Commentary on "Research Design in End-of-Life Research: State of Science"

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Dr. George’s (2002) review of research to date on the end of life was methodical and highly informative. In response, these comments are organized in a similar fashion, but start where she concluded—gaps in the knowledge base. Then, because it is the research question that drives a study design and the methods used, the comments conclude with discussion of design and methods.

Gaps in the Knowledge Base

Role of Comorbidities

Noting that attention is focused often on a single disease, usually the life-threatening index condition, Dr. George (2002) pointed out the likelihood of co-morbidities and their contribution to symptom burden. I agree that much is overlooked about the end-of-life experience when the focus is on the terminal condition. A similar “unifocus” can be found in symptom research, including symptoms at the end of life. Just as there are studies of endstage cancer, HIV, congestive heart failure, and chronic obstructive pulmonary disease, similarly there are studies focused on pain, dyspnea, nausea, and fatigue. If we are interested in understanding the dying process or the effectiveness of various models of care at the end of life, we must include comorbidities and the full range of symptoms experienced by patients. In fact, much can be learned by focusing on the symptoms rather than on the condition(s), including the frequency and severity of symptoms, and importantly, how bothersome the symptoms are for a patient. It is these symptoms, their characteristics, and associated distress that will affect a patient’s daily functioning and quality of life. Furthermore, it is worth investigating the relative contribution of each symptom, or class of symptoms, to explaining variability in the end-of-life experience. If the focus of a study is limited to symptoms associated with the index disease, variability in the dying process might go unexplained. This is a particularly relevant point in relation to the trajectories of dying described by Lunney, Lynn, and Hogan (2001). Consider the case of a woman enrolled in a study of breast cancer who has metastatic disease but also Class III or IV congestive heart failure (American Heart Association, 1994). If quality of life and functional status were outcomes of interest, and one inquired only about cancer-related symptoms, the frequency and severity of dyspnea might go unreported. Yet, the symptoms of dyspnea might explain a good portion of the patient’s functional disability at any point in time. Just as care at the end of life is approached holistically, the assessment of symptom burden should be approached in a similar fashion.

Diversity

Dr. George (2002) pointed out gaps in our understanding of age and race differences in end-of-life experiences, preferences, and care. Despite the recognized diversity of this country’s population, most research to date has been limited to White and African American persons. Not only must end-of-life research extend to Asian and Latino populations, but also to the subgroups in these populations. The differences, for example, between Cuban Americans and Mexican Americans have been established in other fields of research. Similarly, Korean and Japanese cultures are distinct and should be studied separately.

The cultural influence on perceptions of illness and death, personal autonomy, and control will affect how one defines “a good death,” that is, desire for information about illness and prognosis, desire to plan end-of-life care, and preference for place of death. To illustrate this point, the following examples are taken from a thought-provoking work by Mitty (2001):

1. Many African Americans see illness as a test of faith in God. Therefore, one should not put business such as treatment refusal in the way of God’s will.
2. For Korean Americans, autonomy is not empowering; instead, it is burdensome and causes loss of hope. This suggests that Korean Americans would not be receptive to advance directives.
3. Among some Asian and Pacific Islander cultures, death outside the home means that the deceased will be a wandering soul with no place to rest. Place of death obviously will be an explanatory factor in the end-of-life experience.

Although the diversity of such cultural influences merits research attention, the diversity and heterogeneity within cultural groups must also be recognized. Points such as those made by Mitty are intended to underscore the importance of cultural differences across groups. One should not assume that, by virtue of ethnic or cultural identification, all persons within a group hold similar thoughts or beliefs.

Provider Effects

Dr. George (2002) has raised an area of research that receives limited attention, but is very important—provider effects. How much do the characteristics of a provider influence the course, process, and outcome of end-of-life care? Evidence from one study (Mebane, Oman, Kroonen, & Goldstein, 1999) shows that African American physicians are more likely than White physicians to order more aggressive care. This finding is consistent with reports from studies of ethnic differences in patient preferences: for example, that African American patients prefer treatments that extend life, with less consideration of the quality of life than by other groups. Other provider characteristics, such as gender, age or years of practice, and type of specialty training, are all worthy of investigation, because they have been shown to influence clinical decision making (McKinlay, Potter, & Feldman, 1996; McKinlay et al., 1998).

Related to this area of research is attention to communication—content and style—between providers and patients, providers and families, and patients and families. Although it is a growing, cross-disciplinary field of research, work in end-of-life studies has focused primarily around advance directives. More attention should be directed to communication across the span of care. This area also offers opportunities for intervention studies to improve communication at various stages of end-of-life care.

Health Systems

I would like to underscore the importance of investigating health system variables. The small area analysis approach of Wennberg and colleagues (Fisher et al., 2000; Wennberg, 1993) has much to offer this field. What was learned by Christakis and Iwashyna (2000) about the use of hospice provides a good example of information that can be used to better integrate palliative care into the acute and long-term care system.

Beyond Health Care to Supportive Services

The community-based, long-term-care system has received short shrift in the study of end-of-life care. Certainly, for the three death trajectories characterized as (a) a distinct terminal phase, (b) an eventually fatal chronic illness, and (c) a slow, progressive disability (Lunney et al., 2001), the caregiving period is lengthy, and much of it occurs outside the structures of medical care. However, not only do supportive community services merit attention, but also the system of informal care. An extensive literature exists on the role of families and friends in providing care to disabled persons (Tennstedt, 1999).

The majority of community-based care is provided by family caregivers (Doty, 1986; National Alliance for Caregiving and American Association for Retired Persons, 1997; Stone, Cafferata, & Sangl, 1987). Certainly hospice care recognizes family caregivers in the focus and delivery of services. Family outcomes are an important concern. However, empirical studies of end-of-life care, with the exception of studies of caring for persons with Alzheimer’s disease (Alzheimer’s Association and National Alliance for Caregiving, 1999; Gill, Hinrichsen, & DiGiuseppe, 1998; Ory, Hoffman, Yee, Tennstedt, & Schulz, 1999; Yordi et al., 1997), have paid less attention to the types and amounts of care provided by family caregivers and to the interface of informal care with formal care.

Additional Areas of Research

There are two areas of research not previously discussed that I would like to mention: (a) the use of complementary or alternative medicine and (b) telemedicine.

The study of complementary or alternative medicine has been limited until recently. Descriptive studies have documented the use of complementary or alternative medicine in cancer and HIV, and efficacy trials of specific treatments for chronic or terminal conditions are now being conducted (Pan, Morrison, Ness, Fugh-Berman, & Leipzig, 2000). The use of alternative medicine is worthy of more direct attention in end-of-life studies, not only in terms of efficacy but also in relation to comfort palliation, satisfaction with end-of-life care, and well-being.

The emerging field of telemedicine may become more prominent in palliative care. Certainly, its utility has been recognized in the management of chronic conditions, such as the monitoring of hypertension (Friedman et al., 1998) and diabetes (Piette, Weinberger, & McPhee, 2000; Rogers et al., 2001; Tsang et al., 2001; Whitlock et al., 2000). Often, through the use of touch-tone telephones, home health agencies have utilized telecommunications to monitor the physical status of patients between visits (Johnston, Wheeler, Deuser, & Sousa, 2000). An untoward change in status can trigger a phone call or home visit by the staff. A pilot study of one system showed the telecommunications system to be equal to the traditional system of care in terms of patient outcomes and satisfaction but, importantly, with considerable cost savings (Stricklin, Jones, & Niles, 2000). The value of such systems in palliative care is apparent. A telecommunications system can also be supportive for family caregivers (Mahoney, Tarlow, Jones, Tennstedt, & Kasten, 2001).
Design and Methods

Dr. George's (2002) review and discussion of conceptual and definitional issues was comprehensive. I want to underscore the point she made about the dearth of hypothesis-driven studies. This is reflected in the designs of most end-of-life research to date. My comments focus on design and methods. Clearly, if we agree with Dr. George's recommendations that research be theoretically based and driven by hypotheses, attention must be given to appropriate design and methods.

Design

In addition to cross-sectional versus longitudinal studies, observational versus experimental designs should be considered. The choice of a design is driven by the research question or hypothesis. Using Dr. George's (2002) recommendation for studies on the trajectories of dying as an example, if one wanted to understand differences in patient advance preferences for care or family well-being across the different trajectories, one would conduct an observational study. However, to investigate the effect of different treatments, interventions, or models of care within or across the trajectories, an experimental design should be used.

Typically, observational studies are conducted in emerging fields of research. Therefore, it is not surprising that most end-of-life studies are observational and cross-sectional. These studies are informative and lead to longitudinal and more rigorously designed studies. Certainly, if we are interested in trajectories of dying, the process of dying, and how best to intervene across the end-of-life span, longitudinal designs are required.

If the interest lies in testing the efficacy of an intervention or comparing interventions (models of care) for purposes of evidence-based practice, it is generally accepted that randomized, controlled trials are indicated. Yet, the mention of a clinical trial typically raises ethical concerns. Concerns arise with randomization of patients to a control group receiving usual care when one is convinced of the value of a model of palliative care. This can be interpreted as similar to withholding treatment, and it is not obvious that anything beyond usual care at the end of life should be better than usual care? However, the more appropriate questions to ask might be how much better, or for whom, or in what setting? The objective is to appropriately design a study with sufficient scientific rigor to ensure that a question that is worth asking can be answered. This reduces the risk of exposing a vulnerable population to an intervention or measurement protocol—that may or may not be burdensome—that yields little or no useful information.

Expensive randomized clinical trials are best reserved for tests of standardized or well-designed interventions (Rossi & Freeman, 1985). At earlier stages in the development of intervention or models of care, pilot studies can be invaluable. They can assist in understanding an intervention and selecting appropriate outcome measures before starting a trial. Quasiexperimental designs can also be used; for example, a nonrandomized but controlled study using constructed controls. The control group in this study design would be a group of patients comparable in essential respects to those exposed to the treatment intervention. So, for example, if one was interested in evaluating a model of palliative care available in an organization, the control group could be recruited from another similar organization not offering this model of care. Other quasi-experimental designs include pretest/posttest designs in a single group or two groups (Cook & Campbell, 1979) and may be appropriate for preliminary pilot studies.

Methods

The study design will drive the method. The overarching methods are quantitative or qualitative. Although an experimental design generally requires quantitative methods, observational studies can be qualitative or quantitative. Qualitative methods have much to offer to end-of-life research. If dying should be “patient-focused and family-centered” (Teno, Byock, & Field, 1999), then it is critical to understand the experience and the effects of intervention from this perspective. I found it somewhat surprising that there are fewer qualitative studies of patients and families (Steinhauser, Christakis, et al., 2000; Steinhauser, Clipp, et al., 2000) than there are of clinicians (Asai et al., 1997; Calam, Far, & Andrew, 2000; Ekblad, Marttila, & Emilsson, 2000; Fetters, Churchill, & Danis, 2001; Matzo & Schwarz, 2001; Oberle & Hughes, 2001).

Qualitative studies can be an important first step in the appropriate design of quantitative observational and experimental studies. For example, there are sufficient data to know that there are ethnic differences and disparities in the use of palliative care services, yet we do not completely understand the reasons. Qualitative designs can be used to detect whether these disparities are related to patient and/or family preferences, knowledge, attitudes, or beliefs or whether the model of care or mode of delivery is not culturally appropriate.

Another valuable use of qualitative studies is to determine whether available measures are culturally sensitive and appropriate. To cite an example from my own research on informal care, most studies have reported that White caregivers report more distress or burden than do caregivers in other ethnic groups (Caldron & Tennstedt, 1998; Ory et al., 1999; Tennstedt, 1999). Yet, these ethnic caregiver groups were found to provide more care to more severely disabled elders (Tennstedt & Chang, 1998). It was assumed that this reflected a cultural difference (e.g., the strong sense of familism in Hispanic culture or the involvement of fictive kin in African American families).

The possibility occurred to me that this was a measurement artifact. The measures of caregiving burden and distress in these studies were developed with White populations. We conducted a qualitative study probing the experiences of White, African American,
Attrition and Missing Data

A final area of comment relates to longitudinal study designs and particularly clinical trials or experimental studies (i.e., handling study attrition). Unlike many other fields of research, death is an expected outcome in this field, not a source of attrition. However, there is a high likelihood of functional attrition. As study participants become more debilitated, they might be less able or willing to continue participation. One way to handle this is through use of proxy informants, noting the issues raised by Dr. George (2002). Proxies may or may not be able to provide complete measurements. It might not be appropriate to collect certain data by proxy (e.g., attitudes). Or a proxy informant might be unwilling to complete the measurement protocol. Such situations will result in missing data that cannot be ignored without biasing the results of the study. This will happen when the cause or the reason for the missing data is the value of the variable itself. For example, if a study outcome is severity level of a symptom, such as pain, and very severe pain prevents the patient from providing data, then the resulting missing data cannot be ignored.

Advances in analytical procedures for handling missing data provide methods for imputing missing values that will minimize bias associated with traditional procedures, such as complete case analysis (listwise deletion), pairwise deletion, or substitution of the mean. Furthermore, the latter two procedures do not provide the basis for estimating standard errors (Graham & Hofer, 2000). The recent development of multiple imputation procedures offers many advantages for handling nonrandom missing data, and several imputation software programs are available.

In summary, the end-of-life experience and care is an emerging field of research. There are significant gaps in knowledge that can be addressed by a variety of research designs and methods. The significant implications of this field of inquiry for improving the end of life merit careful attention to appropriate methodology to ensure that important questions are answered adequately.

References


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