


A Comparative Case Study of Hospice and Hospital End-of-Life Care for Aging Adults With Developmental Disabilities

Global Qualitative Nursing Research
Volume 9: 1–11
© The Author(s) 2022
Article reuse guidelines:
sagepub.com/journals-permissions
DOI: 10.1177/23333936221087626
journals.sagepub.com/home/gqn


Jacqueline M. McGinley¹  and Christina N. Marsack-Topolewski²

Abstract

Greater attention is being paid to issues surrounding end-of-life care for aging adults with developmental disabilities. The purpose of this qualitative study was to explore the end-of-life experiences of two aging adults with developmental disabilities and life-limiting serious illnesses who received care in settings in the United States. Using a comparative case study design, data from three sources (records, staff, surrogates) were collected sequentially and triangulated via within and cross-case analyses. Although the setting and design limit the generalizability of these findings, the feasibility of delivering high quality care to adults with developmental disabilities as they age and experience terminal illnesses is supported. Insights are presented regarding how nurses can address barriers by adapting policies and practices to accommodate the distinct needs that arise as people with developmental disabilities age, become seriously ill, and reach life's end.

Keywords

developmental disabilities, case study, end-of-life care, serious illness, aging; qualitative research, Northeastern United States

Received May 24, 2021; revised February 18, 2022; accepted February 25, 2022

The leading causes of death for individuals with developmental disabilities are now similar to those found in the general population, including: respiratory disease, heart and circulatory disorders, and cancer (Landes et al., 2019; Tuffrey-Wijne et al., 2016). The life expectancy of individuals with developmental disabilities has increased in recent years, with the average lifespan now ranging from early 50s to 60s with variability often attributable to the severity of the developmental disability and other co-occurring conditions (Lauer & McCallion, 2015). Older adults with developmental disabilities, however, seem to remain at greater risk for premature and avoidable death when compared to the general population (Landes et al., 2021).

This may, at least in part, be attributable to some distinct morbidity and mortality patterns that have been observed among people with developmental disabilities. A study conducted in Sweden identified a higher mortality risk among older adults with developmental disabilities, which was attributed namely to “respiratory, nervous, and circulatory diseases” (Ng et al., 2017, p. 1). There is also evidence to suggest that Alzheimer’s disease and dementia disproportionately affects a subset of the population of people with developmental disabilities (Takenoshita et al., 2020). Approximately 8% of all adults with developmental disabilities have been diagnosed with dementia, and the prevalence

of the disease among those who have been diagnosed with Down syndrome increases to over 50% as individuals reach advanced age (Bishop et al., 2015). Yet, McCallion et al. (2019) have noted that the absence of population data related to chronic conditions and serious illnesses commonly found among individuals with a developmental disability has largely resulted in their needs remaining hidden.

There is, however, a body of research emerging from the United Kingdom, Australia, and the Netherlands to suggest that unique factors disproportionately shape the experiences of people with developmental disabilities at the end of life, including: behavioral health needs, complex comorbidities, and polypharmacy (Nicholas et al., 2017; O’Dwyer et al., 2016; Tuffrey-Wijne et al., 2016). Botsford (2000) noted several other individual-level factors that influence the response of people with developmental disabilities to death and dying, including: past learning and experience, intellect, communication skills, and family and staff perceptions of

¹Binghamton University, NY, USA

²Eastern Michigan University, Ypsilanti, USA

Corresponding Author:

Jacqueline M. McGinley, Department of Social Work, Binghamton University, PO Box 6000, Binghamton, NY 13902, USA.
Email: jmcginle@binghamton.edu



both the person and the circumstances of serious illness. A recent article by Fisher et al. (2020) indicated that nurses are uniquely positioned to offer a holistic approach to care for people with developmental disabilities. This holistic approach can promote self-determination and safeguard against harm at the end of life. Fisher et al. (2020) suggested that the use of a holistic approach is possible when nurses are aware of, trained in, and empowered to uphold their ethical commitments. Agency factors can also influence providers' responses to individuals with a developmental disability who are nearing life's end. These agency factors may include intricacy of staff roles, administrative policies and procedures, funding constraints, and complexity of community networks of service (Botsford, 2000; Nicholas et al., 2017).

Due to many of these factors, people with developmental disabilities experience an underutilization of hospice and palliative care services, as well as high rates of hospital/institutional deaths (estimated at >80%; Friedman & Helm, 2010). Todd et al. (2021) similarly found in their exploratory study in Australasia that the majority of people with developmental disabilities died within a hospital setting. Community-based agencies in the United States often become increasingly reluctant or unable to provide specialized support and services when an individual's serious illness warrants palliative care, hospice, and/or hospitalization due to questions and concerns related to insurance eligibility (Lindley, 2018). Findings from the Institute of Medicine (IOM, 2014) also suggested that for individuals with a developmental disability, mismatches often occur between the needs of patients/families and the services that are available or can be reasonably obtained. The fragmented healthcare delivery system, marred by financial incentives, has often resulted in poor care coordination at the end of life (Harrington, 2018; IOM, 2014).

Research has also suggested a disconnect between disability providers and hospice/palliative care services (Dunkley & Sales, 2014; Hahn et al., 2015). While there is a growing body of research focused on interdisciplinary cross-training and care coordination, these interventions often faced significant implementation barriers, achieved inconsistent outcomes, and lacked strategies for widespread dissemination (Lindley, 2018; Ronneberg et al., 2015). Yet, the IOM (2014) has continued to recommend that "integrated, person-centered, family-oriented, and consistently accessible care" be available to all individuals who experience a serious illness or injury (pp. 2–45). More recently, Clark and Watts (2021) reflected upon a humanistic approach to care. While establishing four typologies for dentistry care, the scholars actually articulated a path for healthcare providers to realize the IOM's vision for high-quality, end-of-life care for people with developmental disabilities, including embracing listening and understanding, improving training, tailoring care, and advocating for equity (Clark & Watts, 2021). If an inclusive end-of-life experience is to be achieved, these features are important to ensure the harmony between individuals' values and the care they receive.

Through a combination of advocacy, legislative efforts, and judicial rulings, the deinstitutionalization movement that commenced in the 1980s has led to a paradigm shift whereby most individuals with a developmental disability no longer receive care and services in large, congregate settings but rather live in a family home or small community residence (Braddock, 2002). As a result of this shift, individuals with a developmental disability have necessarily adopted an increased reliance on mainstream forms of healthcare and services (Heller, 2019). However, current research specific to people with developmental disabilities has rarely explored where and how end-of-life care is rendered. This comparative case study sought to address this gap by exploring the end-of-life experiences of two aging adults with developmental disabilities and life-limiting serious illnesses in the United States, thereby illuminating the myriad factors that inform care across settings.

Methods and Analysis

Study Design

The findings presented in this paper were derived from a larger exploratory study that employed a multiple-case study design to describe features perceived to facilitate or impede "good deaths" among aging adults with developmental disabilities in community residences within the United States. For the purposes of this paper, a comparative case study design was used to describe the complex and dynamic interplay between policy and practice that inform the trajectory of how two individuals with developmental disabilities reached life's end. The aim of this descriptive case study was to retrospectively sequence the events that preceded and proceeded end of life, elucidating the phenomena of death for two aging adults with developmental disabilities.

Study Setting

The study was conducted in collaboration with a not-for-profit organization that provides services to individuals with various disabilities and other specialized needs across one state within the northeastern region of the United States; this organization will be referred to as the "provider agency" throughout this paper. It has been well-documented that research with and about people with developmental disabilities has been fraught with methodological challenges, including barriers to accessing data and recruiting study participants (see: Savage et al., 2015; Swaine et al., 2011). Entree to agencies remains challenging and trust between the PI and key informants is often a prerequisite (Patton, 2015; Savage et al., 2015).

After approaching six agencies with whom the PI had existing professional relationships, two agencies expressed willingness to partner. However, only one agency was able to grant the PI access to decedent records. This particular

Table 1. Study Data.

Data source	Case #1: Susan	Case #2: Rick
Phase I: Record review		
Length of review	7 hours, 19 minutes	5 hours, 41 minutes
Record source	Paper; 1 Box & 3 Folders	Paper & Electronic; 2 Boxes & 9 Folders
Phase II: Staff interviews		
Sample size	N=5 (10)	N=5 (7)
Last/current position held	Direct Care Staff, <i>n</i> =1 (5)	Direct Care Staff, <i>n</i> =1 (2)
With provider agency	Clinical Staff, <i>n</i> =1 (1)	Clinical Staff, <i>n</i> =1 (1)
	Professional Staff, <i>n</i> =2 (3)	Professional Staff, <i>n</i> =2 (2)
	Executive Leadership, <i>n</i> =1 (1)	Executive Leadership, <i>n</i> =1 (2)
Phase III: Surrogate interviews		
Sample size	N=1 (3)	N=1 (4)
Relationship	Mother/Guardian	Sister

Note. Study Data. This table illustrates additional details regarding the three sources of data that were collected and then triangulated for this comparative case study. The numbers in parentheses represent the total number of current/former employees and family members who met the inclusion criteria and were recruited to participate in a semi-structure interview regarding that particular case.

agency had retained all records for those who have died within the last 10 years in accordance with retention policies established by their licensing and regulatory authority. In addition, the agency's executive leadership team agreed to provide the PI with names and contact information for all staff and surrogates associated with these cases. Therefore, the study setting was selected purposively due to its capacity to facilitate access to information-rich cases for study (Patton, 2015).

Study Sample

The unit of analysis, or the "case," was defined as a deceased individual with a developmental disability who resided in a community residence operated by this provider agency within the last 10 years (2007–2017). The two cases presented in this paper were extracted from the larger exploratory study for several reasons. First, these were the only cases within the larger study where the PI was able to triangulate three sources of data (record review; staff interviews; and the elusive surrogate interviews), thus providing the opportunity for rich within-case analyses. In addition, both individuals represented in these cases died while enrolled in hospice care, but one did so in a hospital setting and the other did so at home. These similarities and differences offered an opportunity for rich cross-cases analyses, describing the distinct ways in which two people with developmental disabilities accessed and experienced this often-underutilized service.

Data Collection

To retrospectively reconstruct the last year of life of these two individuals, three phases of sequential data collection were used. Phase I involved a review of all available retained records for the deceased individuals. Phase II included semi-structured interviews with consenting agency staff. These interviews were conducted with former and current direct

care staff (i.e., frontline staff, group home managers, program managers); clinical staff (e.g., behaviorists, nurses, social workers); professional staff (e.g., members of the quality assurance department, human resource officers); and, the executive leadership (i.e., senior leaders within the organization). Finally, Phase III involved semi-structured interviews with consenting surrogates (e.g., family, appointed guardians, caregivers). Data were collected from multiple sources to support analytical triangulation to minimize the limitations of retrospective recall and biases of any specific data source (Charmaz, 2014; Yin, 2014). Table 1 presents additional information regarding each phase of the data collection process for the two cases presented in this paper.

A researcher-developed record review instrument was used to collect data during Phase I; information regarding the illness trajectory, as operationalized by the variables of diagnosis/prognosis, symptoms, and medical care, was collected along with data regarding demographic characteristics and advance directives. Semi-structured interviews were conducted with staff members during Phase II and with surrogates during Phase III. Both interview guides consisted of open- and closed-ended questions, which were researcher-developed and adapted from the Quality of Death and Dying instrument (Downey et al., 2010).

Additionally, a field diary was maintained by the principal investigator (PI) throughout the duration of the study. Handwritten field notes facilitated the PI's ability to document data relevant to the study aims but not otherwise collected on the previously-described instruments. The field notes also served to document periodic self-reflections, early analytical conceptualizations, and important insights throughout data collection and analysis (Corbin & Strauss, 2015; Patton, 2015).

Data Analysis

The first phase of analysis involved the extraction and visual display of data derived from the records reviewed during

Phase I of data collection. Data was double-entered during this phase to ensure accuracy (Miles & Huberman, 1994). Within-case displays were used to describe the chronological progression of each person's last year of life, which included the tabulation of events known to be common in serious illness (Miles & Huberman, 1994; Yin, 2014). For example, these early matrices included the number of days per month during which a person visited the emergency room, spent in the hospital, and was enrolled in hospice care. As the data was displayed, connections between events within each matrix became more apparent; for example, the relationship between primary care appointments, referrals to specialists, and medication changes were visually displayed by a network of nodes or data points with connecting lines on the matrix (Miles & Huberman, 1994). The visual display of these networks also helped to illustrate triggering events for emergency room visits, hospitalizations, and care transitions during the last year of life.

In order to develop rich descriptives for each case within this comparative study, data from the staff member and surrogate interviews were next coded, extracted, and then incorporated into the data displays during the second phase of analysis. This process began with an a priori list of codes derived from the variables and patterns observed in the data displays, including such codes as: diagnosis, interdisciplinary team meeting, and care transition (Patton, 2015; Yin, 2014). Interview transcripts were individually coded by the PI. Additional codes, such as funerals and memorials, were developed and defined in analytic memos for inclusion on the master code list (Patton, 2015). After all transcripts had been coded, the PI created time-ordered descriptive displays for each case (Miles & Huberman, 1994) where the earlier data displays were expanded; for example, a hospitalization that had been previously displayed numerically to represent the frequency in which it occurred during a given month was now displayed with data that included admitting diagnoses, test results, treatment plans, discharge guidelines, and lengths of stay.

During the final phase of analysis, critical incident charts were created for each case to "limit an event listing to those events seen as critical, influential, or decisive in the course of some process" (Miles & Huberman, 1994, p. 115). The charts included direct quotes and summary statements extracted from the record review instrument, interview transcripts, and field notes. These charts served to articulate the series of events that transpired from the onset of signs and symptoms of serious illness through the time period following death. This phase of data analysis enabled the principal investigator to verify conclusions made during the first phase; specifically, the displays were corroborated and refined to give form to the rich case descriptions that are presented in this paper.

Strategies to Support Rigor

In addition to the strategies already noted, an audit trail was maintained and documented in memos to describe

methodological decisions, meetings with members of the research team (i.e., this paper's co-author; a palliative care nurse practitioner), and any changes to the study protocol (Patton, 2015). Preliminary analyses were presented to an interdisciplinary group of end-of-life scholars for review and comment; their feedback served to refine the analytical process used in the creation and interpretation of the data displays. Several months later after analysis was completed, findings were presented at the annual retreat of the provider agency, where members of the executive leadership team and other key informants (i.e., surrogates, staff) were present. To facilitate member checking, attendees were invited to provide feedback, during which time they confirmed (often through tears) the accuracy of the cases described (Patton, 2015; Yin, 2014).

Ethics and Dissemination

The study was approved by the University at Buffalo Institutional Review Board (Study IDs: STUDY00001927; STUDY00002138). Interviews were audio recorded and professionally transcribed with interviewee consent. All data files were de-identified and stored on the secure drive at University at Buffalo. Specific details and/or combination of details about deceased individuals or staff members and surrogates who were interviewed for this study have been changed, redacted, or otherwise withheld in the reporting of results to uphold confidentiality (Yin, 2014).

Findings

The following section describes the end-of-life experiences of two aging adults with developmental disabilities and life-limiting serious illnesses. The demographic characteristics of these individuals are presented in Table 2 and at the start of each case study. Each individual's end-of-life experiences are then described, beginning with diagnosis through hospice enrollment and then to the time period immediately following their death. Throughout this paper, each person will be referred to by a pseudonym, respectively: Susan and Rick.

Case 1: Susan

Susan was a 42 years old woman who enjoyed interacting with others and successfully completing work-related tasks at her adult day services program; she expressed her happiness by smiling, clapping, and laughing. Susan was diagnosed with Down syndrome shortly after birth following a pregnancy marked by complications. Susan was later determined to have sensory impairments, as well. Susan received a terminal diagnosis of metastatic cholangiocarcinoma approximately a month and a half prior to her death.

In addition to her terminal cancer diagnosis and intellectual disability, Susan had multiple comorbidities, including dementia of the Alzheimer's type and several gastrointestinal

Table 2. Case Characteristics.

Characteristic	Case #1: Susan	Case #2: Rick
Gender	Female	Male
Race	Caucasian	Caucasian
Age at death	42 years old	42 years old
Insurance	Medicaid and Medicare	Medicaid, Medicare, Private Insurance
Medical diagnosis	Down syndrome	Cornelia deLange Syndrome
Cause of death	Cancer	Complications from Pneumonia
Documented morbidities	n = 15	n = 14
Documented Behavioral Health Diagnoses	n = 2	None Documented
Supports and services		
Adult day services	Yes	Yes
Prescribed diet	Yes	Yes
Durable medical equipment	Yes; protective eyewear	Yes; dentures, incontinence guards, eye glasses
Behavior tracking and/or supports	Yes	Yes

Note. Case Characteristics. This table illustrates demographic characteristics for each individual represented in this comparative case study.

conditions that required ongoing monitoring and treatment from her primary care physician and specialists. She also had several behavioral health diagnoses, including a documented anxiety disorder, that was managed by her psychiatrist with routine and PRN medications. Susan had a history of receiving behavioral support services for head hitting, grunting and screaming, and pushing of individuals and objects.

Although Susan did not speak, she communicated with others through nonverbal gestures and touch. When she turned 18 years old, Susan was determined to be “incapacitated” through guardianship proceedings. It was at this time that a court appointed her mother as her legal guardian, which was a role she served in for the duration of Susan’s life. Susan lived in multiple community residences beginning in childhood. She transitioned to the provider agency in her late 20s, first as a participant in their adult day services programs and then as a group home resident 1 year later.

Diagnosis. Approximately 44 days prior to her death, staff members observed Susan’s skin to be yellow. Several staff members also observed an increase in the intensity and frequency of Susan’s episodes of screaming both at the group home and the adult day services program. Staff members assisted Susan to her primary care physician where she was diagnosed with jaundice and blood work was requested to rule out liver failure and/or hemolysis. The primary care physician, after reviewing the results of the bloodwork, requested an ultrasound and then a CAT scan. The latter showed that Susan had masses on her lungs and other organs.

Plan of care. Ten days after the initial appointment for jaundice, staff members contacted the primary care physician to inquire about the various test results and treatment options for Susan’s increasingly jaundiced skin. The primary care physician advised that Susan should be immediately transported and admitted to the hospital, bypassing the emergency department, for additional diagnostic testing. After

the testing was complete, an oncologist informed a clinical staff member from the provider agency (who had accompanied Susan to the hospital) that she had terminal cancer. This staff person informed her supervisor of the diagnosis and Susan’s mother/guardian also was contacted.

Over the following 6 days, Susan remained hospitalized as her symptoms were treated and her guardian decided the most appropriate course of action. Susan’s mother ultimately elected not to pursue curative or life-sustaining treatment due to her 2 1/2 to 6 months prognosis and history of situational anxiety specific to medical settings and care. Susan was referred for home hospice care with orders to return to normal activity as tolerated. Susan’s mother and staff members discussed the most appropriate location for home hospice care, either at her home or the group home. They selected the latter, due in part to the familiarity of that setting and the availability of 24-hour staffing and support. The group home staff confirmed the acceptability of this decision with the surrogates of Susan’s housemates, all of whom responded positively and expressed that this would be something they would want for their loved ones should they become seriously ill.

As part of her hospice admission and 1 day prior to her hospital discharge, Susan’s mother signed a Do Not Resuscitate/Do Not Hospitalize order. In coordination with the hospice physician, her primary care provider gave orders to discontinue non-essential medications when their supplies were finished and to acquire durable medical equipment (e.g., incontinence guards, oxygen, wheelchair, hospital bed, bedside commode) as needed. The primary care provider also completed a Physician’s Orders for Life-sustaining Treatment form with the following goals of care handwritten and checked: (a) Keep comfortable and maintain quality of life; (b) Palliative care only; (c) No artificial nutrition by tube; (d) Do Not Attempt Resuscitation and allow natural death; and (e) Consent obtained from or discussed with “Legal Guardian” and “Other.”

Interdisciplinary team (IDT) meeting. A day after her discharge from the hospital, an IDT meeting was convened with Susan's case manager; hospice nurse and social worker; guardian and other involved family; and members of the provider agency's direct care staff, clinical staff, and executive leadership team. The IDT discussed her terminal cancer diagnosis and prognosis. They discussed her hospice evaluation (e.g., observations of her low blood pressure, dehydration, and worsening jaundice) and hospice recommendations (e.g., the continuation of normal activities as able, the discontinuation of specialized care and unnecessary medications, and the use of "Physician's Verbal Orders" to document the hospice physician's recommendations and the dissemination of these orders to the prescribing doctor for authorization). The IDT agreed that hospice would provide a comfort pack of medications with the authorization of her treating physician. The IDT also agreed to discontinue Susan's service plan goals, and staff members agreed to submit an Individualized Habilitation Plan (IHP) modification to confirm this decision. The provider agency sought permission for additional staffing and the opportunity to submit an associated funding request to the state's Division of Developmental Disabilities. The IDT discussed opening and transferring funds from the state payee to a burial account.

The IDT also discussed implications of Danielle's Law (P.L. 2003, c. 191 [A3458]). In the state where the provider agency operated, Danielle's Law requires staff working in facilities that provide services to people with developmental disabilities and/or traumatic brain injuries to call 911 in the event of a life-threatening emergency. Because Susan's mother authorized home hospice care and the IDT agreed with this decision, Danielle's Law became relevant because of the likelihood that Susan would experience a life-threatening event in the group home at the end of her life. The IDT discussed that responding to that emergency by either providing care (e.g., CPR) or calling 911 would be in direct contradiction with her advance directives. To resolve this issue, the IDT (led by a member of the provider agency's executive leadership team, primary care physician, and hospice nurse) discussed the issue with the state's oversight organization, consulted with the state's nurse, and ultimately received written approval authorizing a home death.

Home hospice care. For several weeks, Susan continued to participate in her normal routine. She had visits with family and friends, attended her adult day services program, and participated in daily activities within the group home as tolerated. Hospice nurses visited routinely to monitor her status (including taking vitals), provide practical guidance and emotional support to staff, and facilitate her medication management (e.g., discontinuing non-essential medication, obtaining prescriptions for routine and PRN medications to control pain and other symptoms, dispensing and securing controlled substances in accordance with the state's Division of Developmental Disabilities regulations). Staff members

developed a rotating schedule where clinical staff members and direct care supervisors would be on-site frequently to support and minimize the impact on direct care staff members, many of whom had never witnessed serious illness or experienced a death of someone they knew. Clinical staff members and supervisors also facilitated conversations with Susan's housemates, respective of their capacity and preferred communication mode. Her housemates were informed of the disease trajectory and imminence of Susan's death, as determined appropriate.

Two weeks before her death, Susan went home with her mother to celebrate a holiday. Her mother coordinated with hospice to facilitate this visit, including arranging her home to accommodate Susan's changing needs (e.g., moving beds to the first floor), obtaining her routine and PRN medications, and having an on-call contact number for hospice. The visit went relatively well with Susan appearing to be happy throughout their time together. The only significant challenge of the visit involved the oral administration of morphine, for which Susan's mother needed the help of local family who would soothe Susan while she administered the medication every 4 hours.

End of life. A week before her death, Susan continued to reside at the group home becoming increasingly restless with periods of increased screaming. This change in presentation was addressed with more frequent visits by the hospice nurse. In addition, routine and PRN medications were added to treat possible pain and other symptoms. In the final days of life, Susan became increasingly sedated but was never observed to be comatose. During these final days, family and staff members visited periodically and the hospice nurse remained at the group home 24/7.

Susan died in the morning at the group home after her housemates had left for their day services program. Her death was witnessed by her hospice nurse and approximately 2 to 3 direct care staff members, who followed established protocol for notifying the appropriate individuals of her passing. In the hour following her death, additional staff members arrived on-site as did her mother and another family member. Beginning with the family, each person who arrived to the group home had the opportunity to privately observe Susan and say goodbye. The funeral home arrived shortly thereafter to transport Susan's body. At the request of the staff members, Susan's body was taken out the front door, as opposed to the garage, with all present staff members and her family witnessing. Hospice, in coordination with the provider agency, disposed of Susan's medications and facilitated the return of her durable medical equipment. Susan's family assumed responsibility for her personal belongings.

After her passing. The family coordinated her burial and funeral arrangements, which were funded through her burial account and held at the family's local church. The services

were attended by family, friends of the family, and some agency staff members. A memorial service was also held at Susan's group home several weeks after her passing. Thirty-six individuals were in attendance, including staff members, family, and roommates/friends and some of their surrogates. A document that memorialized Susan was distributed and a poem was read on the back porch in her honor.

Case 2: Rick

Rick was a 42-year old man who was adored by his family, friends, and the staff who came to know him. He had myriad pastimes and passions, including many that were related to his dream of 1 day serving on the local police force. Rick died as a result of complications from non-COVID pneumonia approximately 2 weeks after being admitted to a local hospital with signs of labored breathing. Rick was diagnosed with a rare genetic disorder, Cornelia de Lange Syndrome, shortly after birth, with an initial prognosis of approximately 2 years. He had many of the characteristics associated with this syndrome, including distinct facial features and short stature, hearing and vision impairments, behavioral needs, digestive tract issues, and intellectual disability. Documentation in clinical records was inconsistent, with medical descriptors of "moderate" to "severe" intellectual functioning and adaptive behavior listed.

Rick had multiple comorbidities in addition to his intellectual disability, including dysphagia which had contributed to multiple cases of aspiration pneumonia and ultimately a compromised respiratory system. Prior to his hospitalization, he was receiving primary and specialized care for the following conditions: gastroesophageal reflux disease, asthma, peripheral vascular disease, lymphedema, prediabetes, nocturnal incontinence, vision loss, unsteady gait, and obesity. Rick had no documented behavioral health diagnoses; however, he did have a history of receiving behavioral support services for hitting others and breaking objects.

Rick understood most things that were said to him, and he used single words or phrases, modified American Sign Language, and nonverbal cues to express himself. Rick's parents served as his co-guardians; additionally, arrangements had been made for his sister to assume guardianship responsibilities in the event both of Rick's parents predeceased him. Despite early recommendations to consider institutional care, Rick lived at home with his family until his 30th birthday. He transitioned to a group home with the provider agency in the hopes of addressing his increasing behavioral support needs and providing opportunities for meaningful peer relationships.

Diagnosis. Approximately 13 days prior to his death, Rick returned to the group home following a birthday celebration with his family. As the day progressed, he was observed to be shivering and breathing heavily. He was administered the oral inhalant, Albuterol, as a PRN medication to prevent and

treat difficulty breathing. His symptoms worsened, and he began refusing both food and drink. He was transported to the local emergency room, where he was admitted for a "slight case of pneumonia."

Plan of care. Upon admission, Rick was treated with intravenous antibiotics, periodic suctioning, and breathing treatments by a respiratory therapist approximately twice a day. He was also referred for a swallow study to determine his risk for aspiration; ultimately, a nutrition referral was made for intravenous feeding. Group home staff members and/or Rick's family were with him for the duration of his hospitalization, facilitating communication and providing activities of daily living support. Approximately 3 days after his admission, Rick's coughing and breathing difficulties persisted despite the addition of BiPAP ventilation. At the recommendation of the treating hospitalist and pulmonologist, he was moved to the intensive care unit (ICU) where he was placed on a ventilator.

End-of-life decision making. Two days prior to his death and 11 days after his symptoms prompted the initial visit to the emergency department, Rick's mother/guardian, sister, and another family member convened a meeting with one of his treating physicians to discuss a Do Not Resuscitate order. Although Rick had been responding to the intravenous antibiotics and appeared oriented and comfortable for the duration of his stay in the ICU, attempts to wean him from the ventilator had been unsuccessful. The physician advised on various options with respect to his ongoing care needs during the family meeting, including recommending a permanent tracheostomy to extend his life.

The family expressed concern that Rick may attempt to remove the permanent tracheostomy; they feared he may require extended post-surgical sedation and/or restraint that could significantly diminish his quality of life. They also expressed concern that a permanent tracheostomy may require Rick to transition to an alternative long-term care setting, such as a nursing home, which would be both unfamiliar and offer fewer opportunities for community living. Some agency staff members, however, initially disagreed with the family's decision and felt the agency both could and should continue to provide care to Rick should he have a tracheostomy. The agency's administration, in consultation with involved staff members, considered all medical reports and ultimately affirmed the guardian's legal authority to make the final decisions about Rick's care. As one administrator noted, "While I wished for a different decision, I respected their choice."

Hospital hospice care. Approximately 3 hours after his guardian signed the Do Not Resuscitate and orders to withdraw life-sustaining treatment, Rick's breathing and feeding tubes were removed under the witness of three physicians and in accordance with the state's Division of Developmental

Disabilities' Division Circular #38 (N.J.A.C. 10:48B). He was then admitted to hospice care and transferred to an inpatient hospice unit within the same hospital where he had previously been receiving care. Initially, it was estimated that Rick would live for approximately a few minutes to hours following the withdrawal of life-sustaining treatment. However, he remained in hospice care for approximately 2 days prior to his passing. Staff members and his family perceived him to be both comfortable and peaceful during this time. He had frequent visits from extended family and staff members, with both his mother and sister remaining with him around the clock. At approximately 12:30pm following a telephone call from his eldest nephew, Rick died surrounded by his family. Rick's brother-in-law notified the agency's staff members of his passing. Rick's family assumed responsibility for his possessions, and the agency returned all of his unused medications to the pharmacy.

After his passing. The family coordinated Rick's funeral arrangements and cremation, which were partially funded by his burial account. Memorial services were held at a funeral home local to both Rick's family and his group home. A family friend arranged for police cars to gather outside, while pictures of Rick in a police uniform and some of his favorite objects were displayed in memoriam inside. Family, friends, and community members, along with agency staff members and clients, were in attendance. Several staff members spoke in Rick's honor at the services.

The agency also held a memorial service at the group home shortly after his passing, during which time they presented a plaque and planted a bush in his memory. In addition, the family placed a call out through social media during the holiday season following Rick's death. They asked people to mail his guardian a star in honor of Rick, which they then used to decorate an overflowing "tree of stars" in his memory that Christmas.

Discussion

The purpose of this study was to explore the end-of-life experiences of two aging adults with developmental disabilities and life-limiting serious illnesses. A comparative case study was conducted to analyze data from three sequential sources (records, staff, surrogates), which were then triangulated via data displays and case descriptions to retrospectively reconstruct the last year of each individual's life. Findings suggest the feasibility of quality care for adults with developmental disabilities as they age and become seriously ill, while also elucidating the unique challenges that must be addressed to ensure a humanistic approach to death and dying.

The cases of Susan and Rick serve to illustrate distinct pathways by which individuals with developmental disability can pursue hospice care. In the United States, the federal health insurance (Medicare) hospice benefit and hospice

services are accessible to individuals with developmental disability, including those who are receiving residential services through the Medicaid Home and Community-based Services (HCBS) Waiver (Moro et al., 2017). However, this benefit and other palliative care services are often underutilized by individuals with developmental disability (Stein, 2008; Takenoshita et al., 2020). This underutilization is often attributed to a lack of collaboration between developmental disability providers and hospice/palliative care providers; lack of knowledge (among family, staff, healthcare providers) of available referral services; and fears or concerns regarding the dying process (Dunkley & Sales, 2014; Friedman et al., 2012).

Current research has found that effective interdisciplinary collaboration, system-wide support, and mutual respect are essential for high-quality end-of-life care; yet, little is known regarding how to operationalize these best practices within day-to-day services for individuals with developmental disability (Fisher et al., 2020; Ronneberg et al., 2015). The case of Susan illustrated how home hospice was predicated on the support of the person with developmental disability, their surrogate and/or family, direct care staff and administrators from the provider agency, healthcare professionals, direct care staff and administrators from the hospice agency, and state case managers and administrators. In the case of Rick, consensus about the decision to withdraw life-sustaining treatment and pursue comfort-focused care was reached more slowly but ultimately his healthcare providers and agency staff were deferential to the family's wishes. The emotional, logistical, and political nature of these decisions could have imposed insurmountable barriers had there not been consensus around the individual's care.

While the end-of-life experiences of Susan and Rick were generally indicative of high-quality care, they reinforce the distinct challenges individuals with developmental disability face when they become seriously ill. Timely diagnosis and accurate prognostication can be challenging (Heller et al., 2017), which is especially difficult when communication barriers impede the expression of pain and other symptoms. Healthcare providers, including nurses, often lack training and knowledge specific to the treatment of individuals with developmental disability. Similar to the findings of Nicholas et al. (2017) regarding people with the dual diagnosis of developmental disability and mental illness, system-related barriers are pervasive in end-of-life care. Institutional policies and practices have contributed to extensive waitlists and other barriers to access services (Fisher et al., 2020; Tuffrey-Wijne et al., 2016).

Barriers to timely diagnosis have implications for survival, while delayed discussion about prognosis may impact the quality of the dying experience. In this study, both individuals experienced relatively brief end-of-life trajectories, including short lengths of stay with hospice. Susan died 44 days after the initial symptoms of her serious illness were observed and less than a month after enrolling in

hospice care. Rick died within 13 days of being admitted to the hospital and less than 2 days after transferring to hospice care. However, both Susan and Rick had other life-limiting conditions. Yet, it was not until they became seriously ill that discussions regarding their end-of-life care took place. Their experiences highlight the importance and need for regular advance care planning, which recent research has shown to be easily integrated into the person-centered planning process required of those receiving services through federally-funded health insurance programs in the United States (McGinley et al., 2021).

The events following Susan's and Rick's deaths provide examples of the ways in which individuals can be remembered by the people who cared for them and communities where they lived. Historically, individuals with developmental disability have been excluded from rituals related to death and dying (Friedman, 1998; Friedman & Helm, 2010). Research has also found that surrogates, healthcare providers, and agency staff to people with developmental disabilities can experience disenfranchised grief, especially when the profoundness of the relationship and subsequent loss are not acknowledged (Doka, 1999; Perkins & van Heumen, 2018). In this study, Susan was memorialized during a family-organized funeral service and an agency-organized tribute ceremony. Rick was honored during a funeral service, a memorial service at his residence, and a tribute Christmas tree. This indicates that personalities, preferences, and impact of the individuals can be memorialized by family, friends, caregivers, peers, and the larger communities in which they lived both during and after the dying process.

Limitations

This study had several limitations that are important to acknowledge. The utilization of a case study design has implications for the generalizability of the findings (Tsang, 2014; Yin, 2014). For example, both case studies involved individuals with developmental disabilities who were residing in group home settings with their family members serving as their guardians, features which largely informed both decision making and care at the end of life. Many individuals with DD live independently or with family, while others may be their own guardians or have court-appointed guardians (Braddock et al., 2017). Findings from this study may not be easily extrapolated to these scenarios. These cases do, however, achieve the purpose of the study by offering a meaningful contribution to the extant literature by elucidating the distinctive end-of-life experiences of two individuals with developmental disabilities in community residences. While the current research suggests that hospice care is underutilized by individuals with developmental disability, these case examples serve to illustrate the feasibility of this type of care and the improved end-of-life outcomes that can result (Friedman et al., 2012).

The single study setting was selected purposively due to the level of access to individuals' records, staff members, and surrogates that the provider agency granted to the PI. However, all of the community residences operated by the provider agency were located in one state within the northeastern United States, and their operations were directly informed by federal social welfare programs (e.g., Medicare, Medicaid) and state laws, policies, and procedures (e.g., P.L. 2003, c. 191 [A3458], "Danielle's Law"; Division of Developmental Disabilities Circulars). There were findings from this study that may not be generalizable to other states or nations that operate under their own distinct disability and health policies. More broadly, however, these findings suggest that local policies have implications for individuals' illness trajectories and end-of-life experiences; as such, healthcare providers must be knowledgeable of and attentive to the reciprocal relationships that exist between policies and practices.

Conclusion and Implications

Most states and nations have policies that inform services for individuals with developmental disability, including policies specific to end-of-life decision making and care. In this study, the state's Division of Developmental Disabilities issued Division Circulars that inform and enforce policies across the various settings where individuals with developmental disability receive services, such as Division Circular #38 for "Decision-making for the Terminally Ill." In addition, the legislature passed public laws that offered important safeguards by informing the delivery of care to individuals with developmental disability, such as Danielle's Law. The challenge with these policies, as evidenced particularly in the case of Susan, is that they introduced rules and formalities that could have impeded the timely delivery of services and delayed care transitions. Nurses and other healthcare providers who support people with developmental disabilities must be knowledgeable of, and capable of, working within the constraints of these policies. They also should consider opportunities to advocate for policy change when they feel thwarted in their ability to support individuals with developmental disability to die in accordance with their and their surrogates' expressed wishes. In these instances, healthcare providers may find frameworks, such as the consensus norms that emerged from the European Association of Palliative Care, provide valuable guidance for policy and practice (Tuffrey-Wijne et al., 2016).

Although individuals with developmental disability are living longer, there are subsets of the population who are at increased likelihood for chronic, life-limiting diseases and conditions that can result in shorter life expectancies (O'Leary et al., 2018). Providers should consider early palliative care involvement for those with serious illness diagnoses and who are amenable to supportive care in addition to their regular medical services. In this comparative case study,

both of these patients received late hospice care, within days or weeks of their death. Nurses in primary care, hospital, and other settings are often the first to recognize the possibility and probability of death (American Nursing Association Board of Directors, 2017; Glaser et al., 1965). Individuals with developmental disability and their caregivers would benefit from the articulation of these observations, especially if they can serve as a springboard for discussing end-of-life wishes and establishing advance care plans.

Acknowledgments

The authors also wish to thank Elizabeth S. Hull, MS, FNP for her expert review of early drafts of this manuscript.

Declaration of Conflicting Interests

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The authors disclosed receipt of the following financial support for the research, authorship, and/or publication of this article: This manuscript reflects findings from the first author's dissertation study, which was funded by the University at Buffalo's Mark Diamond Research Fund (Grant Number: SU-17-11).

ORCID iD

Jacqueline M. McGinley  <https://orcid.org/0000-0002-8037-9318>

References

- American Nursing Association Board of Directors. (2017, March 13). *Call for action: Nurses lead and transform palliative care*. Author. <https://www.nursingworld.org/~497158/globalassets/practiceandpolicy/health-policy/palliativecareprofessionalissuespanelcallforaction.pdf>
- Bishop, K. M., Hogan, M., Janicki, M. P., Keller, S. M., Lucchino, R., Mughal, D. T., Perkins, E. A., Singh, B. K., Service, K., & Wolfson, S. (2015). Guidelines for dementia-related health advocacy for adults with intellectual disability and dementia: National Task Group on intellectual disabilities and dementia practices. *Intellectual and Developmental Disabilities, 53*(1), 2–29. <https://doi.org/10.1352/1934-9556-53.1.2>
- Botsford, A. L. (2000). Integrating end of life care into services for people with an intellectual disability. *Social Work in Health Care, 31*(1), 35–48. https://doi.org/10.1300/J010v31n01_03
- Braddock, D. (2002). *Aging and developmental disabilities: Demographic and policy issues affecting American families*. In J. B. Blacher, B. L. Baker, & D. Braddock (Eds.), *Families and mental retardation: A collection of notable AAMR journal articles across the 20th Century* (pp. 345–350). American Association on Mental Retardation.
- Braddock, D. L., Hemp, R., Tanis, E. S., Wu, J., & Haffer, L. (2017). *The state of the states in intellectual and developmental disabilities: 2017*. American Association on Intellectual and Developmental Disabilities.
- Charmaz, K. (2014). *Constructing grounded theory* (2nd ed.). SAGE Publications, Inc.
- Clark, L., & Watts, C. (2021). The journey of humanizing care for people with disabilities. *Qualitative Health Research, 31*(7), 1199–1208. <https://doi.org/10.1177/10497323211000334>
- Corbin, J., & Strauss, A. (2015). *Basics of qualitative research: Techniques and procedures for developing grounded theory* (4th ed.). SAGE Publications, Inc.
- Doka, K. J. (1999). Disenfranchised grief. *Bereavement Care, 18*(3), 37–39. <https://doi.org/10.1080/02682629908657467>
- Downey, L., Curtis, J. R., Lafferty, W. E., Herting, J. R., & Engelberg, R. A. (2010). The quality of dying and Death Questionnaire (QODD): Empirical domains and theoretical perspectives. *Journal of Pain and Symptom Management, 39*(1), 9–22. <https://doi.org/10.1016/j.jpainsymman.2009.05.012>
- Dunkley, S., & Sales, R. (2014). The challenges of providing palliative care for people with intellectual disabilities: A literature review. *International Journal of Palliative Nursing, 20*(6), 279–284. <https://doi.org/10.12968/ijpn.2014.20.6.279>
- Fisher, K., Robichaux, C., Sauerland, J., & Stokes, F. (2020). A nurses' ethical commitment to people with intellectual and developmental disabilities. *Nursing Ethics, 27*(4), 1066–1076. <https://doi.org/10.1177/0969733019900310>
- Friedman, R. I. (1998). Use of advance directives: Facilitating health care decisions by adults with mental retardation and their families. *Mental Retardation, 36*(6), 444–456.
- Friedman, S. L., & Helm, D. T. (2010). *End-of-life care for children and adults with intellectual and developmental disabilities*. American Association on Intellectual and Developmental Disabilities.
- Friedman, S. L., Helm, D. T., & Woodman, A. C. (2012). Unique and universal barriers: Hospice care for aging adults with intellectual disability. *American Journal on Intellectual and Developmental Disabilities, 117*(6), 509–532. <https://doi.org/10.1352/1944-7558-117.6.509>
- Glaser, B. G., Strauss, A. L., & Strauss, A. L. (1965). *Awareness of dying*. Aldine.
- Hahn, J. E., Fox, S., & Janicki, M. P. (2015). Aging among older adults with intellectual and developmental disabilities: Setting national goals to address transitions in health, retirement, and late-life. *Inclusion, 3*(4), 250–259. <https://doi.org/10.1352/2326-6988-3.4.250>
- Harrington, S. (2018). *At peace: Choosing a good death after a long life*. Grande Central Life & Style.
- Heller, T. (2019). Bridging aging and intellectual/developmental disabilities in research, policy, and practice. *Journal of Policy and Practice in Intellectual Disabilities, 16*(1), 53–57. <https://doi.org/10.1111/jppi.12263>
- Heller, T., Scott, H., & Janicki, M. P. (2017, August). *Caregiving and intellectual and developmental disabilities and dementia: Report of the pre-Summit workgroup on caregiving and intellectual and developmental disabilities*. Office for the Assistant Secretary of Planning and Evaluation. <https://aspe.hhs.gov/system/files/pdf/257461/IDD%20PreSummit.pdf>
- Institute of Medicine (IOM). (2014). *Dying in America: Improving quality and honoring individual preferences near the end of life*. The National Academies Press.
- Landes, S. D., Stevens, J. D., & Turk, M. A. (2019). Obscuring effect of coding developmental disability as the underlying cause of death on mortality trends for adults with developmental disability: A cross-sectional study using us mortality

- data from 2012 to 2016. *BMJ Open*, 9(2), e026614. <https://doi.org/10.1136/bmjopen-2018-026614>
- Landes, S. D., Turk, M. A., & Bisesti, E. (2021). Uncertainty and the reporting of intellectual disability on death certificates: A cross-sectional study of US mortality data from 2005 to 2017. *BMJ Open*, 11(1), e045360. <https://doi.org/10.1136/bmjopen-2020-045360>
- Lauer, E., & McCallion, P. (2015). Mortality of people with intellectual and developmental disabilities from select U.S. State disability service systems and medical claims data. *Journal of Applied Research in Intellectual Disabilities*, 28(5), 394–405. <https://doi.org/10.1111/jar.12191>
- Lindley, L. C. (2018). Children with intellectual disability and hospice utilization: The moderating effect of residential care. *The American Journal of Hospice & Palliative Care*, 35(7), 945–950. <https://doi.org/10.1177/1049909117743181>
- McCallion, P., Ferretti, L. A., Beange, H., & McCarron, M. (2019). *Epidemiological issues in intellectual disability and aging research*. In V. P. Prasher & M. P. Janicki (Eds.), *Physical health of adults with intellectual and developmental disabilities* (pp. 9–26). Springer.
- McGinley, J., Marsack-Topolewski, C. N., Church, H. L., & Knoke, V. (2021). Advance care planning for individuals with intellectual and Developmental Disabilities: A State-by-State content analysis of person-centered service plans. *Intellectual and Developmental Disabilities*, 59(4), 352–364. <https://doi.org/10.1352/1934-9556-59.4.352>
- Miles, M. B., & Huberman, A. M. (1994). *Qualitative data analysis: A sourcebook*. SAGE Publications, Inc.
- Moro, T. T., Savage, T. A., & Gehlert, S. (2017). Agency, social and healthcare supports for adults with intellectual disability at the end of life in out-of-home, non-institutional community residences in Western nations: A literature review. *Journal of Applied Research in Intellectual Disabilities*, 30(6), 1045–1056. <https://doi.org/10.1111/jar.12374>
- Ng, N., Wallén, E. F., & Ahlström, G. (2017). Mortality patterns and risk among older men and women with intellectual disability: A Swedish national retrospective cohort study. *BMC Geriatrics*, 17(1), 269–313.
- Nicholas, D. B., Calhoun, A., McLaughlin, A. M., Shankar, J., Kreitzer, L., & Uzande, M. (2017). Care experiences of adults with a dual diagnosis and their family caregivers. *Global Qualitative Nursing Research*, 4, 1–10. <https://doi.org/10.1177/2333393617721646>
- O'Dwyer, M., Peklar, J., McCallion, P., McCarron, M., & Henman, M. C. (2016). Factors associated with polypharmacy and excessive polypharmacy in older people with intellectual disability differ from the general population: A cross-sectional observational nationwide study. *BMJ Open*, 6(4), e010505. <https://doi.org/10.1136/bmjopen-2015-010505>
- O'Leary, L., Cooper, S. A., & Hughes-McCormack, L. (2018). Early death and causes of death of people with intellectual disabilities: A systematic review. *Journal of Applied Research in Intellectual Disabilities*, 31(3), 325–342. <https://doi.org/10.1111/jar.12417>
- Patton, M. Q. (2015). *Qualitative research & evaluation methods* (4th ed.). SAGE Publications, Inc.
- Perkins, E., & van Heumen, L. (2018). *When the journey ends: Parental caregiving and later life planning*. In B. S. Nelson Goff & N. Piland Springer (Eds.), *Intellectual and developmental disabilities: A roadmap for families and professionals* (pp. 216–228). Routledge, Taylor & Francis Group.
- Ronneberg, C. R., Peters-Beumer, L., Marks, B., & Factor, A. (2015). Promoting collaboration between hospice and palliative care providers and adult day services for individuals with intellectual and developmental disabilities. *OMEGA-Journal of Death and Dying*, 70(4), 380–403. <https://doi.org/10.1177/0030222815573724>
- Savage, T. A., Moro, T. T., Boyden, J. Y., Brown, A. A., & Kavanaugh, K. L. (2015). Implementation challenges in end-of-life research with adults with intellectual and developmental disabilities. *Applied Nursing Research*, 28(2), 202–205. <https://doi.org/10.1016/j.apnr.2014.10.002>
- Stein, G. L. (2008). Providing palliative care to people with intellectual disabilities: Services, staff knowledge, and challenges. *Journal of Palliative Medicine*, 11(9), 1241–1248. <https://doi.org/10.1089/jpm.2008.0130>
- Swaine, J., Parish, S. L., Luken, K., & Atkins, L. (2011). Recruitment and consent of women with intellectual disabilities in a randomised control trial of a health promotion intervention. *Journal of Intellectual Disability Research*, 55(5), 474–483. <https://doi.org/10.1111/j.1365-2788.2011.01399.x>
- Takenoshita, S., Terada, S., Kuwano, R., Inoue, T., Cyoju, A., Suemitsu, S., & Yamada, N. (2020). Prevalence of dementia in people with intellectual disabilities: Cross-sectional study. *International Journal of Geriatric Psychiatry*, 35(4), 414–422. <https://doi.org/10.1002/gps.5258>
- Todd, S., Brandford, S., Worth, R., Shearn, J., & Bernal, J. (2021). Place of death of people with intellectual disabilities: An exploratory study of death and dying within community disability service settings. *Journal of Intellectual Disabilities*, 25(3), 296–311. <https://doi.org/10.1177/1744629519886758>
- Tsang, E. W. K. (2014). Generalizing from research findings: The merits of case studies. *International Journal of Management Reviews*, 16(4), 369–383. <https://doi.org/10.1111/ijmr.12024>
- Tuffrey-Wijne, I., McLaughlin, D., Curfs, L., Dusart, A., Hoenger, C., McEnhill, L., Read, S., Ryan, K., Satgé, D., Straßer, B., Westergård, B. E., & Oliver, D. (2016). Defining consensus norms for palliative care of people with intellectual disabilities in Europe, using Delphi methods: A White Paper from the European Association of Palliative Care. *Palliative Medicine*, 30(5), 446–455. <https://doi.org/10.1177/0269216315600993>
- Yin, R. K. (2014). *Case study research: Design and methods* (5th ed.). SAGE Publications, Inc.

Author Biographies

Jacqueline M. McGinley, PhD, LMSW, is an assistant professor at Binghamton University, College for Community and Public Affairs, Department of Social Work, Binghamton, New York.

Christina N. Marsack-Topolewski, PhD, LMSW is an associate professor at Eastern Michigan University in the School of Social Work, Ypsilanti, Michigan and is an Affiliated Research Associate at the MI-DDI.