

OPEN

Defining Future Research Priorities in Donation and Organ and Stem Cell Transplantation With Patients, Families, Caregivers, Healthcare Providers and Researchers Within the Canadian National Transplant Research Program

Fabián Ballesteros Gallego, MSc,^{1,2} Carolina Martin, MSc,¹ Julie Allard, MSc,^{1,2} Samantha J. Anthony, PhD,^{2,3} Vincent Dumez, MSc,^{4,5} David Hartell, MA,² Marie-Josée Hébert, MD,^{1,2,6} Greg Knoll, MD,^{2,7} Lori J. West, MD, PhD,^{2,8} Linda Wright, MHSc,^{2,9} and Marie-Chantal Fortin, MD, PhD^{1,2,6}

Background. Patients, families, and caregivers have a unique understanding of the diseases they live with and provide care for every day. Their experience and expertise are important and should be taken into consideration when determining research priorities. The aim of this study was to gather the perspectives of Canadian patients, families, caregivers, researchers, and healthcare professionals on what research priorities were important to them in the field of organ and hematopoietic cell transplantation (HCT) and donation within the Canadian National Transplant Research Program (CNTRP). **Methods.** The CNTRP developed a national consultation process, which included a Web-based survey and in-person workshop, to ascertain and validate the viewpoints of the Canadian donation and transplant community. The Web-based survey identified 3 principal research priorities (increasing donation, developing better antirejection drugs and developing tolerance), which were further refined and prioritized during the one-and-a-half day national workshop held in Toronto in November 2015. **Results.** A total of 505 participants answered the Web-based survey, and 46 participants (28 patients, 12 researchers and 6 healthcare professionals) participated in the in-person workshop. Workshop participants ranked the following 2 priorities as the most important in the fields of donation, HCT, and solid organ transplantation: methods for developing a culture of donation (within healthcare organizations and throughout society); and methods for improving graft survival and antirejection therapy. **Conclusion.** The CNTRP will use these results to prioritize future research projects and studies in donation, HCT, and solid organ transplantation in the years to come.

(*Transplantation Direct* 2018;4: e360; doi: 10.1097/TXD.0000000000000791. Published online 23 May, 2018.)

Received 2 November 2017. Revision requested 7 February 2018.

Accepted 3 March 2018.

¹ Centre de Recherche du Centre Hospitalier de l'Université de Montréal (CRCHUM), Montréal, Canada.

² Canadian National Transplant Research Program, Canada.

³ Transplant and Regenerative Medicine Centre, The Hospital for Sick Children, Toronto, Canada.

⁴ Direction Collaboration et Partenariat Patient, Faculty of Medicine, Université de Montréal, Montréal, Canada.

⁵ Centre of Excellence on Partnership With Patients and the Public, Université de Montréal, Montréal, Canada.

⁶ Faculty of Medicine, Université de Montréal, Canada.

⁷ Clinical Epidemiology Program, Ottawa Hospital Research Institute, Ottawa, Canada.

⁸ Faculty of Medicine, University of Alberta, Edmonton, Canada.

⁹ Department of Surgery and Joint Centre for Bioethics, University of Toronto, Toronto, Canada.

This research project was funded by a Planning and Disseminating Grant from the Canadian Institutes of Health Research (CIHR) and by the CIHR Institute of Infection and Immunity.

The authors declare no conflicts of interest.

F.B.G. participated in the research design, data analysis and writing of the article.

C.M. participated in the data analysis and writing of the article. J.A. participated in

the data analysis and review of the article. S.A. participated in the research design and review of the article. V.D. participated in the research design, research and review of the article. D.H. participated in the research design, research and writing of the article. M.-J.H. participated in the research design and review of the article. G.K. participated in the research design and review of the article. L.W. participated in the research design and review of the article. L.W. participated in the research design, research and review of the article. M.-C.F. designed the workshop and participated in the research, data analysis and writing of the article.

Correspondence: Marie-Chantal Fortin, MD, PhD, Room 12-418, 900 Saint-Denis St., Montréal, QC, Canada H2X 0A9. (marie-chantal.fortin@umontreal.ca)

Supplemental digital content (SDC) is available for this article. Direct URL citations appear in the printed text, and links to the digital files are provided in the HTML text of this article on the journal's Web site (www.transplantationdirect.com).

Copyright © 2018 The Author(s). *Transplantation Direct*. Published by Wolters Kluwer Health, Inc. This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal.

ISSN: 2373-8731

DOI: 10.1097/TXD.0000000000000791

Reports have shown that research priorities for patients and caregivers are not reflected in research published in medical journals.¹⁻³ One approach to address this problem is engaging patients and caregivers in setting research priorities. There are 3 principal arguments for involving patients in setting research priorities. First of all, patients have experiential knowledge gained from living with an illness and are able to provide valuable insight on which research topics are most relevant for them.⁴ Second, given that a large part of medical research is publicly funded, there is a democratic argument for involving patients in setting research priorities, because they are taxpayers.⁴ Lastly, the patients are the direct beneficiaries of the research outcomes, and research should thus strive to meet their important needs.⁴ Recently, various initiatives and institutes have focused on patient engagement in research, such as the Strategy for Patient-Oriented Research by the Canadian Institutes of Health Research,⁵ the Patient Centered-Outcomes Research Institute⁶ in the United States, and INVOLVE in the United Kingdom.⁷ Patients have also recently been involved in setting research priorities for chronic kidney disease,^{8,9} rheumatology,¹⁰ chronic obstructive pulmonary disease,¹¹ and breast cancer.¹²

In the field of solid organ transplantation (SOT), several priority-setting exercises have been published in recent years. Tong and colleagues¹³ published a systematic review reporting 28 studies with research priority setting in SOT. Among these studies, only 32% of them involved patients in the priority-setting process, and only in the fields of kidney and heart transplantation. In this systematic review, there is no report of a priority-setting activity involving patients, families, caregivers, researchers, and healthcare professionals (HCPs) in the combined fields of donation, SOT, and hematopoietic cell transplantation (HCT). In the fields of HCT and organ and tissue donation, there have been no reported priority-setting activities involving patients and caregivers.

The Canadian National Transplant Research Program (CNTRP) is a national research initiative designed to increase organ and tissue donation in Canada, improve graft survival, and enhance the quality of life of Canadians living with a transplant.^{14,15} The program brings together over 150 funded and active researchers, patients, and trainees in the field of donation and SOT and HCT. The CNTRP aims to increase patient engagement in research and to support novel ways to integrate patients and families as active participants across the research network. For the CNTRP, patient engagement occurs when “patients meaningfully and actively collaborate in the governance, priority setting, and conduct of research, as well as in summarizing, distributing, sharing, and applying its resulting knowledge.”⁵ To achieve this goal of patient engagement across the CNTRP, the latter conducted a French-language pilot workshop with 10 patients and 5 researchers in August 2014, in Montréal.² The objective of the current study was to refine the preliminary research priorities identified during the pilot workshop, elicit new priorities from a broader national cohort of patients, and rank them by order of priority. This was achieved through the development and launch of a national priority-setting survey followed by a workshop with patients, caregivers, HCPs, and researchers to evaluate and rank the research priorities. The final list of research priorities will be used by the CNTRP to determine future research projects/studies.

MATERIALS AND METHODS

The survey and national-workshop participants were asked to use their experiences as patients, family members, caregivers, researchers, and/or HCPs to help identify the top research priorities in transplantation and donation within the CNTRP. The ethics review board of the Centre hospitalier de l'Université de Montréal approved the survey and the workshop, and all participants provided informed consent before answering the survey and participating in the workshop.

Survey

A bilingual (French and English) online survey was conducted with patients (SOT and HCT recipients, transplant candidates and living donors), caregivers, deceased donors' family members, transplant professionals, and researchers in Canada. Findings from the pilot workshop² were used as a guide to develop the survey questions. The team that developed the survey included several researchers in the fields of clinical transplantation, ethics, and social work, as well as 2 patients. The survey questions addressed challenges surrounding various aspects of transplantation, grouped into the following 6 themes: (i) pretransplant, (ii) posttransplant, (iii) biomedical, (iv) psychological and social, (v) ethical and legal, and (vi) healthcare organization-related (refer to **SDC, Appendix 1** <http://links.lww.com/TXD/A101> for survey questions.) The team decided to group the questions into these 6 themes to elicit research priorities from every step of the patients' journey.

The survey included 9 multiple-choice questions, as well as 1 open-ended question allowing respondents to add research themes or priorities. These additional research themes or priorities were read and coded by 2 independent coders from the research group (F.B. and C.M.) and were reclassified into an existing theme where appropriate. To answer the multiple-choice questions, participants were asked to choose the top 3 issues that they felt should be addressed in research within each theme. At the end of the survey, the respondents ranked 14 research priorities from most to least important. This final list of 14 priorities included the 10 original priorities identified during the pilot workshop and 4 additional priorities judged important by the CNTRP leadership (ie, pretransplant organ repair before transplant, improved access to transplant programs and/or transplant waiting list, complications after transplantation, and treatment adherence). The survey questions were pretested, in French and in English, with 15 laypeople, clinicians, and transplant patients. The survey was administered and hosted by the Population Research Laboratory at the University of Alberta (<http://www.prl.ualberta.ca/>).

Recruitment was primarily done through professional and patient associations that had agreed to distribute and promote the survey invitation to their members. These groups included (i) professional organizations, such as the Canadian Society of Transplantation and the Canadian Blood and Marrow Transplant Group; (ii) health charities, such as the Kidney Foundation of Canada and the Canadian Liver Foundation; (iii) patient advocacy groups, such as the Canadian Transplant Association (CTA); (iv) organ donation organizations, such as Transplant Québec, Trillium Gift of Life and Canadian Blood Services; and (v) members of the CNTRP research network. Facebook and Twitter were also used to invite people to take part in this survey, and the survey was

promoted during the launch of the 2016 Transplant Games in Toronto in April 2015. The CTA was used to reach patients and caregivers, since it is the largest national organization that brings together transplant recipients and their family members, transplant candidates, donor families, living donors, and health professionals.

Because the invitations to participate were sent through the various organizations and through social media, it was impossible to accurately estimate the number of invitations sent and to follow up with people who did not answer the survey. Moreover, some participants may have received multiple invitations if they belonged to multiple professional associations. The survey was launched in spring 2015 and was available from April 22 to July 22, 2015. Three follow-up reminders were sent to Canadian Society of Transplantation, CTA, and CNTRP members.

Based on respondents' survey answers, descriptive statistical analysis was performed to examine the prevalence of the respondents' choices. Pearson χ^2 tests were performed to determine the differences in proportion between each group. For the rankings of priorities, a *t* test was performed to compare the means of ranking of each research theme between each group. Statistical analysis was performed using R statistical software.¹⁶ A *P* value less than 0.05 was considered statistically significant.

National Workshop

A facilitated in-person workshop was held in Toronto on November 25 and 26, 2015. To recruit participants, a purposive method (for researchers and HCPs) and a snowball and purposive sample method (for patients and caregivers) were used.¹⁷ To recruit researchers and HCPs, the research team therefore selected individuals who they believed would provide and generate valuable information. For patients, we used a purposive and a snowball strategy that involved inviting patients to suggest other participants.^{18,19} In both cases, we recruited participants from different age groups and regions of Canada, and with different experiences, areas of expertise, and so on.

A total of 75 participants from across Canada were invited, including 36 patients (15 kidney transplant recipients, 6 HCT recipients, 4 waitlisted patients, 4 lung transplant recipients, 3 heart transplant recipients, 3 liver transplant recipients, and 1 multiorgan transplant recipient), 9 caregivers, and 30 researchers and HCPs. Of this total, 46 individuals agreed to participate. Refusals to participate were all due to unavailability at the workshop date. To participate, participants needed to be fluent in English and have experience with and insight into organ and stem cell donation or transplantation. Participants included: (i) *patients* reflecting a broad range of transplant experiences (waitlisted patients and transplant recipients); (ii) *caregivers*, including family members of deceased organ donors and transplant recipients, as well as living donors; and (iii) *researchers and HCPs* involved in organ and stem cell donation or transplantation, including CNTRP researchers involved in various areas of scientific enquiry and with different areas of research expertise (clinical, biomedical, health services, and population health), physicians, surgeons, psychologists, nurses, social workers, dietitians, pharmacists, and physiotherapists. The participants' travel and accommodation expenses were all reimbursed with funding obtained through a CIHR Planning and

Dissemination Grant and funds allocated by the CIHR Institute of Infection and Immunity.

The workshop started in the afternoon of November 25 with a series of introductions and background lectures informing participants of the structure and function of the CNTRP, the basics of patient engagement, and the results of both the pilot workshop and national survey. A dinner was organized that evening, and seating was arranged to encourage networking and informal patient and researcher interactions.

On the second day (November 26), participants were divided into 6 small groups of 8 to 9 people. Each group consisted of 2 researchers, 1 HCP, and 4 to 5 patients and/or caregivers. The first question used to initiate the small group discussions was: "According to your experience, what research priorities should be developed within the CNTRP?" One patient or caregiver was assigned to each group to facilitate these discussions, each of these facilitators having received training before attending the workshop. This training consisted of a 1-hour preworkshop session provided by members of the research team (M.C.F. and V.D.). The facilitators also received a guide detailing the instructions for managing the discussions. The team chose to have patients facilitate the groups to make it easier for patient participants, who might have been too intimidated to contribute or speak openly with a researcher or HCP as a facilitator. To incite and facilitate the discussion, a list of the top 28 research priorities was provided to all participants (SDC, Appendix 2 <http://links.lww.com/TXD/A102>). This list included the 10 priorities identified during the pilot workshop and each of the top 3 issues identified for each survey theme (18 priorities in total). Participants were informed that this list was a starting point and were invited to suggest new priorities.

After the small group discussions, the participants came back together for a plenary session, and each group presented its top 3 research priorities identified. Vincent Dumez, a patient and expert in patient partnership and member of the research team, facilitated the plenary. During this session, participants decided to reclassify the research priorities into 2 categories to help facilitate the priority-setting exercise: 1 category for transplantation and 1 for organ/stem cell donation. Afterward, participants voted anonymously on their top 3 priorities for each category, on individual sheets which were collected by the research team. The audio for the small group and plenary discussions were digitally recorded and transcribed for analysis of the discussion. The transcripts were each read by 2 people to ensure that no research priorities were missing from the final list. Transcripts were analyzed using an inductive content and thematic analysis.¹⁸ NVivo version 11 (QSR International) computer software was used for the analysis.

RESULTS

Survey

Demographics and Characteristic of Survey Respondents

In total, 663 unique responses were received. Among the responses received, 505 respondents identified themselves as belonging to 1 or more of the categories indicated (researcher, HCP, patient, caregiver) and were considered for analysis (76.2% completeness rate). The characteristics of the survey respondents are provided in Table 1. Because the

TABLE 1.**Survey respondent characteristics (n = 505)**

Characteristics	Patients, n = 234 (%) ^a	Caregivers, n = 145 (%) ^a	Researchers and HCPs, n = 210 (%) ^a
Transplant recipient	212 (90.9)		
Kidney	78 (36.8)		
Liver	48 (22.6)		
Heart	42 (19.8)		
Lung	41 (19.3)		
Other (pancreas, multiorgan, stem cell)	19 (9.0)		
Patient on a waiting list	33 (14.1)		
Family member or caregiver		139 (95.9)	
Living donor (kidney, liver, stem cell)		19 (13.1)	
HCP			182 (86.7)
Transplant physician			61 (33.5)
Nurse			37 (20.3)
Transplant coordinator			23 (12.6)
Transplant surgeon			8 (4.4)
Other			72 (39.6)
Researcher in the field of transplantation			91 (43.3)
Age ^b			
<30	13 (5.7)	15 (10.6)	17 (8.3)
30-39	23 (10.0)	21 (14.8)	50 (24.4)
40-49	46 (20.0)	27 (19.0)	58 (28.3)
50-59	74 (32.2)	43 (30.3)	57 (27.8)
>60	74 (32.2)	35 (24.6)	23 (11.2)
Sex ^c			
Female	127 (55.2)	102 (72.3)	128 (62.4)
Male	103 (44.8)	39 (27.7)	77 (37.6)
Primary language ^d			
English	182 (78.8)	118 (87.7)	151 (73.7)
French	43 (18.6)	21 (14.9)	52 (25.4)
Ethnic group ^e			
White	203 (91.9)	124 (89.9)	169 (82.8)
Location of residence in Canada ^f			
Ontario	102 (44.5)	73 (52.5)	60 (30.0)
Prairies	52 (22.7)	31 (22.3)	36 (18.0)
Quebec	43 (18.8)	18 (12.9)	59 (29.5)
Atlantic	18 (7.9)	11 (7.9)	37 (18.5)
British Columbia	14 (6.1)	6 (4.3)	8 (4.0)

^a Because respondents could self-identify in more than 1 category and demographic questions were not mandatory, the totals in each category do not necessarily correspond to their respective denominators.

^b n (patients) = 230, n (caregivers) = 142, n (researchers and HCPs) = 205.

^c n (patients) = 230, n (caregivers) = 141, n (researchers and HCPs) = 205.

^d n (patients) = 231, n (caregivers) = 141, n (researchers and HCPs) = 205.

^e n (patients) = 221, n (caregivers) = 138, n (researchers and HCPs) = 204.

^f n (patients) = 229, n (family and caregivers) = 139, n (researchers and HCPs) = 200.

participants could self-identify as more than 1 category (as a researcher and patient or as a caregiver and HCP, for instance), the total number of participants across all categories exceeds 505. Of the participants, 46.3% were patients, 28.7% were caregivers, and 41.6% were researchers/HCPs. Of the researchers and HCPs, 51.7% had conducted clinical research in the field of organ and stem cell donation and transplantation, and 59.0% of patients and caregivers had been participants in clinical research.

Research Priorities According to the Survey

Table 2 summarizes the 3 most important research priorities identified for each question. For the priorities related to pretransplantation, the 3 groups of respondents were unanimous on the most important priority. In the pretransplant

period, the most important priorities were “increasing the number of organs available for transplantation” (51.1% total, 53.2% patients, 54.9% caregivers, 46.7% researchers/HCPs, $P = \text{NS}$), “the prevention of disease leading to the need for transplantation” (29.6% total, 31.8% patients, 28.9% caregivers and 26.2% researchers/HCPs, $P = \text{NS}$), and “developing new ways to match donors/recipients for improving transplant outcomes” (28.0% total, 25.8% patients, 27.1% caregivers and 29.5% researchers/HCPs, $P = \text{NS}$).

In the posttransplant period, patients and caregivers were in agreement on research priorities, whereas there were differences between researchers/HCPs and patients and caregivers. The most important priority for the 3 groups of participants was “long-term medical complications of transplantation” (53.7% total, 58.6% patients, 48.6% caregivers,

TABLE 2.
Top 3 research priorities by survey category

	All	Patients	Caregivers	Researchers and HCP	Patients vs caregivers	Patients vs researcher and HCP	Caregivers vs researcher and HCP
(1) Pretransplant period	n = 503 (%)	n = 233 (%)	n = 144 (%)	n = 210 (%)	<i>P</i>	<i>P</i>	<i>P</i>
Increasing the number of organs available for transplantation.	257 (51.1)	124 (53.2)	79 (54.9)	98 (46.7)	NS	NS	NS
The prevention of disease leading to the need for transplantation	149 (29.6)	74 (31.8)	41 (28.9)	55 (26.2)	NS	NS	NS
Developing new ways to match donors/recipients for improving transplant outcomes	141 (28.0)	60 (25.8)	39 (27.1)	62 (29.5)	NS	NS	NS
(2) Posttransplant period	n = 501 (%)	n = 232 (%)	n = 142 (%)	n = 210 (%)			
Long-term medical complications of transplantation (eg, diabetes, cancer, infections, cardiac, and vascular disease)	269 (53.7)	136 (58.6)	69 (48.6)	102 (48.6)	NS	0.049	NS
Improved graft survival (having the transplant last longer)	198 (39.5)	81 (34.9)	49 (34.5)	97 (46.2)	NS	0.019	0.030
How to decrease the number of drugs necessary to prevent rejection	175 (34.9)	90 (38.8)	58 (40.8)	57 (27.1)	NS	0.013	0.013
(3) Biological research	n = 500 (%)	n = 232 (%)	n = 144 (%)	n = 208 (%)			
How can we safely decrease or eliminate the need for antirejection drugs?	268 (53.6)	142 (61.2)	83 (57.6)	95 (45.7)	NS	0.001	0.029
How can cancer risk after transplant be minimized?	179 (35.8)	98 (42.2)	54 (37.5)	61 (29.3)	NS	0.006	NS
What blood or urine tests (biomarkers) will indicate or predict the risks of rejection and complications after transplant?	178 (35.6)	78 (33.6)	49 (34.0)	81 (38.9)	NS	NS	NS
(4) Psychosocial issues	n = 500 (%)	n = 232 (%)	n = 143 (%)	n = 209 (%)			
Managing psychological complications such as depression or anxiety	338 (67.6)	140 (60.3)	102 (71.3)	146 (69.9)	0.045	0.048	NS
Psychological side effects of antirejection drugs	241 (48.2)	142 (61.2)	76 (53.1)	70 (33.5)	NS	<0.001	<0.001
Exercise and rehabilitation	241 (48.2)	113 (48.7)	66 (46.5)	95 (47.7)	NS	NS	NS
(5) Ethical and legal issues	n = 498 (%)	n = 232 (%)	n = 142 (%)	n = 207 (%)			
Presumed consent to donation	295 (59.2)	142 (61.2)	92 (64.8)	110 (53.1)	NS	NS	0.041
Family veto	225 (45.2)	122 (52.6)	65 (45.8)	78 (37.7)	NS	0.002	NS
Removing financial barriers or disincentives to donation	213 (42.8)	104 (44.8)	64 (45.1)	79 (38.2)	NS	NS	NS
(6) Healthcare organization	n = 491 (%)	n = 229 (%)	n = 142 (%)	n = 204 (%)			
Implementation of new organ donation programs	255 (51.9)	118 (51.5)	82 (57.7)	91 (44.6)	NS	NS	0.016
Posttransplant follow-up	249 (50.7)	122 (53.3)	72 (50.7)	95 (46.6)	NS	NS	NS
Telemedicine	199 (40.5)	93 (40.6)	61 (43.0)	87 (42.6)	NS	NS	NS

NS, not significant.

48.6% researchers/HCPs, $P < 0.05$ patients vs researchers/HCPs). “Improved graft survival” was more important for researchers/HCPs than for patients and caregivers (39.5% total, 23.9% patients, 34.5% caregivers, 46.2% researchers/HCPs, $P < 0.05$ patients vs researchers/HCPs and caregivers vs researchers/HCPs), whereas “how to decrease the number of drugs necessary to prevent rejection” was more important for patients and caregivers than for researchers/HCPs (34.9% total, 38.8% patients, 40.8% caregivers, 27.1% researchers/HCPs, $P < 0.05$ patients vs researchers/HCPs and caregivers vs researchers/HCPs).

In the biological field, the most important topic was “how to safely decrease or eliminate the need for antirejection drugs” (53.6% total, 61.2% patients, 57.6% caregivers, 45.7% researchers/HCPs). This research priority, however, was ranked significantly higher by patients and caregivers than by HCPs and researchers ($P < 0.05$ patients vs researchers/HCPs and caregivers vs researchers/HCPs). The second most important priority for patients was “how

cancer risk after transplant [can] be minimized” (35.8% total, 42.2% patients, 37.5% caregivers, 29.3% researchers/HCPs, $P < 0.05$ patients vs researchers/HCPs). All were in agreement about the third priority, “what biomarkers will indicate or predict the risks of rejection and complications after transplant,” (35.6% total, 33.6% patients, 34.0% caregivers and 38.9% researchers/HCPs, $P = NS$).

There was some disagreement about the most important priority in research related to psychosocial issues. For all participants, the most important priority in psychosocial research was “managing psychological complications (depression and anxiety)” (67.6% total, 60.3% patients, 71.3% caregivers, 69.9% researchers/HCPs), but more caregivers and researchers/HCPs than patients selected this issue ($P < 0.05$ for patients vs caregivers and patients vs researchers/HCPs). “Psychological side effects of antirejection drugs” and “exercise and rehabilitation” were chosen as important priorities for 48.2% of participants. However, “psychological side effects of antirejection drugs” was more important

for patients and caregivers than for researchers/HCPs ($P < 0.001$ patients vs researchers/HCPs and caregivers vs researchers/HCPs).

In the research on legal and ethical issues, participants identified “presumed consent to donation” as the most important priority (59.2% total, 61.2% patients, 64.8% caregivers, 53.1% researchers/HCPs, $P < 0.05$ caregivers vs researchers/HCPs). The other top priorities identified were “family veto” (45.2% total, 52.6% patients, 45.8% caregivers and 37.7% researchers/HCPs, $P < 0.05$ patients vs researchers/HCPs) and “removing financial barriers or disincentives to donation” (42.8% total, 44.8% patients, 45.1% caregivers, and 38.2% researchers/HCPs, $P = NS$).

In the field of healthcare organization research, the 3 leading priorities were “the implementation of new organ donation programs” (51.9% total, 51.5% patients, 57.7% caregivers and 44.6% researchers/HCPs, $P < 0.05$ caregivers vs researchers and HCPs), “posttransplant follow-up” (50.7% total, 53.3% patients, 50.7% caregivers and 46.6% researchers/HCPs, $P = NS$) and “telemedicine” (40.5% total, 40.6% patients, 43.0% caregivers and 42.6% researchers/HCPs, $P = NS$).

When asked to rank a list of 14 priorities (1 being most important and 14 being least important), all 3 categories of participants identified “increasing organ donation” as the most important priority. The second priority, “the development of better antirejection drugs,” was ranked higher by patients and caregivers than by researchers/HCPs ($P < 0.001$ patients and caregivers vs researchers/HCPs). Lastly, the third priority, “the induction of tolerance,” was ranked higher by patients than by researchers and HCPs ($P < 0.05$ patients vs researchers/HCPs). Table 3 summarizes the rankings.

NATIONAL WORKSHOP

Demographics and Characteristics of Workshop Participants

Forty-six participants from 4 Canadian provinces agreed to participate in the workshop. One patient left before the

end of the workshop due to medical problems. Among the remaining 45 participants, 18 (40.0%) were patients, 10 (22.2%) were family members and caregivers, 11 (24.4%) were researchers, and 6 (13.3%) were HCPs. The characteristics of the participants are summarized in Table 4.

Research Priorities Identified During the Workshop

After the small group and plenary discussions, participants voted on a list of priorities related to both organ/hematopoietic cell donation and transplantation. A final list was generated using all the research priorities identified in each small group. In the posttransplant period, participants identified the following as the 5 most important research questions: (i) how to improve graft survival, (ii) how to achieve personalized use of antirejection medication through the development of immune monitoring, (iii) how to reduce the adverse effects of antirejection medication and long-term complications, (iv) how to improve psychological support and the management of psychological complications, and (v) how to improve quality of life. Table 5 presents the questions and the votes for each priority, along with excerpts from the discussions.

For organ donation, participants identified the following as the 5 most important research questions: (i) how to educate the public and raise awareness about organ donation, (ii) how to improve donor management and graft quality, (iii) how to foster a culture of donation in hospitals and in society at large, (iv) how to support living donors and families (long-term impact and psychological care), and (v) how to increase the number of deceased and living donors. Table 6 presents the questions and the votes for each priority, along with excerpts from the discussions.

DISCUSSION

The most important research priorities identified through the survey remained consistent with those identified during the workshop. Using the survey, participants ranked increasing organ donation, developing better immunosuppressive drugs (with fewer adverse effects), developing tolerance,

TABLE 3.
Research priority rankings

Research theme	All, n = 505	Patients, n = 234	Caregivers, n = 145	Researchers and HCP, n = 210	Patients vs caregivers	Patients vs researcher and HCP	Caregivers vs researcher and HCP
	Mean ^a ± SD	Mean ± SD	Mean ± SD	Mean ± SD	P	P	P
(1) Increasing organ donation rates.	3.7 ± 4.0	3.4 ± 3.9	3.3 ± 4.0	4.3 ± 4.1	NS	NS	NS
(2) Better antirejection drugs	4.2 ± 3.6	3.7 ± 3.6	3.6 ± 3.5	5.0 ± 3.6	NS	<0.001	<0.001
(3) Tolerance to organ grafts	4.6 ± 3.8	4.3 ± 3.8	4.4 ± 4.0	5.1 ± 3.7	NS	0.028	NS
(4) Improving graft survival	5.0 ± 4.0	4.9 ± 3.9	4.8 ± 3.9	5.1 ± 4.0	NS	NS	NS
(5) Complications after transplant	5.1 ± 3.7	4.9 ± 3.8	4.8 ± 3.7	5.6 ± 3.6	NS	NS	NS
(6) Organ repair before transplant	5.2 ± 3.7	5.0 ± 3.8	5.1 ± 3.9	5.4 ± 3.8	NS	NS	NS
(7) Improving access to organ transplantation	5.2 ± 4.0	4.9 ± 4.0	4.2 ± 3.9	5.9 ± 3.8	NS	0.008	<0.001
(8) Alternatives to transplantation	5.4 ± 3.9	4.9 ± 3.8	5.3 ± 4.1	6.0 ± 4.0	NS	0.005	NS
(9) Patient-physician communication	6.0 ± 4.4	5.4 ± 4.5	4.8 ± 4.2	7.1 ± 4.3	NS	<0.001	<0.001
(10) Lifestyle (exercise, diet)	6.1 ± 4.0	5.9 ± 4.1	5.5 ± 3.8	6.5 ± 3.9	NS	NS	0.022
(11) Treatment adherence	6.1 ± 4.1	6.5 ± 4.4	5.6 ± 4.0	5.9 ± 4.0	NS	NS	NS
(12) Psychological follow-up	6.2 ± 3.8	6.1 ± 3.9	5.4 ± 3.8	6.6 ± 3.6	NS	NS	0.007
(13) Alternative medicine	7.9 ± 4.8	7.1 ± 4.8	6.9 ± 4.7	9.2 ± 4.6	NS	<0.001	<0.001
(14) Safe pregnancy after transplantation	8.2 ± 4.1	8.1 ± 4.3	7.5 ± 4.1	8.4 ± 3.9	NS	NS	NS

^a Average of ranking allocated to each priority. 1 is the most important and 14 is the least.

TABLE 4.
Workshop participant characteristics

Characteristics	N = 45 (%)
Patients	17 (40.0)
(1) Transplant recipient	15
•Kidney	4
•Stem cell	3
•Heart	2
•Liver	2
•Lung	2
•Multiorgan	2
(2) Transplant candidate	2
Caregiver, family member of deceased organ donor and/or living donor	10 (22.2)
Researcher and/or HCP	17 (37.8)
(1) Researcher	11
•Kidney transplantation	4
•Biomedical science	3
•Heart transplantation	2
•Multiorgan transplantation	1
•Stem cells transplantation	1
•End-of-life/donation	1
•Ethical, economic, legal, and social	1
(2) HCP	6
•Physician (nephrologists)	2
•Nurse (multiorgan unit)	1
•Transplant coordinator	1
•Psychologist	1
•Organ procurement organization member	1
Sex	
Male/female	18 (40.0)/27 (60.0)
(1) Patients (18)	8/10
(2) Caregivers (9)	1/8
(3) Researchers and HCPs (18)	9/9
Province of residence	
•Ontario	22 (48.8)
•British Columbia	12 (26.6)
•Quebec	6 (13.3)
•Alberta	5 (11.1)

and improving graft survival as the most important research priorities. During the workshop, participants ranked organ donation focused on increasing the number of organs available and improving the quality of organs and grafts for transplantation as the most important research priorities. With respect to transplantation, the most important priorities were the improvement of graft survival, personalized use of antirejection drugs, and the reduction of adverse effects and long-term complications. Of course, the research priorities identified, such as increasing organ donation, are somewhat vague. It will be up to the research teams and patient research partners to develop research projects that might provide answers to the questions identified.

In contrast to the first workshop, the national workshop participants elicited priorities that had not been mentioned or prioritized during either the survey or the pilot workshop, such as the development of strategies to improve communication between medical teams and patients and families, how to address cultural issues in organ donation and transplantation, how to incorporate alternative medicine, and how to make

pregnancy safe and possible after transplantation.² One possible explanation for this discrepancy could be that the national workshop was attended by researchers and HCPs, who might have involuntarily framed the research priorities in the small group discussions. Indeed, during the pilot workshop, patients and caregivers had time to express themselves freely before researchers and HCPs were included in the discussion.

The results of the survey show that, most of the time, patients and caregivers agree. However, there are some disagreements between patients and researchers/HCPs, and between caregivers and researchers/HCPs. It is unsurprising that patients and caregivers share the same research priorities and that they could disagree with researchers and HCPs given that they do not have the same experience of transplantation and donation. For instance, patients and caregivers prioritized reducing long-term complications after transplantation and decreasing the number of drugs necessary to prevent rejection over improving graft survival. That being said, for 8 of the 18 priorities in the 6 themes, and 6 of 14 priorities to rank, there were no significant differences between the 3 groups of participants.

Previous studies have described priority-setting exercises in the field of SOT^{8,9,13,20,21}; however, this is the first study to present a research priority-setting study that combines the fields of donation, SOT and HCT (patients, caregivers, researchers, and HCPs). Bringing these 3 communities together for this exercise reflected the scope and mandate of the CNTRP, the only national program in the world that unites these 3 clinical research areas together with patients.¹⁵ Combining these patients in this research priority-setting study also makes sense from a disease management perspective, because HCT and SOT patients share some similarities, such as life-threatening conditions, immunosuppressive drugs, psychosocial challenges, rejection (or graft-versus-host disease), and infectious, cardiovascular, and neoplastic complications.

Tong and colleagues¹³ recently conducted a systematic review of 28 priority-setting exercises in SOT. Only 9 of these studies included patients. Research priorities identified in that review were related to organ donation, waitlisting and allocation, histocompatibility and immunology, immunosuppression, complications (graft- and recipient-related), re-production, psychosocial and lifestyle issues, and disparities in access and outcomes.¹³ These priorities bear some similarities to those identified during this priority-setting exercise.

Recently, a British team reported the results of a patient partnership priority-setting exercise in kidney transplantation.²¹ That team used a survey with open-ended questions, with 183 survey respondents, and a consensus workshop to identify the top 10 priorities in kidney transplantation. The 3 most important research priorities identified during the survey were: (i) the best treatment for improving graft survival, (ii) the development of reliable tools to assess the suitability of organs for transplantation and predict outcomes, and (iii) the development of treatment for vascular rejection. During the workshop, participants identified the top 10 research priorities, all of which were biomedical, including the treatment of vascular rejection, the tailoring of immunosuppression for an individual patient, the prevention of sensitization after the failure of a first transplantation, the development of tolerance and the best combination of

TABLE 5.**Posttransplant care research priorities**

Research priorities and discussion excerpts	N = 43 (%)
How to improve graft survival "Make transplants last longer."	25 (58.1)
How to achieve personalized use of antirejection medication (immune monitoring, decreased use of medication) "Consider side effects for new medication or reduce medication."	20 (46.5)
How to reduce adverse effects and long-term medical complications "How can long-term medical complications of transplantation (eg, diabetes, cancer, infections, cardiac and vascular disease) be minimized?"	19 (44.2)
How to improve psychological support and management of psychological complications "Support posttransplantation and psychosocial support for families and patients." "There is [a] need for more education and support at work, at school and [in] everyday life."	13 (30.2)
How to improve quality of life "How can long-term medical complications of transplantation be minimized to improve quality of life?"	12 (27.9)
How to develop early markers of rejection "How to develop tools, immune monitoring to reduce rejection safely?"	10 (23.3)
How to improve management of pediatric patients and conduct research on pediatric issues "How to improve healthcare support for patients from pediatrics to adult services [...] is a 'tremendous transition.'"	8 (18.6)
How to improve the management of chronic complications "How to improve rehabilitation/chronic management (healthcare organizations, psychosocial, communication between HCPs, etc). Partnership and models of care."	7 (16.2)
How to deal with the feeling of survivorship and family burden "How to help with the difficult transition to home and posttransplant care plan." "What would be the best strategy to fund research and support families of transplant patients?"	6 (14.0)
How to improve social, cultural, and economic support "How to address cultural issues in organ/stem cell donation and transplantation." "How to improve [the] psychosocial experience for donors, families of donors, caregivers and recipients, including cultural issues."	5 (11.6)

TABLE 6.**Organ donation research priorities**

Research priorities	N = 43 (%)
How to educate the public and raise awareness about organ donation "How do we normalize donation (solid organs, tissue and stem cells), including in end-of-life discussions, with the ultimate goal of increasing donors? How [can we] improve [the] identification of donors? Organ donation awareness?"	19 (44.2)
How to improve donor management and graft quality "What are the main means to use the organs we are not using today? Best way[s] to increase use of organs that are not being used." "Medical intervention to use organs not used before" "Optimizing organs useful for transplantation" "Use of new technologies to allow the use of organs [that] have not been used so far."	19 (44.2)
How to foster a culture of donation in hospitals and in society at large "How to improve organ donation awareness... everybody can become a potential donor."	18 (41.9)
How to support living donors and families; understand long term impact and psychological care "How can we [more effectively] support donors, families and recipients in a transplant process? Psychosocial, economic, medical, etc." "How to improve follow up [for] living donors and long-term medical outcomes with higher-risk donors."	14 (32.6)
How to increase the number of donors (deceased and living) "Increasing donation includes many ideas: presumed consent, donation programs, medical alternatives to use organs not used before, donor expert focus, increase the pool of possible donors (DCD), education on opting out, health organization facilitating organ donation, reduce workup time, [reconditioned] organs, better criteria for marginal organs."	13 (30.2)
How to develop alternatives to transplantation (artificial organs, xenotransplantation) "We need to [develop] technologies and new interventions to use more organs."	12 (27.9)
How to increase the number of transplantations (how to make optimal use of organs) "More organs getting to recipients. Use of new technologies to allow the use of organs [that] have not been used so far, etc."	9 (20.9)
How to improve access to organ transplantation "How can we improve national and universal access to all parts of donation and transplantation?"	8 (18.6)
How to mitigate the burden (economic and social) for donors and their families "How can we [more effectively] support donors, families and recipients in a transplant process (psychosocial, economic, medical, etc)? How to improve [the] psychosocial experience for donors, families of donors, caregivers and recipients, including cultural issues."	6 (14.0)
How to improve requests for organ donation and family consent "How to approach families (how to deal with family veto, legal issues)."	4 (9.3)

immunosuppressive drugs. Psychological complications and organ donation were not mentioned as research priorities in this study.²¹

The research priorities identified during the survey and the national workshop are similar to the unmet clinical needs in transplantation identified by Stegall et al,²² namely, those surrounding improving long-term graft survival, increasing the number of organ donors, immunosuppressive drugs with fewer adverse effects, induction of specific immunologic unresponsiveness, prevention and treatment of long-term medical complications (viral and cancer), and rehabilitation.

Recently, the National Health Institute has convened working groups to determine the research needs for HCT patients. No priority-setting exercises involving patients and caregivers have been reported in the field of HCT. One of the working groups conducted a scoping review to look at the research needs related to patient-reported outcomes and health-related quality of life. The priority domains of research are sexual dysfunction, fatigue, sleep disruption, nonadherence, health behaviors, and psychological dysfunction.²³ Other working groups published reviews presenting the current knowledge and unmet needs in the field of HCT for inherited bone marrow failure syndromes,²⁴ for severe combined immunodeficiency syndromes, and for sickle cell disease and thalassemia.²⁵ Improving long-term survival and morbidity, alternative donor sources, the conditioning of HCT, and chronic graft-versus-host disease were reported as unmet needs in research.^{24,25} There are some similarities with the research priorities elicited during the survey and the workshop.

One strength of our priority-setting exercise was the number of survey respondents, with 505 participants completing the survey, of which 46% identified themselves as patients. Furthermore, patients and caregivers—SOT and HCT patients, transplant candidates, living organ donors and family members of deceased donors—with an extensive and diverse range of personal experiences with transplantation and donation were included in the national workshop. That being said, this project also had its limitations. The research team recognizes that the survey questions were geared toward SOT and may have lacked relevance to HCT recipients. This could be explained by the fact that there was no one representing the HCT community on the research team during the survey's creation. In addition, patients from ethnic minorities were underrepresented in the national workshop (vis-à-vis the national demographics, considering that 19.1% of the Canadian population identifies as a visible minority),²⁶ although 1 patient from the Aboriginal community did participate. There is therefore a need to validate the research priorities with Canada's different ethnic groups, including Aboriginals, particularly because there are disparities in access to kidney transplantation among Aboriginals and patients from ethnic minorities.²⁷

CONCLUSIONS

This is the first national research priority-setting exercise in donation, SOT, and HCT in Canada. The priorities identified are the result of discussions with patients, family members, caregivers, HCPs, and researchers, and they will inform the identification, prioritization and funding of projects, and the future strategic directions of the CNTRP. This

priority-setting exercise enabled a dialogue between patients, researchers, and HCPs within the CNTRP, as well as the development of the CNTRP patient-partnership strategy. In the coming years, the research team will assess how this exercise has shaped research projects within the CNTRP. The priorities identified will continue to be reassessed in the future to ensure that they are still relevant and have not become obsolete and to determine whether new priorities have emerged.

REFERENCES

1. Jun M, Manns B, Laupacis A, et al. Assessing the extent to which current clinical research is consistent with patient priorities: a scoping review using a case study in patients on or nearing dialysis. *Can J Kidney Health Dis.* 2015;2:35.
2. Allard J, Durand C, Anthony S, et al. Perspectives of patients, caregivers and researchers on research priorities in donation and transplantation in Canada: a pilot workshop. *Transplant Direct.* 2017;3:e127.
3. Crowe S, Fenton M, Hall M, et al. Patients', clinicians' and the research communities' priorities for treatment research: there is an important mismatch. *Res Involv Engagem.* 2015;1:2.
4. Caron-Flinterman JF, Broerse JE, Bunder JF. The experiential knowledge of patients: a new resource for biomedical research? *Soc Sci Med.* 2005;60:2575–2584.
5. Canadian Institutes of Health Research. Strategy for Patient-Oriented Research—Patient Engagement Framework. <http://www.cihr-irsc.gc.ca/e/48413.html>. Updated July 2, 2014. Accessed August 15, 2014.
6. Fleurence RL, Beal AC, Sheridan SE, et al. Patient-powered research networks aim to improve patient care and health research. *Health Aff (Millwood).* 2014;33:1212–1219.
7. NHS National Institute for Health Research. Involve. <http://www.invo.org.uk/>. Updated 2015. Accessed October 21, 2016.
8. Manns B, Hemmelgarn B, Lillie E, et al. Setting research priorities for patients on or nearing dialysis. *Clin J Am Soc Nephrol.* 2014;9:1813–21.
9. Tong A, Crowe S, Chandon S, et al. Research priorities in CKD: report of a national workshop conducted in Australia. *Am J Kidney Dis.* 2015;66:212–222.
10. de Wit M, Kirwan JR, Tugwell P, et al. Successful stepwise development of patient research partnership: 14 years' experience of actions and consequences in outcome measures in rheumatology (OMERACT). *Patient.* 2016:1–12.
11. Caron-Flinterman JF, Broerse JEW, Teerling J, et al. Patients' priorities concerning health research: the case of asthma and COPD research in the Netherlands. *Health Expect.* 2005;8:253–263.
12. McCormick S, Brody J, Brown P, et al. Public involvement in breast cancer research: an analysis and model for future research. *Int J Health Serv.* 2004;34:625–646.
13. Tong A, Sautenet B, Chapman JR, et al. Research priority setting in organ transplantation: a systematic review. *Transpl Int.* 2017;30:327–343.
14. Canadian National Transplant Research Program. Transforming transplant research in Canada. <http://www.cntrp.ca/#!about/c4nz>. Accessed December 9, 2016.
15. Hébert M, Hartell D, West L. Transdisciplinary tour-de-force: the Canadian National Transplant Research Program. *Transplantation.* 2016;1000:466–470.
16. R Core Team. R: a language and environment for statistical computing. <https://www.r-project.org/>. Accessed October 26, 2017.
17. Poses RM, Isen AM. Qualitative research in medicine and health care. questions and controversy. *J Gen Intern Med.* 1998;13:32–38.
18. Green J, Thorogood N. *Qualitative methods for health research.* Third ed. London, UK: SAGE; 2014.
19. Barbour R. Checklists for improving rigour in qualitative research: a case of the tail wagging the dog? *BMJ.* 2001;322:1115–1117.
20. Tong A, Sainsbury P, Carter SM, et al. Patients' priorities for health research: focus group study of patients with chronic kidney disease. *Nephrol Dial Transplant.* 2008;23:3206–3214.
21. Knight SR, Metcalfe L, O'Donoghue K, et al. Defining priorities for future research: results of the UK kidney transplant priority setting partnership. *PLoS One.* 2016;11:e0162136.
22. Stegall MD, Morris RE, Alloway RR, et al. Developing new immunosuppression for the next generation of transplant recipients: the path forward. *Am J Transplant.* 2016;16:1094–1101.

23. Bevans M, El-Jawahri A, Tierney D, et al. National Institutes of Health hematopoietic cell transplantation late effects initiative: the patient-centered outcomes working group report. *Biol Blood Marrow Transplant*. 2017;23:538–551.
24. Heimall J, Puck J, Buckley R, et al. Current knowledge and priorities for future research in late effects after hematopoietic stem cell transplantation (HCT) for severe combined immunodeficiency patients: a consensus statement from the second Pediatric Blood and Marrow Transplant Consortium International Conference on late effects after pediatric HCT. *Biol Blood Marrow Transplant*. 2017;23:379–387.
25. Shenoy S, Angelucci E, Arnold S, et al. Current results and future research priorities in late effects after hematopoietic stem cell transplantation for children with sickle cell disease and thalassemia: a consensus statement from the second Pediatric Blood and Marrow Transplant Consortium International Conference on Late Effects After Pediatric Hematopoietic Stem Cell Transplantation. *Biol Blood Marrow Transplant*. 2017;23:552–561.
26. Statistics Canada. Immigration and Ethnocultural Diversity in Canada. <http://www12.statcan.gc.ca/nhs-enm/2011/as-sa/99-010-x/99-010-x2011001-eng.cfm>. Updated September 15, 2016. Accessed January 15, 2018.
27. Mucsi I, Bansal A, Famure O, et al. Ethnic background is a potential barrier to living donor kidney transplantation in Canada: a single-center retrospective cohort study. *Transplantation*. 2017;101:e142–e151.