Biol Blood Marrow Transplant 25 (2019) 1416–1423

Biology of Blood and Marrow Transplantation

journal homepage: www.bbmt.org

Supportive Care

A Conceptual Framework and Key Research Questions in Educational Needs of Blood and Marrow Transplantation Patients, Caregivers, and Families

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Article history:
Received 7 January 2019
Accepted 11 February 2019

Keywords:
Blood and marrow transplantation
Education needs
Self-management
Patients
Caregivers

A B S T R A C T

Patient, caregiver, and family education and support was 1 of 6 key areas of interest identified by the National Marrow Donor Program/Be The Match 2-year project to prioritize patient-centered outcomes research (PCOR) goals for the blood and marrow transplantation (BMT) community. PCOR focuses on research to help patients and their caregivers make informed decisions about health care. Therefore, each area of interest was assigned to a working group with broad representation, including patients, caregivers, and clinicians. Each working group was charged with identifying gaps in knowledge and making priority recommendations for critical research to fill those gaps. The report from this working group presents a conceptual framework to address gaps in knowledge regarding patient and caregiver education in BMT and recommendations for priority research questions on this topic.

INTRODUCTION

Advances in blood and marrow transplantation (BMT) outcomes are increasing the number of survivors, with an estimated one-half million survivors by 2030 in the United States [1]. Several care models are being used to address the specific needs of this population [2], many relying on patient-reported outcomes of self-management techniques. Emerging literature suggests that including patients as active team members in the recovery process has a positive impact on transplantation outcomes [3-5]. The National Marrow Donor Program(NMDP)/Be The Match developed an initiative to engage patients in setting a patient-centered BMT research agenda that was supported by the Patient-Centered Outcomes Research Institute (PCORI) [6]. This initiative was coordinated through 6 different working groups. The Patient, Caregiver, and Family Education and Support Working Group ("Working Group" hereinafter) was based on the postulate that BMT is a highly complex therapy requiring lifelong follow-up and care, which naturally implies that patients, caregivers, and their families must understand what is at stake to play active roles at all stages of the recovery process. The Working Group's aims were to review the current evidence and identify gaps in knowledge relative to patient/caregiver BMT education and to design and prioritize future relevant research questions on this topic.

METHODS

The Working Group comprised volunteers recruited during symposia and informal focus groups [6]. It included 8 BMT health professionals

Financial disclosure: See Acknowledgments on page 1422.

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https://doi.org/10.1016/j.bbmt.2019.02.017
1083-8791/

(5 physicians, 2 registered nurses, and 1 social worker), 3 BMT recipients, 1 BMT caregiver, and 2 NMDP staff liaisons. For Working Group purposes, "education and support" covered all aspects relevant to patient education, defined as the procedure in which health care providers impart information to patients/caregivers about their health status and needs [6,7]. Other types of support, such as psychosocial support, were addressed by other working groups [6].

Several meetings were held, both in-person and through 27 teleconferences and webinars over a 2-year period [6]. The Working Group first exchanged personal thoughts and experiences about education. Then an extensive review of the English literature was performed (by A.R.G. and J.F.) for the years 1995 to 2017 using the following key words: patient education, family education, methods, research, counseling, how do patients learn, problem solving, patient expectations, provider expectation, autologous stem cell transplant, allogeneic stem cell transplant, cord blood transplant, stem cell transplant, bone marrow transplantation, family/caregiver education, palliative care, research, timing of education, survey, resources, and questionnaire.

A total of 230 articles were identified and screened for relevance using titles and abstracts by Working Group members organized in dyads. All but 1 dyad included one health professional paired with 1 patient/ caregiver; the remaining dyad was composed of 2 health professionals. This effort was coordinated by a review dyad formed by 2 health professionals (K.S.P. and A.R.G.), who participated in all discussions and reviewed systematic reviews available on the topic.

The 79 most relevant articles, based on content and methodology, were then fully analyzed to identify relevant issues pertaining to patient/caregiver education. Within each dyad, articles were assigned a primary reviewer and a secondary reviewer. Primary reviewers read the articles and documented the populations studied, type of research, primary research objective, gap(s) identified, and recommendation(s) for future research. Secondary reviewers provided additional critiques. These issues were discussed with all Working Group stakeholders and used to create a conceptual framework on the impact of education on healthcare outcomes. Relevant research questions based on the identified gaps in evidence were discussed until consensus was reached.

RESULTS

Conceptual Framework

The Working Group adapted the conceptual framework of Lorig et al [8] on the impact of self-management on healthcare outcomes to include specific aspects pertaining to BMT education and outcomes (Figure 1). The first step was to conduct an assessment of patient/caregiver status before initiating the education process. The education process followed, split into 3 major components: the educator (the individual transmitting the information), the educational material (resources used to transmit information), and the delivery method (techniques used to transmit information, including format, timing, and setting). The Working Group hypothesized that the initial patient/caregiver baseline status and the education provided impact transplantation outcomes at the level of patient-reported health knowledge, satisfaction, and sense of control. This in turn can affect such endpoints as survival, return to work or school, and quality of life.

Baseline Assessment

Education is not a "one size fits all" process, but rather requires a close fit between all phases of the process, starting with an optimal context evaluation [8]. Education needs to be tailored to stages of the BMT process (pre, inpatient, immediate post, and long-term follow-up) and the general well-being/health at the time of education (eg, fatigue, coping, distress, readiness) [8,9]. The ability to learn is influenced by cultural background, motivation, and barriers to learning, as well as preferential learning styles; although most people are visual learners, others respond better to auditory or kinesthetic methods [8]. Furthermore, health literacy, which has been identified as a positive predictor of transplant outcomes in renal transplantation [9], should be taken into account. It is essential that patients/caregivers be allowed to express their preferences and expectations early in the treatment of

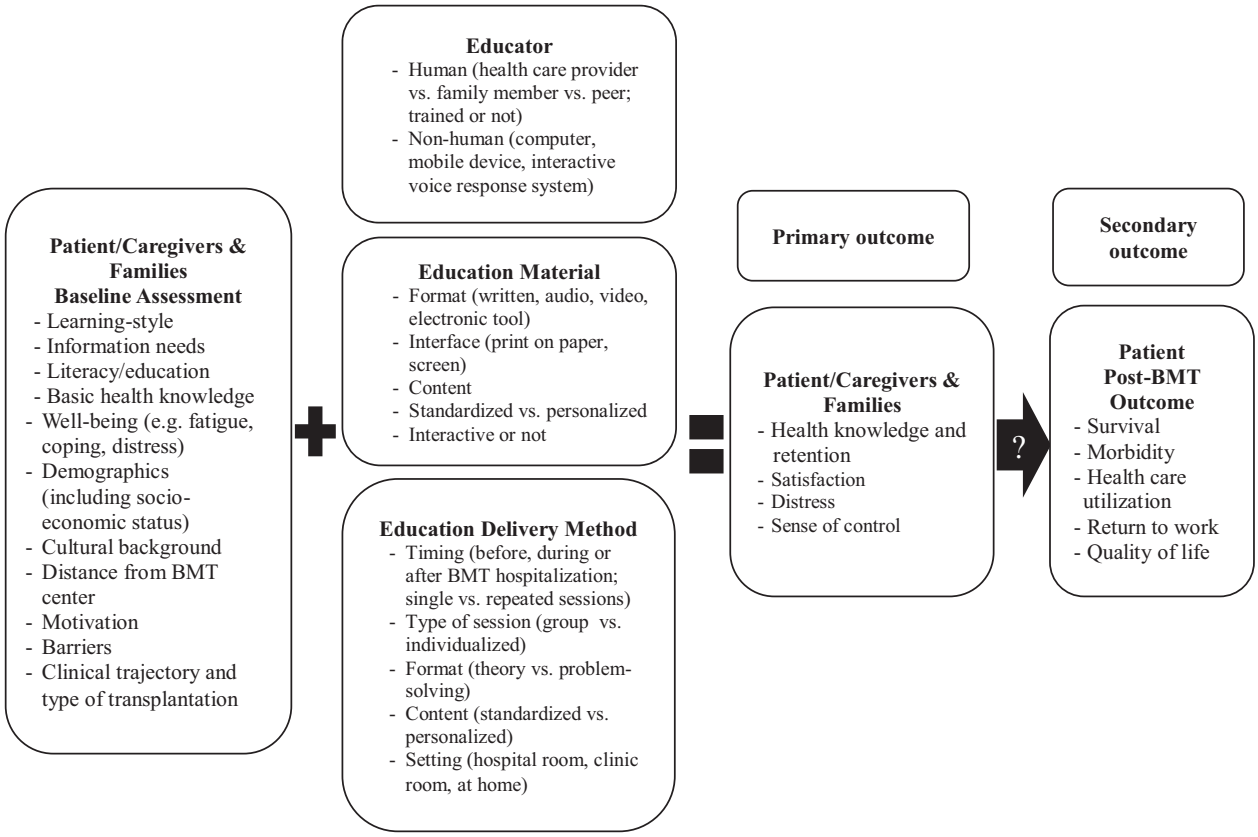

Figure 1. Conceptual model of the impact of patient/caregiver education on BMT outcomes. (Adapted from the self-management model of Lorig et al [8].)

hematologic disease [10,11], and that the information provided be tailored to address what really matters most to them [8].

Education Process

Educator. Although the Merriam-Webster Dictionary defines an educator as someone skilled in teaching [12], it does not elaborate on how to acquire that skill. In BMT, there are often multiple and varied educators, including physicians, nurses, home care aides, patient and caregiver peers, and technology experts who develop online modules. Some educators may have gained teaching skills from formal courses, whereas others may have learned from experience. The best type of educator for BMT patients and patient/caregiver preference remain unclear.

In our highly connected world, the use of eHealth (electronic and web-based health resources) as an education medium is becoming increasingly popular. Interactive response programs are attractive alternative devices to offer education, allowing for multiple presentations using skip-logic and repetition of information while respecting the patient's agenda with the ability to be accessed on demand. Such systems require an initial financial investment yet can be more cost-effective than hiring extra personnel to deliver information for larger groups of patients. Voluntary Web-based eHealth interventions have also been shown to decrease cancer symptom distress in medical and radiation oncology patients [13].

No matter who the educator is, care must be taken to engage all patients. When family practice physicians engaged with high-participation patients (defined in this study as frequently seeking/verifying information and using assertive models of communication), the communication tended to be more patient-centered. However, patients with low participation received more paternalistic education and were not as actively engaged. Regardless of the patient's behavior, a bidirectional flow of information should be sought with all patients, and educational conversations should remain patient-centered [14].

Educational material. Educational material is the material used to transmit relevant information to patients/caregivers to improve the knowledge and understanding of their health status and potential health hazards. This covers any format (ie, written, audio, video, and electronic tool) or interface (ie, printed material or screen). Important characteristics are its content (ie, the aspect of the BMT process/health status to which it relates) and whether it is personalized (adapted to the patient's age and situation) and/or interactive (ie, when the information displayed is responsive to patients).

Choosing the right information format depends on preferences and learning styles of patients to optimize information retention and cost-effectiveness [15]. Most transplantation centers provide printed education material; however, several teams are investigating other interfaces [15-19]. Cancer survivorship programs have successfully explored telephone counseling [17]. Going even further, a virtual electronic interface, such as the Inspire project (https://inspire4survivorship.org) created by the Fred Hutchinson Cancer Research Center as a resource for long-term BMT survivors regarding long-term complications, health screening, and psychoeducation, has the advantage of being ubiquitously accessible, easily updated, personalized, and interactive [18,19]. This type of interface can also offer attractive opportunities, such as adapting information to specific patient situations or adding gamification aspects (ie, point scoring, competition with others, or rules of play, to encourage user engagement) to education [16,20,21]. The BMT Roadmap project, another virtual interface based on the patient's own electronic health record, showed that using an electronic interface was well accepted by patients [16,20] and corroborated the observation of Russell et al [22] in cancer patients that personalized information is more likely to be retained.

Educational material should provide objective, reliable, and accurate information, because patients tend to overestimate the benefits of BMT [23]. Medical information should be kept up to date with recent scientific advances, a challenge to achieve. Although the optimal content of educational materials has not been extensively researched, potentially due to its variability and lack of standardization, several studies have supported the importance of education about both early and long-term effects of BMT [19,24,25] and psychosocial issues, such as changes in family roles, homesickness, care for pets, and effects on caregivers [24,26].

Several studies also have advocated approaching patients about alternative themes, including advanced care planning and early palliative care issues (eg, coping of dependent children, advance directives, defining care preferences, personal coping techniques), relevant topics not discussed in more than one-fourth of BMT recipients studied [27,28]. Loggers et al [28] recommend privileging the use of generic terms such as "supportive care" to promote patient discussions of these confronting issues. Another subject often overlooked by healthcare professionals is sexual health [24,29]. In a Swedish study, more than 80% of participants did not recall being approached about this issue, despite the high prevalence of these problems in BMT survivors [29-31]. Fertility is also repeatedly reported as an underdiscussed transplant issue [32].

It is a challenge to offer "universal" educational material to patients/caregivers worldwide. Psychosocial issues related to transplantation survivorship are closely linked to several aspects of the healthcare system [24]. Similarly, how information is conveyed needs to be adapted to local language and cultural preferences to optimize information uptake [24]. Finally, scientific evidence pertaining to minority groups (including rare diseases or ethnic groups, nonmalignant disease, relapsed disease, and geriatric patients) and the impact of BMT on non-BMT patient groups (caregivers, dependent children, and stem cell donors) is largely lacking, making it difficult to offer reliable information concerning transplantation outcomes [24,34].

Education delivery. A simple definition of education delivery is the transfer of knowledge, skills, and information from educator to student. However, in practice effective education delivery is a complex process dependent on appropriate timing (before, during, or after hospitalization; single versus repeated sessions), type of sessions (group sessions versus individual sessions), delivery formats (theory versus problem-solving), content (standardized versus personalized), and setting (hospital room, clinic room, or at home).

The initial consultation within a BMT clinic is often the first point of education delivery [33], but studies describe several other options in the timing of education delivery. Hospital discharge is consistently identified as the time of greatest anxiety for patients/caregivers and the time of greatest demand for education [35,36]. A recent study of BMT survivors showed that patients continue to seek information from a variety of sources, including health care providers and publications, with a preference for online information [37]. Internet-based programs provide broad education delivery reaching large numbers of transplantation recipients; however, broad-scale information can also be a paradoxical barrier to education

delivery, often failing to target individual needs of BMT recipients [19] and cancer patients [38,39].

In the solid organ transplant setting, it has been shown that patients/caregivers require repeated education sessions for optimal information retention and satisfaction [40]. However, time is a limiting factor for delivering the desired extent and content of information. Even after a 3-hour initial consultation, only 66.7% of BMT recipients at Loyola University reported having received adequate information to make decisions, although this could still be considered an improvement compared with the percentage before the consultation (23.2%) [37]. Group sessions are a method to optimize the cost-effectiveness of teaching moments, but a study in breast cancer survivors showed that sessions were not well attended, with just under one-half of survivors attending a minimum of 1 group session and attendance correlating with more advanced cancer and recent completion of treatment [41].

The most direct form of education delivery from health care providers is verbal teaching and discussions, which unfortunately is the least effective stand-alone format [42,43]. Some teams are now exploring interactive problem solving education sessions, which instead of offering "ready-made" solutions help individuals identifying the definition of the problem they face, generate potential solutions, envision practical implementation strategies, and plan an evaluation of the results. Core aspects of self-management to facilitate such a problem-solving approach are (1) the availability of appropriate information to allow for decision making (such as being aware of which alarm symptoms should trigger contact with health professionals, or normal thresholds when using self-monitoring devices, for example), (2) the ability to identify and use reliable sources of information, (3) the ability to partner with health professionals to report health issues timely and actively participate in the decision making process, and (4) the capacity to generate concrete action plans with specific and realistic short-term goals [8]. This approach seems to be more efficacious than traditional teaching strategies in populations afflicted with chronic diseases [8], and Bevans et al [35,44] have shown that such sessions also had a positive impact on patient/caregiver satisfaction and self-efficacy in the context of BMT.

It is likely that patients/caregivers require personalized education, because satisfaction of kidney and BMT recipients improves when patients are engaged in their own healthcare [20,45-47]. Personalized education delivery is facilitated by providing multiple information platforms, including written materials, Web-based resources, and/or group discussions in person or through videoconferencing [18,19,34,42,48-50]. Telehealth for education delivery has proven feasible and well-liked by BMT patients for learning about mindfulness, cancer symptoms, and body changes, including sexual health [50]. A common barrier to videoconferencing is sourcing equipment and independent time commitments by participants [49,50]. Preferences vary, however; in a group of kidney transplant candidates and potential living donors given tablet-based video education and group videoconferencing, transplant candidates preferred the variety of media formats, whereas donors preferred to hear from other living donors or a peer group [49]. With the advent of eHealth, it will be necessary to evaluate the different virtual channels (eg, apps, websites, YouTube videos, social platform discussion groups) available to patients in a rigorous manner to better understand where to invest for further developments.

Finally, it is an obvious but often overlooked fact that the setting and context in which patients receive information has an impact on information retention. For example, patients who have a good front desk experience and bring questions to appointments were more likely to report knowing more about their health after doctor visits, suggesting that respectful workforce and patient engagement through questions are important to patient knowledge and health behavior [51]. Frank-Bader et al [40] also showed that solid organ transplant recipients were more receptive to teaching when time was scheduled in advance with clear learning objectives and took place in a quiet environment.

Outcomes

The primary outcome reported in the majority of health education studies is whether or not respondents demonstrate improved health knowledge and make better choices about their health after intervention [42,51]. However, successful education delivery also improves patient/caregiver self-efficacy, decreases distress through preparation for the transplantation process, and increases patient satisfaction [3,26,35,44,47,52]. More long-term education outcomes could include changes in survival and transplant morbidity, health care utilization, patient/caregiver ability to return to work, and quality of life [24,53,54], but none of these has formally been incorporated in education studies to date.

KNOWLEDGE GAPS AND PRIORITY RESEARCH QUESTIONS

There is a general paucity of available evidence in the current literature regarding patient/caregiver education in the context of BMT. Gaps in knowledge include a lack of differentiation between patient- and caregiver-specific educational needs, optimal time points and methods of education, and the impact of the different components of educational interventions. These topics represent high-yield areas for the development of educational strategies and future research considerations (Table 1). Based on the literature reviewed, our conceptual framework (Figure 1) and group discussions (selected individual comments from patients/caregivers are summarized in Table 2), 9 priority research questions were developed according to the PICO format [55]: (P) population or sample participants, (I) intervention of interest, (C) comparator or control group and (O) outcomes or results to measure the effectiveness of intervention (Table 3).

Three common themes emerge from these questions. First, we recognize the importance of involving all potential actors in the population of interest: not only the patient, but also his or her caregivers and family. Second, whichever the intervention planned, it is likely that implementation will be challenging, given the numerous factors likely to play direct or indirect roles in the educational experience of the patient/caregiver. Therefore, future protocols may need to incorporate a specific intervention tailored to the real-world setting, acknowledging the presence of multiple confounding variables. Third, during Working Group discussions, it was noted that the outcomes most important to healthcare providers were improvements in patient/caregiver knowledge, whereas patients/caregivers favored improvement in self-efficacy and decreased distress. Thus, we decided to include both as outcomes of interest.

Our multidisciplinary experience in this PCOR project has shown the importance of patient and caregiver input in research initiatives. Patients and caregivers were invariably present at all steps of the project and intensively participated in group discussions, steering decisions and putting the results of published studies into perspective. Consequently, as the top priority question, we propose the addition of a standardized self-directed education program (supported by patient advocacy groups) to the standard education package of the BMT center. We believe that this would have the advantage of tying in principles of autodetermination and self-management to provide a more personal approach to the education program,

Table 1
Recommendations and Research Considerations

Conceptual Model Domain	Available Evidence	Educational Strategies and Research Considerations
Baseline assessment	The ability to learn is influenced by cultural backgrounds, motivation and barriers to learning, as well as preferential learning styles [8].	Develop algorithms to tailor education to serve individual patient and caregiver needs [45-47].
	Patients and caregivers appreciate personalized education [20].	Assess the general well-being/health of the patient at the time of education (eg, fatigue, coping, distress, readiness to learn) [8,9].
	Health literacy is a positive predictor of outcomes in renal transplantation [9].	
	Patient/caregiver engagement in education can greatly vary [14,23].	
Educator	eHealth methods have been shown to be noninferior to conventional education in the setting of lung transplantation [18].	Consider the use of eHealth-based technologies as an education medium [16,18,19,20].
	In BMT patients, several virtual interfaces are well accepted by patients [16,18-20].	Compare satisfaction with education with in person versus remote delivery [49] or eHealth methods [16,18-20].
	eHealth methods can be more cost-effective than hiring extra personnel to deliver information for larger groups of patients [18].	
Education Material	BMT patients tend to overestimate the benefits of transplantation [23].	Develop standards for patient and caregiver education curriculums [19,24-26,48].
	Many transplant topics are overlooked during educations including sexual health, fertility, financial toxicities, advance care planning, and palliative care [24,28-32,48].	Train providers to include difficult “alternative” topics in education curricula, including sexual health issues, advance care planning, and the role of palliative care.
Education delivery method	Patients and caregivers report the highest demand for BMT education at or near hospital discharge from their transplant. [35,36,47].	Spread information during transplantation hospitalization to decrease anxiety and demand at time of discharge [34] and plan education moments after discharge to help implement discharge recommendations to the reality of the home setting [47].
	Patients and caregivers require repeated sessions of education for satisfactory retention of information [40], and BMT patients continue to seek information long after transplantation [37].	Consider eHealth to facilitate demands for repeated delivery while respecting the patient/caregiver's own agenda.
	Teaching patients problem-solving skills is more efficacious than traditional teaching methods [8,35,44].	Train providers to teach problem solving and self-efficacy skills to patients and caregivers [8].

Publications reporting evidence and results specific to BMT patients are the following: 16, 19, 20, 23-32, 35, 45, 47 and 48.

Table 2
Selected Individual Patient/Caregiver Comments During Group Discussions

Patients
“Get all the info you can, as soon as you can because [after transplantation] there is no part of your life goes unchanged.”
[Talking about quality of life questionnaires and numerical scales] “I'm done comparing my life now to my life before transplantation, and I'm changing some two's into eight's.”
“There's a lot of information and resources available, but it's all about the access and timing.”
“Up to the famous day 100, I felt prepared.”
“It's hard to get back on your feet socially.”
“I didn't understand what they meant by ‘getting back to the new normal’; if you ask me, there is no such thing as ‘the new normal.’”
“Somewhere in the transition, you lose friends.”
“It is important to ask people what is most challenging for them.”
“Both at the time of transplantation and for a long time afterwards, I was surprised at how differently my caregiver and I heard the same information.”
Caregivers
“In every hospital, there should be a separate soundproof room with cushion for caregivers to throw against the walls to ventilate their frustration.”
“I would have loved to have had someone to talk to at the time.”
“We define ourselves by our responsibilities.”
“Someone once said to me, ‘Prepare for the worst and expect the best’.”

by using the first-hand experience of individuals having gone through the BMT process themselves. Such an initiative would obviously require careful planning and training of “peer coaches” to ensure that patients and caregivers have the necessary tools to assist others by sharing their experience while realizing that each individual story is different.

Limitations of this project are linked to the selection bias inherent to this type of initiative. Both healthcare professionals and patients/caregivers participating in this work had an intrinsic interest in these topics and tended to have similar perspectives on the importance of patient participation in research projects. Future efforts need to identify how to involve less outreaching and articulate patients/caregivers to allow for a better representation of the whole population of BMT recipients. This could possibly be addressed by designing surveys to reach out to a large number of patients through existing patient advocacy groups. It should also be noted that whereas participating healthcare professionals were at least partially compensated for their time investment in the project (considering that this type of scientific activity is expected from healthcare professionals working in an academic setting), patients and caregivers gave their input without any financial incentive or compensation. This indirectly selects highly motivated, altruistic individuals who are also healthy enough to have the energy to invest time in advancing the BMT field and have the socioeconomic latitude to do so. Therefore, we need to acknowledge that we probably still missing the voice of the most vulnerable patients/caregivers.

Finally, it is our duty as health care providers to equip our patients/caregivers with the tools they need to serve as real experts in research project collaborations, allowing them to critically assess projects using both their experience and their understanding of the general principles of clinical research. Such an initiative is sponsored by, for example, the European Patients' Academy (https://www.eupati.eu). Following such

Table 3
Potential Priority Research Questions Related to Education in the BMT Process for Patients and/or Caregivers and Families before, during, and after BMT

Research Question*	Population	Intervention	Comparator	Outcome	Conceptual Model Domain	Current Evidence and Potential Implementation Challenges
1	Patients/care-givers & families before, during, and after BMT	Offering provider-directed education together with self-directed education, using a specific peer-group assisted education program	Offering provider-directed education only	Self-efficacy, satisfaction, distress, and/or health knowledge retention	Educator	It might be challenging to identify a standardized peer-group assisted education program, but several US-based patient advocacy groups (eg, nbmtLINK.org; BMTinfonet.org) already offer one-on-one conversations with trained peer support volunteers.
2		Assessing how individual (demographics, socioeconomics, health literacy), clinical (type of transplant), and environmental (distance from transplantation center) factors impact outcome	Not applicable		Baseline subject characteristic	Some of the baseline assessment characteristics involve potentially sensitive information to be collected directly from the patient/caregivers/ families (eg, socioeconomic resources, literacy).
3		Assessing learner readiness (fatigue, stress, and coping) before starting a standard educational session	No prerequisite to start a standard educational session		Baseline subject characteristic	No standard tools/cutoff values are available to assess an ideal readiness to learn. Education sessions also need to be standardized to allow for comparison.
4		Using specifically trained health professionals to give education sessions	Education sessions given by untrained health care professionals		Educator	Health professionals are likely to have different baseline teaching abilities depending on their personality and professional experience. Furthermore, there is no standard validated training program, which can currently be offered to health professionals to improve their teaching skills, and it might be challenging to ask busy professionals to invest time in training programs.
5		Making use of an electronic interface to provide information in addition to the standard printed material	Standard printed material		Education material	It will be difficult to deal with the 'spill over' effect of patients/ caregivers actively looking for information through other channels than those offered by the transplant center.
6		Making use of a standardized education plan	No standardized education plan		Education material	Most transplantation programs probably have some basic education program for all transplantation recipients and their caregivers.
7		Increasing the frequency of education moments	Standard education moments		Delivery method	It is challenging to define 'standard education' and to avoid the confounding effect of patients actively seeking information from other sources (internet, books, peers).
8		Dedicating special education moments for caregivers	Standard patient-only education moments		Delivery method	It is not easy to formally segregate information moments for caregivers or patients, considering their close proximity during hospital stays.
9		Offering education in the home setting in addition to the hospital setting	Offering education in the hospital setting only		Delivery method	The home setting is likely to vary widely from subject to subject, introducing uncontrolled variation in the intervention group.

* Except for question 1, considered the priority research question, other questions are ordered based on their relation to the conceptual model presented in Figure 1, not according to priority.

training initiatives is likely to help patients/caregivers provide more meaningful feedback in discussions and overcome any reluctance to challenge healthcare professionals during discussions. However, this again requires an active time and financial investment by patients/caregivers, which should be recognized and supported by our scientific community.

SUMMARY

To our knowledge, this project is the first to engage BMT patients and caregivers in identifying knowledge gaps and research questions to address their own unique educational needs. The use of a conceptual framework for education such as the one proposed here can assist physicians and researchers in designing studies to address these issues in BMT patients/caregivers and evaluate the impact of patient/caregiver education on long term outcomes.

Our approach had the advantage of being broad scale thanks to the large number of volunteers who participated in this project. Because of the scope of the project and the limited availability of BMT-specific literature on the subject, it was impossible to perform a systematic literature review on the state of patient/caregiver education in BMT. However, this novel multidisciplinary approach broadened our perspective of optimal patient/caregiver education. Patients and caregivers shared personal educational experiences during the transplantation process and long-term follow-up. Some patients/caregivers admitted being intimidated at first, but all agreed that this was a unique opportunity to participate and share their thoughts. Their input was informative and enlightening and helped prioritize the research questions.

This project has opened our eyes not only to the available evidence, but also to how this evidence relates to the experience of those who need it most. We hope that this type of initiative will grow and open the way for more projects where the voice of patients and their caregivers can be heard by the whole healthcare community.

ACKNOWLEDGMENTS

We thank the PCORI Program Officer, Yasmeen Long, for her assistance throughout the project. We thank all the patients, their caregivers and family members who participated in this project, with a special thank you to Everett Lee and his family for their dedication to the completion of this project.

Financial disclosure: This project was supported in part by a Patient-Centered Outcomes Research Institute Eugene Washington Engagement Award (PCORI EAIN-2956).

Conflict of interest statement: There are no relevant conflicts of interest to report.

Authorship statement: L.J.B. designed the project and recruited the members of the project. All authors participated in the discussion, critically reviewed the literature and the manuscript. H.M.S. suggested the conceptual model and manuscript structure. H.M.S., L.F., L.J.B., and K.S.P. wrote the manuscript. All authors approved the final version of the manuscript before submission. H.M.S and L.F. contributed equally to this work and should be considered co-first authors.

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