





Journal of Pain and Symptom Management

Available online 2 October 2022

In Press, Journal Pre-proof 

Review Article

Layers of Loss: A Scoping Review and Taxonomy of HD Caregivers' Spiritual Suffering, Grief/Loss and Coping Strategies

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<https://doi.org/10.1016/j.jpainsymman.2022.09.010>

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Abstract

Context

Huntington's disease (HD), an incurable, multi-generational, autosomal dominant disorder, creating unique challenges and a myriad of spiritually-related stressors in those affected and their familial caregivers. Spiritual suffering, experiences of grief/loss, and coping strategies have not been systematically studied in HD caregivers.

Objectives

To comprehensively define spiritual suffering, grief/loss, and coping strategies used by HD caregivers.

Methods

A PRISMA-ScR scoping literature review was conducted. Data from included research articles were organized thematically using induction and open coding. A grounded, deductive approach was used to delineate a demarcated taxonomy of themes, which encompasses all three over-

arching domains. Four reviewers, employing a modified Delphi approach, ascertained which themes were demonstrated by research participants in each study.

Results

36 of 583 articles met the review criteria; none were published in the palliative care literature. Investigations primarily focused on intrapersonal (self-image) distress and existential angst; only rarely looked deeper into divine/transpersonal suffering, disrupted religious relationships, or meaning distress. HD caregivers experience profound grief/loss, expressed as disenfranchised grief that is associated with the ambiguous loss of their loved one, loss of family structure, social connectedness, and personal losses. Half of the studies reported maladaptive HD caregiver coping strategies—characterized by dysfunctional escape schemes; in contrast, transcendent/creative strategies were often unexplored.

Conclusion

HD caregivers experience prolonged grief and other forms of spiritual suffering as they progressively lose their loved ones and disruption to their own lives. With an improved assessment tool, teams with spiritual and palliative care experts will better be able to support HD family caregivers.

Introduction

Huntington's disease (HD) is an incurable, autosomal dominant neurodegenerative disorder; which slowly strips away a patient's independence, demarcated by cognitive, psychiatric, and executive decline, behavioral and personality changes, affective problems as well as crippling motor impairments.^{1, 2, 3} Moreover, HD presents with a painfully long decline, which eventually leads to a patient's death over 20 years after diagnosis and 35 years after symptom onset.⁴ While many neurodegenerative disorders present in old age, HD typically presents in patients' 30s and 40s when careers are developing and family concerns are paramount. Given the young age of onset, the hereditary risk to offspring of 50%,^{1, 2, 3} and the prolonged disease course, HD presents a unique challenge to patients, families, and clinical care teams alike.^{5, 6, 7} HD familial care partners often experience prolonged emotional distress and caregiver burden as the disease slowly robs them of their loved ones.^{8, 9, 10} The very nature of the continuous caregiver experience of progressive loss in HD, where their loved one does not die for many years,¹¹ generates a context for chronic grief¹² or prolonged grief disorder.¹³ Studies show high rates of depression, anxiety, and other psychotic symptoms among care partners.^{14,15} Some HD patients even begin manifesting the disease as children.^{16,17} Not surprisingly, studies have demonstrated high rates of family dysfunction, adverse parenting, abuse, and stark impacts on all members of a family coping with HD.⁷ This continuous trauma experienced by HD care partners has detrimental effects on caregivers who often struggle to maintain their sense of self;^{8,10} one

seminal article infers that HD “partners have at least as much psychological distress as [patients], but partners have the tendency to draw back.”⁹ Furthermore, they observe that “the grief of [patients’] partners is often ‘disenfranchised’, or not socially recognized, as if they have no right to mourn.”⁹ Thus, in addition to all the physical and practical burdens of caregiving in HD, family members must personally deal with overwhelming psycho-socio-spiritual stressors.

Despite excellent primary palliative care provision by neurology teams^{18, 19, 20} and the support of multiple national advocacy organizations,^{21,22} we hypothesize that significant gaps remain—both in the knowledge of the breadth of HD caregiver suffering and in how other providers might help. Managing these gaps could potentially be addressed by palliative care (PC) experts; unfortunately, there is limited data on the role of PC in these caregivers. PC involvement rates with HD patients and their caregivers have been reported as low as 4%, even when they are at the end of their lives.²³ In addition, spirituality and spiritual concerns are ignored by the instruments usually used to measure caregiver quality of life (as well as by most non-palliative clinicians in family conferences),²⁴ and yet spirituality as a source of support and/or suffering is ubiquitously experienced by patients with devastating serious illnesses.^{25,26} Public and medical awareness of HD, including understanding the experiences, burdens, and suffering of families is wanting. This lack of understanding extends to the PC community. We aim to evaluate the current state of HD caregivers' emotional and spiritual suffering by asking: “what is known about the spiritual suffering, grief, and coping strategies for caregivers of HD patients?” We hope to provide a framework for palliative care and spiritual support experts to better address and support caregivers of HD patients over the unrelenting disease course of HD.

Section snippets

Methods

This scoping review was conducted following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses for Scoping Reviews (PRISMA-ScR) guidelines.²⁷ ...

Results

Of the 583 unique study articles originally identified, 36 were included in the final thematic analysis. Journal focuses of the examined studies were nursing (8), sociology (7), genetics (6), neuroscience (6), psychology (4), medical (3), quality of life (1), and thanatology (1). None of the included studies were published in journals with a focus on palliative care (Table 3)...

Discussion

This study confirms the hypothesis that HD patient caregivers' burdens are high and that they have significant unmet needs. As one participant said, "*Caregivers have enough responsibilities on their hands to spend hours researching for help.*"⁹⁶ Variables such as income, economic pressure, and social support predict life quality among caregivers of people with progressive neurologic diseases,⁹⁷ and yet these factors only begin to scratch the surface of the multifaceted forms of suffering that...

Conclusion

The burdens of caring for a patient with HD encompass every aspect of humanity, including spiritual suffering and grief. We can safely assume, based on this review, that spiritual suffering and grief are ubiquitously experienced by caregivers of patients with HD. These sources of prolonged pain and loss are rarely seen by the public or healthcare professionals.¹⁶⁴ Attendance to spiritual suffering, grief and loss, and supporting adaptive coping are areas where PC experts excel, and yet...

Conflicts of Interest/Competing Interests

All authors have completed and submitted the ICMJE form for Disclosure of Potential Conflicts of Interest. Dr. Gabbard is supported by the National Institute on Aging of the National Institutes of Health under Award Number K23AG070234. No other disclosures were reported....

Acknowledgments

The authors thank Mark McHone, Librarian, Coy C. Carpenter Library, Wake Forest University School of Medicine, for initial assistance in designing and conducting the literature searches....

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