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What They Want: Inclusion of Blood and Marrow Transplanation Survivor Preference in the Development of Models of Care for Long-Term Health in Sydney, Australia

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A B S T R A C T

Four hundred forty-one adult allogeneic blood and marrow transplantation (BMT) survivors participated in a cross-sectional survey to assess long-term follow-up (LTFU) model of care preference. Survey instruments included the Sydney Post BMT Survey, Functional Assessment of Cancer Therapy-BMT, Depression Anxiety Stress Scales 21, the Chronic GVHD Activity Assessment—Patient Self Report (Form B), the Lee Chronic GVHD Symptom Scale and the Post-Traumatic Growth Inventory. We found most BMT survivors (74%) would prefer LTFU with their transplantation physicians alone or in combination with transplantation center–linked services (satellite clinics or telemedicine) Over one-quarter indicated a preference for receiving comprehensive post-transplantation care in a “satellite” clinic staffed by their BMT team situated closer to their place of residence, with higher income, higher educational level, and sexual morbidity being significant social factors influencing this preference. Regular exercise was reported less often in those who preferred telemedicine, which may reflect reduced mobility. The factor most strongly associated with a preference for transplantation center follow-up was the severity of chronic graft-versus-host disease. Full- and part-time work were negatively associated with transplantation center follow-up, possibly implying decreased dependency on the center and some return to normalcy. This study is the first to explore the preferences of BMT survivors for long-term post-transplantation care. These data provides the basis for LTFU model of care development and health service reform consistent with the preferences of BMT survivors.

INTRODUCTION

Advances in transplantation technologies, better patient and donor selection, and improved supportive care over the past 2 decades have significantly improved outcomes of bone marrow transplantation (BMT) such that 70% to 80% of those who are alive at 2 years can expect to live long term [1,2]. Unfortunately, many of these survivors experience significant late morbidity and mortality. A collective effect of underlying disease and comorbidities, prior treatment, toxicity of conditioning therapies and immunosuppression, and effects of graft-versus-host disease (GVHD) [3-5] results in a 59% cumulative incidence of developing a chronic health condition by 10 years after transplantation [6], a 3.5-fold increased risk of developing a severe or life-threatening condition compared with siblings [7], and a 30% lower life expectancy in adult BMT survivors [8]. Each of these

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long-term and late effects are even more profound in adult survivors of childhood BMT [9-11]. Life-long follow-up is, therefore, essential to optimize the benefit and minimize the prevalence and impact of the adverse late effects of BMT [12].

Consensus guidelines for screening and preventative practices for long-term survivors of BMT have been available for almost a decade [13,14]. These guidelines, agreed to by 7 international BMT organizations, outline the surveillance tests, clinical assessments, and preventative care that BMT survivors require at regular intervals—for life—to monitor for recurrent and secondary malignancies; chronic GVHD; infections; respiratory, cardiovascular, renal, musculoskeletal, ocular, oral, gastrointestinal, dermatological, and endocrine dysfunction; and psychosocial issues, among others. Given the range of morbidities experienced by BMT survivors, it is unsurprising that a BMT survivor receiving follow-up care according to these guidelines would require up to 34 assessments annually; including health history, clinical examinations, laboratory analysis, diagnostic imaging, psychosocial assessments, health counseling and education; and involve at least 6 clinical specialties [14]. This demand is likely to increase in coming years as the indications for BMT expand, more recipients of BMT survive [15], knowledge of late effects increases, and the BMT physician workforce plateaus [16,17]. Although there is broad agreement about the necessity for comprehensive follow-up of BMT survivors, the demand for long-term follow-up (LTFU) is placing an overwhelming demand on the capacity of transplantation centers (TC) that have historically been responsible for such care. Given the diverse needs of transplantation survivors and the variable capacity of TCs to provide LTFU [18], different models for delivery of long-term health care for BMT survivors have been developed. Drawing on experience in both cancer survivorship and chronic care, these models of care include variations of specialized LTFU clinics at BMT centers, referral back to local hematologists and/or primary care providers, shared care models, telemedicine, and videoconferencing [12,19-25].

Patterns of BMT activity, BMT survival, and issues with BMT LTFU in Australia mimic international trends [26]. BMTs are only performed in selected major urban tertiary centers that have the necessary expertise, training, resources, and accreditation. BMT recipients who live in rural and regional areas must relocate to metropolitan areas for the pre, peri-, and acute post-transplantation period. Returning to their homes, many BMT survivors experience difficulties with access to and cost of specialist services, fragmentation of care, and poor communication in a complex health care system, which includes public and private services, and are easily lost to follow-up, particularly as time from transplantation increases. This has meant large variations in care and long-term outcomes, particularly for BMT units that perform fewer than 50 allogeneic transplantations per year. Establishing an effective model of long-term care is essential to reduce late effects and prevent premature mortality [12]. We report the results of a cross-sectional study of long-term survivors of BMT in New South Wales (NSW), Australia to identify their preferences for long-term care; to examine the demographic, socioeconomic, and transplantation factors and sequelae associated with different preferences for follow-up; to identify gaps in service provision provided to this vulnerable and high-risk patient group; and to support clinical and health policy decision-making around long-term care.

METHODS

Background to NSW BMT Service

NSW is Australia's most populous state, with a population of ∼ 7.5 million, and covers an area of 800,628 km [2]. Over one third of the residents live outside the greater Sydney area [27]. At the time of study commencement, there were 4 adult allogeneic centers in NSW, all based in Sydney and collectively performing approximately 175 BMTs annually [26]. A survey of BMT survivors was undertaken to explore survivors' health status, demographics, service utilization, and follow-up preferences.

Patients and Procedures

Potential participants were identified from allogeneic transplantation databases from all adult allogeneic TCs in NSW. Participants were eligible if they were $\geq$18 years of age (at the time of survey) and had undergone an allogeneic BMT at an adult BMT center between January 1, 2000 and December 31, 2012, were $\geq$17 years at the time of transplantation, could read and write English, and could provide consent. Names and phone numbers were provided to the research team. Consenting participants were given the option to self-complete the questionnaire or complete it via a phone interview with 1 of the researchers. A second round of telephone calls was made to 178 participants who had not returned the survey within 1 month. All authors had access to primary clinical trial data. The study protocol was approved by the Northern Sydney Local Health District Human Research Ethics Committee (NSLHD Reference: 1207-217M).

Instruments

The Sydney Post-BMT Study Survey was developed by the research team from a review of the literature and discussion with patients attending BMT LTFU clinics. The survey comprised 402 questions grouped into 20 domains and included questions relating to specialist referrals and LTFU preferences with respect to location and provider. Other relevant domains included demographics, medical complications, tests and assessments, medications and therapies, infections, vaccinations, complementary therapy use, cancer screening, relationship status income, and lifestyle factors after allogeneic BMT. The questionnaire used tick-box responses, short-answer questions, and 5-step Likert scales measuring attitudes and other factors and took approximately 1 hour to complete. The questionnaire was piloted with 6 BMT survivors in clinic and phone interviews to assess face and content validity and to check for comprehension. For each consenting participant, data were collected on dates of diagnosis and transplantation, stage/remission status at transplantation, transplantation conditioning, GVHD prophylaxis, stem cell source, and donor type.

Preference for LTFU for specialist care and health service utilization were analyzed according to a range of demographic, transplantation, psychosocial, and lifestyle variables assessed using the Functional Assessment of Cancer Therapy—Bone Marrow Transplant (FACT-BMT Version 4) [28,29], anxiety stress and depression (the DASS 21) [30-32], chronic GVHD (Chronic GVHD Activity Assessment—Patient Self Report [Form B] [33] and the Lee Chronic GVHD Symptom Scale) [34], and the Post-Traumatic Growth Inventory score [35,36]. For ease of completion, all instruments were combined into 1 booklet.

Statistical Analysis

Categorical responses were summarized using frequencies and percentages. Parametric continuous variables were summarized using means and standard deviations, and nonparametric variables using medians, interquartile ranges, or ranges. Odds ratios (OR) and 95% confidence limits (CI), Pearson chi-square test, or Fishers exact tests were used for comparative analysis of dichotomous categorical variables. Adjusted OR (AOR) to account for potential confounding effects were determined using multivariable logistic regression analysis. Two sample comparisons of parametric and nonparametric data were determined using the independent *t*-test, and Wilcoxon rank-sum tests, respectively; greater than 2 sample comparisons were determined using 1-way analysis of variance and Kruskal Wallis tests. A 2-tailed P value $< .05$ was used as the level of statistical significance.

Statistical analysis was performed using STATA version 12.1 statistical package (StataCorp, College Station, TX).

RESULTS

A total of 1475 allogeneic BMT were performed in the study period. Of the 667 recipients known to be alive at study sampling, 581 (87%) were contactable and were sent study packs. Four hundred forty-one (66% of total eligible, 76% of those contacted) returned the completed survey. Three percent declined participation.