifida. In fact, one of the physicians interviewed stated that over 90% of parents decide to terminate a pregnancy when their child is diagnosed with spina bifida.

At first, I remembered feeling shocked. They were talking about me. Although I had spina bifida, I did not see myself as "severely handicapped" or "crippled," as described on the show. I had a career. I was "normal," or at least I was trying to be. I had never written a letter in response to an issue in the news. After the show, a video clip depicted various people stating their opinions about previous episodes. I wrote the following and mailed it the next day:

Dear Sir,

I am writing in response to your segment on spina bifida. I was quite distressed by your one-sided view of children with this congenital problem. Your emphasis was on problems and handicaps, such as the money needed for various therapies and doctors, and the example of a child without bladder control who needed to be relieved of urine through a tube. There is another more positive side which I would like to share with you. I have spina bifida. When I was born, my mother was told I might not walk and to watch for other abnormal signs such as hydrocephalus (increased fluid on the brain). Fortunately, I walked but at a later age than most. I have had numerous surgical procedures, however, to correct deformities of my foot, and due to a lack of sensation I have a chronic problem with my foot that periodically needs surgery. I also did not have bladder control but had an operation many years ago called an ostomy which has allowed me to live a normal life. I work as an occupational therapist in a major New York hospital, I am a part-time university instructor, and I am almost finished [with] a doctorate degree in my field. I am single, live in Manhattan, have many friends, date, go to the theatre and movies, and I especially enjoy dining out. You see, I enjoy my life. I enjoy helping other people in my work, and I enjoy loving and intimate relationships. My life has not been easy, but I feel I have an understanding of the human condition that would not have been possible without my experiences. Listening to your show, I became aware that I might not exist if such a test was available 31 years ago. I'm glad my parents did not have to make this choice. I am not against abortion, but I feel your show portrayed a very negative picture about this issue. I do not feel you presented a balanced view for one to make an informed choice for or against aborting a spina bifida fetus. I hope you will give this attention on your show.

Two days later, I received a call from a producer of the show. He asked me if I was a "right to lifer." He wanted to know if my reason for writing had to do with an agenda against abortion. He appeared reassured when I said no, and he then asked me if I would be willing to come to the TV studio and tape a video commenting about the show. Without hesitation I said I would do it.

What an experience! Early one morning I arrived at NBC studios, located just across the street from Radio City Music Hall. I met a young woman at the elevator who was to produce my 15-second segment. She guided me through a large room with many desks—reminding me of a newsroom—into a smaller room. A light was mounted above me and a camera person was off to the side. The producer gave me my letter (see above) with several sentences underlined. She told me just to read it. The camera rolled continuously. She asked me questions and commented on my mood to get me into the frame of mind with which I wrote the letter. She said, "Aren't you angry about this?" After a few more "takes," she said, "I think we have it." We went into a video room to look at the tape and she seemed pleased. All week I worried about how my comments would be placed. On the Friday evening, as I sat with friends (they had a color TV; I didn't), I waited. Finally the anchor said:

Time now for your comments. Last week, I did a report we called "Terrible Choice." It was about a new medical technology that can tell whether a fetus inside the womb has a defect known as spina bifida, or open spine. And we reported that when faced with the knowledge that their child will be crippled, over 90% of the parents are choosing to have an abortion. (Utley, 1982)

Then came the viewers' responses. The first comment was from a parent who indicated that his 10-year-old son watched

the program and "was outraged." His son ". . . began to cry, because he was never told that it was this hopeless a situation" (Utley, 1982). The second comment was from another parent of a child with spina bifida who said that her daughter has a happy life but she thought the show "did a fine job of informing the public about the availability of this new testing procedure" (Utley). I was the last to comment. I said:

I have spina bifida and I lead a very normal and productive life. I feel that if the test was available 31 years ago, I might not exist today. And that's quite upsetting to me. I'm glad my parents didn't have to make this choice. (Utley, 1982)

Because my comments were last I hoped that my words would leave people with more of a balanced impression about spina bifida. The show had been so overwhelmingly negative. One of my coworkers commented that my perspective would make it more difficult for parents as they decide about whether or not to abort a fetus with spina bifida. I can only imagine how difficult that choice must be.

In the 1980s and 1990s, prenatal testing became more efficient and less risky with the introduction of choronic villus sampling and enhanced ultrasonography (Powell, 2000). A newer blood test, called a triple screen (Graves, Miller, & Sellers, 2002) measured two other enzymes or hormones as well as alpha fetal protein and had a better detection rate than MAFP. The triple screen calculation also included a woman's age, weight, race, and whether she was diabetic. Research continues to pursue less invasive testing that can be performed at an earlier stage of pregnancy. According to the International Federation for Spina Bifida and Hydrocephalus (IFsbh) policy statement, an estimated 90% of women in Western countries interrupted a pregnancy after a positive screening for spina bifida (International Federation for Spina Bifida and Hydrocephalus, n.d.). For those who continue a pregnancy after a diagnosis of spina bifida, there are benefits to screening. For example, transfer to a tertiary neonatal unit may be indicated as these facilities are more capable of managing high-risk pregnancies and increase the chance of survival for the newborn. Also, knowing ahead of time that a child will have a particular birth defect allows parents time to prepare physically and emotionally for the child's birth (Merkens, 1997).

I married at age 34. My very first pregnancy lasted 5 months. Although my AFP test was negative, it was recommended that I have an amniocentesis due to my age and my history of spina bifida. I had a 2–3% risk of having a child with spina bifida and an even higher risk of having a child with Down syndrome. Two weeks after the amniocentesis, I developed an infection that resulted in losing our child. She did not have spina bifida but I later learned that because I had an ostomy, the bacteria present on my abdomen could not be removed by simply swabbing with betadine. Putting the needle into my abdomen most likely introduced bacteria into the fetal sac.

After several failed pregnancies, I thought I might have to give up on the experience of childbirth, my last "normal" developmental task that I was driven to accomplish. Finally, my husband and I adopted an infant from China. However, shortly after we returned home, I discovered that I was once again pregnant. For my husband and I, prenatal screening was filled with anxiety. By the time I became pregnant at age 44, the "triple screen" was recommended. I'll never forget that Friday afternoon when I received a call from my obstetrician. He said that my triple screen test was "positive." I asked him what that meant and he said, "It means you should have an amniocentesis." I was frightened. Was there a chance this amniocentesis could once again create the risk of a bacterial infection? And even if that disastrous outcome didn't occur, what about the results of the test itself? What if we had to make that "terrible choice"? What would we choose?

The following Monday, I had my physician fax me the numerical results of the triple screen. Armed with the results, I went to the university bookstore and sat with the latest edition of a textbook on prenatal diagnosis. I discovered that because of my age, it would have been impossible for me to have a negative result on the test because age was weighted in the equation. Rather than an amniocentesis, I sought an ultrasound from a local expert in this technique. Genetics testing was part of the package. We learned that in my case of spina bifida (my spine was not open), my mother's AFP would not have been elevated. The MAFP test would not have revealed my spina bifida. I hadn't known that.

The next part was the ultrasound. We held our breath as we scanned the ultrasound screen and listened as the physician rattled off measurements as he examined the fetus. I watched his face to see if I could detect a worried look. He finally indicated there was no evidence of Down syndrome or spina bifida. He said, "I do not recommend an amniocentesis." On further look, we learned I was carrying a boy, as the ultrasonographer said, "There's the turtle!"

Despite these results, my obstetrician still felt I should have an amniocentesis. I switched physicians over a disagreement with him about this. A group of physicians, who only treated high-risk patients such as myself, took over my pregnancy management and agreed with me that I did not need an amniocentesis. Nine months after adopting our daughter, I gave birth to a 7 lb 12 oz boy, born naturally.

So far, I've avoided the question, "What would we have done if the tests were positive for spina bifida or Down Syndrome?" Although I don't think I could say categorically until faced with the situation, at that time, our goal in seeking testing was to be prepared for a child with impairments.

Into the 21st Century

In 1998, the first intrauterine surgery was performed to close the spinal lesion prior to birth. This procedure is currently being evaluated through a prospective, randomized

clinical trial funded by the National Institutes of Health. In the United States, there are four centers that are currently participating in the study. So far, the surgery has decreased the need for a shunt at birth (Bruner et al., 1999).

> *Looking back on my TV appearance 23 years ago, I realized this was my first experience as a disability activist. At the time, it was not a conscious effort at taking a political stand, but a natural one, moved by my personal experience. Today, I am more firmly against selective abortion but consider myself pro-choice. Reading the disability literature and attending a seminar led by disability activist and political scientist Harlan Hahn has strongly influenced my thinking and awareness. This happened not just by reading the disability literature, some of which I had previously read; it was hearing Professor Hahn's vivid personal stories that moved me to refine my viewpoint and self-image. The seminar was a transformative experience through which I began to view myself as disabled, rather than a person with spina bifida striving to be "normal." I realized I also had stories to tell and began to do so in my writing. For example, in a recent article in the* American Journal of Occupational Therapy *(Neville-Jan 2003), I challenged several widely accepted views about chronic pain. In another paper, published in* Disability & Society *(Neville-Jan, 2004), I used autoethnography to connect the personal experience of pain more directly with political action.*
>
> *Being a relative newcomer to the field of disability studies, I naively thought I could write this paper from an apolitical stance. However, to truly "get" disability studies is to realize that "an apolitical disability stance" is an oxymoron. Disability studies by its nature is political. bell hooks (1989), a radical feminist writer, provided a cogent way to understand the process involved in realizing the true meaning of the statement "the personal is political." She states:*
>
> *Politicization of the self can have its starting point in an exploration of the personal wherein what is first revolutionized is the way we think about the self. To begin revisioning, we must acknowledge the need to examine the self from a new, critical standpoint. Such a perspective, while it would insist on the self as a site for politicization, would equally insist that simply describing one's experience of exploitation or oppression is not to become politicized. It is not sufficient to know the personal but to know—to speak it in a different way. Knowing the personal might mean naming spaces of ignorance, gaps in knowledge, ones that render us unable to link the personal with the political. (p. 107)*

In the discussion that follows, I will problematize prevention and spina bifida and speak of prevention in a different manner.

Discussion

Prevention

Typically we think about prevention as a good thing. In fact, much government spending in the United States targets prevention programs. For example, research results coupled with educational campaigns alert us that attention to diet, weight, and physical activity level can potentially reduce our risks for cardiovascular disease, diabetes, and stroke (U.S. Department of Health and Human Services, n.d.). According to the U.S. Preventative Services Task Forces' *Guide to Clinical Preventive Services* (1996) these examples represent primary prevention activities that ". . . refer(s) to the complete prevention of disease, often through methods that inhibit exposure to risk factors" (p. 1). Primary prevention aims to completely eradicate disease. Because a heavy economic and social burden results from diseases such as cancer and cardiovascular disease, programs focused on primary prevention incur the least cost to society. The list of primary preventive actions that are recommended by government and health officials are innumerable and constantly changing, based on biomedical and genetic research into the causes of disease. Several emerging areas of occupational therapy practice have addressed primary prevention, such as assessments and interventions for preventing school violence and workplace injury (Malugani, 2004). The "Well Elderly" program has received wide acclaim and given birth to a variety of targeted applications of the prevention-oriented *lifestyle redesign* genre of intervention (Clark et al., 1997).

Secondary prevention aims to detect the early onset of disease and limit disease effects (U.S. Preventative Services Task Force). Screening tests done on persons who are at risk for or who exhibit initial symptoms of a disease can potentially alter the course of a disease such as breast cancer or diabetes. Occupational therapists are increasingly involved in promoting healthy lifestyles in people screened for or who are already diagnosed with diabetes, arthritis, and heart disease.

Tertiary prevention involves treatment and rehabilitative activities in order to prevent further impairment. The goal is to restore a higher level of functioning in someone already with a diagnosis (U.S. Preventative Services Task Force, 1996). This level of prevention is the most costly to society, as it takes place after a person has already acquired some degree of impairment. Tertiary prevention continues to be a major practice component of occupational therapy within the rehabilitation paradigm.

Prevention and Spina Bifida

Medical technological advances have greatly aided primary and secondary prevention of spina bifida. Primary prevention actions are those timed prior to conception and include recommendations for women of child-bearing years to supplement their diet with folic acid (Geisel, 2003). In fact, folic acid campaigns are ubiquitous. The Centers for Disease Control and Prevention (CDC), the March of

Dimes, the SBAA, and the National Council on Folic Acid have developed extensive educational materials and prevention activities informing and motivating women to use a diet supplement with folic acid prior to conception to help prevent neural tube defects. These efforts have been successful, as evidenced by a decrease in the incidence of spina bifida (Centers for Disease Control and Prevention [CDC], 2002).

Secondary prevention of spina bifida includes activities that occur prenatally, such as screening, terminating pregnancy (also called *selective termination* or *selective abortion*), and, most recently, intrauterine fetal surgery. Intrauterine surgery, currently being tested in clinical trials, appears to reduce at birth the impairments related to hydrocephalus. Among secondary prevention activities, intrauterine surgery produces a child with spina bifida but with fewer and less severe impairments. However, prevention associated with prenatal screening, selective abortion, or withholding treatment at birth has been the subject of much controversy.

Technological advances, such as the development of the shunt, have also influenced tertiary prevention, in that during the past 50 years, of those infants that are born with spina bifida, more have survived into adulthood (Brown, 2001; Dillon, Davis, Duguay, Seidel, & Shurtleff, 2000; Hunt, Oakeshott, & Kerry, 1999). Nonetheless, due to their high cost, tertiary prevention efforts are far less supported today by insurance companies and government agencies. The fact that only a few studies have examined the life experiences of individuals with spina bifida supports this view (Liptak, 2003).

Associations and government offices involved in policy decisions about spina bifida express the dual aims to both prevent spina bifida and to improve the quality of life for persons with spina bifida (tertiary prevention). In most cases, prevention is listed first. Can the government, or any other public health organization, support both? Can education about spina bifida portray both a positive view of growing up as well as the difficulties of managing life with severe impairments? Is secondary prevention such as prenatal screening with subsequent termination really a disguise for euthanasia, or does prenatal screening provide an opportunity for informed decision making?

Reflections on Prevention

Over the past 4 years, I have been immersed in the literature about spina bifida. From my standpoint as a person with spina bifida, primary and secondary prevention efforts seem to be based primarily on negative views of spina bifida. For me, the messages conveyed are the following: (a) spina bifida is a severe and devastating birth defect, (b) spina bifida negatively impacts the quality of life of a child and his or her family, and (c) the social and economic burden of spina bifida is substantial.

Spina bifida is a severe and devastating birth defect.

Although it cannot be disputed that folic acid campaigns have been successful in reducing the number of infants born with neural tube defects (CDC, 2002), as a person with spina bifida, I find descriptions of spina bifida in the medical literature that are associated with these campaigns to be overwhelmingly negative. For example, the CDC, a major national promoter of folic acid, provides an educational booklet for "campaign partners" (CDC, 2001). The booklet profiles a family that has two children born with spina bifida. One child has ". . . a severe form of spina bifida" and the other child ". . . a milder form of spina bifida." More alarming to me than the words are the pictures that accompany the text. The first picture depicts the infant's back with the unaltered sac prior to surgery. Next are photographs that show the scars left from surgery. More photos illustrate gastrostomy tubes, a breathing apparatus, splints, crutches, and a wheelchair. Both children are pictured smiling, but the text describes a negative perspective that ". . . most affected children have some learning disability. Many of these children have problems with bowel and bladder control, a source of major concern and embarrassment to them" (p. 8). Although the information presented is accurate, it is overwhelmingly negative. It reminds me of the news program in the '80s, "The Terrible Choice" (Utley, 1982).

Bannister (2000) presented arguments for and against intrauterine surgery for spina bifida. At the end of the article, he stated:

> It cannot be contested that spina bifida aperta is anything other than a major disaster affecting the whole of the central nervous system, and minor improvements in the neurological status do not make the outcome significantly better. (p. 112)

I disagree. For me, spina bifida has not been a "major disaster." Peter Singer (2002), a controversial bioethicist recently appointed to Princeton University as a Professor of Bioethics at the Center for Human Values, describes spina bifida. He states:

> . . . the infant is born with its spinal cord exposed. Three out of every thousand babies have this condition, which adds up to a large number of babies. Although treatment is possible, with the more severe cases even immediate surgery and vigorous treatment will not result in successful rehabilitation. The children will grow up severely handicapped, both mentally and physically and they will probably die in their teens. (p. 226)

After further descriptions of a child with spina bifida as incontinent, paralyzed, blind, retarded, spastic, and with

hydrocephalus so severe as not to be able to hold his or her head up, Singer stated that "the obvious alternative" to bringing up such a child, namely "a swift, painless death for the infant," is against the law. Although his perspective does not represent all bioethicists, Singer's status and position give him a powerful voice in this debate. Not Dead Yet (www.notdeadyet.org), a disability activist organization, has challenged mercy killing and views it as the ultimate means of discrimination.

Spina bifida negatively impacts the quality of life of a child and his or her family.

As presented earlier, Lorber (1971) justified the nontreatment of infants with spina bifida based on several prognostic criteria. Infants not treated and left to die (actually, some infants took 9 months to die) were those who possessed severe handicaps. Treatment was withheld based on the imagined future quality of life for these infants. Shaw (1977), a pediatric surgeon, provided a "rule of thumb" for determining quality of life in these life and death situations: QL = NE x (H + S). This means that quality of life results from the interaction of the infant's natural endowment and contributions of home and society. Hahn (2002) is highly critical of this formula, stating that according to this calculation, ". . . non-disabled rich people enjoy a higher 'quality of life' than persons who are poor and disabled" (p. 174). Quality of life arguments for withholding treatment have been criticized as being too vague and unclear about who is being served. Are decisions made about the well-being of the infant or about the family and society? Also, and most importantly, where do you draw the line in determining whose quality of life is unacceptable? Disability studies writers (Kittay & Kittay, 2000; Mostert, 2002) refer to the "slippery slope" related to these arguments. The genocide in Nazi Germany began with exterminating those who were deemed "defective." Will selective termination and prenatal testing result in attempts to get rid of all people society labels as "undesirable"? How often is treatment withheld in the neonatal intensive care unit today? Singer (2002) reports of a "recent investigation" in the nursery at Yale-New Haven. He states:

> . . . over a 2 1/2-year period, 43 deaths, or 14 percent of all deaths in the nursery, were related to withholding treatment. The decision to withhold treatment at this nursery was in each case made by parents and physicians together on the basis that there was little or no chance for a meaningful life for the infant. (p. 225)

Singer (2002) contends that needless suffering occurs because society fails to see that ". . . the case of a dying horse is really quite parallel to the case of a dying infant" (p. 225). Would we let a horse die in agony rather than kill it? Then why do we do that with infants who will probably die? Further, he states, "The burden on the family can easily be imagined, and it is doubtful if the child's life is a benefit for him." (p. 226)

The social and economic burden of spina bifida is substantial.

The SBAA (n.d.) describes the economic costs to society of spina bifida to exceed $200 million per year. As Kielhofner stated in his opening paper (2005), rehabilitation was driven by the need to reduce the economic burden to society of persons with disabilities. Today, economists calculate the cost of a disability. Medical researchers use this information as a rationale for support or contest policy decisions. The social and economic consequences for an individual parent raising a child with an impairment are also considered in the decision to terminate a pregnancy.

Singer proposed "active euthanasia" as a "rationale basis" for managing the resource allocation problem in health care. Similar views are expressed by Buchanan, Brock, Daniels, and Wikler (2000). They focus primarily on prenatal testing and reproductive freedom and believe that a fetus is not self-conscious and therefore not a person. In response to critiques from the disability community that their views devalue persons with disabilities, they state:

> Advocating the fourth model of intervention (selective abortion) is tantamount to saying that people like you (who have disabilities) have no right to exist only on the highly controversial assumption that fetuses are persons. (p. 280)

Buchanan et al. (2000) further assert that the disability rights critique is the "familiar objection to the age-old practice of abortion" (p. 280). However, many in the disability community are pro-choice but against selective abortion (Parens & Asch, 2000).

The disability rights movement has been very vocal in the worry that the new genetics will become the new eugenics. Bioethicists such as Peter Singer at Princeton University and Dan Brock at Brown University have prominent positions through which to transmit their views. People in the disability community feel that their unique perspective about their own quality of life is mostly disregarded by the medical community, including genetics counselors, genetics researchers, and bioethicists (Anstey, 2003; Wertz, 2000).

Summary

Many myths and negative conceptions exist in society about persons with impairments. These preconceptions direct many decisions both at the beginning and end of life, and reflect a lack of exposure to the viewpoints and lived experiences of persons with impairments. Each situation is

different, highly complex, and poses its own moral dilemmas. Parens & Asch (2000) present an excellent discussion of the disability rights critique.

Implications for Occupational Therapy

Disability studies calls for dismantling the stereotypical thinking that exists in society about disability. However, in many occupational therapy academic programs, disability studies is only beginning to become an integral part of the curriculum. Wright (2004) proposed that occupational therapy programs include disability studies through the incorporation of the World Health Organization's *International Classification of Functioning, Disability and Health (ICF)* (WHO, 2001). The ICF aims to incorporate both a medical model and social model of disability. Occupational therapists need to gain an increased awareness of the multiple perspectives that exist within the community of persons with impairments. Occupational therapy researchers need to add to the literature with both qualitative and quantitative research, allowing the voices of study participants with impairments to be heard and their lived experiences to be explored and understood. Occupational therapists need to become aware of the current status of topics such as euthanasia. For example, according to an Associated Press report (2004, November 30), a study by Dutch physicians found that, since 1997, 22 newborns with severe spina bifida were killed. In the Netherlands, euthanasia is legal for adults. However, it is illegal to euthanize persons who are unable to make informed decisions such as terminally ill infants and persons in a coma. The report stated that one hospital has already developed a protocol for infant euthanasia and applied it to euthanize four newborn infants. The guidelines suggest that the medical team, including independent physicians and parents, make the decision based on amount of unrelieved pain and lack of future improvement. The report pointed out that this practice goes largely unreported and is hidden in medical practice. The Gronigen Protocol, named after the hospital where this practice takes place, reminds me of Lorber's criteria used in the '70s to determine which infants with spina bifida should live (Lorber, 1971).

Harlan Hahn (personal communication, March 2004) has suggested that parents who are predicted to give birth to a child with a disability be offered the opportunity to meet a person with the same disability so they can see for themselves the reality of their lives. Carla Verpoorten (2000), in an overview of the international literature about termination of pregnancy in the case of spina bifida, states: "The pessimistic public opinion has to be changed before we can assure prospective parents that they and their future child will be welcomed whether or not the child has a disability" (p. 3). Only then can truly informed choices be made. Disability studies provides a way to reframe thinking about persons with impairments. Armed with this information, occupational therapists can act as advocates, and, in a more informed manner, support or contest policy initiatives.

Conclusion

IFsbh hosted its 12th conference in 2000 in Toulouse, France. The conference produced "Resolution Toulouse 2000" concerning the theme, "The Right to be Different." The following statements summarize the resolution and were endorsed by adults and parents of children with spina bifida:

- People with spina bifida and hydrocephalus live a full life with equal value to that of any other citizen
- Adults with spina bifida stated very clearly that their quality of life is not a reason for abortion
- Parents and adults state that the impairment is not the burden. The burden is the constant struggle against inaccessible environments and inadequate systems, which should be allies and should be supportive of them http://www.ifglobal.org/pregnancy.asp?lang=1&main=8&sub=3

This resolution provides an alternate view of spina bifida, not as the disaster and tragedy presented in the medical literature, but as a positive experience for the person and their family.▲

Acknowledgments

I wish to thank the research team of "Boundary Crossing: An Ethnographic & Longitudinal Study" including, Dr. Mary Lawlor (Principal Investigator), Dr. Cheryl Mattingly (Co-Principal Investigator), Dr. Lanita Jacobs-Huey (Co-Investigator), Melissa Park, Nancy Bagatell, Amy Buffington, Erica Angert, Susan Stouffer, Jeanine Blanchard, and Jeanne Gaines for their valuable comments. I thank Melissa Park for her persistence in finding the NBC tape. Faryl Reingold was an expert editor and commenter. This paper draws on experiences gained through funded projects from the National Center for Medical Rehabilitation Research, National Institute of Child Health and Human Development of the National Institutes of Health grant #RO1 HD 3887801A1 and disability supplement #RO1 HD 3887801A1S.

Endnote

[1]In this paper, I prefer to use person-first language and use the term "impairment" rather than "disability." For example, I say "women with physical impairments" rather than "disabled women" or "women with a

disability." Those in the disability movement have recommended using disability-first language as a means of resistance to and rejection of the dominance of nondisability in society. Activists in the disability movement felt that the term "impairment" tended to focus on "cure" and directed money towards medicine ". . . rather than on barriers which make it possible to participate in the dominant culture with an impairment" (Russell, 1998, p. 16). This so-called *social model of disability* has achieved some success in challenging ". . . disablism, oppression and exclusion in society" (Hughes & Paterson, 1997, p. 325). However, several writers (Hughes & Paterson; Stineman, 2001) have called for a recognition of the embodied nature of disability and thus more attention be devoted to impairment. My language preference reflects this embodied perspective of disability. I feel my language choice does not detract from my message that is equally about medicine and attitudinal barriers in society.

References

Adams, K. L. (2000). Genetics breakthroughs shape OT intervention. *OT Practice Online*. Retrieved August 11, 2004, from http://www.aota.org/featured/area2/links/link16ay.asp

Andren, E., & Grimby, G. (2000). Dependence and perceived difficulty in activities of daily living in adults with cerebral palsy and spina bifida. *Disability and Rehabilitation, 22*, 299–307.

Anstey, K. W. (2003). Prenatal testing and disability: The need for a participatory approach to research. In S. Y. Song, Y. M. Koo, & D. Macer (Eds.), *Bioethics in Asia in the 21st century* (pp. 347–356). Tsukuba Science City, Japan: Eubios Ethics Institute. Retrieved March 2005, from http://www.biol.tsukuba.ac.jp/~macer/ABC4/abc4347.htm

Appleton, P. L., Minchom, P. E., Ellis, N. C., Elliot, C. E., Boll, V., & Jones, P. (1994). The self concept of young people with spina bifida: A population based study. *Developmental Medicine & Child Neurology, 36,* 198–215.

Associated Press. (2004, November 30). Hospital carries out procedure on few ill infants. MSNBC News: Children's Health. Retrieved March 2005, from http://www.biol.tsukuba.ac.jp/~macer/ABC4/abc4347.htm

Bannister, C. (2000). The case for and against intrauterine surgery for myelomeningoceles. *European Journal of Obstetrics & Reproductive Biology, 92*(1), 109–113.

Bergman, J. (1994). Protecting the mobility of the aging persons with cerebral palsy or spina bifida. In D. Lollar (Ed.), *Preventing secondary conditions associated with spina bifida and cerebral palsy: Proceedings and recommendations of a symposium, February 17–19, 1994, Crystal City, Virginia* (pp. 54–64). Washington, DC: Spina Bifida Association of America.

Bradbury, R. (1950). *Fahrenheit 451*. New York: Random House.

Brown, J. (2001). Orthopaedic care of children with spina bifida: You've come a long way, baby! *Orthopaedic Nursing, 20,* 51–58.

Bruner, J. P., Tulipan, N., Paschall, R. L., Boehm, F. H., Walsh, W. F., Silva, S. R., et al. (1999). Fetal surgery for myelomeningocele and the incidence of shunt-dependent hydrocephalus. *JAMA, 282,* 1819–1825.

Buchanan, A., Brock, D., Daniels, N., & Wikler, D. (2000). *From chance to choice: Genetics & justice*. Cambridge, England: Cambridge University Press.

Centers for Disease Control and Prevention. (2001). Living with spina bifida: A glimpse for prevention campaign partners. Retrieved August 10, 2004, from http://www.cdc.gov/Images_-_Video_and_Audio/Images/Birth_Defects/Folic_Acid/LivingwithSB_Eng.pdf

Centers for Disease Control and Prevention. (2002). Spina bifida and anencephaly prevalence—United States, 1991–2000. *Morbidity and Mortality Weekly Report*, *51*(RR-13), 9–11.

Clark, F., Azen, S. P., Zemke, R., Jackson, J., Carlson, M., Mandel, D., et al. (1997). Occupational therapy for independent-living older adults. A randomized controlled trial. *JAMA, 278*, 1321–1326.

Davidoff, M., Petrini, J., Damus, K., Russell, R., & Mattison, D. (2002). Neural tube defect-specific infant mortality in the United States. *Teratology, 66,* S17–S22.

Dennis, M., & Barnes, M. (2002). Math and numeracy in young adults with spina bifida and hydrocephalus. *Developmental Neuropsychology, 21,* 141–155.

Dennis, M., Fletcher, J., Rogers, T., Hetherington, R., & Francis, D. (2002). Object-based and action-based visual perception in children with spina bifida and hydrocephalus. *Journal of the International Neuropsychological Society, 8,* 95–106.

Dennis, M., Rogers, T., & Barnes, M. (2001). Children with spina bifida perceive visual illusions but not multistable figures. *Theoretical and Experimental Neurolopsychology, Tennet XI,* 108–113.

Dillon, C., Davis, B., Duguay, S., Seidel, K., & Shurtleff, D. (2000). Longevity of patients born with myelomeningocele. *European Journal of Pediatric Surgery, 10*(Suppl.), 33–34.

Dunne, K., Gingher, N., Olsen, L., & Shurtleff, D. (1986). *A survey of the medical and functional status of members of the adult network of the Spina Bifida Association of America*. Rockville, MD: Spina Bifida Association of America.

Ellis, C., & Bochner, A. (2000). Autoethnography, personal narrative, reflexivity. In N. K. Denzin & Y. S. Lincoln (Eds.), *Handbook of qualitative research* (2nd ed., pp. 733–768). Thousand Oaks, CA: Sage.

Farley, T., Vines, C., McCluer, S., Stephans, V., & Hunter, J. (1994). Secondary disabilities in Arkansans with spina bifida. *European Journal of Pediatric Surgery, 4* (Suppl. 1), 39–40.

Feuchtbaum, L., Currier, R., Riggle, S., Roberson, M., Lorey, F., & Cunningham, G. (1999). Neural tube defect prevalence in California (1990–1994): Eliciting patterns by type of defect and maternal race/ethnicity. *Genetic Testing, 3*, 265–272.

Geisel, J. (2003). Folic acid and neural tube defects in pregnancy: A review. *Journal of Perinatal & Neonatal Nursing, 17*(4), 268–279.

Graves, J. C., Miller, K. E., & Sellers, A. D. (2002). Maternal serum triple analayte screening in pregnancy. *American Family Physician, 65*, 915–920.

Hahn, H. (2002). Academic debates and political advocacy: The U.S. disability movement. In C. Barnes, M. Oliver, & L. Barton (Eds.), *Disability studies today* (pp. 162–189). Cambridge, England: Polity Press.

Harper, J. C. (2001). Introduction. In J. C. Harper, J. D. Delhanty, & A. H. Handyside (Eds.), *Preimplantation genetic diagnosis* (pp. 3–12). London: Wiley.

Holmbeck, G. N., & Faier-Routman, J. (1995). Spinal lesion,

shunt status, family relationships and psychosocial adjustment in children and adolescents with spina bifida myelomeningocele. *Journal of Pediatric Psychology, 20,* 817–832.

hooks, b. (1989). *Talking back: Thinking feminist, thinking black*. Boston: South End Press.

Hughes, G., & Paterson, K. (1997). The social model of disability and the disappearing body: Towards a sociology of impairment. *Disability & Society, 12,* 325–340.

Hunt, G. (1990). Open spina bifida: Outcome for a complete cohort treated unselectively and followed in to adulthood. *Developmental Medicine and Child Neurology, 32,* 108–118.

Hunt, B., Oakeshott, P., & Kerry, S. (1999). Link between the CSF shunt and achievement in adults with spina bifida. *Journal of Neurology, Neurosurgery & Psychiatry, 67*, 591–595.

Hunt, G., & Poulton, A. (1995). Open spina bifida: A complete cohort reviewed 25 years after closure. *Developmental Medicine and Child Neurology, 37,* 19–29.

Huxley, A. (1932). *Brave new world*. New York: HarperCollins.

International Federation for Spina Bifida and Hydrocepahalus. (n.d.). *IFsbh policy statement*. Retrieved March 5, 2005, from http://www.ifglobal.org/pregnancy.asp?lang=1&main=8&sub=3

Kielhofner, G. (2004). *Conceptual foundations of occupational therapy* (3rd ed., pp. 238–253). Philadelphia: F. A. Davis.

Kielhofner, G. (2005). Rethinking disability and what to do about it: Disability studies and its implications for occupational therapy practice. *American Journal of Occupational Therapy, 59,* 487–496.

Kittay, E. F., & Kittay, L. (2000). On the expressivity and ethics of selective reproductive decision making and policies. In E. Parens & A. Asch (Eds.), *Prenatal testing and disability rights* (pp. 165–195). Washington, DC: Georgetown University Press.

Kinsman, S. L., & Doehring, M. C. (1996). The cost of preventable conditions and adaptive behavior with spina bifida. *European Journal of Pediatric Surgery, 6*(Suppl. 1), 17–20.

Lary, J. M., & Edmonds, L. D. (1996). Prevalence of spina bifida at birth—United States, 1983–1990: A comparison of two surveillance systems. *Morbidity and Mortality Weekly Report, 45*(SS-22), 15–26.

Liptak, G. S. (2003). Summary of findings. In G. S. Liptak (Ed.), *Evidence-based practice in spina bifida: Developing a research agenda* (pp. 10–12). Washington, DC: U.S. Department of Health and Human Services & U.S. Department of Education.

Lollar, D. (1994a). Encouraging personal and interpersonal independence. In D. Lollar (Ed.), *Preventing secondary conditions associated with spina bifida and cerebral palsy: Proceedings and recommendations of a symposium, February 17–19, 1994, Crystal City, Virginia* (pp. 17–25). Washington, DC: Spina Bifida Association of America.

Lollar, D. (1994b). *Preventing secondary conditions associated with spina bifida or cerebral palsy: Proceedings and recommendations of a symposium, February 17–19, Crystal City, Virginia.* Washington, DC: Spina Bifida Association of America.

Lorber, J. (1971). Results of treatment of myelomeningocele: An analysis of 524 unselected cases, with special reference to possible selection for treatment. *Developmental Medicine & Child Neurology, 13*, 279–303.

Lowry, L. (1993). *The giver*. New York: Laurel Leaf Books.

Malugani, M. (2004). *Emerging areas in occupational therapy*. Retrieved March 2005, from http://www.otjoblink.org/links/link09.asp

Marge, M. (1994). Toward a state of well-being: Promoting healthy behaviors to prevent secondary conditions. In D. J. Lollar (Ed.), *Preventing secondary conditions associated with spina bifida or cerebral palsy: Proceedings and recommendations of a symposium, February 17–19, Crystal City, Virginia* (pp. 87–93). Washington, DC: Spina Bifida Association of America.

Mazur, J., Shurtleff, D., Menelaus, M., & Colliver, J. (1989). Orthopaedic management of high-level spina bifida. *Journal of Bone and Joint Surgery, Incorporated, 71-A,* 56–61.

Merkens, M. J. (1997). Care of the child with neural tube defect: The spina bifida program at CDRD. Retrieved March 2005, from http://cdrc.ohsu.edu/clinical/portland/spinabifida/neuraltube.html

Mostert, M. (2002). Useless eaters: Disability as genocidal marker in Nazi Germany. *Journal of Special Education, 30,* 155–168.

Neville-Jan, A. (2003). Encounters in a world of pain: An autoethnography. *American Journal of Occupational Therapy*, *57*, 88–98.

Neville-Jan, A. (2004). Selling yourself to the devil: An autoethnography of pain, pleasure, and the quest for a child. *Disability & Society, 19*(2), 113–127.

Query, J., Reichelt, C., & Christoferson, L. (1990). Living with chronic illness: A retrospective study of patients shunted for hydrocephalus and their families. *Developmental Medicine and Child Neurology, 32,* 119–128.

Parens, E., & Asch, A. (2000). *Prenatal testing and disability rights.* Washington, DC: Georgetown University Press.

Peterson, P., Rauen, K., Brown, J., & Cole, J. (1994). Spina bifida: The transition into adulthood begins in infancy. *Rehabilitation Nursing, 19,* 229–238.

Powell, C. M. (2000). The current state of prenatal genetic testing in the United States. In E. Parens & A. Asch (Eds.), *Prenatal testing and disability rights.* Washington, DC: Georgetown University Press.

Reed-Danahay, D. (1997). Introduction. In D. Reed-Danahay (Ed.), *Auto/ethnography: rewriting the self and the social*. New York: Berg.

Rekate, H. (1991). *Comprehensive management of spina bifida*. Boca Raton, FL: CRC Press.

Russell, M. (1998). *Beyond ramps: Disability at the end of the social contract*. Monroe, ME: Courage Common Press.

Shaw, A. (1977). A short formula for quality of life. *Hastings Center Report, 5*(1), 37–43.

Singer, P. (2002). *Unsanctifying human life*. In H. Kuhse (Ed.), Unsanctifying human life. Malden, MA: Blackwell.

Singh, D. (2003). Families of children with spina bifida: A review. *Journal of Developmental and Physical Disabilities, 15,* 37–55.

Smith, G. K. (2001). The history of spina bifida, hydrocephalus, paraplegia, and incontinence. *Pediatric Surgery International, 17*, 424–432.

Spina Bifida Association of America. (n.d.). *About spina bifida*. Retrieved March 3, 2005, from http://www.sbaa.org/site/PageServer?pagename=about_sb

Stineman, M. G. (2001). Defining the population, treatments, and outcomes of interest: Reconciling the rules of biology with meaningfulness, *American Journal of Physical Medicine & Rehabilitation, 80,* 283–290.

Swank, M., & Dias, L. (1992). Myelomeningocele: A review of the orthopaedic aspects of 206 patients treated from birth with no selection criteria. *Developmental Medicine Child Neurology, 34,* 1047.

Turk, M. (1994). Attaining and retaining mobility: Clinical issues. In D. Lollar (Ed.), *Preventing secondary conditions associated with spina bifida and cerebral palsy: Proceedings and recommendations of a symposium, February 17–19, 1994, Crystal City, Virginia* (pp. 42–53). Washington, DC: Spina Bifida Association of America.

U.S. Department of Health and Human Services. (n.d.). *Safety and wellness*. Retrieved March 6, 2005, from http://www.hhs.gov/safety/index.shtml

U.S. Preventative Services Task Force. (1996). *Guide to clinical preventative services* (2nd ed.). Baltimore: Williams & Wilkins.

Utley, G. (Reporter). (1982). The terrible choice. *NBC News Magazine* [Television broadcast]. Retrieved from archives in 2002. New York: National Broadcasting Corporation.

Verpoorten, C. (2000). *Termination of pregnancy when the unborn child has spina bifida and/or hydrocephalus: An overview on international literature*. Retrieved August 20, 2004, from http://www.ifglobal.org/human_rights.asp?lang=1&main=9&sub=4

Wertz, D. (2000). Drawing lines: Notes for policymakers. In E. Parens & A. Asch (Eds.), *Prenatal testing and disability rights* (pp. 261–287). Washington, DC: Georgetown University Press.

World Health Organization. (2001). *International classification of functioning, disability and health (ICF).* Geneva, Switzerland: Author.

Wright, B. A. (2004, Fall). Redefining disability to promote equality: The role of disability studies in educating occupational therapists. *Disability Studies Quarterly, 24*(4). Retrieved March 2005, from http://www.dsq-sds.org/2004_fall_toc.html