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Philadelphia College of Osteopathic Medicine
Graduate Program in Biomedical Sciences
School of Health Sciences

**A Systematic Review of Late effects, Current Models of Follow-up in Three
Pennsylvania Health Network Settings, and Survivor Care Plans for Brain
Neoplasm Survivors**

A Capstone in Public and Population Health by Megan Elizabeth Donlick
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Submitted in Partial Fulfillment of the Requirements for the Degree of
Master of Science in Biomedical Sciences, Public Health Concentration

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ABSTRACT

Introduction: Brain neoplasm survival rates have increased substantially in developed countries due to modern medical interventions, research, and enhanced therapeutic options. As the second leading cause of pediatric cancers, this has been a topic of interest within the field of public health. With mortality decreasing, scientists are now focusing the lens on the long-term outcomes of survivors, coined “survivorship” and subsequent late effects. These effects are the combined neurocognitive, psychological, and social deterioration and deficits experienced due to the brain pathology itself or exposure to associative treatments. This phenomenon is now appearing frequently in the literature. The aim of this review is to examine standards of care, planning for survivors, post-diagnostic interventions, and lapses in follow-up for brain tumor survivors. Models of care were observed in three major hospital systems and compared to the literature. Furthermore, a comprehensive examination of common late effects associated with the various treatment modalities, and the process for continued patient care has been provided for scope. *Methods:* Relevant literature was derived from publicly available databases for scholarly articles. Peer-reviewed studies were slated for consideration. *Discussion:* The extent of neurocognitive, psychological and social decline is individual and lapses in care come from lack of standardization in the follow-up process. Also, with current models of care, there is a substantial patient initiative is required, leaving patients and caretakers, already heavily burdened, to fend for themselves. This review contributes to the knowledge base afforded to survivors, thereby informing patients, caregivers, and practitioners, such that preemptive decisions can be made to bolster late effects and improve quality of life.

INTRODUCTION

Cancer is a major public health concern. According to the CDC, it is the second leading cause of death in the United States, just after heart disease (CDC, 2021). For children, brain cancer is the most common, and pediatric brain tumors (PBTs) account for most cases of solid cancer in this age category (Satariano, 2016). Due to medical developments, interventions, research, and innovative therapeutic options the five-year survival rates have increased to around 80% (Lönnerblad et al., 2019). With mortality decreasing, scientists and medical professionals are now focusing the lens on the long-term outcomes of survivors.

Around 700,000 Americans live with a primary brain tumor, and 85,000 are projected to be diagnosed in the coming year (Quick Brain Tumor Facts, 2020). Survivors will have been exposed to various treatments, such as chemotherapy, radiation, or surgical resection, among other therapeutic options, which all have associative hazards. Also, the brain pathology in and of itself can have deleterious effects. Researchers are just beginning to understand the role of maladaptive health behaviors, the development of psychological conditions, and the interplay of subsequent disease states that come after the initial diagnosis. Michael Feuerstein, a cancer survivor, describes this experience as such: “There are many unmet needs and concerns of patients and their caregivers further complicated by high symptom burden, often at disease onset, which increases over time due to tumor, treatment effect, or most often, an admixture of both” (2009). These incumbrances all fall under the category of late effects.

BACKGROUND

Late effects

Late effects are the combined neurocognitive, psychological, and social deterioration and/or deficits experienced due to the brain pathology itself or exposure to associative treatments. Late effects are highly individual and come with varying ranges of severity due to the diverse group of aetiologies, treatment options and subsequent modality-specific toxicities, care options, and disease occurrence. While there is no setlist of what a patient will experience, there are common problems that have been reported, and new connections are coming to light through more research and clinical trials.

Cognitive impairment is just one of many possible late effects, but 50-90% of brain tumor survivors will experience this to some degree (Alemany et al., 2020). Cognitive impairment could include the considerable decline in executive functioning, attention, working memory, and processing speed. Diminished IQ and mental fatigue are also reported (Winter et al., 2014). Fatigue has been associated with other somatic and psychological late effects as well. Endocrinopathies and depression, for example, can be the culprit. However, fatigue can also display as a stand-alone symptom and manifest as concentration problems, irritability, and physical deficits. The interplay of the late effects is not well studied but is a valid concern.

Problems in the area of executive functioning would include the inability to regulate behaviors and exert control, goal setting, and willpower. In a study by Alias et al., processing speed has been listed as a “core deficit.” The forfeiture be related to white matter loss or a consequence of neurotoxicity due to treatments. Included among

available data were metrics describing the extent of a neuropsychological processing deficit which comprised of: “alertness, sustained attention, focused attention, working memory capacity, executive visuomotor control, and cognitive flexibility.” Survivors are susceptible to distractions, have lower levels of inhibition, and are less alert, which is in concurrence to available research (2018).

The presence of these neurocognitive late effects has been thought to predict subsequent psychological and social difficulties. Coping through the lifecycle of survivorship has been listed as an essential idea when considering the mitigation of late effects and the eventual dilapidation in health. Cognitive impairment and decline in executive functioning, as aforementioned, can result in the decline of processing, attention, working memory, etc. This is likely to affect the ability of the survivor to utilize appropriate coping mechanisms and can consequently exacerbate psychological and social effects – the proverbial domino effect. (Robinson et al., 2015).

The literature is scant on psychological effects experienced by survivors, as few studies have truly addressed these effects or focused solely on them. Yet, among the body of evidence, these effects are noted across the board. Psychological corollaries encompass the mental, emotional, and behavioral aspects of the patient experience and regularly overlap with the neurological effects. Likewise, social deficits experienced by brain tumor survivors consistently coincide; hence they are often referred to as psychosocial effects. Behavioral and emotional regulation are important factors to consider as deficits in these areas could affect school and subsequent work performance in later years. Hindrances in these settings might mean a limited scope of options for future occupational prospects, do to perceived incapability.

From a behavioral standpoint, attention-deficit/hyperactivity disorder (ADHD) is prevalent in the survivor population, especially for those diagnosed at a younger age (Shabason et al., 2019). Social competence, a facet of self-esteem, has been reported as a deficit for PBTS, with anticipated continual decline as the patient ages (Ruiter et al., 2015). Indications of post-traumatic (PTSD) stress are not uncommon for survivors and appear to gradually intensify with age, with 20.5 % of adult survivors meeting the criteria for PTSD in one studies sampling, as cited in a study by Cousino et al. (2017). Depression, anxiety, and antisocial behavior have all been consistently conveyed throughout the trials. Suicide ideation was noted in a study from *Cancer Epidemiology*, with had a reference to the Childhood Cancer Survivor Study (CCSS). Somatization, where psychological concerns manifest as organic symptoms, was described in the study as well. This phenomenon increases progressively as survivor health deteriorates. Within the same study, a cited Swiss Childhood Cancer Survivor Study (SCCSS) listed psychotic tendencies and aggression among self-reported late effects, especially prevalent in the female survivor population (Erdmann et al., 2020).

External factors such as family stress, coping, and function seem to play a role in the mental and emotional state of the survivors – though adaption related studies are insufficient. Competence and attention either by the caregiver or the survivor appear to have an effect in the direction of long-term outcomes, which range from positive to detrimental (Barakat et al., 2015). Survivors of brain tumors have been found to leave their parent homes later, especially males., and there is some evidentiary support for a lower likelihood of marriage and cohabitation with a partner. Further, survivors are less likely to have children. These aberrant social consequences are assumed to be attributable

to psychological late effects, biological repercussions from somatic late effects, fertility problems, and concerns about the future (Erdmann et al., 2020)

These outcomes are often measured through self-reporting or report by proxy, via a teacher, parent, caregiver, etc. Health related quality of life, HRQOL, is a comprehensive evaluation that is frequently referred to in the literature. This subjective measure looks at illness from a multidimensional perspective noting general, physical, spiritual, and social aspects. Other significant features of this assessment are that it looks at resilience and body image, which is an often overlooked component of psychological health. Another common reported measure is the Intensity of treatment rating, which increases based upon treatment type and number of treatment modalities. Supplementary objective scales surrounding health and wellbeing are the Bakas Caregiver Outcomes Scale (BCOS), Family Assessment Device (FAD), Brief Symptom Inventory (BSI). These look at various aspects of social, psychological, and physical functioning to provide scope. The Pediatric Oncology Quality of Life Scale looks at HRQOL's physical and emotional components and rates them on a Likert scale, with lower POQOL being indicative of high HRQOL (Barakat et al., 2015).

While most late effects are considered damaging, there are reported measures that indicate certain positive outcomes. Resilience is the ability to recover from undesirable situations, a measure of grit. This is often increased in survivors. Further, increased social connectivity is a consequence of late effects and brain neoplasms in general. This "posttraumatic growth," as it is called, has positive connotations, and increased interpersonal relationships, resiliency, spiritual and religious beliefs, body image are among these positively reported measures (Satariano, 2016). Despite reported limitations

in social interactions, one study reported friendships as being a motivating factor in continuing schooling and work, though there may be difficulties in organizational settings due to late effects. An aspect of hopeful future social growth is shown in survivors as well (Boydell et al., 2008).

Somatic disease burden can manifest in many ways, and these late effects can influence any body system. Severity and type appear to have some relation to age at clinical onset, anatomical location, presence of secondary tumor, grade, and treatment modality than the psychological and social effects. One study by Bhakta et al. found that by fifty years of age, survivors suffered up to 17 chronic health conditions, on average (Erdmann, 2020). Typically reported sequelae across the board include seizures, generally partial complex seizures, and headaches. Physical functioning and athletic performance are often at a deficit. Both hearing and vision loss have been included among possible neuropathies, as well (Alemany et al., 2020). Among possible visual neuropathies is a dysconjugate gaze, a lack of visual coordination that will affect the survivor into adulthood in both the educational setting and in reading tasks (Janss, Mazewski, Patterson, 2019).

Additionally, endocrine disorders are often diagnosed and well documented among survivors. Hypothyroidism, hypogonadism, and hyperprolactinemia are among possible late effects. Excess weight and obesity can also present for brain tumor survivors, nonspecific to gender (Ramanauskiene et al., 2014). Hypersomnia, sleep apnea, and autonomic dysregulation are also possible (Janss, Mazewski, Patterson, 2019). Pituitary hormone (PH), Growth hormone (GH), luteinizing hormone (LH), follicle stimulating hormone (FSH), and adrenocorticotrophic hormone (ACTH) deficiency are

also implicated in many survivors. Adrenal axis functioning was often seen as deficient and endocrine dysfunction was seen within a decade in one study of a neuro-oncology database (Lawson et al., 2019).

Endocrinopathies are deleterious and associated with negative prognosis including, but not limited to life-long treatment, decreased quality of life, and frequent symptom management. Endocrine dysfunctions can take many years to develop after initial diagnosis and treatment. Thus, early detection is key to improve morbidity and prospective health for survivors. For young children and adolescents, these neuroendocrine late effects can adversely affect growth and puberty, with both expedited and delayed puberty described in studies. Furthermore, because these disorders can have late-onset, they may not be diagnosed right away, as aforementioned, which can have long-lasting consequences for patients and higher associative costs for chronic disease management (Maciel et al., 2021). It is difficult to discern whether these hormone deficiencies are caused by the tumor or the treatment, although they likely have a compounding effect, and tumor location appears to play a major role as well across the various studies.

Metabolic syndromes and complications have also been listed as potential late effects. Maciel et al. described impaired cardiac functioning, abnormal lipid profiles, higher body fat levels, insulin sensitivity reduction, and atypical glucose tolerance as a direct result of hormonal late effects, most notably in GH deficiency. Early-onset atherosclerosis and bone density symptomology was also discussed (2021).

Ramanauskienė et al. similarly show through analysis that cytostatic drugs used in treatments damage endothelium and can also cause cardiovascular problems through the

promotion of atherosclerotic plaques, atherogenesis. As in the Maciel et al. study findings, the group reported dyslipidemia, diabetes, osteoporosis, and hyperinsulinism in brain tumor survivors, resulting from treatment and endocrine dysfunction (2014).

Other possible somatic late effects have included pulmonary disease. Risk factors for pneumonia are higher, and survivors of CNS tumors have the “highest excess risk” for respiratory death in the Erdmann et al. epidemiological study. French and British Childhood Cancer Survivor Studies, FCCSS and BCCSS respectively, cited within the study both indicate diabetes mellitus, as well as difficulties with pregnancy and labor. Survivors are also at an increased risk for stroke (2020).

The neurocognitive, psychosocial, and somatic late effects described within this review are just a sampling of conceivable symptom affliction suffered by patients. The breadth of prospective late effects experienced by survivors is extensive and highly complicated, as previously stated, but nonetheless a valid statement that bears iteration. Deficiencies and dysfunctions frequently take time to develop. Oftentimes, survivors will not follow up, even if a late effect is identified (Mellblom et al., 2020). There is a call throughout the literature to standardize and structure models of care to better support survivors and provide sufficient access to said care programs. For children and adolescents, it is suggested that “pedagogical assessment and individualized support” be offered, regardless of tumor grade, as it has been determined in studies that brain tumor survivors perform at a deficit in the educational setting, irrespective of malignancy, and so “require the same attention and support” (Lönnerblad et al., 2019).

With such noble intentions, these ideas appear to have merit and it is possible that educational settings could slate students for supportive care. However, for the average adult, past the formative educational years, there is no such system in place to control for preemptive screening, cognitive training, and symptom management. Hardly falling within recognized traditional company benefit obligation, the responsibility falls to the patient and/or caregiver. Moreover, time taken from employment and costs of screening outside the educational setting provide another complex set of considerations for the brain tumor survivor. Such luxuries are not afforded to all.

Survivorship

In 2006, the Institute of Medicine published a bellwether report: *From Cancer Patient to Cancer Survivor: Lost in Transition*, whereby the idea of “survivorship,” and what that meant, began to take a tangible form. The report defines survivorship as a “distinct phase of the cancer trajectory,” Providing such designation takes the metaphysical ideology of what it means to “survive” and bestows both shape and scope. A clear delineation of the term allows for advocacy and awareness. These are essential functions in fostering momentum and a trajectory towards a goal, namely individualized clinical standards, comprehensive research, and accessible, perpetual coordinated care. Furthermore, the report elucidated essential components of what survivorship care is comprised of, as seen in Figure 1.

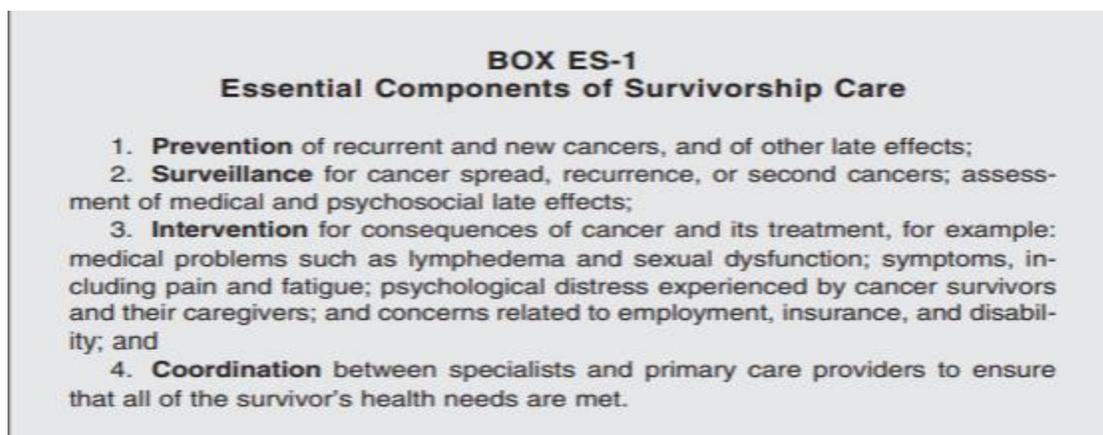


Figure 1. Essential Components of Survivorship Care, as outlined by the Institute of Medicine Report, *From Cancer Patient to Cancer Survivor: Lost in Transition* (2006)

The authors of this review speculated that care of quality would include these four components and imagined a system wherein attention and awareness afforded both providers and patients a more complete understanding of late effects, earlier diagnosis, and a plausible detection of trends (National Research Council, 2006).

With the advent of a new survivorship definition, there came a “strong impetus to take clinically meaningful steps toward broadening the focus of neuro-oncological care” (Leeper, Milbury, 2018). The growing consensus was that there was a need to focus on individual patient care plans, as well as an overhaul at the systems level (see Appendix A). A model of quality care would take a village, or rather a high qualified, interprofessional team that can take the patient from the acute phase at clinical diagnosis, through follow-up, and into long-term management. An identification of the “stakeholders” was invaluable to carving out a purposeful plan (National Research Council, 2006). In 2012, the American College of Surgeons Commission of Cancer (CoC) issued a directive that patients be provided survivorship care plans. This was a crude first attempt at standardizing care as outlined by the 2006 report. These early plans were to include information surrounding the patient-specific treatment modality and recommendations for follow-up, mostly to be utilized in future primary care. The

primary care physician (PCP) was projected to play a significant role in the early models of care as the thruput between necessary intervention and continued management of health. Other mid-level were expected to fill in the gaps (Leeper, Milbury, 2018).

RESEARCH STRATEGIES

Applicable background information on late effects, the survivorship concept, and data surrounding general survivorship programming were derived by conducting a systematic review of publicly available databases and search engines for scholarly work, including EBSCO host, PubMed, and Google Scholar. The year of publication was noted, with newer, pertinent studies taking precedence in the selection of relevant literature. Moreover, peer-reviewed articles were slated for consideration as a means of quality control to provide rigor to the data and ideas presented herein.

Three large hospital systems in Pennsylvania with an associative survivorship program were chosen for comparison: University of Pennsylvania Hospital (HUP), Thomas Jefferson University Hospital (TJUH), and University of Pittsburgh Medical Center (UPMC). The Sidney Kimmel Cancer Center, an Affiliate of Thomas Jefferson University Hospitals and Jefferson Health, offered the Neu Center for Supportive Medicine and Cancer Survivorship as well as the Brain Tumor Center support group. The Abramson Cancer Center is the oncology center for the University of Pennsylvania Hospital and has the Abramson Cancer Center Survivorship Program. University of Pittsburgh Medical Center is associated with the Hillman Cancer Center and offers the LiveWell Survivorship Program.

Reviews of the various program offerings were completed using information available from the relevant group websites and general details through operators of the programs. Qualitative assessments of the survivorship plans were made based upon the model, perceived efficacy, and consistency of care. Patient follow-up timelines were taken into account, with respect to acute phase, just after rendered diagnosis and treatment, versus long-term management of care. Coordination of services and appointments via a primary care physician, a team, or a nurse navigator was observed. Furthermore, attention was paid to the patient and caregiver responsibilities within the survivorship planning, as it was perceived to affect compliance with a care plan.

DISCUSSION

As illustrated throughout the review, survivorship has become a topic of increased interest within the research realm and in the field of public health. For brain cancer survivors, the initial assault of diagnosis and treatment are typically just a blip on the radar, with the actual battle existing in the future unknown and the array of potential late effects. Information is power, and the access to pertinent material surrounding late effects, warning signs, supportive care, and mitigation strategies is thought to be the best way to bolster survivors for what is to come. Cancer programs, research teams, and major hospital systems have begun to implement programs and policies aimed to meet the essential components of survivorship care (Figure 1). Care models have been developed accordingly in an attempt to meet the various and individual needs of patients and their families. In an effort to provide a meaningful contribution to this body of knowledge, this

review looks at a comparison between three major hospital systems in Pennsylvania and respective survivorship care models.

Penn Medicine

The University of Pennsylvania Health System (UPHS), colloquially referred to as Penn Medicine, is associated with the Abramson Cancer Center. U.S. News & World Report lists it among the top 15 hospitals in the nation and within the top 20 for Neurology and Neurosurgery (U.S. News, 2020). As the forerunner in the group to be studied, it was initially assumed this system would provide the most thorough and comprehensive model. Indeed, upon examination, UPHS had a very well outlined program run in large part by nurse navigators who coordinated care and services with other providers within the UPHS system. The Abramson Cancer Center interestingly boasted the first adult survivorship care program in the nation, founded in 2001. This is not insignificant, as most therapeutic interventions, research, and survivorship models centered around pediatric tumors and adolescent late effects (Survivorship Program, 2021). This model had specific teams for cancer type, with a nurse navigator leading each team. For neuro-oncology specifically, there is a brain tumor support team and integration of services to meet individual needs (Brain Tumor Support, 2021).

New patients gain access to the system through a referral from a primary care provider, a specialist or can recommend themselves through a call center. Patients could include local residents, children aging out of the Children's Hospital of Philadelphia (CHOP – pediatric affiliate of UPHS), or even out-of-state residents willing to pay out-of-network premiums for advanced therapies and the UPHS name. The navigator will

assess current state, schedule necessary treatment based upon provider recommendation, and then proceed to connect the patient with various subspecialties for continued care. Contact is largely achieved through email and phone calls for the convenience of the patient and to alleviate the heavy patient load of the navigators. When a patient passes the acute phase, which is the immediate diagnosis and treatment, they are slated for follow-up scans and appointments as recommended by the oncologist and managed care team that is part of the patient's individualized care plan (Brain Tumor Support, 2021). Insurance is not likely to be an issue, for if it is accepted once, it will be accepted anywhere within UPHS. Due to the extensive network within UPHS, there is no barrier in coordination of services in-house, and there is no lack of sub-specialists available to provide world-class care.

The positive aspects of UPHS speak volumes to the program. The nurse navigator's role coordination of care in the acute phase alleviates patient and caregiver burden to a certain degree, which is critical under the duress of the circumstance. The broad network of providers and subspecialists means patients can be confident symptoms and late effects will be addressed by a highly qualified individual within the system. The neuro-oncology brain tumor support team tends to keep patients until the follow-up scans trail off, whereupon there is the Survivorship Program, which is available to any adult survivor. This program will provide a questionnaire to identify late effects, but again – after the presentation. Information and strategies for late effect management are given as well, and referrals are made to nutritionists for metabolic complications, therapists, or specialists such as an endocrinologist for somatic symptoms. The University of Pennsylvania is a major research center; as such, there are a wealth of studies and clinical

trials being conducted with convenient access. Patients can receive information about these services through the generalized survivorship program as well, but it is not required. Survivors are given a choice to continue coordination with the neuro-oncology team (Brain Tumor Support, 2021).

The UPHS model of survivorship has good bones. The effort aimed at quality and access to care is evident. The scaffolding for a truly efficacious model is present, but there are some negative aspects to the UPHS model. For one, navigators make calls to patients in the acute phase for coordination of services and treatments. After such time, it is the burden of the patient or caregiver to contact the navigator or primary care physician for follow-up scans, symptom discussions, and questions about late effects. Doctors may or may not have discussions with their patients about late effects, but these tend to happen as symptomology presents, meaning there is only secondary or tertiary intervention of late effects, at best. There are no prescription services, and the use of the navigator or provider to coordinate care does not necessarily ensure a timely visit. Patients and caregivers still must wait patiently for appointments, as nothing is expedited by virtue of utilizing the survivor care plan. There is also limited capacity, especially for psychosocial treatment. Should a patient need more care, there will be a local referral, but the responsibility falls to the patient again to set up and manage appointments and symptoms. The model is focused more on access for new patients, as opposed to case management. There are limited resources and staff, even for such a large system. As such, patients still fall through the cracks.

Jefferson Health

Thomas Jefferson University Hospitals (TJUH), or Jefferson Health, is another nonprofit organization in the city of Philadelphia. While lacking the ivy name, TJUH has no shortage of hospitals, 14 to be specific, and is not lacking in available services for brain tumor patients and survivors. In fact, there is a recent merger proposal with Einstein Healthcare Network, which would greatly expand the Jefferson services, provide access to the underserved populations in the Philadelphia area, and further the interests of community health initiatives (Brand, 2021). Within the Jefferson hospital system, there is the Sidney Kimmel Cancer Center and the Brain tumor support division. More recently, the Neu Center for Supportive Medicine and Cancer Survivorship was founded, but it is currently only available at the Center City Campus (Jefferson University Hospitals, 2021).

The Brain Tumor Support Center is facilitated by specialists from the Vickie and Jack Farber Institute for Neuroscience, as mentioned in the Support and Resources Tab of the Jefferson Hospital webpage. Offerings include a Brain Tumor Support Group every second Thursday of the month, where patients and families can attend to receive emotional support from other survivors, creating a sense of community. Through the Cancer Center are generalized oncology social workers, support groups, and nutrition counselors available to patients. Just as with UPHS, there is access to clinical trials and research studies, as well as educational opportunities to learn more about brain tumors and treatment options. The group also affiliates with the Reproductive Medicine Associates of Philadelphia for fertility counseling. During the pandemic, the Cancer

Support Center at Jefferson wanted to maintain supportive care options, and so provided mindfulness activities to help ensure cancer patients did not feel alone, such as free Headspace Application access, guided art, and recorded yoga (Supportive Care for Survivors, n.d)

The Neu Center for Supportive Medicine and Cancer Survivorship offers informative services to newly diagnosed patients. Greg Garber, Director of the Cancer Support and Welcome Center, explained the program offerings as such: “This can be anything from work and family-related issues, to the management of distress and other symptoms, education around survivorship-related topics, and guidance on nutrition, finances and insurance, sexuality and intimacy, and self-care,” (Supportive Care, n.d.). This program aims to specifically address psychosocial late effects in an impactful way. The spiritual, psychological, social, and economic needs of survivors and their caregivers are taken into consideration. Appropriate counseling and assistance are rendered based upon continued evaluation of the survivor population. Garber purported “aggressive” management of symptoms in “as many areas as possible” would mean better outcomes for survivors (Supportive Care, n.d.). Through the program, there is also the Jefferson Buddy Program, that connects patients with a similar diagnosis.

Positive aspects of the Jefferson Program definitely include the strong psychosocial late effect support component. The Neu Center is unique in the rigor of this feature, and it can only serve to help survivors better handle late effects as they arise. Furthermore, the range of supportive care groups, specific to interventions, and integrated mindfulness activities provide a well-rounded program for survivors choosing the

Jefferson System. Yoga, meditation applications, and educational services can help patients better cope, which is an important facet of survivorship, as previously mentioned. Whereas fertility management and aggressive psychosocial late effect exploration help mitigate somatic and psychological symptom burden. Moreover, research from the current patients allows for further survivor care plan development, as this program is in its infancy. This is indicative of better patient outcomes as the program matures.

The Jefferson program attributes are worth noticing. However, there are still deficits. For one, the plethora of counselors and the use of the generalized Support and Welcome Cancer to get access to the brain tumor-specific services could be confusing for survivors. While it may not seem significant, extra calls, more hurdles, and less help equal more burden for patients and caregivers, already exhausted mentally, physically, and emotionally. Also, there is little information pertaining to the active pursuit of patients for supportive follow-up, meaning they likely fall through the cracks, just as with UPHS.

UPMC

The University of Pittsburgh Medical Center, or UPMC, is a mammoth nonprofit healthcare organization on the western side of Pennsylvania, with satellite locations nestled all over Pittsburgh. U.S. News & World Report lists UPMC Presbyterian Shadyside as the number two hospital in Pennsylvania, above TJUH, and just behind UPHS, which holds the coveted first place position (U.S. News & World Report, 2021). The population served by UPMC is much smaller than Philadelphia, as cited by the

United States Census Bureau, with some 300, 286 residents. The lack of ethnic diversity is a point of interest, excepting a larger Hispanic population than Philadelphia. Education and cost of living, and availability of housing are higher for Pittsburgh, too. This means the health landscape of the city looks different, based upon the social determinants of health (Quickfacts, 2019). Suffice it to say, UPMC serves a vastly different population with different sets of needs. However, the system is equally matched in neurosurgery and neurology, just neck-and-neck with the University of Pennsylvania Hospital System for rankings.

UPMC offers the Hillman Cancer Center and the Cancer LiveWell Survivorship Program to address late effects and patient requirements throughout the life cycle as a survivor. According to the available data: “Our goal is to make cancer survivorship as easy as possible by addressing a wide range of needs and concerns” (Survivorship Program, n.d.). Just as with TJUH and UPHS, the LiveWell program grants patients access to clinical trials and participation information for research studies, support group connections, and newsletters. The rest of the services are aimed to augment current and prescribed care. Consults are offered with midlevel providers upon treatment completion. The website lists coordination of care as a possible service to integrate future care with the doctor’s therapeutic forecast. Observation for reoccurrence is assured, and management of both short- and long-term effects is mentioned but not explicitly stated. Genetic counseling, fertility services, nutrition, and rehabilitative services are available at patient or provider request (Livewell Survivorship Program, n.d.).

As with the other programs, the UPMC model of survivorship care seeks to address late effects and help provide services surrounding treatment become a reality through care coordination in the acute phase. The website outlines these services in no specific terms but asserts the provider consults would manage follow-up care. With nutritional, fertility, and genetic counseling options, UPMC is up to date with competitors in provisional tools available to survivors, too. The hospital system is diverse, so availability of practitioners is not a problem, nor is accessing services, provided the insurance was accepted upon treatment. Similar to the case with UPHS. Patients have a plethora of specialists to choose from but must wait for appointments, as with UPHS.

The biggest pitfall to the UPMC LiveWell program is, again, the patient/caregiver incumbrance to coordinate. Even accessing the initial coordination services must fall to the patient or caregiver after a doctor has given a referral. Symptoms are addressed as they come about with providers, not before, and that does not necessarily mean a call to the program will be made. Primary care physicians and specialists make recommendations, and it is up to the patient to follow up. Also, the generalized nature of cancer services is less targeted than the Jefferson or Penn models. Brain tumor survivors are afforded more individualized care through the lifecycle of the Philadelphia brain tumor survivor-specific programs.

A Utopian Model of Survivorship

From the comparison, it can be discerned that the three systems had three vastly different programs, despite all working towards the collective goal of quality care, tailored services, cohesive coordination, and better prognosis for survivors. Across the board, the

eyesore in each of the presented programs was the responsibility placed on patients and caregivers to access services. This hardly seems reasonable, considering the constraints and stress a patient and their loved ones are already experiencing just by virtue of diagnosis and subsequent treatment.

The lack of available professionals to ensure adequate follow up is understandably a cause for concern. Mid-level providers play an important role, bridging the gaps and alleviating the strain on primary care and specialist services, and navigating follow up. UPHS does this best with the nurse navigator system. The other two models make some effort to coordinate between the specialist treatment plan and the primary care providers. Still, the outline of these models in action was vague, fragmented, and vapid in approach. The previously mentioned Institute of Medicine Report, describes this as an “absence of a locus of responsibility” (National Research Council, 2006). When there aren’t enough nurses and navigators, the responsibility shifts to caregivers and patients. This is inappropriate and irresponsible from an ethical standpoint.

A proposed survivorship model would feature end-to-end integration of services. See Appendix A for a process flow diagram depicting an example of a proposed survivor care model. A streamlined, standardized process where survivors and caregivers take a back-seat approach and follow along for the journey would be the gold standard. Specialists and primary care providers would sit at the forefront, determining treatment modalities and individualized follow-up plans based on clinical diagnosis, treatment type, and patient history. Future scanning and primary care visits would have an outlined schedule that could be adjusted for patient needs. Nurses act as the navigators within the utopian model for all

coordination of care, including scheduling scans, services, prescriptions, and late effect tracking. A system like this would require advanced record keeping, registry-based systems, and separate systems to keep track of all patients, such that no patient is left behind.

Much of the literature cited the importance of preventative care. For neurocognitive and psychosocial late effects, this includes catching symptoms early for a better prognosis. In an ideal model, all patients undergoing treatment would receive supportive services throughout the acute phase and then receive a baseline assessment upon entering into a survivorship program. Best practices would dictate each patient be assigned a counselor or social worker, along with the nurse navigator, who could see the patient, caregiver, and family through the next phases of care. Patient fears and concerns would need to be assessed and periodically addressed through virtual health sessions, visits, surveys, phone calls, or emails. It would fall to the social worker to document, record, and relay any concerns to the primary care physician or nurse navigator for follow up, and it would be the job of the liaison to continue to check-in.

Speech, occupational and physical therapy baseline assessments could be made upon entrance into the survivorship program. Furthermore, it would be useful to garner a baseline for genetic, fertility, and nutrition as well. Late effect deficiencies in these areas are well known, which is why most programs offer services for them. The best model screens late effects habitually. Supportive psychosocial services, neurocognitive testing, screening for various somatic and psychological effects could all be described by the liaison and set up, on a standardized time schedule, by the nurse navigator.

For children, many of these services could be shared with specialized school officials. It has already been determined that pediatric brain tumors are the second leading cause of cancer (Lönnerblad et al., 2019). With this statistic in mind, there is certainly no lack of need for trained, specialized workers to handle this kind of care. School counselors could be informed when a child has been diagnosed, set them up for screening, therapy, appropriate testing, and work with the parents to ensure better educational outcomes and future prospects for pediatric brain tumor survivors. One such study, by Northman et al., describes a plan that includes “school-hospital liaisons” that help with home services, ensuring a more supportive environment at school with regard to neurocognitive deficits. In this model and in other proposed plans, the researchers advocate the importance of continued surveillance to more adequately address late effects, ensure rapid diagnosis at onset, begin application of the appropriate treatment, and see that testing is administered in a timely fashion. Another big push is for medical staff, school, caregiver, and social worker communication. This ensures all the relevant people involved in patient care are all on the same page, treatment decisions can be agreed upon, and swift action taken in decline (2015).

From a public health perspective, community initiatives specific to survivors could manage the support aspect of the survivor programs. Support groups, social determinants assessments based upon socioeconomic condition, life cycle position, risk factor groups etc. could be controlled by local public health offices. Resources, programming, education, pamphlets for late effects, and prevention services could also fall under the public health sphere and are among the ten essential services of public, under the assessment and policy development functions (CDC, 2021). Epidemiologists and public health officials keep

many records and databases current with relevant health data. Utilizing an observation and surveillance system to track late effects, chronic conditions, and a survivor record system would be an effective measure. Further, a registry system would catch non-compliant patients and allow public health workers, primary care physicians, counselors, and social workers to slate this population for assistance and service access. For example, if transportation or economic decline are issues, these patients could utilize aid available through nonprofits and public health organizations to meet their needs and therefore be able to seek necessary treatment and testing.

This kind of model would be more feasible in big healthcare systems or wealthier for-profit hospitals. However, it would be best if the bigger systems collaborate with smaller systems and hospitals, thereby bolstering some of the cost and sharing the work equally. This is easier said than done, and these registries would have to comply with HIPAA standards. Nevertheless, ensuring adequate record-keeping, utilizing national registries, and having a solid tracking system would help patients get the best possible continued care, no matter where care is received. At the end of the day, keeping patients up to date with checkups, screening, and follow-up is paramount.

External to the standardized model are extraneous factors that act as barriers to care, such as cost, health insurance, availability of providers, and employment concerns of patients. More should be done for hiring adequate numbers of counselors, navigators, trained oncology nurses, and social workers. For an integrated system to be set in place, organizations would have to be on board with appropriate headcount, hiring, and recruiting for services. When the three hospital systems were discussed, one of the redeeming

qualities of each of the programs was the broad access to a network of subspecialists and providers. If insurance is accepted, there is likely no issue going forward with care, as the network all fall under the same umbrella. However, this does not address the elephant in the room, which is the American Healthcare System as a whole. Not every American is covered, and even if there is a plan, the insurance company may not always agree to care and follow up. Also, many insurance plans are based on employer coverage. Throughout treatment and with future suffering of late effects, survivors may lose access to employer provided benefits, including insurance. Even if survivors are covered, the premiums, copays, coinsurance, cost of services not covered may be financially crippling.

The proposed model would increase the number of visits and services utilized, but follow-up is a complex process. Though a large portion of this might be considered preventative care, in which case insurance may cover it, the boundaries and lines in the convoluted insurance system are blurred. The 2006 report addressed some of these concerns stating that Medicare can be an option for those over 65, but a large portion of the survivor population does not fall into that category. Within the report, there was a mention that an Institute of Medicine committee that addresses insurance pushed for Universal Healthcare in 2004, controlled costs for services, and increased funding to other programs that could provide coverage, such as COBRA (Consolidated Omnibus Budget Reconciliation Act). In the interim, continuous health care for cancer survivors is invaluable, and many of these interventions can be achieved through policy writing. Funneling federal money into high-risk pools was proposed, which would lower costs for those under managed care plans. Additionally, there was mention of Medicaid eligibility

criteria changing for breast and cervical cancer patients in 2000, but no such exception was made for brain neoplasm and other cancer survivors (National Research Council, 2006).

The biggest takeaway from the literature review is that this is a multifaceted process. With more healthcare systems acknowledging the experience of the survivor and crafting plans to offer supportive care, there is likely no one size fits all model. Existing plans, such as those in the Pennsylvania health systems, are built upon current knowledge and research. Local health care systems are making concerted efforts, utilizing available resources, to provide their definition of quality care. It's a step in the right direction. A six-year survivor of malignant brain cancer, Michael Feuerstein, described his survivor care plan as a "living document" (2009). The process of defining a plan, staying on top of late effects, managing chronic conditions, integrating teams and providers, getting insurance on board is clearly a dynamic process. With advocacy and awareness, these themes can be explored further. Best practices can and should be continually reassessed, with new information and upgraded care plans displacing outdated archetypes.

The survivorship model is a "simple and logical dimension of care," but it's easy for patients to feel lost when it comes to the long-term outcomes (Feuerstein, 2009). As new research is conducted surrounding survivor care models and quality, changes can and should be implemented aimed at alleviating patient and caregiver burden. Attempts should also be made at the policy level to provide a standardized version of care as a springboard to further the efficacy and patient perceived value of supportive care plans. Taken together, these endeavors would afford brain tumor survivors a safety net, some semblance of normalcy, and a better quality of life.

RECOMMENDATIONS FOR FUTURE STUDIES

Future recommended studies should investigate standardized care plans, costs, efficacy, and troubleshooting. These plans can be implemented or overhauled if solid research supports a better model of care. Additionally, the interplay between neurocognitive, psychosocial, and somatic late effects is only beginning to be expounded, and should be studied more thoroughly. More studies on adults should be performed, as much of the literature focused on children due to brain plasticity and likely due to readily available funding for pediatric research. Further, tracking of late effects to discover patterns and potential markers should be considered. Much of the literature could only speculate the causal relationship of late effects and assumed shared causation of treatment modality and the neoplasm pathology. Research to better understand these mechanisms would help providers and patients understand the direct implications of a specific treatment modality and the effects that could ensue. Responses in the education system for children are beginning to be studied, but more could be done. Also, further research concerning adult survivors in the workforce and complications surrounding late effects should be done.

Public health interventions and community based survivor care models could also be researched. How best to disseminate information to patients, providers, and the public on long-term effects and survivorship is an important question. Also, determining what survivorship looks like in different stages of the life cycle, by zip code, in different ethnic backgrounds is a worthy goal. Information surrounding these topics is important to provide scope to a complete and equitable care plan, with appropriate access and

consideration. Cancer is a major public health concern, and it would behoove the public to be knowledgeable of these outcomes, as many will likely encounter what it means to be a “survivor.”

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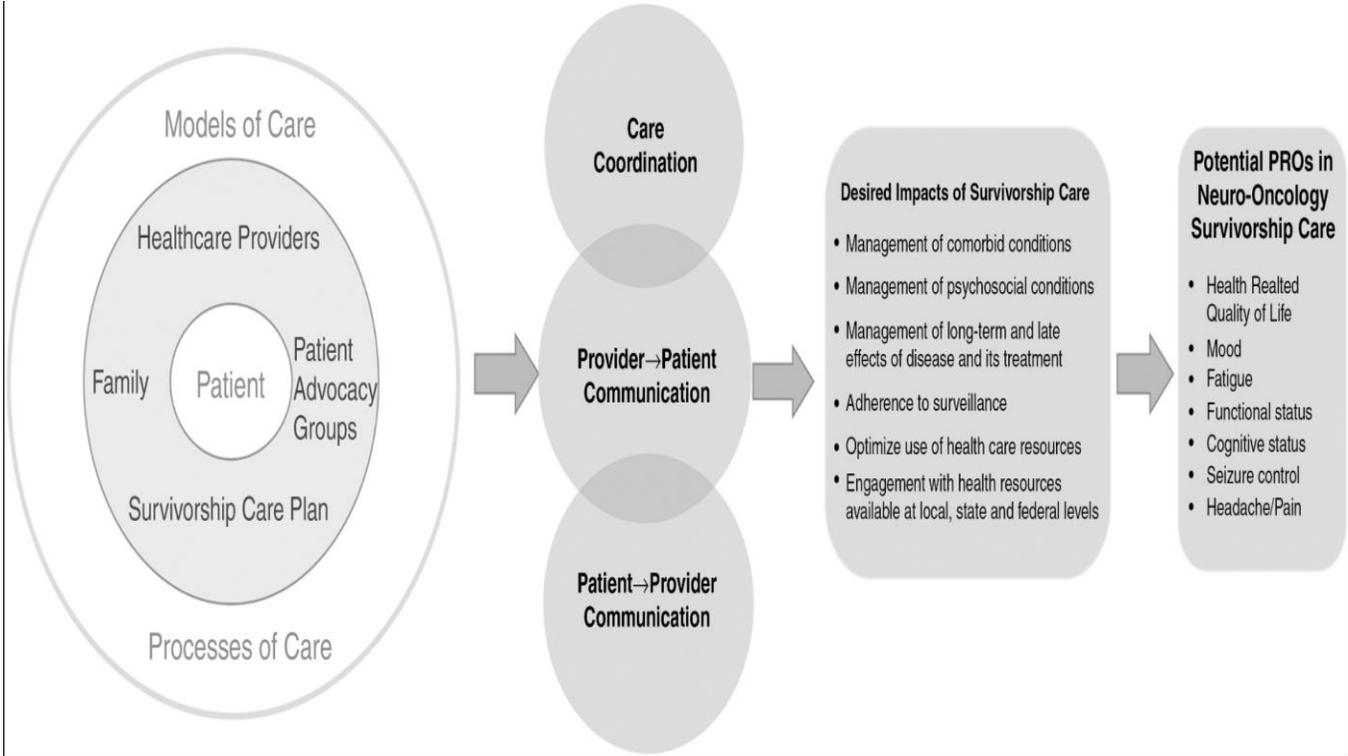
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APPENDIX



Appendix A. An Example of Survivorship Care - An Integrated Process Flow Map for a Plausible Model of Quality Care (Leeper, Milbury, 2018).