Establishing an evidence-based framework for involving patients in research about chronic kidney disease

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A thesis submitted in fulfilment of the requirements for the degree of

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Declaration

This is to certify that to the best of my knowledge, the content of this thesis is my own work.

This thesis has not been submitted for any degree or other purposes.

I certify that the intellectual content of this thesis is the product of my own work and that all the assistance received in preparing this thesis and sources has been acknowledged.

Signature: ……………………. Date: 29 April 2021

Talia Margot Gutman

Author’s Contribution

The work presented in this thesis has been carried out by the author under the supervision of Professor Allison Tong of the Sydney School of Public Health, The University of Sydney, Professor Jonathan Craig of the College of Medicine and Public Health, Flinders University, and Associate Professor Shilpanjali Jesudason of the Faculty of Health and Medical Sciences, University of Adelaide.

The author planned the research, designed the studies, obtained ethics approval, collected, managed and analysed the data, interpreted results, drafted and revised the manuscripts for submission to peer-reviewed journals, and wrote and compiled this thesis.

As supervisor for the candidature upon which this thesis is based, I can confirm that the authorship attribution statements above are correct.

Signature: ……………………. Date: 29 April 2021

Professor Allison Tong

Ethical Clearance

The studies presented in Chapters 4 and 6 received ethics approval from The University of Sydney.

The study presented in Chapter 7 received ethics approval from The Royal Melbourne Hospital.

The study presented in Chapter 8 received ethics approval from The University of Sydney, British Columbia Children's Hospital, The University of British Columbia Research Ethics Board, Alberta Children's Hospital, The University of Calgary Research Ethics Board, Texas Children's Hospital, The Institutional Review Board for Baylor College of Medicine and Affiliated Hospital, The Children's Hospital at Westmead, The Sydney Children's Hospital Network Human Research Ethics Committee, The Royal Children's Hospital, The Melbourne Children's Campus Research Ethics and Governance, Lady Cilento Hospital, and The Children's Health Queensland Hospital and Health Service Research Governance.

All participants gave written informed consent for participation in these studies.

Chapters 2, 3 and 5 did not require ethics approval.

Abstract

Involving consumers (defined as patients and informal caregivers/family members) in research as more than ‘subjects’ is now globally advocated to improve the relevance, importance, and quality of research. Growing evidence in the general population and some specific disease groups continues to demonstrate the benefits of consumer involvement in research, including alignment of research priorities, better recruitment/retention, unique insights in data analysis, and broader dissemination and translation into policy and practice. Reporting on consumer involvement is now mandated by some journals (e.g. British Medical Journal) and funding organisations (e.g. National Health and Medical Research Council, Cancer Australia, Patient-Centered Outcomes Research Institute, National Institute for Health Research).

Despite this, consumer involvement in research in chronic kidney disease (CKD) remains scarce as researchers are uncertain about approaches and often lack resources to undertake it. Patients with CKD and their families face unique challenges that may hinder their ability or willingness to be involved in research, such as the lifelong nature of CKD, periods of prolonged illness, time-consuming therapies and burnout from ongoing caring responsibilities. Despite the enormous burden on consumers to self-manage their care, including medications and complex therapies such as dialysis, they are often disempowered and excluded from important treatment decisions due to health system operations, limited education and time with clinicians, and urgent clinical needs. Distancing consumers from being actively involved in their own treatment decisions further hinders their potential to be involved in research. The lack of patient involvement in research can limit the relevance of research to patients and their caregivers, with a recent study showing that 80% of clinical

research did not address the top 10 priorities identified by patients with CKD. Such limited conducting, reporting and publishing of consumer involvement activities in this population means that the evidence for best practice remains in its infancy, and researchers remain uncertain of how to involve consumers in meaningful ways.

This thesis aims to summarise the existing evidence, generate new evidence to address gaps in best practice and synthesise the data to develop a practical evidence-based framework for the meaningful, impactful and sustained involvement of consumers in CKD research. The first part of this thesis (chapters 2-5) provides a comprehensive overview of the literature for consumer involvement in research and shared decision-making in published CKD research. It develops a conceptual understanding of the benefits, challenges and gaps of consumer involvement in this population, based on the literature and perspectives from consumers and health professionals. The second part of this thesis (chapters 5, 6, 7) contains applied empiric studies demonstrating and evaluating consumer involvement in research and decision-making in real world settings. The final chapter integrates the key findings and recommendations to provide a practical framework for researchers to guide best practice in involving consumers in all types of research in CKD.

Acknowledgements

This thesis could not have been produced without the support, guidance and expertise of many wonderful and wildly intelligent people whom I would like to sincerely thank.

First, I would like to thank my primary supervisor, Professor Allison Tong. Allison's extraordinary research talent and skills, particularly in elevating qualitative research to high quality biomedical journals, patience and diligence in training and mentoring, and genuine care for her staff, students, research participants and every potential end-user of the research she is involved in, have imbued this process with meaning and truly made me feel that what we are doing matters for patients and their families. Her ever caring and compassionate nature has guided me through a difficult personal journey in this time, ensuring that my health was always a priority, but supporting me in my goal to finish this thesis. I would also like to thank my auxiliary supervisors Professor Jonathan Craig and Associate Professor Shilpanjali Jesudason for their continued expert guidance and insights. I would like to thank Jonathan for his big picture thinking and oversight of this thesis. I would like to thank Shilpa for her clinical insights and constant connection to the practical implications and implementation of this work in the clinical, research and consumer communities.

To all the wonderful patients, family members and caregivers that have given of their time, expertise and emotional energy to contribute to both the data collection and the outputs of this work, thank you. I hope that having your voices shared in this context shows the huge value and impact your involvement has. A very special thank you to Nicole Scholes-Robertson, patient partner extraordinaire, for your constant support as a friend, researcher and consumer.

A big thank you to all of my colleagues and friends at the Centre for Kidney Research that I have been lucky enough to meet and work with along the way. I have learnt something from each one of you and have valued your support and feedback through these last few years. A special thank you to Karine Manera, Angela Ju and Ayano Kelly for your friendship, support and guidance as fellow PhD students.

Finally, a mammoth thank you to my incredible husband Ilan. His unwavering support and belief in me are unmatched. Thank you for all the reviews and edits, your expert formatting, the PowerPoint and Excel lessons, your endless emotional support and guidance and for picking up the slack in the tough times. You have carried me through this, and I am forever grateful.

This thesis is dedicated to all the patients and families living with chronic illness – may this go some way towards giving you a voice in the research community, and to my soon to be born Baby Boy – may we start a new chapter together!

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First Author Publications Arising from this Thesis

This thesis is presented for examination as a thesis containing published work. Chapters 5, 6, 7 and 8 have been published in international peer-reviewed journals. Chapters 2 and 4 have been submitted for publication. I am the first and corresponding author of each of these papers. Chapter 3 is a book chapter accepted for publication. I am the last author, however all authors shared equal responsibility in the writing of this manuscript.

Chapter 2	**Gutman T,** Scholes-Robertson N, Tong A, Jesudason S, Guha C, White DM, Levin A, Craig JC. Consumer involvement in chronic kidney disease research: a systematic review. American Journal of Kidney Diseases. Submitted March 2021.
Chapter 3	Schell J, Cavanaugh K, **Gutman T.** Shared decision-making. Evidence Based Nephrology. Editors: Molony DA, Strippoli G, Craig JC. Wiley. West Sussex, United Kingdom; 2021. Accepted for publication.
Chapter 4	**Gutman T,** Kelly A, Scholes-Robertson N, Craig JC, Jesudason S, Tong A. Patient and caregiver experiences and attitudes towards involvement in research in chronic kidney disease: an interview study. Clinical Journal of the American Society of Nephrology. Submitted April 2021.
Chapter 5	**Gutman T**, Tong A, Howell M, Dansie K, Hawley CM, Craig JC, Jesudason S, Chapman JR, Johnson DW, Murphy L, Reidlinger D, Crowe S, Duncanson E, Muthuramalingam S, Scholes-Robertson N, Williamson A, McDonald S. Principles and strategies for involving patients in research in

	chronic kidney disease: report from national workshops. Nephrology Dialysis Transplantation. 2020;35(9):1585-94.
Chapter 6	**Gutman T**, Manera KE, Baumgart A, Johnson DW, Wilkie M, Boudville N, Craig JC, Dong J, Jesudason S, Mehrotra R, Neu A, Shen JI, Van Biesen W, Blake PG, Brunier G, Cho Y, Jefferson N, Lenga I, Mann N, Mendelson AA, Perl J, Sanabria RM, Scholes-Roberston N, Schwartz D, Teitelbaum I, Tong A. “Can I go to Glasgow?” Learnings from patient involvement at the 17th Congress of the International Society for Peritoneal Dialysis (ISPD). Peritoneal Dialysis International 2020;40(1):12-25.
Chapter 7	**Gutman T**, Lopez-Vargas P, Manera KE, Craig JC, Howell M, Tunnicliffe DJ, James LJ, MacGinley R, See E, Wong J, Voss D, Saunders J, Menachem S, Jesudasen S, Tong A, de Crespigny PC. Identifying and integrating patient and caregiver perspectives in clinical practice guidelines for percutaneous renal biopsy. Nephrology. 2019; 24(4):395-404.
Chapter 8	**Gutman T**, Hanson CS, Bernays S, Craig JC, Sinha A, Dart A, Eddy AA, Gipson DS, Bockenhauer D, Yap HK, Groothoff J, Zappitelli M, Webb NJA, Alexander SI, Goldstein SL, Furth S, Samuel S, Blydt-Hansen T, Winklemayer WC, Currier H, McTaggart S, Walker A, Ralph AF, Ju A, James LJ, Tong A. Child and parental perspectives on communication and decision-making in pediatric CKD: a focus group study. American Journal of Kidney Diseases. 2018; 72(4):547-59.

Conference Proceedings Arising from this Thesis

2019

Selected abstract: Gutman T, Lopez-Vargas P, Manera KE, Craig JC, Howell M, Tunnicliffe DJ, James LJ, MacGinley R, See E, Wong J, Voss D, Saunders J, Menachem S, Jesudasen S, Tong A, de Crespigny PC. Identifying and integrating consumer priorities in the development of renal biopsy guidelines. International Society of Nephrology World Congress of Nephrology, 12-15 April 2019, Melbourne Australia.

Selected abstract: Gutman T, Tong A, Howell M, Dansie K, Hawley CM, Craig JC, Jesudason S, Chapman JR, Johnson DW, Murphy L, Reidlinger D, Crowe S, Duncanson E, Muthuramalingam S, Scholes-Robertson N, Williamson A, McDonald S. Principles and strategies for involving patients in research in CKD. International Society of Nephrology World Congress of Nephrology, 12-15 April 2019, Melbourne Australia.

Selected abstract: Gutman T, Lopez-Vargas P, Manera KE, Craig JC, Howell M, Tunnicliffe DJ, James LJ, MacGinley R, See E, Wong J, Voss D, Saunders J, Menachem S, Jesudasen S, Tong A, de Crespigny PC. Identifying and integrating consumer priorities in the development of renal biopsy guidelines. Public Involvement in Health Research Symposium, 21 February 2019, Sydney Australia.

Selected abstract: Gutman T, Tong A, Howell M, Dansie K, Hawley CM, Craig JC, Jesudason S, Chapman JR, Johnson DW, Murphy L, Reidlinger D, Crowe S, Duncanson E, Muthuramalingam S, Scholes-Robertson N, Williamson A, McDonald S. Principles and strategies for involving patients in research in CKD. Public Involvement in Health Research Symposium, 21 February 2019, Sydney Australia.

2018

Selected abstract: Gutman T, Lopez-Vargas P, Manera KE, Craig JC, Howell M, Tunnicliffe DJ, James LJ, MacGinley R, See E, Wong J, Voss D, Saunders J, Menachem S, Jesudasen S, Tong A, de Crespigny PC. Identifying and integrating consumer priorities in the development of renal biopsy guidelines. NHMRC Symposium on translational research, 27-28 November 2018, Sydney Australia.

Selected abstract: Gutman T, Tong A, Howell M, Dansie K, Hawley CM, Craig JC, Jesudason S, Chapman JR, Johnson DW, Murphy L, Reidlinger D, Crowe S, Duncanson E, Muthuramalingam S, Scholes-Robertson N, Williamson A, McDonald S. Principles and strategies for involving patients in research in CKD. NHMRC Symposium on translational research, 27-28 November 2018, Sydney Australia.

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Selected abstract: Gutman T, Lopez-Vargas P, Manera KE, Craig JC, Howell M, Tunnicliffe DJ, James LJ, MacGinley R, See E, Wong J, Voss D, Saunders J, Menachem S, Jesudasen S, Tong A, de Crespigny PC. Identifying and integrating consumer priorities in the development of renal biopsy guidelines. American Society of Nephrology Annual Scientific Meeting, 23-28 October 2018, San Diego United States.

Invited plenary: Gutman T, on behalf of the BEAT-CKD investigators. Strategies for involving patients in research in kidney disease. Australian and New Zealand Society of Nephrology Annual Scientific Meeting, 8-12 September 2018, Sydney Australia.

Selected abstract: Gutman T, Hanson CS, Bernays S, Craig JC, Sinha A, Dart A, Eddy AA, Gipson DS, Bockenhauer D, Yap HK, Groothoff J, Zappitelli M, Webb NJA, Alexander SI, Goldstein SL, Furth S, Samuel S, Blydt-Hansen T, Winklemayer WC, Currier H, McTaggart S, Walker A, Ralph AF, Ju A, James LJ, Tong A. Shared decision-making in paediatric CKD. Australian and New Zealand Society of Nephrology Annual Scientific Meeting, 8-12 September 2018, Sydney Australia.

Selected abstract: Gutman T, Lopez-Vargas P, Manera KE, Craig JC, Howell M, Tunnicliffe DJ, James LJ, MacGinley R, See E, Wong J, Voss D, Saunders J, Menachem S, Jesudasen S, Tong A, de Crespigny PC. Identifying and integrating consumer priorities in the development of renal biopsy guidelines. Australian and New Zealand Society of Nephrology Annual Scientific Meeting, 8-12 September 2018, Sydney Australia.

Selected abstract: Gutman T, Tong A, Howell M, Dansie K, Hawley CM, Craig JC, Jesudason S, Chapman JR, Johnson DW, Murphy L, Reidlinger D, Crowe S, Duncanson E, Muthuramalingam S, Scholes-Robertson N, Williamson A, McDonald S. Principles and strategies for involving patients in research in CKD. Australian and New Zealand Society of Nephrology Annual Scientific Meeting, 8-12 September 2018, Sydney Australia.

Invited speaker: Gutman T. Patient engagement in research. BEAT-CKD Research forum, 23-25 July 2018, Brisbane Australia.

Selected abstract: Gutman T, Hanson CS, Bernays S, Craig JC, Sinha A, Dart A, Eddy AA, Gipson DS, Bockenhauer D, Yap HK, Groothoff J, Zappitelli M, Webb NJA, Alexander SI, Goldstein SL, Furth S, Samuel S, Blydt-Hansen T, Winklemayer WC, Currier H, McTaggart S, Walker A, Ralph AF, Ju A, James LJ, Tong A. Paediatric shared decision-making in chronic kidney disease. European Renal Association and European Dialysis and Transplant Association Congress, 24-27 May 2018, Copenhagen Denmark.

Awards Arising from this Thesis

2019

NHMRC Postgraduate Research Scholarship: Awarded NHMRC scholarship to support PhD research on consumer involvement in research in chronic kidney disease. Amount: $66,501.50

Research Student Grants Scheme, Sydney School of Public Health, The University of Sydney: Awarded to fund travel to present at an international conference. Amount: $480.00

Postgraduate Research Support Scheme, The University of Sydney: Awarded to fund travel to present at an international conference. Amount: $2,140.22

Centre for Kidney Research Top- Up Scholarship: Awarded to supplement NHMRC PhD scholarship. Amount: $3,000

2018

Research Student Grants Scheme, Sydney School of Public Health, The University of Sydney: Awarded to fund travel to present at an international conference. Amount: $1,100

Postgraduate Research Support Scheme, The University of Sydney: Awarded to fund travel to present at an international conference. Amount: $1,272

Better Evidence and Translation – Chronic Kidney Disease (BEAT-CKD) PhD Scholarship: Awarded scholarship to support PhD research on consumer involvement in research in chronic kidney disease. Amount: $30,000

Educational Workshops Arising from this Thesis

2020

Consumer Involvement in Research: Workshop for the Matilda Centre, 28 May 2020, University of Sydney, Sydney Australia.

2019

Consumer Involvement in Clinical Practice Guideline Development: Workshop for the Guidelines International Network (GIN) Conference, 31 October 2019, Adelaide Australia.

Consumer Involvement in Research: Workshop for Researchers and Health Professionals for the Patient Centred Research Network (PACER) Network and the University of Sydney, 14 August 2019, The Children's Hospital at Westmead, Sydney Australia.

Consumer Involvement in Research: Workshop for the Better Evidence and Translation in Chronic Kidney Disease (BEAT-CKD) Research Forum, 12 August 2019, University of Sydney, Sydney Australia.

Consumer Involvement in Research: Workshop for Clinical Epidemiology Students, 28 May 2019, University of Sydney, Sydney Australia.

Left: Nicole Scholes-Roberts and me co-presenting our workshop on Consumer Involvement in Research for health professionals at The Children's Hospital at Westmead, August 2019

Consumer Involvement Initiatives Arising from this Thesis

2018 | Establishment of BEAT-CKD consumer advisory board

Following the national workshops conducted and published in Chapter 5, a consumer advisory board consisting of more than 40 members across Australia was established to oversee high level strategic decisions in the BEAT-CKD program of work. Since then this group has also overseen the development of consumer sessions for the Australian and New Zealand Society of Nephrology Annual Scientific Meetings, and members are now involved in tactical capacities as research partners and advisors in all BEAT-CKD research projects through a process of self-nomination based on interests and expertise. Members of this advisory board have also been responsible of onboarding and mentoring new consumer research partners at a project level.

Top: Consumer delegates ANZSN ASM 2018

Bottom Left: BEAT-CKD Consumer Advisory Board virtual meeting 2020

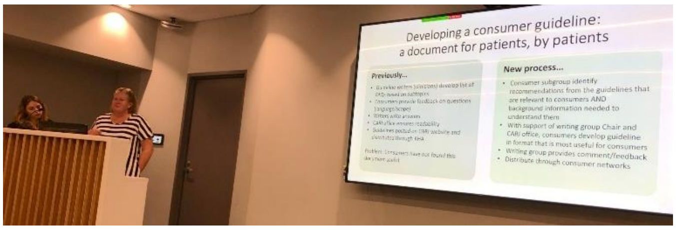
Bottom Right: Consumer presenters at BEAT-CKD Research Forum 2020

BEATCKD @BEAT_CKD · Aug 4, 2020

Day 2 kicks off with a consumer-focused session! @shyamrith @scholes_nicki @CharmaineJGreen @ChandanaGuha @KidneyHealth @PKD_Australia @talia_gutman @JonathanCCraig @NephMcDonald @DWJohnsonNeph @a_viecelli @shilpa_jes @Kidney_Trials @cariguidelines @CochraneKidney #beatckdforum20

2019 | Development of the strategy for involvement in the Caring for Australians and New Zealanders with Kidney Impairment (CARI) Clinical Practice Guidelines

The results of this thesis have informed a new strategy for consumer involvement in the entirety of the guideline process. Until 2019 consumers were involved in workshops to identify and prioritise topics and outcomes for inclusion in the guidelines, provided comment and feedback on the consumer versions of the guidelines, and in public review of the draft guidelines. Currently, three consumers sit on the Steering Committee in the same capacity as the other expert members, consumers are involved in the guideline writing groups as equal members of the team, two consumers are employed as part of the CARI office team, consumers lead the development of the consumer guidelines supported by CARI staff and still participate in workshops and public review processes.

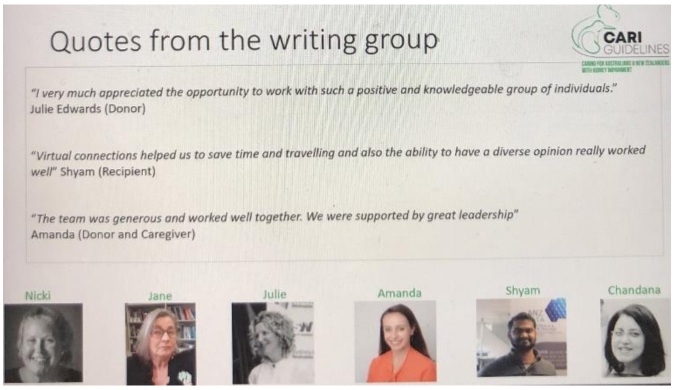


Developing a consumer guideline:
a document for patients, by patients

Previously...

New process...

- Consumer subgroup identify recommendations from the guidelines that are relevant to consumers AND background information needed to understand them
- With support of writing group Chair and CARI office, consumers develop guideline in format that is most useful for consumers
- Writing group provides comment/feedback
- Distribute through consumer networks



Quotes from the writing group

"I very much appreciated the opportunity to work with such a positive and knowledgeable group of individuals."
Julie Edwards (Donor)

"Virtual connections helped us to save time and travelling and also the ability to have a diverse opinion really worked well" Shyam (Recipient)

"The team was generous and worked well together. We were supported by great leadership"
Amanda (Donor and Caregiver)

Nicki
Jane
Julie
Amanda
Shyam
Chandana

Top: CARI Steering Committee, including three consumer members, 2019

Middle: Nicole Scholes-Robertson and me co-presenting the new strategy for consumer involvement for the CARI guideline, including the new process for developing consumer guidelines, 2019

Bottom: Consumer guideline writing group for consumer biopsy guideline, 2020

List of Abbreviations

ACP	Advance care planning
AIDS	Acquired immunodeficiency syndrome
AKTN	Australian Kidney Trials Network
ANZDATA	Australian and New Zealand Dialysis and Transplant registry
ANZSN	Australian and New Zealand Society of Nephrology
ASM	Annual Scientific Meeting
ASN	American Society of Nephrology
BEAT-CKD	Better Evidence and Translation in Chronic Kidney Disease
BMJ	British Medical Journal
Can-SOLVE CKD	Canadians Seeking Solutions and Innovations to Overcome Chronic Kidney Disease
CARI	Caring for Australians and New Zealanders with Kidney Impairment *(formerly KHA-CARI, Kidney Health Australia – Caring for Australasians with Renal Impairment)*
CCC	Comprehensive Conservative Care
CKD	Chronic Kidney Disease
CIHR	Canadian Institutes of Health Research
CME	Continuing Medical Education
CONSORT	Consolidated Standards of Reporting Trials
COREQ	Consolidated Criteria for Reporting Qualitative Studies
CT	Computerized Tomography
DDAVP	Desmopressin Acetate
FDA	Food and Drug Administration

GRIPP	Guidance for Reporting Involvement of Patients and Public
HIV	Human Immunodeficiency Virus
ICU	Intensive Care Unit
IPDAS	The International Patient Decision Aid Standards
ISN-ACT	International Society of Nephrology's Advancing Clinical Trials initiative
ISPD	International Society for Peritoneal Dialysis
JLA-PSP	James Lind Alliance Priority Setting Partnership
KHA	Kidney Health Australia
KHI	Kidney Health Initiative
KPIN	Kidney Patient Involvement Network
NHMRC	National Health and Medical Research Council
NIHR	National Institute for Health Research
OMERACT	Outcome Measures in Rheumatology
PACER	Patient Centred Research Network
PD	Peritoneal Dialysis
PCORI	The Patient-Centered Outcomes Research Institute
PICOM	Population, Intervention, Comparator, Outcomes, Methodology
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-Analyses
RCT	Randomised Controlled Trial
RPA	Renal Physicians Association
SDM	Shared Decision-Making
SONG	Standardised Outcomes in Nephrology
SPOR	Strategy for Patient Oriented Research

STROBE	Strengthening the Reporting of Observational Studies in Epidemiology
TCU	Transitional Care Unit
TYPE	The Typology of Youth Participation and Empowerment pyramid
UK	United Kingdom
US	United States
WHO	World Health Organisation

Chapter 1: Introduction

1.1 Overview

"Consumer engagement should be designed by choice rather than chance"

Shyamsundar Muthuramalingam, Patient, Consumer Engagement Project Officer ANZDATA
BEAT-CKD Consumer Advisory Board[1]

This thesis is presented as a series of chapters with the overall aim of establishing a 'best-practice' framework for involving consumers in research in chronic kidney disease (CKD) that is grounded in the empiric studies which form the content of the following chapters. This chapter provides an overview of the history of consumer involvement in research more broadly, the specific nature, challenges and opportunities for consumer involvement in research related to CKD, and the potential for improved communication and shared decision-making among patients and their clinicians, which can support consumer involvement in research. This chapter also includes justification for this program of work and outlines the aims of each study included in the thesis.

The research presented in this thesis spans several national and global initiatives and collaborative partnerships including: The Standardised Outcomes in Nephrology (SONG) initiative[2] (*a global initiative that aims to establish core outcome sets in nephrology, based on the shared priorities of all stakeholders)*; and The Better Evidence and Translation in Chronic Kidney Disease (BEAT-CKD) program[3] funded by the Australian National Health and Medical Research Council to generate high quality evidence to inform practice and policy in CKD. BEAT-CKD brings together the Australian and New Zealand Kidney Trials Network (AKTN)[4], the Australian and New Zealand Dialysis and Transplant (ANZDATA) registry[5], Cochrane Kidney and Transplant[6], and Caring for Australians and New Zealanders with Kidney Impairment (CARI) clinical practice guidelines[7]. BEAT-CKD is underpinned by a Consumer Advisory Board contributing to research activities in this program[8].

1.2 Consumer involvement in research

"Nothing about us without us"[9]

The slogan "nothing about us without us" originated in the 1500s in central European politics, as kingdoms transitioned into democracies[10]. This statement has fittingly come to define a movement that empowers patients and their caregivers (consumers) to have their voices heard and included in all aspects of care, policy and research that impacts them as end-users. The slogan was first adopted in this context in the early 1990s by patient advocacy groups - first in disability activism, and then in the HIV consumer community - coinciding with the beginning of an era of democratising medicine, and shift away from the traditional relationships and roles of doctor as expert and patient as passive participant[11,12]. Limitations with this paternalistic model, particularly the belief that the patient and clinician hold similar values and priorities for treatment and care, were identified as the capacity and expertise of people with lived experience began to be recognised[13].

Definitions

The terminology used to define involvement is varied, and some terms are often used interchangeably but can denote different things. For example, the United Kingdom and Australia tend to use "involvement" to describe the broad inclusion of consumers in research activities as more than research subjects, while Canada and the United States tend to use "engagement" to describe the same concept, which for other nations has a more discrete meaning of selecting and recruiting consumers to become involved[14,15]. Ultimately consumer involvement is about meaningful contributions to research as more than 'subjects' or data points.

For the purposes of this thesis I follow INVOLVE, the peak body in the United Kingdom funded by the National Institute for Health Research (NIHR) and global leader in supporting active public involvement in health and social care research, which defines consumer involvement as *"research being carried out 'with' or 'by' members of the public rather than 'to', 'about' or 'for' them"*[14]. This definition is echoed by the National Health and Medical Research Council[16]. Additional definitions can be found in Box 1.1.

Box 1.1 Definitions

Patient: refers to a patient with lived experience of chronic kidney disease

Carer/Caregiver: refers to an informal caregiver or family member of a patient with lived experience of chronic kidney disease

Consumer: refers to a patient, caregiver, family member or community member with lived experience of chronic kidney disease

Involvement: refers to sustained and meaningful contributions to the research process as more than a research subject or participant

Engagement: refers to the establishment of a relationship between patients and researchers and is the first step of involvement.

Participation: refers to consumers involved in research as subjects in trials or studies, with no involvement or decision-making power in the planning, execution or translation of the study.

Opportunities and benefits for researchers and consumers through research involvement

Involving consumers (including patients, their families, and members of the general community) helps to reduce research waste and improve the relevance and quality of research through alignment of research priorities, better patient recruitment/retention, unique insights in data analysis, and broader dissemination and translation into policy and practice[17-22]. Consumers have also reported personal benefits from being involved, including increased knowledge of their disease and the research process, greater access to information, developing social relationships with other consumers and health professionals outside of a clinical context and an opportunity to 'give back' to the medical community and 'pay it forward' to other consumers[19,23,24].

Top-down advocacy and recommendations

Global, national and local organisations now advocate for, and some even mandate, the involvement of consumers in research. The World Health Organisation (WHO) states that "the people have the right and duty to participate individually and collectively in the planning and implementation of their health care", including research[25]. The NIHR's INVOLVE in the United Kingdom was established in 1996 to support active public involvement across all stages of research, and has developed resources to assist both researchers and consumers in doing this[14]. In Australia, the National Health and Medical Research Council (NHMRC) advocates "the right and responsibility" to involve patients in all stages of research in order to receive funding and encourages researchers to "consider the benefits of actively engaging consumers in their proposed research"[26]. The Patient-Centered Outcomes Research Institute (PCORI) in the United States drives research guided by consumers to ensure they have access to relevant evidence to inform decision-making[15]. The Canadian Institutes of Health Research

(CIHR) formally recognises the value of the unique knowledge that comes from lived experience of a condition and states that involvement in research increases the quality of care[27]. Many major journals now require researchers to report whether consumers were involved, with the British Medical Journal stipulating the inclusion of a statement in the methods section under the subheading ‘patient involvement’, outlining how consumers were involved in the selection of the research question and outcome measures, the design of the study, recruitment, conduct, and how the results will be disseminated to study participants. Nephrology journals have been slower to follow suit, however the Clinical Journal of the American Society of Nephrology now publishes ‘Patient Voices’ pieces and has patient editors, and the American Journal of Kidney Diseases requires a ‘non-technical summary’ following the abstract. While there is an evident shift towards recommending the involvement of consumers in all aspects of research, the substantial variability in the strength and particulars of these recommendations demonstrates the need for more evidence.

Consumer involvement in other disease groups

Groups representing consumers with disabilities and HIV and have paved the way for consumer involvement in advocacy and care, leading to involvement in research through community-based participatory research, largely focused on implementation of research through program and service development[11,28]. Participatory Action Research has also become a pillar of Indigenous Health Research, where the focus is on co-designing research projects with the community and community elders and to share the ownership and benefits of research with the communities[29,30]. In cancer, consumers have been contributing to research for decades and some groups have developed formal mechanisms for involving consumers, such as the development of consumer advisory boards[31]. Cancer Australia has developed the “National Framework for Consumer Involvement in Cancer Control” which identifies key elements of committed organisations, capable consumers, inclusive groups and shared focus[32]. Despite this, evidence of impact often focuses on discrete activities with limited decision-making power, such as impact on recruitment processes or patient information leaflets[33]. There remains a paucity of evidence across all health areas about the nature, impact and outcomes of consumer involvement in research that support practical recommendations for researchers[19-21,24].

Challenges and limitations

While many of the benefits are now well established, there are challenges that prevent researchers from involving consumers in their research[19,20,34,35]. Frequently cited are the resources required to effectively support meaningful involvement, including funding, time and personnel to support training and logistics. With research budgets already stretched, and involvement often occurring only after grants have been awarded, it can be difficult to prioritise consumer involvement in allocating the resources needed. Other key challenges are the lack of ‘best-practice’ guidelines for how to involve consumers in research, and inconsistencies in the conducting, reporting, publishing, and synthesising of consumer involvement activities that make it difficult to learn from and improve upon what has been done in the past. Also, while the culture of research practice is changing, involvement of consumers still challenges the long-held notion that the clinician/researcher is the true expert. Researchers may not recognise consumer involvement as being useful or necessary, and lived experience is often undervalued, leading to tokenistic, and therefore suboptimal, involvement strategies.

1.3 Chronic Kidney Disease

"[CKD] is time consuming. It's scary… all of a sudden it's permeating [your] whole life"

Patient[1]

It is estimated that CKD has a global prevalence of between 11-13% and is a key determinant of poor health outcomes of major non-communicable diseases, including an up to tenfold increase in cardiovascular mortality[36-38]. More than two million individuals receive life sustaining kidney replacement therapy worldwide, although it has been suggested that this could be as little as 10% of those who need it[36]. In Australia, one in three adults is at risk of CKD, with one in ten showing early signs of the disease, and CKD contributes around 17,500 deaths per year, more than the combined deaths from breast cancer, prostate cancer and road accidents[39]. Patients with CKD face a life-long condition with mortality rates of up to 100 times higher than the general population, and worse quality of life than patients with other chronic diseases[40,41]. The relentless symptom and treatment burdens across the spectrum of CKD (i.e. CKD, dialysis, transplant, end-of-life care) mean that patients are faced with many critically important treatment decisions over the course of their life. For patients with end-stage kidney disease there is an enormous burden to provide their own care, including managing medications and complex therapies such as doing dialysis on their own, often at home. The medical, surgical and technical interventions (e.g. dialysis) have profound impacts on the patient's physical and psychosocial well-being and quality of life. Given these responsibilities and impacts, research should form a critical part of a consumer's decision-making toolkit, yet a recent review found that 80% of clinical research in CKD does not address the top 10 research priorities identified by patients, and most trials do not report outcomes which have been consistently identified to be critically important to patients, such as fatigue, life participation and mortality[41,42].

1.4 Consumer involvement in chronic kidney disease

Consumer involvement in research in CKD is needed to align research efforts with their priorities, and thus improve decision-making, satisfaction, and clinical and quality of life outcomes[9,43-45]. However, the unique challenges CKD consumers face may impact their ability and willingness to contribute to research. For patients, the burden of self-management responsibilities and time-consuming and invasive life-sustaining therapy (i.e. dialysis), and prolonged periods of illness, including symptoms of fatigue and impaired cognitive function, might make involvement exceptionally challenging, while for caregivers, the burden of caring responsibilities can become all-consuming and may lead to burnout.

There has been a recent shift within government and professional organisations to systematically involve patients with CKD in research. The Kidney Health Initiative (KHI), a partnership between The American Society of Nephrology (ASN) and the United States Food and Drug Administration (FDA), have established a Patient and Family Partnership Council, supported by several patient organisations, to give consumers a voice in CKD research in the United States[46]. The Australian Government Department of Health has identified the need to establish a National Kidney Consumer Research Hub in their National Strategic Action Plan for Kidney Disease[39]. Can-SOLVE CKD in partnership with the Strategy for Patient Oriented Research (SPOR) currently involves 75 patient partners across 18 research projects in Canada[47,48]. The BEAT-CKD program formed a national Consumer Advisory Board to inform high level strategy and project specific involvement across Australia’s national kidney trial, registry, clinical guidelines, and Cochrane groups[3,49]. Under the BEAT-CKD umbrella, The SONG Initiative has involved thousands of consumers and health professionals globally to establish consumer driven core outcome sets for trials in kidney disease[50,51].

The International Society of Nephrology's Advancing Clinical Trials (ISN-ACT) initiative Patient Engagement working group recently conducted a survey with 177 clinician members to understand the nature of consumer involvement in kidney trials globally, and found an "absence of formal mechanisms" for involvement and did not explore the nature of involvement including research stage, activities or consumer roles[52]. The Kidney Patient Involvement Network (KPIN) in the United Kingdom has involved 80 clinicians in identifying pros and cons of consumer involvement, and currently has a survey open for consumers to share their experiences being involved in research[52,53]. Despite these initiatives, there is scant evidence to inform or support best-practice for involving consumers with CKD in research, particularly from the consumer perspective.

1.5 Patient needs and priorities for shared-decision making

Shared decision-making in a clinical context involves "the meeting of two experts"[13] – the patient and the clinician, to determine the best course of action for the patient considering the evidence, clinician expertise and the priorities and preferences of the patient. Over the last few decades this practice has become more acceptable and even expected as medicine shifts "from paternalism to partnership"[13]. In many ways this mirrors consumer involvement in the research setting. This thesis shows the bidirectional nature and impact of shared decision-making on consumer involvement in research. There is vast potential to engage consumers in their own clinical care and outcomes by activating them as decision-makers in their care, leading to more informed consumers with better outcomes that are more closely aligned with their individual priorities. Consumers who are living well with CKD and have positive experiences of health systems may have improved capacity and desire to become involved in research[1]. Conversely, consumers involved in research develop a better understanding of their illness, health systems and strengthen relationships with clinicians, leading to improved engagement in their care and better outcomes[20,34].

However, implementing shared decision-making in CKD remains challenging due to the unpredictable and sometimes urgent nature of the disease and often conflicting priorities of patients and clinicians, particularly with vulnerable populations, including children[13,44,54]. These populations are also typically underrepresented with regards to being involved in research[17,19]. Thus, promoting effective communication and shared decision-making in clinical care has the potential to improve engagement and involvement in research related activities and promote diverse inclusion of consumer research partners.

1.6 Justification for this thesis

Despite the steps taken towards empowering patients as partners in research by major international initiatives such as INVOLVE, the James Lind Alliance, PCORI, and kidney-specific initiatives such as Can-SOLVE, and growing advocacy for patient partnership in all stages of the research cycle[20,43,55], involving patients in research remains challenging. Sparse data are available to develop evidence-based best practice to support researchers in implementing involvement[20,43,55]. Researchers remain uncertain about how to effectively communicate opportunities for involvement and provide necessary training and support, and little has been done to evaluate existing methods[19]. Patients often face a systemic power imbalance, uncertainty about their role and must contend with a significant illness burden that may impair their physical and cognitive abilities[34]. More work is needed to understand the role patients can play, and their potential to impact and shape the future of health research.

1.7 Aims of this research

This thesis focuses on developing evidence-based best practice for consumer involvement in CKD research. The specific aims of this thesis are to:

1. Describe the ways in which consumers have been historically involved in CKD research (Chapters 2,4 and 5)
2. Describe the ways in which consumers have been involved in shared decision-making in CKD treatment and care (Chapter 3)
3. Describe consumer perspectives on involvement in decision-making (Chapter 8)
4. Identify effective methods to engage and involve consumers in CKD research (Chapters 2,4 and 5)
5. Describe the impact of consumer involvement on research output (Chapters 6 and 7)
6. Evaluate consumer involvement projects (e.g. conferences) and consumer contributions to research (Chapters 6 and 7)
7. Develop a practical framework for involving consumers in CKD research (Chapter 9)

1.8 Research methods used in this thesis

In this thesis, I used the following research methods: systematic review with a synthesis of qualitative data (Chapter 2), literature review with narrative summary (Chapter 3), semi-structured interviews (Chapters 4 and 6), and focus groups (Chapters 5, 7 and 8).

A systematic review of consumer involvement in published CKD research was conducted (Chapter 2), using an inclusive approach that placed no limits on study/publication type, language or population, in order to thoroughly scope the existing literature in this area. Quantitative methods were employed were possible to demonstrate the dearth of CKD research reporting consumer involvement and to quantify aspects of the included studies. Since most of the data were limited and inconsistently reported, a qualitative synthesis using an a priori coding framework was conducted to provide a systematic and transparent approach to analysis to gain detailed insights into the ways in which consumers had been involved in research and allow for the generation of practical recommendations. Since best practice is in its infancy in this area it was not possible to conduct a quality appraisal of the included studies. Of note, no systematic reviews, randomised controlled trials or cohort studies were identified, justifying the need for further qualitative research.

Semi-structured interviews were conducted with consumers (patients and caregivers) to ascertain detailed insights of their personal and unique experiences in being involved in research more broadly (Chapter 4) and specifically in the scientific meeting of the International Society for Peritoneal Dialysis (Chapter 6).

Simultaneous focus groups were conducted in a workshop setting with consumers (Chapters 5 and 6) and health professionals (Chapter 5) in order to encourage dynamic and broad discussions whereby participants could question and challenge one another, allowing for more complex discussion and justification of their perspectives. Separate focus groups were held with children and their parents (Chapter 8) to allow for rapport building and open conversations about their experiences with decision-making and communication in their clinical care.

Analysis of the qualitative data in this thesis involved techniques adapted from the grounded theory approach and thematic analysis[56-58]. Grounded theory is a methodological approach used in qualitative health research, whereby the theory is derived directly from the data and analysed using a systematic and inductive process[56]. This process is iterative, with data collection and analysis often occurring concurrently, allowing for purposive sampling to ensure diverse perspectives are sought, and for the question guides to be adapted as needed to maximise the breadth and depth of data collected (data saturation)[56]. Thematic analysis is a systematic approach to ‘meaning making’ with qualitative data whereby concepts are identified through line-by-line coding of the data and grouped into themes and subthemes that reflect the attitudes, perspectives and experiences of the participants[58]. Relationships between themes and subthemes can also be illustrated, providing deeper interpretations of the data.

Participant selection

Selection processes for each study are described in detail in the respective chapters, however this thesis included patients with a wide variety of clinical (e.g. diagnosis, CKD stage, treatment modality) and demographic (e.g. country, age) characteristics. For Chapters 4 and 5, consumers were purposively selected for their specific experience and expertise with consumer and research organisations.

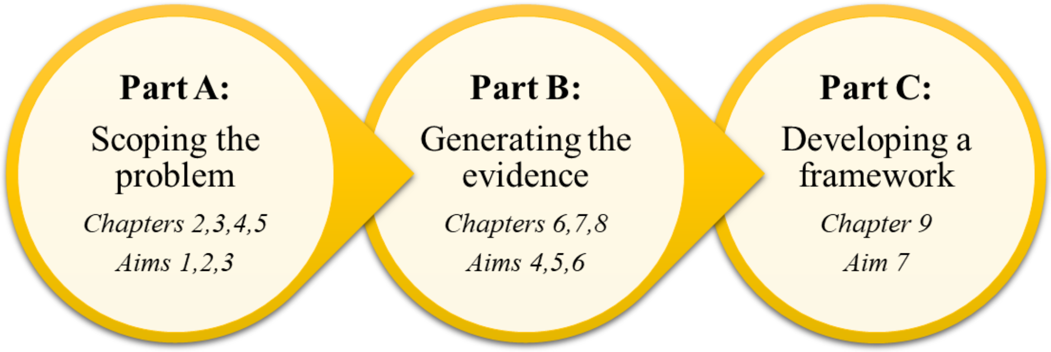
1.9 Consumer involvement in this thesis

Consumers were involved in many aspects of the development of this thesis. Consumers were involved as research partners and co-authors in Chapters 2, 4, 5, and 6. Consumers in these chapters were involved from the inception of the research idea and questions through the conducting, analysis, and reporting of the research. Consumer participants for studies in Chapters 5, 7 and 8 were acknowledged for the contribution of their perspectives and experiences as impacting research processes and outcomes. Specifically, consumer research partner and co-chair of the BEAT-CKD Consumer Advisory Board, Nicole Scholes-Robertson, has served as a constant connection to the consumer voice, providing feedback on all aspects of this thesis. This research program has also been guided by the adjacent BEAT-CKD Consumer Advisory Board.

1.10 Structure of this thesis

This thesis is structured in three parts: A) conceptual development of consumer involvement in research and shared decision-making, B) applied studies on consumer involvement in research and decision-making in real world settings, C) establishing a practical and evidence-based framework for involving consumers in CKD research (Figure 1.1). Chapters 5,6,7 and 8 have been published in international peer-reviewed journals. Chapter 3 is accepted for publication as a book chapter in Evidence Based Nephrology. Chapters 2 and 4 have been submitted for publication with international peer reviewed journals.

Figure 1.1 Thesis structure: Establishing a framework for involvement



Part A: Conceptual Development (Chapters 2,3,4,5)

The first two chapters provide a comprehensive overview of the literature on consumer involvement in research and shared decision-making in published CKD research. Chapter 2 is a mixed-methods systematic review using quantitative and qualitative methods to summarise current practice of consumer involvement in published CKD research. Chapter 3 provides a narrative overview with practical examples and applications of evidence-based shared decision-making in CKD.

Chapters 4 and 5 are primary studies addressing conceptual aspects of consumer involvement rooted in real-world experiences. Chapter 4 is an interview study exploring experiences and attitudes on consumer involvement from the consumer perspective. Chapter 5 reports experiences, perspectives and discussions of both consumers and health professionals from focus groups held at workshops around Australia.

Part B: Applied Empiric Studies (Chapters 6,7,8)

Chapters 6, 7 and 8 provide novel insights into the application, implementation and evaluation of consumer involvement in research and decision-making practices in practical settings.

Chapter 6, an interview study with consumers and health professionals, describes and evaluates experiences involving consumers as partners in scientific meetings including identifying successful elements and opportunities for improvement. Chapter 7 utilises consumer discussions from focus groups to understand from their perspective their priorities and preferences for clinical guideline development for kidney biopsies. Chapter 8 describes the experiences and tensions in clinical decision-making from the perspectives of children with CKD and their caregivers.

Part C: Framework Development (Chapter 9)

Chapter 9, the concluding chapter of this thesis, synthesises the evidence from the preceding studies to present a practical, evidence-based framework for the meaningful, impactful and sustained involvement of consumers with CKD (all stages, diagnoses and ages) across all stages and types of research. It highlights key elements for best-practice and emphasises the need for evaluation, reporting and publishing of consumer involvement activities going forward to strengthen and develop the evidence.

1.11 Thesis summary

This integrated program of work presented in my thesis takes a practical approach to understand the landscape of patient involvement in CKD research, generating novel insights and valuable recommendations from both the patient and health professional perspectives about the potential risks, challenges and benefits of involvement, the mismatch in priorities between patients and health professionals, successful involvement practices and suggestions for improvement. Principles and strategies for successful patient engagement and involvement are identified, culminating in the development of a practical and feasible framework with specific and tangible actions for involving consumers with CKD in research.

Collectively, the findings from these studies generate comprehensive, in-depth and new insights about consumer involvement in research from their own perspective and provide an urgently needed response to the demand from the kidney community to bridge the gap between patients and researchers. The framework could be used to inform strategies to improve the way in which consumers can meaningfully and effectively contribute as partners in research in CKD, which will benefit both consumers and researchers. It will accelerate the culture shift towards patient-centred research in Australia and internationally by providing the means to implement “best practice in consumer involvement” in trials (e.g. Australian Kidney Trials Network, International Society of Nephrology (ISN)), conferences (e.g. Australian and New Zealand Society of Nephrology (ANZSN), International Society for Peritoneal Dialysis (ISPD)) and broader research. It will also empower consumers and consumer organisations such as Kidney Health Australia (KHA) and the BEAT-CKD Consumer Advisory Board through the delivery of evidence-based tools. Additionally, this framework contains highly transferable knowledge that could be valuable in other chronic disease settings.

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Chapter 2: Consumer involvement in chronic kidney disease research: a systematic review

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This chapter is structured as per the journal article.

2.1 Abstract

Rationale and objectives: Limited consumer involvement in published chronic kidney disease (CKD) research may reduce its relevance, impact, and transferability into clinical policy and practice. We describe the landscape of consumer involvement in CKD research.

Study design: Systematic review.

Setting and study populations: Consumers involved in published CKD research.

Selection criteria: Any article type from inception of relevant databases to 2019 that described consumer involvement in CKD research.

Data extraction: Descriptive quantitative data were extracted using a standard form. All text were imported into HyperRESEARCH software for line-by-line coding.

Analytical approach: Descriptive synthesis of the following domains: defining involvement, purpose of involvement, selection of consumers, stages of the research, resources, and evaluation.

Results: Fewer than 0.01% of 792,568 indexed CKD papers were eligible to be included. These 49 articles involved over 2789 consumers from 10 countries. No randomized controlled trials or systematic reviews explicitly discussed consumer involvement. Consumers were identified through clinical and patient networks based on demographic/clinical characteristics and personal experience/attributes. Consumers were least likely to be involved as drivers of research (2%) and most likely to be involved as informants (47%) with limited decision-making power. They were also more likely to be involved in priority setting (55%) and research design (49%) and least likely to be involved in data collection (18%). Qualitative data show involvement resulted in increased recruitment and retention, richer data, and more useful outputs for end users. However, barriers including limited resources and need for tailored solutions exist.

Limitations: Reporting of consumer involvement may be incomplete and not reflect actual conduct.

Conclusions: Around one CKD article in 14,000 reported any consumer involvement. Consumers were most likely to be involved in discrete activities with limited decision-making power. Increasing the provision of financial, logistical and training resources to consumers may support more meaningful involvement. Ongoing evaluation of processes and impacts of consumer involvement, including reporting and publishing, is needed to strengthen evidence and practice of consumer involvement in CKD research.

2.2 Introduction

The misalignment of priorities between researchers and consumers (defined as patients and informal caregivers or family members) and the absence of involvement of consumers in research are well recognized and may limit the relevance and impact of research[1]. Despite increasing evidence that consumer involvement benefits research, for example, by improving recruitment, and increasing the uptake of study findings and patient outcomes[2-5], consumer involvement in chronic kidney disease (CKD) research has been more limited than in other health disciplines, and the value of consumer involvement is not widely understood[6].

A recent review found that 80% of clinical research in CKD does not address the top 10 research priorities identified by patients[7]. Additionally, outcomes consistently prioritized by consumers, such as mortality and fatigue, are infrequently reported in trials[1,8], indicating that finite resources may not be optimally allocated. Patients with CKD face unique challenges that may impact their ability and willingness to contribute to research, including the burden of self-managed and time-consuming essential therapy (i.e. dialysis), and prolonged periods of illness, while the burden of caring responsibilities may lead to burnout for carers.

Many consumer involvement projects and initiatives are not adequately evaluated or reported[5,9-12], and there remains uncertainty around best practice[2,4,11,13,14]. Thus, a better understanding of successful processes and strategies for involving consumers in this population is needed. This review aims to describe consumer involvement in published CKD research to provide a comprehensive understanding of current practice and inform ways to strengthen consumer involvement in future CKD research.

2.3 Methods

We used the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) checklist (where possible) and flow diagram (Figure 2.1) to report this study[15].

Consumer involvement in this review

Three consumers (NSR, DW, CG) with varied experience and expertise (patient and caregiver, CKD stage (including underlying condition, and treatments experienced), prior involvement in research) from two countries that were already connected with the research team were emailed and invited to be involved. Consumers were involved in all stages and aspects of this project in the same capacity as the rest of the research team. Including multiple consumers with different backgrounds allowed for inclusion of diverse perspectives and experiences. One consumer co-researcher (NSR) worked with the lead author to develop the search strategy, screen identified articles, develop the coding framework and conduct the synthesis. All consumer co-researchers provided feedback on multiple versions of the draft manuscript along with the other co-authors, which was used to improve the manuscript and integrated into the final version.

Literature search

Selection criteria: Articles of any type (e.g. randomized trials, observational studies, qualitative research, basic science) that described any consumer involvement in CKD research (across any stage of CKD), in any language, were eligible. Consumer involvement was defined as any activity relating to a research project or program that involved one or more consumers in any aspect or stage of the project[16]. Publications were excluded if they described consumer involvement in clinical care or involved patients as research subjects/participants only. Two reviewers (TG, NSR) independently screened the search results.

Search strategy: We conducted a search of electronic databases followed by a manual search of relevant sources. We first searched for terms relating to CKD and then combined these with an existing search filter for terms related to consumer involvement[17] (Appendix B.1). We searched MEDLINE, Embase and PsychINFO, from inception to October 2019. Additionally, we searched all articles that cited the Guidance for Reporting Involvement of Patients and Public (GRIPP)[18] or GRIPP2[19] checklists, journals on consumer involvement

(Research Involvement and Engagement, The Patient, Health Expectations), and peak organization websites for articles relating to consumer involvement in CKD (Appendix B.1).

Data extraction and synthesis framework

Descriptive data, including year of publication, article type, country, number and type of consumers, consumer activities, stages of research and consumer roles, were extracted using a standard form (Appendix B.2). We imported the complete text of these articles into HyperRESEARCH software for analysis. Using descriptive synthesis, author TG coded the articles using a framework that included the following domains: defining involvement (definitions for involvement, involvement activities, levels of involvement); stating the purpose of involvement (alignment with project aims, purposeful involvement); selecting consumers (selection, number, type, representation, roles); stages of the research; resources (financial, training/education); and evaluation of impact (challenges, benefits, impacts)[6,16,18-20].

2.4 Results

Search results and study characteristics

We identified a total of 470,702 citations from Embase, 313,313 citations from Medline, and 8,553 citations from PsychINFO that were CKD related. Combining these 792,568 citations with search terms for consumer involvement, and a manual search of additional relevant journals and grey literature (n=126), yielded 1192 articles (excluding duplicates). After title and abstract screening of these 1192 articles, 120 full text articles were screened, and 49 (38 from databases and 11 from other sources), met the eligibility criteria (Figure 2.1). While we were unable to export and screen all 792,568 titles for duplicates to determine an exact aggregate denominator, 34 included articles were identified through Embase (0.007%), 26 through Medline (0.008%) and 3 through PscyhINFO (0.04%), allowing us to calculate that a weighted average of 0.007% of all indexed CKD research reported any consumer involvement. Furthermore, these accounted for only 78% of the articles included in the qualitative synthesis as 22% were uniquely identified through other sources.

The included articles (n=49) were from ten countries: Australia (n=15 publications), Canada (n=11), United States (n=9), United Kingdom (n=7), Denmark (n=2), Netherlands (n=2), Ireland (n=1), France (n=1), Korea (n=1) and Sweden (n=1) (Table 2.1). Studies were predominantly qualitative (n=22, 45%), but also consisted of protocols/reports (n=8, 16%), narrative reviews (n=6, 12%), editorial/commentary (n=5, 10%), James Lind Alliance - Priority Setting Partnerships (JLA – PSP) (n=4, 8%), pilot intervention (n=1, 2%), survey (n=1, 2%), mixed methods study (n=1, 2%) and a webpage (n=1, 2%). All were published since 2008. Of note, there were no randomized controlled trial (RCT), systematic review or cohort study identified that reported on consumer participation, and no study from low-income settings. Among the 37 articles that reported numbers of consumers, more than 2789 consumers were involved (including 1124 identified as patients and 237 identified as caregivers) (Appendix B.2). Eight (16%) publications included a designated section/figure to describe consumer involvement in the study[21-28]. Only two (4%) studies used the Guidance for Reporting Involvement of Patients and Public (GRIPP/GRIPP2)[18,19] in their reporting[23,25].

Descriptive synthesis

In the following paragraphs we use the coding framework to describe the involvement of consumers in published CKD research. Where possible, for each of the domains we report the number and proportion of included articles (n=49) that addressed the domain, describe the relevant reported consumer involvement actions, and summarize author reflections and recommendations.

Defining involvement

Eight (16%) articles provided definitions for seven consumer involvement terms: community-based participatory research, patient-researcher partnership, patient engagement, patient/consumer/public involvement, co-researcher, co-production (Appendix B.3). Two referenced the INVOLVE definition[25,29]. Some projects involved consumers in a single activity (e.g. priority setting workshop[30-33]), while others involved consumers in multiple activities across the research process (e.g. consumer partners involved in all activities[9,21,23,25,34-37]). Consumer activities and level of decision-making power are shown in Figure 2.2.

Stating the purpose of involvement

Seventeen (35%) articles addressed the reasons for involving consumers in the research[23,24,29-32,34,37-46]. Four (8%) provided general reasons including improved relevance, importance, quality, knowledge translation, public accountability, and improved health outcomes[29,38,39,45]. Six (12%) involved consumers to identify and/or prioritize topics and outcomes for research[30-32,37,43,46]. Seven (14%) described project-specific purposes including eliciting patient specific knowledge, solutions, feedback and advice; developing consumer materials, involvement strategies, and products; and generating consensus on critically important outcomes[23,24,34,40-42,44].

Identifying and selecting consumers

Nineteen (39%) publications described specific methods and settings for identifying consumers, which included hospitals/clinics[23,32,34,47-51], patient organizations/advocacy groups/charities [9,32,36,37,42,43,48,50-52], social media[36], research networks/professional societies[36,42,43,50], posters/flyers[23,30,49], health care professionals/researchers[9,30,32,34,48], study participants (e.g. for embedded substudy)[24,53] and known consumer research partners/leaders [9,26].

<u>Criteria and considerations for selection:</u> Some studies involved consumers with experience in research[9,33,35,48] (n=4, 8%), with a health or academic background[25,35,37], (n=3, 6%) who held advocacy roles[33,54] (n=2, 4%), or consumers with whom researchers had an existing working relationship[9,48] (n=2, 4%). Others sought consumers with no connection to the research team[23,48] (n=2, 4%) and/or little to no prior experience in research[53,55] (n=2, 4%). Some targeted consumers with specific experience of a treatment or disease [23,31,34,44,47,49,51] (n=7, 14%), who were from disadvantaged and vulnerable groups (n=4, 8%)[26,40,48,52], those who spoke languages other than the country’s official language[26,48] (n=2, 4%), or those who could provide perspectives broader than their own experience (consumer advocates)[25,37] (n=2, 4%). Some researchers were conscious about the disease and treatment burden of CKD and involved more patients to allow for absences or periods of sickness[48] (n=1, 2%). Consumer motivation, education and relevant skills were also considered[25,48] (n=2, 4%).

<u>Reporting characteristics of consumers:</u> Thirty-seven (75%) articles gave details about the number of consumers involved, and of these 36 (97%) included more than one consumer (Table 2.1)[9,23-28,30-38,41-44,46,47,49-63]. Thirty-six (73%) provided one or more characteristics of

consumers they involved including gender, disease stage and ethnicity (Appendix B.2)[9,23-28,30-38,40-44,46,47,49-53,55-64].

Representation and inclusion: While some authors described seeking diverse consumers based on age, employment status, CKD stages/diagnoses[28,30,43,48,53,55], some indicated that vulnerable and minority ethnic groups (including Asian, African American, Indigenous Peoples) were difficult to reach and engage[43,46,50]. The need for demographic "representation" among consumers was challenged, noting that this was not required for other members of the research team[41].

Defining consumer roles: Consumers held various roles ranging from observing, informing and engaging with researchers from their personal experience (e.g. information sessions, surveys, focus groups) to driving research projects (e.g. patient led research network). Table 2.2 shows the range of consumer roles with descriptions and examples of responsibilities for each. Some consumers felt there was a lack of clarity about their roles and responsibilities - "there was no profile of what is expected of a lay co-researcher, and no guidance on the activities I would need to take part in"[25] and suggested the need for a structured job description or terms of reference.

Stages of the research

Consumer involvement across the stages of research is shown in Table 2.1.

Involvement in setting priorities: Twenty-seven (55%) articles addressed consumer involvement in setting research priorities[26,28,30,31,35-40,43,45,46,48-53,55,58,59,61,62,65-67], with most using the JLA - PSP approach (n=7)[32,38,43,50,53,55,59]. Consumers were also involved in prioritizing topics for clinical guidelines (n=6)[28,31,40,51,52,58]. Priorities were elicited through involvement as members of the steering committees, surveys, and workshops.

Involvement in research design: Consumers contributed to protocols, advised on acceptability, feasibility and practical aspects of interventions and study design, and provided input on recruitment and retention (n=24, 49%)[9,21,22,25-28,34-36,39,42,44,45,47-49,53,55,58,60,62,63,65]. Consumers were primarily involved through advisory or reference groups, steering committees, or in workshops and provided insights and ideas that led to changes and improvements in study/intervention design (e.g. preference for overnight dialysis rather than continuous wearable device[42], selection of outcomes and outcome measures (including core outcomes for trials)[44,47,48], developed recruitment materials[23], improved readability of materials[39,62,63]). Consumers helped to ensure that study design and materials were culturally safe and inclusive for minority groups[26,35,58,60,62].

Involvement in data collection: Consumers contributed to the development of questionnaires and interview guides, and conducted interviews and focus groups, (n=9, 18%)[22,23,25,26,35,37,39,48,63]. They established rapport whereby participants felt “no barriers in explaining their stories”[37], and patients identified ways to ensure consumers felt safe participating (e.g. having a native speaker/translator in the interview[26]). Consumers identified strategies to address barriers to recruitment (e.g. provide a patient navigator/advocate[35]).

Involvement in data analysis: Fourteen (29%) articles described involving consumers in data analysis[21-26,37-39,48,53,55,58,63]. Consumers were predominantly involved in thematic analysis of qualitative studies (n=7)[21,23-26,37,55]. Consumers provided feedback on preliminary findings, worked with researchers to analyse the data, or led analyses. Authors noted that partnering with consumers in analysis yielded more nuanced findings and unique insights of the consumer's context not captured by researchers, for example related to the burden of dialysis[37].

Involvement in disseminating findings: Consumers presented findings at conferences, gave lectures at meetings and events, authored papers, produced lay summaries, developed educational materials (i.e. videos, brochures) and attended conferences as delegates[9,21-23,25,35-37,39,45,48,68] (n=12, 24%). Twenty-two (45%) articles included consumer co-authors[9,25,30,33,35-37,41-44,47,50,52,53,55,57,59,60,65,67,68], including four consumer first authors[9,25,57,68]. Fourteen (64%) of these also acknowledged additional consumer contributors at the end of the paper[25,30,33,36,37,41-44,47,53,55,60,65]. Nine (18%) articles included this acknowledgement without consumer co-authors[24,27,28,32,34,46,48,51,62].

Involvement in implementation of findings: Consumers implemented study interventions through pilot testing, providing feedback and identifying perceived barriers to the interventions (n=10, 20%)[23,24,26,33,35,42,48,51,57,63]. As an example, consumer advisory council members modified the implementation of an intervention to ensure it "reflected the unique culture and circumstances of the community"[26]. Consumers were involved in knowledge translation activities (e.g. videos, educational materials), guiding implementation of a new model of care, enacting legislation (access to dialysis[57]), streamlining regulatory decision-making processes (e.g. medical device approval with FDA), and selecting topics/outcomes for guidelines.

Involvement in Evaluation: Thirteen (27%) articles involved consumers in evaluating the project/intervention. Methods included informal discussions[9] (example output: role descriptors were useful to set expectations), workshops/meetings[26,27,48,58] (e.g. identified consumer educational needs/interests; meaningful intervention for First Nations people), consumer testimonials[42] (e.g. increased machine portability, automatic transmission of treatment data, and remote monitoring), surveys/written feedback[28,36,42,60,62,64] (e.g. need for simpler language) and interview studies[24,53] (e.g. preference for in-person over online participation). Consumers also communicated the impact of the project back to participants and the broader community, using plain language and promoted future projects.

Resources

Some studies described the resources necessary for the involvement of consumers, including financial support and compensation to aid involvement, and human resources needed to provide education, training, and logistical support.

Financial support: Thirteen (27%) articles mentioned financial support for consumers, including travel grants to consumers to attend a workshop[42], reimbursement for travel[9,25,26,28,32,46,47], and a stipend/honorarium[9,25,48,58,62]. Two included budgets for consumer involvement[22,25] and one reported specific funding for a consumer co-researcher[25]. One stated no remuneration was provided[56]. It was recommended that consumers be reimbursed for expenses incurred (time off work, transport, accommodation and childcare costs)[39], and providing compensation would allow for more diverse involvement[41].

Education/training: Training included formal workshops (for consumers and researchers)[9,23,26,34,59], informal discussion-based learning/feedback tailored to specific tasks[25,36], and learning-by-doing approaches[25,37], and focused primarily on research methods and subject matter content. Other support provided for consumers included induction/orientation programs, mentoring, peer-to-peer support and liaisons who provided consumers with practical, logistical and technical support (e.g. accessing videoconferencing, supporting mobility needs)[23,48]. Providing education/training was found to enable consumers to make more meaningful contributions[41,42].

Evaluation of impact

Sixteen (33%) publications reported evaluation of the consumer involvement activities and most were informal and brief[9,22,23,25,26,28,31,34,36,37,43,48,55,60,63,66]. The benefits, challenges and long-term impacts from the researcher and consumer perspectives are provided in Table 2.3. The principles and strategies identified as leading to beneficial and meaningful involvement are shown in Table 2.4.

2.5 Discussion

Fewer than 1 in 10,000 indexed CKD research papers included any description of consumer involvement. Most articles that reported any consumer engagement were qualitative studies, and no RCT, systematic review, or cohort study was able to be included, meaning that the most highly cited and influential studies in CKD that are used to inform decision-making do not involve consumers. While the frequency of articles describing consumer involvement do appear to be slowly increasing over time, in most occasions when consumers are engaged in research, their involvement is limited and rarely positioned to impact meaningfully on design and conduct. Those involved at higher levels (e.g. co-researcher) were often highly educated, with some medical or academic experience, further marginalising already disadvantaged and vulnerable groups who are often more impacted by CKD. Research leadership by consumers was scarce, emphasizing a need for mentorship and training of consumers by the kidney research community.

Included studies do demonstrate to the kidney research community that involvement is feasible. Consumers were involved in varied activities and roles, with different levels of decision-making power, across all stages of the research cycle. Authors described diverse methods for identifying potential consumer partners and pathways to involving them in research, supported by clear justification for their inclusion. Authors also described principles and strategies employed to maximize benefits and mitigate challenges, and outlined the resources needed to support consumers, including financial, logistical and personnel. Reported outcomes of these consumer involvement initiatives included improved communication, increased recruitment, retention and adherence, richer data collection, validation of findings, improved knowledge translation, more useful outputs for end users and increased agreement on priorities between stakeholders.

We noted some differences by research stage in terms of the activities and number of consumers involved. The majority of consumers were involved in single large group activities (e.g. workshops, surveys) in the preparation phases (i.e. priority setting and design) with minimal opportunity to provide feedback or ensure that their voices have been carried through to the execution and translation phases, which has been echoed in other reviews[2,14,69]. Although consumer involvement in data collection and analysis in qualitative research yielded better quality data[2], in these stages fewer consumers were involved, and this was typically through a co-investigator role or as members of a consumer advisory group.

Whilst we conducted a comprehensive search of articles on consumer involvement in published CKD research and a descriptive synthesis according to an explicit framework, there are some potential limitations. There is inconsistent and variable reporting of consumer involvement in clinical research, leading to potential ascertainment bias. Of the included studies, few focused solely on consumer involvement processes/activities - most embedded a brief description of consumer involvement. All articles were from high income, predominantly English-speaking countries, echoing the need for further documentation of consumer involvement in research in low-income settings to establish whether this imbalance is due to a paucity of awareness, activity or of reporting in low-income settings[70].

This review highlights opportunities to strengthen consumer involvement based on evidence from published studies for the consideration of the kidney research community, including: identifying and selecting consumers; setting expectations; support and capacity building; and evaluating processes and impact (including consistent reporting and publishing of involvement).

Researchers often report difficulties in identifying potential consumers to involve[71]. From the evidence, we suggest that consumers be included to provide diverse perspectives, and selected based on their relevant experience of CKD, capacity to fulfil the specific roles as required by the research project, and personal attributes (e.g. skills, experience, motivations, knowledge, and expertise)[71,72]. Due to the practicalities and purpose of involving consumers, and the relatively small number involved, diversity should take precedence over representativeness[13,71]. Also, care should be taken to ensure inclusion of vulnerable and disadvantaged groups, requiring financial and logistical support, and special knowledge of the relevant community. Of note for researchers working with patients with CKD or carers, prognoses of CKD can be unpredictable and may impact upon their motivation or ability to be involved, and mechanisms to provide additional support and flexible approaches may be required.

Developing a role description or terms of reference clearly outlining the responsibilities, tasks, and the time required can clarify expectations regarding involvement[11,14]. It is recommended that consumers be inducted to the research project/team and be provided any necessary information and materials, and that researchers work with consumers to determine mutually acceptable schedules and explain timelines to avoid conflict or disappointment. Having mechanisms in place at the start to resolve conflict or concerns that may arise and providing feedback to consumers on the value and impact of their contribution may also help to provide support and reassurance.

Providing financial and logistical support may be useful for sustaining ongoing and meaningful involvement[14] and may allow for broader participation[29,41]. Costs that might be considered include reimbursement for expenses incurred to participate (e.g. travel, childcare, accommodation, meals) and payment for their time. INVOLVE provides useful policies, tools and calculators to help budget for these needs, and recommends compensation of USD$200 per person per day for consumer involvement activities[73,74]. Logistical support, particularly with technology (e.g. videoconferencing) or access (e.g. building access, online resources), and practical support for any impairments consumers may have may also be required. Having a ‘research buddy’ can help consumers feel supported and integrated in the research team. Longer term, building reciprocal relationships that are not tied to an individual project (e.g. salaried co-researchers, advisory boards), and research training programs/pathways (e.g. research methods, postgraduate courses) are needed to build capacity to support active involvement in the execution and translation phases of research[14,71].

Formal evaluation of consumer involvement processes and outcomes has been limited. In future studies evaluation outcomes could be quantitatively assessed (e.g. impact on recruitment and adherence, community knowledge/awareness). However, it is difficult to develop best practice for consumer involvement while reporting and publishing remains scant. While the GRIPP/GRIPP2[18,19] checklists have been developed to improve reporting, only two (4%) studies used this guidance in their reporting[23,25]. More is required to strongly encourage or mandate the reporting of consumer involvement as has been done by some journals (e.g. BMJ). We note that it may not be possible to include all the reporting items from GRIPP2 in a manuscript, however these details could be included in supplementary files or warrant a separate publication. Alternatively, existing frameworks for individual study types (e.g. CONSORT, STROBE) could be amended to include important items relating to consumer involvement in a table or figure, or require justification for not involving consumers.

Historically, consumer involvement has rarely been reported in published CKD research. Fewer than 1 in 10,000 indexed CKD research papers were eligible to be included, thus limiting potential to influence decision-making. However, the included studies demonstrate that involving consumers in all aspects of with CKD research is possible and beneficial. To build on what has been done, future research could aim to include diverse perspectives and support patients’ health needs, focus on developing organisational processes to adequately prepare consumers and researchers for their roles, and provide financial and logistical support, training, and mentorship to consumers to develop ongoing and meaningful involvement, including in later stages and with more decision-making power. In a broader context, formal and published evaluation of consumer involvement processes and impacts are required to strengthen the evidence for best practice in kidney research, alongside effort from the kidney research community to adapt to the evolving science of best practices for the inclusion of consumers as it develops in other health disciplines.

Acknowledgements

We thank the consumer co-authors (NSR, CG, DW) for their important contributions to this work. We would also like to acknowledge the thousands of consumers who have participated in the included research projects.

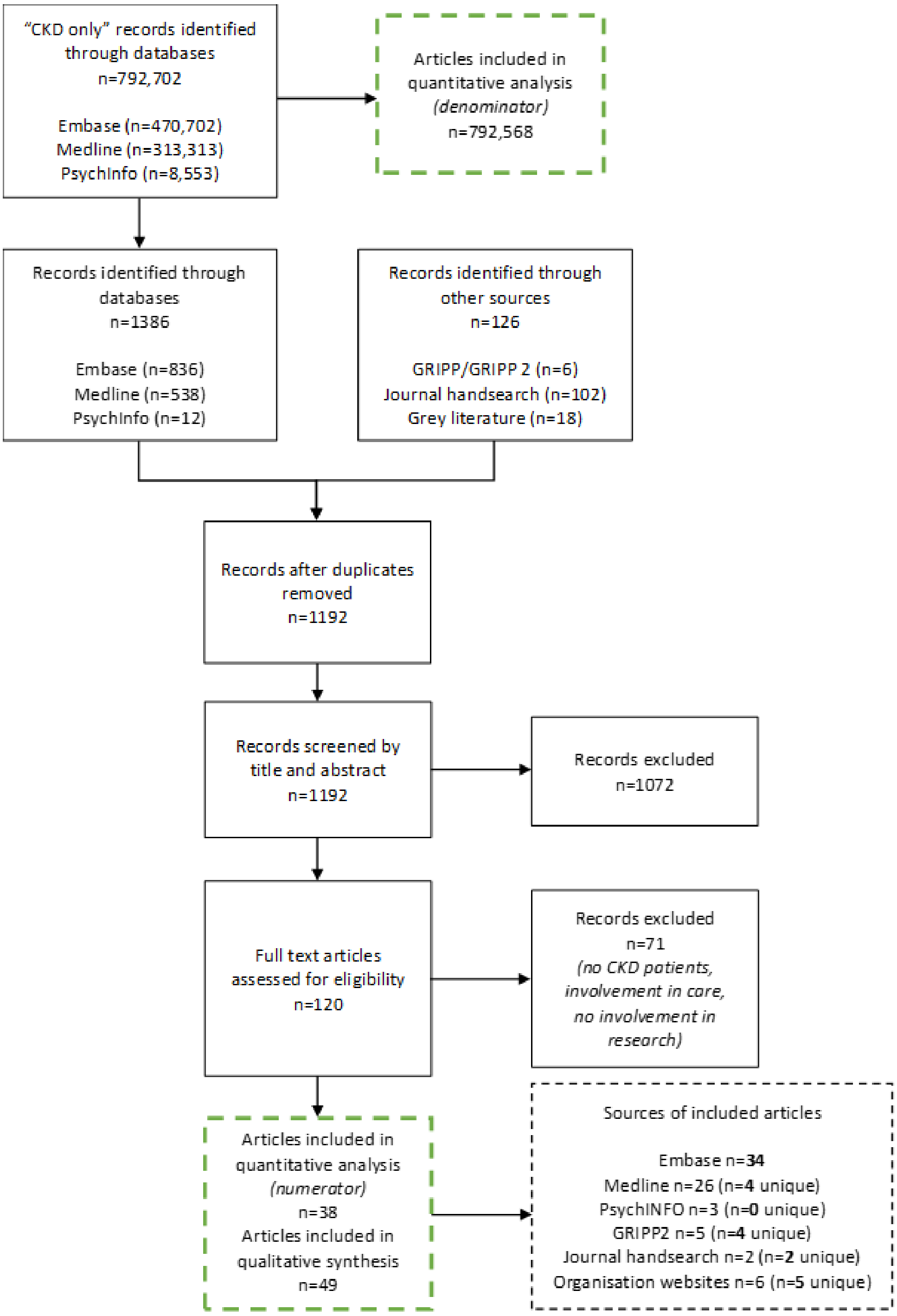

Figure 2.1 Search results

The PRISMA flow chart shows the records identified in the literature search. 792,702 records relating to CKD research were identified through Embase, Medline and PsychINFO. Of these 1386 records also related to consumer involvement in research. An additional 126 records were identified through other sources. After screening, 39 records from the databases and 10 records from other sources were eligible for inclusion.

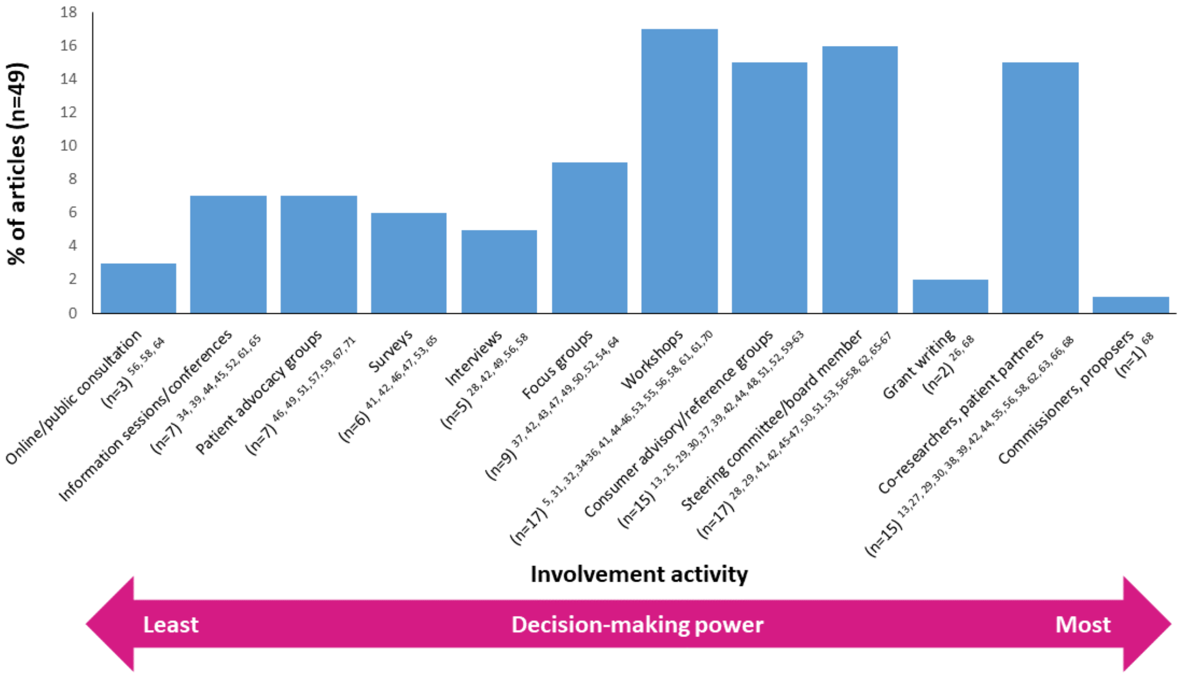

Figure 2.2 Involvement activities

Figure 2.2 shows the range of activities that consumers were involved in in the included articles, as well as the proportion of articles that described each activity, and the relative decision-making power consumers held, from least to most.

Table 2.1 Characteristics of included articles (n=49)

Characteristic	N (%)
Year published	
<2010	1 (2)
2010-2015	14 (29)
>2015	33 (67)
Not reported	1 (2)
Article type	
Qualitative methods	22 (45)
Protocol/report	8 (16)
Review	6 (12)
Editorial/commentary	5 (10)
Priority setting partnership	4 (8)
Other*	4 (8)
Number of consumers included^	
1-10	12 (24)
11-30	12 (24)
31-100	6 (12)
101-200	2 (4)
>200	5 (10)
Not reported/applicable	12 (24)
Country	
Australia	12 (24)
Canada	11 (22)
United States	11 (22)
United Kingdom	9 (18)
Other**	12 (24)
Consumer roles	
Attendees	3 (6)
Informants	23 (47)
Advocates	10 (20)
Advisors	18 (37)
Experts	11 (22)
Partners	17 (35)
Drivers	1 (2)
Stage of the research cycle	
Priority setting	27 (55)
Design	24 (49)
Data collection	9 (18)
Data analysis	14 (29)
Dissemination	12 (24)
Implementation	10 (20)
Evaluation	13 (27)

*Pilot study, survey, mixed methods, web page; **Denmark, France, Ireland, Korea, Netherlands, Sweden, International; ^consumer numbers not comprehensively reported, numbers indicate minimum number of consumers involved.

Table 2.2 Consumer roles and responsibilities

Role*	**Definition***	**Example responsibilities**
Attendees	Receive information from researchers/experts with limited opportunity to contribute *(i.e. educational seminar, information session)*[36,49,62]	Participate in online forums Attend meetings, conferences
Informants	Provide personal perspectives/experiences of condition/treatment/care *(i.e. surveys, interviews, focus groups)*[24,28,30,31,34,37,38,40,42-47,50-53,55,58,59,61,62]	Relay and relate personal experiences, provide testimony (regulatory) (e.g. managing fluid intake, usefulness of devices) Identify and prioritize (rate/rank) research questions, outcomes, topics Provide preferences (e.g. aims, topics, formats, outcomes, trade-offs)
Advocates	Represent broad range of perspectives/experiences *(i.e. patient organisations, patient representatives)*[30,33,48,52,54,56,57,64,67,68]	Provide guidance around cultural safety and sensitivity, diversity Apply personal experiences to broader community/population needs Liaise between research and consumer communities
Advisors	Provide advice based on experiential knowledge *(e.g. advisory group)*[21,25,26,28,36-38,41,45,48,49,52,56-58,60,63,64]	Attend meetings/phone calls/teleconferences Monitor and review progress Provide feedback – written/verbal (e.g. search terms, protocols, recruitment/enrolment strategies) Advise on communication strategies with participants/consumers Provide advice and guidance on recruitment strategies Develop study/patient materials (e.g. interview guides, educational materials/sessions, support tools, consent forms, lay summaries, newsletters, progress reports, cultural messages)
Experts	Provide high quality knowledge and expertise as end user *(i.e. board member with voting rights)*[9,24,25,27,33,38,43,44,47,48,54]	Attend meetings/phone calls/teleconferences Alert research team to potential risks, ethical issues Provide advice, insights and guidance on implementation (e.g. core outcomes) Participate in strategic and practical decision-making
Partners	Participate as full members of the research team *(i.e. co-researcher)*[9,23,25,26,35-37,41,42,50,52,53,55,59,60,62,63]	Attend meetings/phone calls/teleconferences Connect the research team with the greater consumer community (e.g. community coordinator, consumer liaison, buddy) Contribute to study design/methods (e.g. study question, outcome selection, outcome measures, recruitment materials, power calculations, data collection tools, intervention design) Conduct interviews

		Contribute to/conduct analysis, interpretation and presentation of results (e.g. qualitative analysis, validation of themes, contextualizing quantitative data) Write recommendations Disseminate research findings (e.g. present at meetings/conferences, write papers for patient/academic publications)
Drivers	Lead, propose and drive research projects/agendas *(i.e. patient led research network)*[65]	Apply for grants, funding Identify project aims and objectives, define scope Monitor and review progress Govern networks/bodies/organizations (i.e. identify and prioritize research questions, allocate funding Conceptualize, design and implement sub studies (e.g. PROM sub study)

** Adapted from National Framework for Consumer Involvement in Cancer Control, Cancer Australia 2011*[6]

Table 2.3 Challenges, benefits and impacts

Evaluation component	**Examples from included papers**	
	Predominantly affecting consumers	**Predominantly affecting researchers**
Benefits to the research project	Improved content and clarity of information[22,60] Improved communication (type, language, format and timing of information)[9,34,36] Encouraged adherence to timelines[23] Improved knowledge translation activities (e.g. educational videos, lay summaries) [23,28,55] More useful outputs for end users (clinicians, consumers)[22,28] Increased agreement on priorities between stakeholders[66] Cultural safety, sensitivity[26]	Increased recruitment rates[22,66] Increased retention/adherence rates[22,63] Improved relevance/importance of topics, interventions[28,31,36,60] Validation of findings[22] Richer data collection (interviews)[23,37] Captured research findings missed by researchers (questions, themes, topics, nuances) [23,31,37,43]
Challenges/ barriers	Lack of shared language (lay versus scientific/medical jargon)[48] Limited research training[48,55] Timing/scheduling, need for flexibility and time commitment [48,55] Transparency around contributions/impact[48] Navigating different timeline expectations, time between meetings, adherence to timelines, time to implementation [23,25]	Navigating dynamics between researchers and consumers (including consumer partners and consumer participants)[34,37] Understanding ethics requirements for including consumers as partners in research[23,48] Resistance of researchers towards questions generated by non-scientists[48] Involving broad/diverse consumers ("engaged predisposition")[55] Documenting/recording PPI activities[25]
Long term impacts	Enhanced patient advocacy (consumers representatives, researchers, clinicians)[25,55] Continuity of involvement in future projects[55] More empathy/compassion for and understanding of one another (consumers and researchers/clinicians) [9,23,26,34,55] Sense of empowerment/meaning for consumers[48] Support/perspective/community derived from meeting other consumers[55] Enhanced knowledge and understanding for consumers (disease progression, symptoms, treatment options, general kidney health, research process)[9,55] Confidence for consumers to advocate themselves in clinical settings[9,55]	Shift in research interests/focus to be more patient-centred[55] Enhanced knowledge and understanding for researchers (patient-oriented research, how to involve consumers, value of involvement) [9,55] New/modified clinical approaches (e.g. including consumer priorities in clinic questionnaire, guidelines)[28,31,55] Connection between the research team and the broader community[26] Patient Reported Outcome substudy development[63]

Table 2.4 Strategies and principles for successful involvement

Principle	Strategies
Avoiding tokenism	Build ongoing, trusting and authentic relationships[35,39,41,65] Encourage open dialogue throughout[37,39,48,68] Engage in meaningful activities[68] *(e.g. involve consumers early and in upstream decision-making with the rest of the research team)*
Respecting consumers' time and capacity	Clarify roles, responsibilities and deliverables (include in study protocol)[35,39,41,68] Develop a Terms of Reference/ Memorandum of Understanding[9,26,39] Minimise burden on consumers (time, financial)[25,26,40,41,68] Schedule activities on days/times that enable attendance[25,26,36] Schedule activities in locations accessible to consumers (or consider access e.g. university/hospital buildings)[9] Elicit preferences for communication modalities - offer multiple modes of attendance/participation, where possible allow for face-to-face meetings[9,36,41,53]
Valuing consumer knowledge and expertise	Provide regular project updates/results[41] Demonstrate impact of involvement[42,48,53] *(e.g. provide written feedback to consumers to show how their contributions impacted the research)* Build capacity (e.g. training, peer mentoring)[41,42] Reimbursement for time and expenses[9,39,41]
Consideration for the patient journey	Be sensitive to patient wellness/treatment stage[41,63] Allow for varied degrees of involvement according consumer preferences[48] Involve more than 'needed' with the understanding that attendance may fluctuate ('over-recruit')[48,63] Involve consumers in determining timelines[26]
Considering the context of the 'whole' person	Understand their skills and interests, aside from being a 'patient' or 'carer'[35] Work within their availability – consider existing work, personal and family/social commitments[25,41,68] Accommodate language/learning needs[48] *(e.g. provide translator; conduct in own language; ensure appropriate communication for level of education)* Respect diversity and differences[37] "Remember the humanity of the person"[68] *(e.g. remember that being a patient is only one aspect of a person's life and they will have competing priorities including family, career, social)*
Sharing of power and providing support	Provide orientation/induction[39] Buddy/liaison role[39,48,63] Provide practical support where needed (e.g. technical – videoconferencing, transport)[25] Involve consumers in decisions regarding training desired/required[9,48] Use plain language[41,42] Training for researchers[41]

	Provide 'safe' environments to empower contributions[37] *(e.g. ensure proper consent and confidentiality; establish relationships with community leaders)* Involve more than one consumer in any group[37,51,63]
Developing consumer networks/systems to support continued involvement	Establish a board/panel/council of consumers who are ready to become involved[41,48] Identify opportunities for future involvement[35] Cultivate partnerships/networks with patient advocacy organizations and health care professionals[41,42,48] Explore opportunities for web-based platforms/networks[41]
Reciprocity	Facilitate bi-directional knowledge exchange and translation[41,52] Provide access to education/training/tools[42] Empower health ownership[41] *(e.g. provide education/training about disease management)*
Inclusion of perspectives	Involve diverse consumers[65] Where possible and relevant, look to include vulnerable and minority groups[48]
Valuing the local	Engage local community members as 'cultural brokers'[40,52] *(e.g. employ a community leader/member as a co-researcher and support them to liaise between the community and the research team)*
Cultural safety and competence	Understand the distinct needs and expectations of community groups[40] Acknowledge importance of spiritual beliefs[26] *(e.g. engage with the community to understand how their beliefs can be acknowledge and accommodated)* Provide regular updates to and seek feedback from the community, include contact information and photographs of the research team[26] Establish ground rules for confidentiality[9]

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Chapter 3: Shared decision-making

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This chapter is structured as per the book chapter.

3.1 Introduction

The chronic kidney disease journey, from diagnosis to end of life care, includes many complicated treatment decisions that require patients and physicians to consider the risks and potential benefits of life extending treatments and therapies. The impact of treatment decisions on a patient’s life vary significantly from minor changes in their daily routine, for example having to begin daily medication to control blood pressure, or limiting salt intake in their diet, to life-altering changes that accompany life prolonging therapies, such as thrice weekly hemodialysis. The trade-off between burdens and benefits of a given treatment will differ for each patient depending on their values and preferences. Many patients rely on their clinicians for guidance on treatment decisions that are not only in their best prognostic interests, but also align with their values and preferences. Over the last few decades, the clinician’s role in treatment decision-making has shifted from paternalism to partnership, and it is now recognized that clinicians should engage in shared decision-making (SDM) to determine the most appropriate course of treatment for each patient.

Box 3.1 Case example

Ms. K is a 75 year-old wheelchair-bound woman with progressive stage 4 kidney disease due to heart failure and diabetes who is seen in nephrology clinic for management of advanced kidney disease. She requires assistance for bathing and dressing and recently moved into a personal care home. The nephrology clinician has cared for Ms. K over three years and notes a progressive decline in function and health. Reflecting on the treatment decisions that she and Ms. K will address, the clinician asks herself:

- How do I help Ms. K make the right decision to manage her advanced kidney disease?
- How do I prepare Ms. K to make the right decisions to manage her future end of life care?

Nephrology clinicians frequently face situations such as in the example (Box 3.1) above in which they inform and guide patients to make treatment decisions about their kidney disease and how to manage accompanying setbacks. In addition to dialysis modalities and transplantation, comprehensive conservative care (CCC) is an elected treatment path that emphasizes quality of life and symptom management. For older patients especially those with advanced conditions, dialysis may not offer meaningful survival or quality of life[1-5]. Patients managed conservatively are more likely to have their symptoms alleviated, spend survived time out of the hospital and receive dedicated end of life care with services such as hospice[6,7]. Yet, patients are less likely to understand these prognostic trajectories, nor the option to forego dialysis and choose CCC[8].

Shared decision-making (SDM) in nephrology care is an essential component to ensuring patients make informed decisions that reflect their priorities. In SDM, clinicians and patients work together to make decisions that balance the potential risks and expected benefits with patient preferences and values. This chapter provides a summary of the evidence for SDM in nephrology care, particularly for older patients with advanced comorbidities facing difficult decisions about treatment of advanced kidney disease and preferences for end of life care. This chapter includes a definition of SDM, communication strategies to facilitate clinicians in engaging in SDM, and patient decision aids aimed to prepare patients to consider their values and preferences with the treatment decision.

3.2 Definition

Shared Decision Making has been defined as the meeting of two experts – the patient and clinician - with the goal of working together to make the best health care decision for the patient. The concept of SDM is countered by traditional models including paternalistic and informed models[9] (Figure 3.1). Within the paternalistic model, the clinician is viewed as the expert and so use their skills to determine the best treatments based on the patient's medical condition and circumstances. The paternalistic model assumes that the clinician knows what is in the patient's best interest and hence the flow of information and deliberation is one-directional with minimal input from the patient. A major limitation of this model is the assumption that the clinician and patient espouse similar values and views of what qualifies as benefit.

The countering model on the other end of the decision-making spectrum is the informative model. Sometimes referred to as the consumer model, the informative model shifts the role of decision-making to the patient. Within this model, the clinician provides the necessary information about the disease process, possible treatments and expected risks and benefits with the expectation that the patient decides what is best[9]. This model lacks essential elements of the clinician-patient relationship including helping patients to deliberate and recommending treatment decisions based on their values and preferences.

SDM responds to the limitations of these traditional models and addresses real world challenges to decision-making within health care. In the case above, the right choice for how Ms. K manages her kidney disease and future end of life is not straight-forward and depends on her individual values and priorities.

Charles and authors define SDM as requiring four necessary elements to address these preference sensitive decisions in which no best answer exists[10]. First, at a minimum the treatment decision-making process must involve the clinician and patient. Second, the process is bidirectional, meaning both the clinician and patient share information with each other. Third, both the clinician and patient actively participate in the decision-making process by sharing treatment preferences. Fourth, the final treatment decision is made and agreed upon by both the clinician and patient.

3.3 SDM in Nephrology

Frameworks & Processes

The RPA and Choosing Wisely Campaign recommend nephrology clinicians engage in SDM to address complex treatment decisions such as dialysis initiation versus comprehensive conservative care and preferences for end of life care[11,12]. Yet patients describe insufficient preparation and involvement in treatment decisions. In one qualitative study of US dialysis patients over the age of 65, no patient felt they made the choice to initiate dialysis[13]. Rather these decisions were either influenced by the perception of imminent death without dialysis or by the clinician. Among 197 dialysis patients in Canada with an average age of 53 years, 48% reported that treatment decisions were made by their clinician, with only 25% reporting shared responsibility[14].

A fundamental component of SDM in advanced kidney disease involves providing patient specific prognostic information. Patients report rare prognosis discussions which influence how decisions for end of life care are made. In an interview study, incident hemodialysis patients who reported an overly optimistic prognosis were more likely to elect life-prolonging treatments if they were seriously ill, even if this meant suffering or discomfort[15]. Although there are emerging efforts to develop and validate various risk predictions models into usable forms for clinicians such as online calculators or integrated within the electronic health record, these most often focus on mortality risk and do not provide individualized information about other outcomes important to patients[16]. This omission of relevant prognostic information may in part explain why dialysis patients are more likely to receive intensive therapies at end of life and less likely to receive quality end of life care such as hospice services[7,17].

Another component of the SDM process is for the patient and clinician together understand the patient's values and priorities (Table 3.1)[18]. In a survey study conducted in Australia and Canada, hemodialysis patients and their caregivers prioritized important outcomes in hemodialysis patients. They defined survival in terms of living well, with mortality ranking only 14th in terms of importance[19]. These findings reinforce that clinician focus on treatment decision-making discussions must address more than just survival alone[20].

SHARE Approach to SDM

Multiple SDM models exist, however all reinforce the importance of prognostic information and the elicitation of patient values and priorities to guide treatment decisions[10,21-23]. The SHARE Approach is a five-step process for SDM endorsed by the Agency of Healthcare Research and Quality[23]. These steps include 1) Seek the patient's participation; 2) Help the patient explore and compare treatment options; 3) Assess the patient's values and preferences 4) Reach a decision with the patient and 5) Evaluate the patient's decision. These steps are best accomplished through patient-centered communication strategies. The communication framework Ask-Tell-Ask supports a two-way conversation in which the clinician elicits information from the patient (Ask) before giving relevant information (Tell) and then assesses how the information impacted the patient's understanding (Ask).

Seek the patient's participation (Step 1): The first step of the SHARE Approach is to invite the patient's participation in SDM. This can involve establishing rapport, reinforcing patient's autonomy, and the patient's preferences for decision-making. Both clinician and patient are expected to collaborate proactively in this decision-making process by exchanging information about the illness, diagnosis and treatment from their areas of expertise; outlining explicit values and preferences in the context of care pathways and/or lifestyle; reasoning together to determine the best option for the patient; and agreeing and implementing the choice that aligns best with clinical evidence and patient preference. Establishing rapport and trust is an important aspect of seeking patient participation. This involves gaining an understanding of the patient's experience and culture which may involve traditions, beliefs and values based on cultural norms[24,25]. In addition, seeking participation also involves determining the patient's desired level of involvement in the decision-making process. Some patients may prefer a passive role deferring decision-making to a loved one or may elect a clinician-centered approach that is more directive[26]. This preference to limit their involvement or defer the final decision to the clinician can be respected and the SHARE approach can be adapted to support these decision-making preferences and needs.

The first *Ask* of the *Ask-Tell-Ask* framework starts with gaining the patient and/or family's input rather than reflectively disclosing information. Open-ended questions, such as 'What have the doctors told you about your kidney disease?', allow the clinician to explore what the patient understands about their condition. These exploratory questions also allow the clinician to learn decision-making preferences, cultural experience and belief system (Box 3.2).

Box 3.2 Seek patient's participation (Step 1)

The clinician caring for Ms. K wants to learn about her experience with her disease and learn must how much involvement Ms. K wants in her treatment decisions, who, if any, are the other people that will be involved in the decision making process. To determine these important aspects, the clinician asks Ms. K:

- Can you tell me about the impact your treatment has had on your life so far?
- As your clinician, what would be helpful for me to know about you and your life?
- How much involvement have you had in other medical decisions? Has this worked for you? Why/Why not? Do you want to do this differently?
- Who would we include in these discussions and decisions?

Help the patient explore and compare treatment options (Step 2): This step involves detailed explanations and discussions of the potential treatment options available to the patient, including the expected benefits and risks associated with each one. This step addresses the *Tell* of the *Ask-Tell-*Ask framework. This *Tell* should include information about treatment options, including prognostic information, and should be given in a way that is clear without medical jargon.

Discussions about prognosis are often avoided until forced, either in the setting of decline or after prompted by the patient. This may be due to the inherent uncertainties regarding prognosis. While prognostic tools exist to help predict how a patient might respond to a given treatment, significant uncertainty remains for how a patient will actually do[27-29]. This uncertainty challenges the clinician’s skills in guiding treatment decision-making and helping prepare patients for living with a life prolonging treatment. Yet, many patients do want information about what to expect in the future, and prefer that this information be given voluntarily by the clinician[30].

Understanding what prognostic information the patient desires can influence the kinds of information the clinician should provide[31]. Prognostic information should include both survival and quality of life to ensure the patient receives information that addresses their personal concerns. During discussions of treatments options, it is helpful describe the different treatment paths in terms of the expected benefits or potential risks (“Choosing to start dialysis may extend your life, but it may also mean that you spend more time in hospital and depend on others for help” (Box 3.3).

Box 3.3 Help explore and compare treatment options (Step 2)

Ms. K’s nephrology clinician considers the relevant treatment options available and tailors the prognostic information based on Ms. K’s clinical condition to include both survival and quality of life:

Option 1 – Trial of dialysis: “For patients who are older with other advanced conditions, dialysis may add little additional time and survived time will likely include setbacks such as loss of function and more hospitalizations.”

Option 2 – CCC: “For patients similar to you, choosing conservative care may lead to shorter time however time lived is focused on managing your symptoms and quality of life, without life prolonging treatments such as dialysis.”

Assess the patient’s values and preferences (Step 3): After sharing prognostic information and relevant treatment options, the conversation should shift from giving information (*Tell*) toward eliciting goals and values (*Ask*). This process ensures the decision is guided by what matters most to the patient, such as quality of life, level of independence, and preferences for care at the end of life. This also may be integrated throughout the therapeutic relationship between patient and clinician over time. In a small study of 99 dialysis patients reflecting on their recent experience with selecting a treatment option for advanced kidney disease, 20% reported that their doctor asked about their values and preferences[8]. In a recent study of 271 patients with stage 4 or 5 non-dialysis dependent CKD from one center, patients most commonly identified maintaining independence as their top priority (49%). However, when the clinician was asked about the top priority of the patient only 31% (95% CI: 26% to 37%) were in agreement[20]. There continues to be significant opportunity to increase the activities in clinical care in support of eliciting and understanding values and preferences.

Learning what matters most to patients involves learning patient goals and values. These ‘big picture’ goals and values are best elicited with open-ended questions that explore hopes for, as well as worries about, the future[32-34]. By asking about hopes for the future, the patient can consider the values and priorities that are most important and whether these values and priorities support a treatment focused on quantity or quality of life. In addition to understanding what the patient hopes for in the future, it is as important to explore what the patient hopes to avoid. These concerns may be related to physical suffering, psychological distress and being a burden to loved ones. Asking the patient about their worries provides insight into the conditions or health states that a patient hopes to avoid. For patients whose goals are focused on comfort, worries may include going back to the hospital, being away from the family or living in dependent condition (Box 3.4).

Box 3.4 Assess the patient’s values and preferences (Step 3)

After giving Ms. K all the relevant treatment information it is now time to elicit her priorities for treatment:

- Given this news, what is most important to you?
- As you think about the future, what concerns you the most?
- What do you want to avoid in the future?

It may be helpful to give an example comparing the two treatment options with specific potential consequences:

“Some people want to live as long as possible and choose dialysis even at the risk of frequent hospitalizations and less independence. Other people wish to focus on the quality of their lives and prefer treatments that are focused on their symptoms without dialysis even if this meant life could be shorter. Do you have a sense of how you feel about this?”

Reach a decision with the patient (Step 4): The fourth step involves offering a treatment recommendation that incorporates the prognostic data and the patient goals. For patients with older age and advanced comorbidities, the relevant treatment options are trial of dialysis (hemodialysis or peritoneal dialysis) versus CCC. Many patients desire a recommendation from the clinician while a minority of patients do not. Therefore, it is helpful to *ask permission* to provide a recommendation. If the patient agrees, the clinician can proceed with a recommendation that is grounded in the patient's own goals and values (Box 3.5).

Box 3.5 Reach a decision (Step 4)

The nephrology clinician asks Ms. K's permission prior to offering a suggested treatment plan based on what she has shared with you:

- From our discussion I have a sense of what things are most important to you. Would it be ok if I offer my suggestion about a plan moving forward?

Ms. K agrees, and the clinician offers a recommendation that is supported by Ms. K's goals and priorities:

"Based on your goals to spend as much time with your family and away from the hospital even if this means shorter time, I would recommend we pursue conservative care with a focus on your quality of life and we will not do dialysis as this would not achieve your goals. How does that sound to you?"

Evaluate the patient's decision (Step 5): After the treatment path is elected, the SDM process continues and includes opportunities to reassess and revisit the care plan. First, it is difficult to prepare a patient for exactly what to expect living with dialysis or CCC. Words often fall short compared to the lived experience. Second, uncertainty makes it difficult to adequately prepare patients for what to expect even with prognostic guidance. Third, many patients may choose dialysis as a better alternative compared to death. Therefore patients, especially those who elect dialysis, benefit from evaluation of the decision and anticipatory guidance for future end of life needs[35] (Box 3.6). A recent study noted that it is equally important not to continually question a decision once it has been made, as this may be perceived as badgering and threaten the trust previously developed in the clinician-patient relationship[36]. Determining the intervals of re-evaluation of the treatment decision should be a part of the collaborative plan.

Box 3.6 Evaluate the decision (Step 5)

Patients who elect trial of dialysis especially benefit from evaluation of the decision to assess whether the treatment is meeting the patient's goals without causing unwanted burdens:

- "It's been two months since we started dialysis, what has been your experience?"
- "When we started dialysis, we hoped that it would help you get stronger and back home. Have you been able to accomplish this goal?"
- "What concerns do you have with continuing dialysis?"

Evaluating the decision also involves preparation for future setbacks and end of life. Advance care planning (ACP) is a continuous process through which providers engage in discussions with patients and their families to learn their goals and preferences toward future medical treatments[37]. ACP conversations allow patients to reflect on their care preferences to inform decision-making in future declining health states. Patients who had have an ACP are more likely to receive care that is consistent with their care goals including end of life care such as hospice[38]. Dialysis patients are less likely to have engaged in advance care planning conversations. Thus, end of life is often experienced in the hospital following life sustaining treatments with less opportunity for quality hospice care[7]. In U.S. nursing home patients with dementia on dialysis, those who engaged in early ACP by completing an advance directive and naming a surrogate decision-maker had fewer hospitalizations, less intensive care, and fewer invasive procedures compared to those who lacked both[39].

The SHARE Approach is generally supported by efforts to increase clinician communication skills and self-efficacy in the provision of SDM. Educational materials in kidney disease can be used in Step 2 when delivering information about treatment options, however decision tools specific to the SDM process that aim to deliver prognosis information aligned with values (Step 3) are helpful to reach a collaborative treatment decision (Step 4).

3.4 Decision Tools

Decision aids are informational documents that present educational and prognostic information about the health condition. Information then is presented about the treatment options, and when available information about outcomes including side effects (Figure 3.2). Decision aids do not make recommendations about a specific treatment and they do not replace evaluation and counseling from a clinician. Optimized decision aids present the capability to customize the information presented to the patient using the decision aid. The International Patient Decision Aid Standards (IPDAS) Collaboration has developed criteria to assess the quality of patient decision aids using a checklist with 28 different criteria[40].

Although there has been a growing interest in the development and provision of decision aids in nephrology, few have been tested in trials to determine their impact on outcomes. The most common outcomes influenced by decision aids include addressing decisional uncertainty, knowledge and understanding, and congruence between the decision and patient values[41]. A recent review of decision aids for renal replacement therapy described 10 decision aids, and another 4 that were more specific to conservative care[42]. Most decision aids had yet to be rigorously tested for efficacy on either decisional or clinical outcomes. The reports available support that the various decision aids are generally accepted by patients (Table 3.6). One small study examined the online self-directed decision aid Chooosingdialysis.org which provides information about all treatment options but focuses on dialysis modality decisions (in-center hemodialysis or peritoneal dialysis). It was tested in 63 people (average age 59 years) and demonstrated increased kidney knowledge and less decisional conflict after its use[43]. There was no change in decisional self-efficacy score. The largest studies reported are in the context of increasing the use of kidney transplantation as a therapy option, and have shown variable results. None of the studies describe the process of using a decision aid during the clinical encounter compared to self-directed use. There are substantial gaps in knowledge regarding the use of decision aids to facilitate SDM in kidney disease care, and importantly critical gaps in understanding their influence on decisional, behavioral and health outcomes in diverse patient populations.

3.5 Barriers and Opportunities

A review of the evidence reveals potential barriers to SDM, many of which can be attributed to poor communication strategies, highlighting opportunities to address these barriers in future.

Perceived power imbalance with emphasis on lack of information: A systematic review of qualitative studies found that patients believed that the health care team were considered to “own” the knowledge and it was up to them to decide what patients should know[44]. A study that included interviews of 35 nephrologists from 9 U.S. states found that they generally attributed failed discussions to factors related to the patient rather than self-reflection of their own skills[45]. Participants in an interview study identified the use of medical jargon by health professionals as confusing[46]. This perception may be amplified when the patient has low health literacy which is associated with lower perceived knowledge (r_s=0.19; $p<0.0001$) among patients with advanced chronic kidney disease[47], and patients with higher perceived knowledge express greater satisfaction with their clinician's communication (Adjusted Odds Ratio: 2.13 (1.59, 2.86); $p<0.0001$). Training to improve clinicians' communication skills may directly address these potential barriers, and this may become a priority as nephrologist communication metrics have been incorporated into value-based payment systems in the United States[48].

Patients identified a lack of information about all available treatment options as a barrier to actively participating in their treatment decisions[49]. Delivery of information was labeled as "inconsistent" and "accidental", leaving patients feeling that they did not have all the information they needed to participate in SDM[44]. Complete and systematic delivery of information, using effective communication strategies, early in the diagnosis may help to address these issues. A recommended communication strategy to ensure that Step 2 is effective and that patients understand the content of the information is to perform a teach-back whereby as an example the clinician asks the patient to share how they would explain the concept or skills to their family member. This method has effectively improved self-care in kidney disease hypothesized to be due to better patient knowledge, self-efficacy and practical skills[50].

<u>Medical crisis:</u> In some cases patients believed that there was no time or opportunity to engage in SDM when there is a need for urgent treatment or because of a physiological

contraindication (e.g. no vascular access)[18,44,49]. While urgent treatment decisions may be necessary in a small proportion of patients, for most there is time to present the treatment options to the patient and determine their preferences before proceeding. Also identified as a barrier was the timing of when the information was presented – some explained that it was left too late and they were too unwell or did not have enough time to absorb the information in order to make a decision[49]. Although it is important to determine if a patient is cognitively suitable to participate in SDM, once this is confirmed using the SHARE Approach steps include the option for patients to decline SDM at the time of invitation by the clinician, as well as opportunities to process the informational content while performing Step 4 to collaborative reach a treatment decision.

A Transitional Care Unit (TCU) program is a novel healthcare system delivery approach to address the concern regarding those who have advanced to end-stage kidney disease, who have not yet developed a treatment decisional plan, but now their medical status precludes them from fully participating in SDM[51]. This program emphasized stabilization of medical status paired with a comprehensive educational curriculum that purposefully employed SDM techniques and decision aids. While the programs efficacy is yet to be evaluated it demonstrates that with leadership buy-in SDM can be core to the mission of integrated kidney care.

<u>Lack of treatment options:</u> Clinicians may perceive dialysis to be the only relevant treatment options and may therefore guide the discussion with dialysis as a default decision. In a study of Veterans Administration clinician documentation from the electronic health record, clinicians were more likely to push dialysis even when patients strongly verbalized a wish to forego dialysis[52]. Three themes that emerged described: 1) dialysis as a norm in which clinicians felt obligated to repeat or reexamine the patient’s decision to forego dialysis; 2)

when clinicians decided dialysis was not a viable option, this decision was justified based on biomedical factors rather than patient goals and values; and 3) when it was determined dialysis would not be pursued, clinicians felt they had little to offer patients beyond dialysis and so often signed off from their care. Increasing education and comfort in providing CCC are strategies to overcome this ‘all or nothing’ view of dialysis[53].

Other barriers identified were no perceived benefit of participating in SDM, lack of time during typical clinical visits, a lack of family support and limited health care resources (e.g. availability of dialysis chairs)[46,49]. Opportunities for SDM are emerging within the context of kidney transplantation organ allocation. One study found that 63% of patients wished to participate in decisions about quality of the organ[54]. This may become even more relevant as other biomarkers, including genetic information, are used for estimating risks related to graft survival.

3.6 Conclusions

Shared decision making is a patient-centered approach to ensure patient values and priorities inform their treatment decisions. SDM is especially relevant for treatment decisions for advanced kidney disease as well as end of life care. The SHARE Approach provides a framework for supporting SDM that ensures treatment decisions reflect relevant prognostic information and what is most important to the patient. Decision tools can help patients consider the treatment options in terms of their values and priorities. Barriers to SDM can be overcome through two-way communication, avoidance of medical jargon and providing patient with necessary decision tools to guide the process.

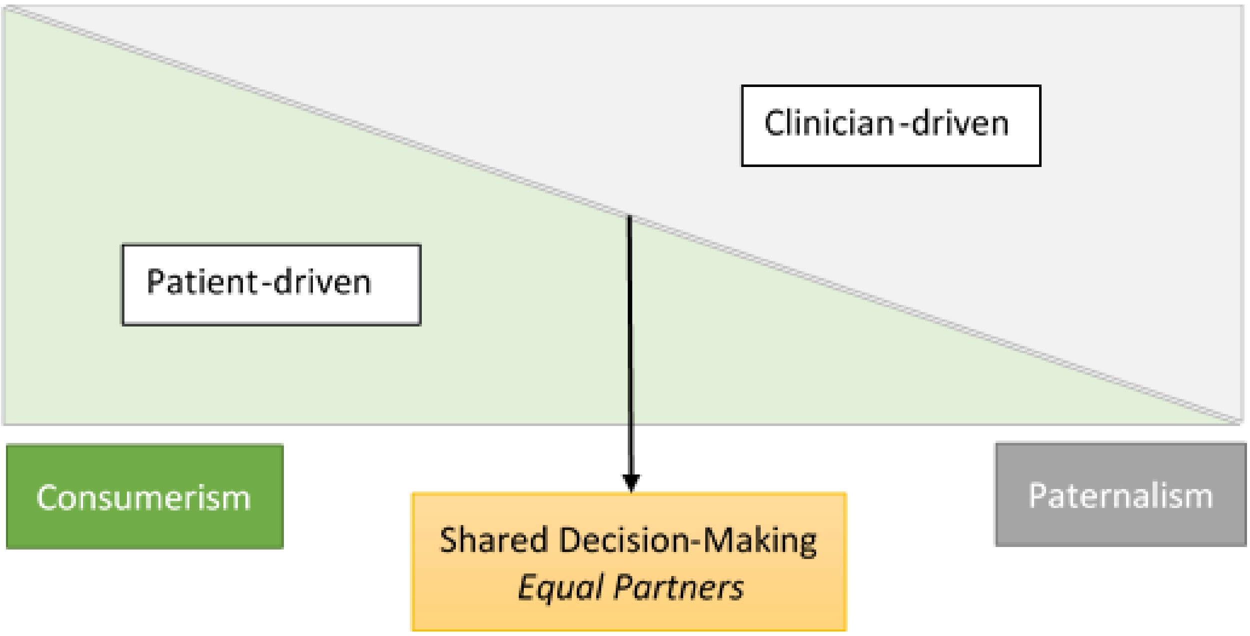
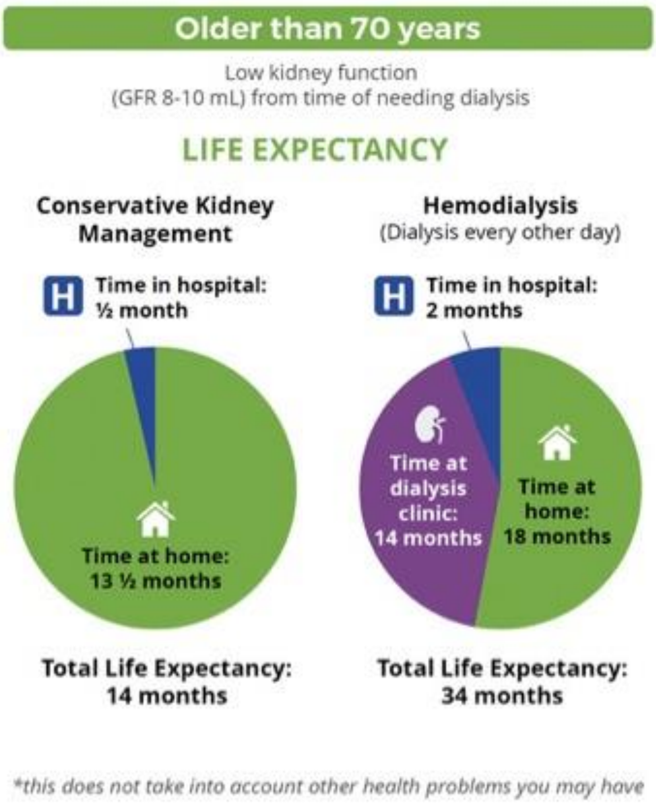
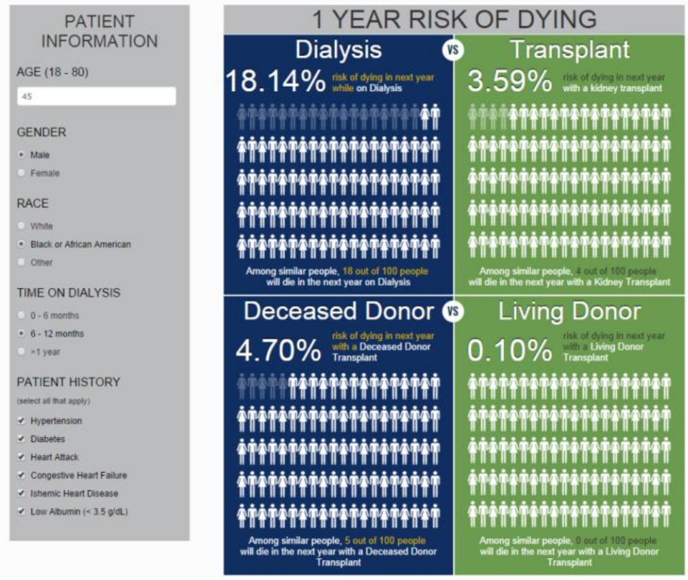


Figure 3.1 Continuum of Patient-Clinician Decision-Making Models



(a) www.ckmcare.com[55]


PATIENT INFORMATION

AGE (18 - 80)

45

GENDER
● Male
○ Female

RACE
○ White
● Black or African American
○ Other

TIME ON DIALYSIS
○ 0 - 6 months
● 6 - 12 months
○ >1 year

PATIENT HISTORY
(select all that apply)
☑ Hypertension
☑ Diabetes
☑ Heart Attack
☑ Congestive Heart Failure
☑ Ischemic Heart Disease
☑ Low Albumin (< 3.5 g/dL)

1 YEAR RISK OF DYING

Dialysis VS Transplant

18.14% risk of dying in next year while on Dialysis

Among similar people, 18 out of 100 people will die in the next year on Dialysis

3.59% risk of dying in next year with a kidney transplant

Among similar people, 4 out of 100 people will die in the next year with a Kidney Transplant

Deceased Donor VS Living Donor

4.70% risk of dying in next year with a Deceased Donor Transplant

Among similar people, 5 out of 100 people will die in the next year with a Deceased Donor Transplant

0.10% risk of dying in next year with a Living Donor Transplant

Among similar people, 0 out of 100 people will die in the next year with a Living Donor Transplant

(b) ichoosekidney.emory.edu

Figure 3.2 Examples of Kidney Decision Aid Content

Table 3.1 Priorities identified as important to patients with advanced kidney disease[18]

Maintain independence	Physical appearance
Flexibility in daily schedule	Social relationships (family & friends)
Privacy	Burden on others
Comfort of environment	Safety related to treatments
Reliability of treatment timing & schedule	Work or school

Table 3.2 Characteristics of included studies

Study ID	N	Study design	Setting	Participants	Topic
Dahlerus et al 2016	180	Semi-structured Interviews	United States	18 years or older, eGFR < 25ml/min/1.73m3 or on dialysis for >3 months	Perspectives on modality choice
Davison et al 2006	24	Ethnographic study with interviews	Canada	Purposive sample: 44-88 years (mean 64), 50% women, n=1 first nations Aboriginal, n=23 white	Perspectives of salient features of effective facilitated advance care planning
Fortnum et al 2014	NA	Development of decision aid	Australia and New Zealand	NA	Shared decision-making: dialysis vs. conservative care
Galla et al 2000	NA	Clinical practice guideline	United States	NA	Shared decision-making in the appropriate initiation of and withdrawal from dialysis
Harwood et al 2012	410	Qualitative synthesis	Six databases	>18 years with chronic kidney disease	Dialysis modality decision-making
Holley et al 2007	614	Observational survey – 2 timepoints	United States	Nephrologists	Changing clinician attitudes and practices of end of life decision-making
Hussain et al 2015	270	Qualitative synthesis	Four databases	Patients aged 26-93 years, health professionals aged 26-61 years	Factors affecting patient or health professional decisions to commence or withdraw from dialysis
Joly et al 2003	146	Cohort study	France	Consecutive pre-ESRD octogenarians referred to nephrology unit	Factors affecting clinician and patient choice of dialysis or conservative care in octogenarians, and impact on survival
Moattari et al 2012	48	Randomized controlled trial	Iran	Patients on hemodialysis	Effect of empowerment on self-efficacy and quality of life (including decision-making)
Morton et al 2010	462	Qualitative synthesis	Six databases	Patients with chronic kidney disease and caregivers	Decision-making and choice for dialysis, transplantation or palliative care

Table 3.3 Impact on patient-centered care and outcomes

Study ID	Impact on patient-centered care and outcomes
Dahlerus et al 2016	More informed/engaged patients who were active in decision-making chose options that better fit their priorities. Important outcomes that impact decision-making were quality of life, convenience of at home therapy, ability to work, risk of infection and medical expertise.
Davison et al 2006	Most patients believed health care providers wanted to know their preferences for advanced care planning and believed this would influence outcomes of treatment. Patients based their decisions on the impact of the care plan on their personal lives and wanted to know how interventions could help achieve personal goals e.g. avoiding hospitalization.
Holley et al 2007	Significantly more nephrologists in 2005 were willing to stop dialysis in a severely demented patient (53% versus 32%, P<0.00001) and to withhold dialysis from a permanently unconscious patient (90% vs. 83%, P<0.001). Most nephrologists in both surveys would respect the request of a competent patient to stop dialysis. When asked whom they would consult to assist in decision-making for each of these hypothetical cases, <98% would consult the patient’s family in each scenario in both 1990 and 2005. In 2005, compared with 1990, nephrologists are more likely to honor a dialysis patient’s wish for DNR status
Hussain et al 2015	For many patients the effects of treatment on their quality of life was more important than medical effectiveness – quality of life was prioritized over longevity/survival. As life on dialysis deteriorated, patients questioned its worth and considered withdrawal. Knowing they had this option gave them control. Health professionals depended on biomedical criteria to guide decision-making rather than patient preference, delaying withdrawal until medically necessary.
Joly et al 2003	Aside from patient refusal, late referral, social isolation, low functional capacity and diabetes may have oriented the medical decision to withhold dialysis.
Moattari et al 2012	A significant change was observed between the groups in terms of stress reduction (adjusted mean difference −3.6 (-4.4 to -1.57), p<0.02) as well as the decision-making dimension (adjusted mean difference −3.21 (-4.39 to-2.03), p<0.001) and overall self-efficacy scores (adjusted mean difference −12.02 (-18.41 to -5.53), p<0.001). Also, a significant difference was found in the overall mean score of quality of life and in all dimensions of the quality of life between the groups (adjusted mean difference −2.93 (-4.39 to -1.47), p<0.001).
Morton et al 2010	Medical outcomes of treatment were considered less important than the effect of treatment on patient's lifestyle. Longevity was viewed as less important than quality of life.

Table 3.4 Communication

Study ID	**Communication – re: prognosis, preferences**
Davison et al 2006	Patients wanted more information earlier in the disease process – this promoted self-reliance, alleviated fear/uncertainties, helped prepare them for future and gave them the knowledge to make decisions that were compatible with their values and beliefs. Patients wanted clear honest discussions in lay language. They explained that jargon caused confusion and misunderstanding. They believed physicians were responsible for initiating discussions around end of life care.
Hussain et al 2015	More information about their future was considered better than none by patients, despite fears/uncertainty this may raise. It was important to patients that information was delivered by a trusted person.
Morton et al 2010	Framing of treatment/interventions by physicians impacted patient decisions e.g. Transplant framed as a six-hour operation with risk of death was seen as not worth it by some patients, despite increased rate of survival.

Table 3.5 Barriers to SDM

Study ID	Barriers to SDM
Dahlerus et al 2016	Medical crisis/need was prioritized over patient choice. Physician control over decision limited patient involvement in decision-making.
Davison et al 2006	Patients less likely to actively engage in decision-making process if they did not perceive any benefit (e.g. aligned priorities). The use of medical jargon was confusing for patients and discouraged them from participating in decision-making. A lack of family support also prevented patients from participating in shared decision-making.
Hussain et al 2015	The perceived power and dominance of the health care team hindered patient involvement in medical decision-making. The health care team were considered to "own" the knowledge and they decided what the patient should know. Lack of time for acutely unwell patients meant decisions were made without including them. Inconsistent and "accidental" delivery of information left patients unable to participate in shared decision-making as they did not understand their treatment options and the respective consequences of each.
Morton et al 2010	Lack of information about all available treatment options limited patients' involvement in their treatment decisions. Physiological contra-indications (e.g. no vascular access) eliminated treatment options in the eyes of physicians in favor of biomedical outcomes. Limited health care resources (e.g. availability of chair/kidneys) meant patients options were also limited. Problematic timing of presenting information to patients by physicians meant they were sometimes too sick or did not have enough time to absorb information

Table 3.6 Nephrology decision aids in support of SDM with peer-reviewed evidence

Study ID	Description of Decision Aid	Summary of Decisional Outcomes
Kidney Supportive Care Research Group[55]	Title: Conservative Kidney Management: Is Dialysis Right for Me Format: Web-based, printing capabilities Topic focus: Comparing dialysis options and conservative care Values clarification exercise: Yes Availability: ckmcare.com/Resources/details/PDA	Not available
Fortnum et al 2015[56]	Title: My Kidneys, My Choice Format: online Topic focus: Comparing dialysis options and conservative care Values clarification exercise: Yes Availability: https://mydialysischoice.org/	N=97 Pre- Post design Increase in perceived kidney knowledge Increase in worry
Boulware et al. 2018[57]	Title: PREPARED Format: brochure, video Topic focus: Comparing dialysis, transplant and conservative care using checklists Values clarification exercise: No Availability: http://ckddecisions.org/prepared-materials/	N=30 DA; N=31 usual care; N=31 DA + financial assistance No change in transplant preparation behaviors at 6 months 68% reported they there considering which pros and cons were more important to them
Kapell Brown et al 2018[58]	Title: CPR-VDA Format: Video & CPR decision worksheet Topic: Code status considerations in end-stage kidney disease Values clarification exercise: Yes Availability: http://vimeo.com/48147363	N=49 patients; N=8 family Pre- Post design Knowledge score increase (p<0.0001) Decisional self-efficacy increase (p=0.005) 72% expressed no decisional uncertainty
Subramanian et al 2018[43]	Title: Choosingdialysis.org: empowering patients for choices on renal replacement therapy Format: Web-based, printing capabilities Topic focus: Peritoneal Dialysis, In-center Hemodialysis; brief information/links other options Values clarification exercise: Yes Availability: free; choosingdialysis.org	N=63 DA; N=70 control Decisional Conflict Score: DA group: Pre 44.3 (16.0); Post 29.1 (13.7); p<0.0001 Kidney knowledge: DA: 90.3 (11.9); Control: 765 (15.3); p<0.0001 No difference decisional self-efficacy
Patzer et al 2017[59]	Title: iChoose Kidney Format: online, printing capabilities Topic: Mortality risk related to dialysis and transplant, incorporating patient variables Values clarification exercise: No Availability: http://ichoosekidney.emory.edu/	Randomized trial N=226 DA; N=216 control Knowledge improvement greater in DA than in control (1.1+ 2.0 versus 0.4 + 1.8; p<0.0001) Transplant access similar (74.3% versus 70.5%)

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Chapter 4: Patient and caregiver experiences and attitudes towards involvement in research in chronic kidney disease: an interview study

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This chapter is structured as per the journal article.

4.1 Abstract

Background and objectives: While patient and caregiver involvement in research is widely advocated to improve the relevance and uptake of study findings, barriers and uncertainties in achieving this goal remain. This study aimed to describe patient and caregiver experiences and perspectives of involvement in research to inform strategies to strengthen patient involvement in research.

Design, setting, participants and measurements: Semi-structured interviews were conducted with 23 adult patients with chronic kidney disease (CKD) and caregivers from Australia, United States, United Kingdom and Denmark. Transcripts were analyzed thematically.

Results: We identified six themes: Grappling with CKD diagnosis (overwhelmed by the burden of illness, silenced by stigma and shame, absence of advocacy and awareness), bearing the responsibility for involvement (autonomy in their own care, forced to be proactive to access opportunities, infrastructural support to connect researchers and patients), battling big agendas (struggling in a system of disincentive, changing research culture, becoming equals), seeing the person behind the patient (harnessing broader knowledge, expertise, skills and interests, understanding patient needs, bolstering impetus), sensitivity to complexities of payment (accounting for individual circumstances, denoting value, enabling diverse involvement), and championing the patient voice (links to important stakeholders, drivers of innovation, responsibility to end-users).

Conclusions: The burden of CKD, limited opportunities, and power asymmetry power between patients/caregivers and researchers were challenges to meaningful involvement in

research. Building trust, awareness of opportunities, and recognizing the broad expertise and value of patients/caregivers, including and beyond their illness experience, may better support patient and caregiver involvement in research in CKD.

4.2 Introduction

Involving patients, their families and caregivers as more than ‘subjects’ or ‘participants’ in research is now widely advocated by funders, regulators, patient and professional organizations, and journals and to align research with the priorities and needs of patients/caregivers and researchers[1-7]. Patient involvement in study goals, design and conduct can improve recruitment and retention, and the reliability and uptake of study findings[8-11]. While there are increasing efforts to involve patients and caregivers in research in CKD, only 20% of clinical research addresses the top 10 patient priorities[12]. Barriers and challenges include the need for resources to provide logistical and financial support, and adequate and appropriate training for both patients/caregivers and researchers[9,10,13,14].

Patients with chronic kidney disease (CKD) depend on time-consuming and invasive treatments with inflexible schedules and contend with many, symptoms, complications, and unpredictable events which can impede capacity for involvement[15,16]. Caregivers also have considerable responsibilities that may limit their ability to be involved in research. These burdens amplify the difficulties faced in identifying and navigating research systems.

There is limited evidence on the perspectives of patients and caregivers with CKD on being involved in research[15-17]. This study aims to describe patients’ and caregivers’ experiences and attitudes to being involved in research in CKD across the research cycle[6] (including priority setting, designing research, collecting and analysing data, disseminating and implementing study findings and evaluating studies), to strengthen and inform best practice approaches for involvement in research in CKD[18].

4.3 Methods

Participant selection

People with lived experience of CKD (including patients and caregivers), who have been involved in at least one stage of the research process[6] (e.g. priority-setting, study design) in any role (e.g. Steering Committee member, investigator), and were English-speaking and aged 18 years or over, were eligible to participate. We applied a purposive sampling strategy to capture a diverse range of demographic (i.e. age, gender, country, profession) and clinical characteristics (i.e. CKD stage, diagnosis), and experience being involved in research (e.g. elected position, paid role, volunteer). Participants were identified through professional and patient networks (Appendix C.1) and invited by email to participate. Informed consent was obtained from all participants. Ethics approval was granted by The University of Sydney.

Data collection

TG and AK conducted semi-structured interviews with participants face-to-face or through videoconference (Zoom Video Communications, Inc) from September 2017 to March 2021. The interview guide was developed based on a review of the literature[10,13,14,19], and discussion among the research team. All interviews were audio recorded and transcribed verbatim. We conducted interviews until we reached data saturation.

Analysis

TG read the transcripts and inductively identified preliminary concepts and grouped these into themes using thematic analysis[20]. Themes were revised based on discussion with AT, AK and NSR, to ensure the coding structure captured the full range and breadth of the data[21]. The transcripts were coded line-by-line to the revised themes using HyperRESEARCH software. We conducted member checking by emailing the preliminary findings to participants for feedback, and additional concepts were integrated into the final analysis[22].

4.4 Results

We contacted 32 patients or caregivers and 23 were interviewed (nine did not respond or were not available). Of the 23 participants, 18 (78%) were patients and 5 (22%) were caregivers from Australia (n=10), United States (n=10), United Kingdom (n=2) and Denmark (n=1). The participant characteristics are shown in Table 4.1. The duration of interviews ranged from 35 to 85 minutes. Of note, participants most frequently reported being involved in priority setting (87%), reviewing documents (78%) and recruiting participants for studies (61%).

We identified six themes: grappling with the CKD diagnosis, bearing the responsibility for involvement, battling big agendas, seeing the person behind the patient, sensitivity to complexities of payment, and championing the patient voice. The following section describes the subthemes. Selected quotations for each theme are available in Table 4.2. Figure 4.1 shows the relationships between themes and subthemes.

Grappling with CKD diagnosis

Overwhelmed by the burden of illness: Living with CKD was “all encompassing” and treatments, such as dialysis, were “time-consuming” and “scary”. Some felt unable to participate fully when they were “wiped out” or only had “one or two good days a week”. With “the goal to go back to your normal life”, patients did not “want to spend 24 hours a day thinking about [their] health” and wanted to “distance themselves from an all-pervasive disease” and were thus reluctant to be involved. Becoming involved in research sometimes meant facing their own mortality. Some found it difficult to share their experiences as they were “cognitively challenged”, “fragile emotionally” and had lost “confidence” in themselves, and downplayed symptoms such as pain, depression and anxiety to “present a brave front”, which were identified as barriers to being involved in research. Caregivers felt “wired” into their roles and found it difficult to take on responsibilities over and above keeping the health of their loved one stable.

Silenced by stigma and shame: Participants noted the stigma related to identifying as a patient and were thus hesitant to share their experiences due to the shame and blame from others - “oh you did drugs and you used alcohol”. Misunderstanding from the broader community that kidney failure was self-inflicted or communicable, and blame from dialysis healthcare professionals around compliance and fluid overload, caused patients to “shut down”, preventing further involvement in research.

Absence of advocacy and awareness: Participants explained that “kidney disease isn’t sexy” and were frustrated by the low level of public awareness compared with other diseases such as cancer and HIV, which downplayed the impact and severity of CKD. Diagnosis at a late stage limited personal awareness and was a barrier to earlier involvement. Some suggested that public advocacy by well-known individuals such as “Shaquille O’Neal” or “Selena Gomez” to “get the word out” would encourage more involvement in research initiatives.

Bearing the responsibility for involvement

<u>Autonomy in their own care:</u> Participants, particularly from the United States, believed health systems pushed patients to “check [their] independence at the door”, discouraging further engagement and leading to an “inactivated population”. They viewed involvement in their own care and learning to advocate for themselves in a clinical setting as a critical first step to being involved in research and noted this was more successfully achieved in other health disciplines (e.g. hemophilia). This also allowed them to develop trusting relationships with their clinicians, opening opportunities to become involved in research.

<u>Forced to be proactive to access opportunities:</u> Participants “scoured the web” or social media for opportunities to be involved in research and had to “learn a new language” to decode jargon and acronyms. Some “internally driven” participants contacted researchers or research groups, but noted “people like me are very unusual” and “not everyone can be as proactive”. Some gave up looking for opportunities as they repeatedly never heard back from researchers.

<u>Infrastructural support to connect researchers and patients:</u> Participants preferred to be invited by researchers rather than searching out research opportunities. Yet they were frustrated at researchers “having to recruit again from scratch” for each project and failing to “re-engage the people who are already interested”. Participants suggested researchers develop databases to disseminate opportunities or match researchers and patients/caregivers based on interest and experience.

Battling big agendas

Struggling in a system of disincentive: Participants, particularly in the United States, explained “the system is designed to be paternal. It pacifies the patient”. Large dialysis companies and hospital systems prioritized “profit” over “health”, and participants empathized with the pressure this placed on doctors who were incentivized to stick to strict schedules and quotas, having no time to educate patients. Some surmised that dialysis companies put up “roadblocks” to stop researchers accessing their patients, who were “captives of a system” who had to “sit down, shut up”.

Changing research culture: Participants wanted researchers and clinicians to acknowledge the paternalistic attitude to involvement – “they don’t want us playing in their sandbox”. They noted that some clinicians/researchers were “protective of their power” but that this was “generational” and younger researchers were opening a “two-way street”, recognizing patients/caregivers as valuable contributors. Social media aided a shift in power, allowing patients/caregivers to start “a dialogue instead of a one-sided monologue”. Government and professional organisations and journals (e.g. British Medical Journal) were praised for efforts to involve patients/caregivers.

Becoming equals: Some participants initially experienced “role confusion” and struggled as they viewed themselves to be “on a much lower level” than other members of the research team (i.e. clinicians). Over time and with encouragement from the researchers they developed confidence to “speak up”. Some felt valued and integrated in collaborative research teams, while others described a chasm between “us (patients/caregivers) and them (clinicians/researchers)” and experienced “a real pushback against treating the patient as an equitable partner”.

Seeing the person behind the patient

Harnessing broader knowledge, expertise, skills, and interests: Patients/caregivers had more to offer than their lived experience, and participants felt researchers failed to “harness” their expertise in other disciplines. They wanted opportunities to contribute to research that was of interest to them, and where they could apply their skills. For example, when designing an infographic: “What if you went out to your consumers and said ‘is anyone a graphic designer?’”.

Understanding patient needs: Having someone check “did you feel like you could speak freely? Was there anything you didn’t understand?” and a contact person they could raise concerns or ask questions was valued. Participants emphasized the need for plain language and detailed information about the research project, the tasks and time required of them, and a timeframe. They appreciated updates on unexpected delays, and being informed of the outcomes of the research. Some participants were involved in developing and delivering research training programs to patients/caregivers, which were viewed as critical to building capacity in researcher partners.

Bolstering impetus for involvement: Participants were driven to become involved in research to “pay it forward” or “give back to the community”. Involvement empowered them to become “proactive” and turn their experiences with kidney disease into “something positive”. Some gained a sense of “purpose” after experiencing so much loss. Others found it “rewarding” to share new knowledge with other patients. Participants enjoyed travelling and meeting other patients and some viewed involvement as an opportunity for professional development, implementing skills and knowledge they cultivated in other areas of their lives.

They conceptualized their involvement in research as improving patient experiences and outcomes.

Sensitivity to complexities of payment

Accounting for individual circumstances: Participants emphasized the need to consider each person's individual needs and preferences regarding reimbursement and payment for involvement in research. Some were frustrated with not being financially compensated for their work – "It's the hardest job I never got paid for", while others were content to contribute without payment because they could "afford to" and found it "rewarding". Some preferred not to receive payment as they felt this could compromise their impartiality, particularly if the money came from a pharmaceutical company. Some cautioned that receiving payment could cause them to lose government benefits such as disability or healthcare, but agreed for their expenses to be reimbursed.

Denoting value: For some, payment demonstrated that researchers valued their contributions as "equal to the other people participating" and made them feel "part of the team". Some noted that they were asked "to do an awful lot for free" and thought this was "disrespectful". One explained that they were met with "silence" every time they raised the issue of payment and felt that patients/caregivers were viewed as "suckers" who would continue to contribute without payment.

Enabling diverse involvement: Participants were concerned that involvement was "heavily dependent on a few highly motivated individuals who give up a lot of their own personal time and personal cost to get involved". Providing payment for time and covering expenses in advance were considered enablers to involve a broader demographic, as some may not be able to afford transport or time off work. Budgeting for patient/caregiver involvement was seen to avoid “the tendency to go for the person that’s geographically closer” and rather allow for purposive involvement.

Championing the patient voice

Links to important stakeholders: Participants viewed themselves as advocates for research, both on the patient/caregiver and researcher sides – “connection is key”. They used their influence within patient and professional organizations to “vouch for researchers”, sway policymakers, lobby government, and disseminate findings broadly. They engaged, trained and empowered new patient/caregiver partners through networking, using “exponential leadership”.

Drivers of innovation: Unique first-hand insights from participants allowed them to contribute compelling and creative suggestions for researchers such as developing a device to detect fluid volume similar to an insulin pump. Some initiated and led new projects – e.g. developing an evidence-based information resource to help transplant recipients return to work.

Responsibility to end-users: As lone voices in research groups, participants reminded the team of “why they’re doing this research” by sharing firsthand insights of their lived experience. Participants considered themselves as advocates for other patients/caregivers, and felt a responsibility to use the opportunities they were given to represent the broader patient voice, particularly for specific groups that were often underserved, such as children with CKD.

4.5 Discussion

Patients with CKD and their caregivers contended with a multitude of treatment and symptom burdens and caring responsibilities, which hampered their capacity to contribute to research. Initially they lacked confidence and struggled with their role as an ‘expert’ among health professionals and required support from researchers, and in some cases financial compensation, to feel accepted as equal partners. Patients and caregivers felt burdened with the responsibility to be proactive in seeking out limited opportunities to become involved, often at personal cost, which they felt hindered diversity and inclusiveness. They also faced stigma of their ‘patient status’ due to poor understanding and awareness of CKD by the general public, and even by some health professionals who laid blame on them for their disease, discouraging involvement. Historical power dynamics of health systems and research culture, including minimal time with their clinicians limited opportunities to build trusting relationships and better understanding of their illness, which they believed would lead to greater involvement in research. Broader skills and expertise from their personal and professional lives were overlooked, and critical connections to important stakeholders, particularly the patient community to whom they felt a responsibility to educate and advocate for, were often ignored.

Whilst the findings were broadly consistent across the participants, there were some differences noted by country. Shame and stigma associated with CKD diagnosis was particularly notable in participants from the United States, who identified this as a major barrier to involvement in research. They were also concerned that clinicians were discouraged from educating their patients due to financially motivated time constraints and thus were unable to develop ongoing trusting relationships. Patients receiving in-center dialysis did not seem to be supported to take ownership of their treatment decisions and care.

Both led to uninformed patients with little understanding or interest in becoming involved in research. In contrast, participants from Australia and the United Kingdom more frequently described how they were positively supported to be involved in research projects.

Patients with HIV/AIDS have similarly reported discrimination and stigma associated with disclosing their patient status as a major barrier to being involved in research[23,24]. Participants from our study believed better education and public awareness could serve to reduce the shame and stigma associated with patient status. Studies in patients with HIV/AIDS and cancer have previously identified the power imbalance between patients and researchers, highlighting patients' lack of trust in the research community and confidence in themselves as contributors[24,25]. Holding meetings in community venues and sharing benefits directly with the community were suggested to avoid "drive by"[24] research, shift the focus to implications and outcomes for the community and build confidence and trust, which could be effective in the CKD population. A further power imbalance identified in our study was the challenge to include the unwell patient. Due to the lifelong nature of CKD, patients were more commonly involved in periods of good health such as when dialysis was been well established, or when their transplant was functioning well. However, participants noted that this may mean researchers are missing perspectives from patients at their most unwell times when they may have valuable insights to share, and care should be taken to ensure inclusion of this subset of the population.

This study provides in-depth insights from patients/caregivers from four countries and with varied experiences of CKD and involvement in research. We reached data saturation and used investigator triangulation to ensure the themes reflected the breadth and depth of the data. However, there are some potential limitations. All participants were from high income countries, English-speaking, and with high educational attainment. While this may limit the transferability of findings to low-income settings or disadvantaged groups, their collective experience in CKD research lends power to their opinions and strength to their recommendations. The dearth of Indigenous voices and cultural diversity needs to be addressed in future work, with a focus on co-design with these communities. Most of the findings related to involvement in priority setting, study design, recruitment and reviewing research. There were little data on involvement in the analysis, evaluation, and implementation stages of research (Table 4.1 and Table 4.4). This mirrors findings from reviews on patient involvement in research more broadly that demonstrate most involvement takes place in the planning and preparation phase[8,17,26,27]. When patients/caregivers are not involved in the execution and implementation phases of research, they are unable to ensure their contributions and priorities are carried through the research project, limiting their value and impact.

Novel to this study are suggestions and considerations from the participant perspective based on their experiences being involved in research (Table 4.3). We suggest that researchers 1) acknowledge the inherent power imbalance patients and caregivers encounter in being involved in research, and 2) implement formal mechanisms co-designed with patients/caregivers to counterbalance this. Often-invisible challenges patients/caregivers face when becoming involved in research include limited experience in the research setting, unfamiliarity with jargon and acronyms and intimidation by experts. These have been similarly identified in other health disciplines, and can discourage open and honest contributions[24,28]. Formal induction processes, a terms of reference outlining expectations and roles for patients/caregivers and researchers, a dedicated contact for patients/caregivers, and leaders encouraging inclusivity and ensuring time and opportunity for patients/caregivers to be heard among professionals can help optimize their involvement[29].

Educating and involving patients in shared decision-making “decreases informational and power asymmetry”[30] between patients and clinicians, helping to establish trusting relationships and set up a dynamic whereby patients can develop confidence in themselves as experts and equal contributors to their care[31]. More research is needed to better understand ways in which shared decision-making in clinical settings can be leveraged to develop future relationships with researchers, and allow patients to conceive their role and value as contributors to research. There is potential for clinics or hospitals to connect patients/caregivers with researchers through information sessions that provide education to patients/caregivers and opportunities for them to get involved in research.

There is also a need for more consistent involvement of patients/caregivers across the research cycle[27]. More work is needed to understand how to better support meaningful involvement in the later stages, particularly in evaluation and implementation of research findings, and disseminating results back to the patient community.

The burden of CKD, limited opportunities, and power asymmetry between patients/caregivers and researchers were challenges to meaningful involvement in research. Building trust, awareness of opportunities, and recognizing the broad expertise and value of patients/caregivers, including and beyond their illness experience, may better support patient and caregiver involvement in research in CKD. Optimizing involvement in these ways may serve to better align research priorities, produce higher quality research of greater importance to all stakeholders and enhance implementation of findings to practice, resulting in better care and health outcomes for patients.

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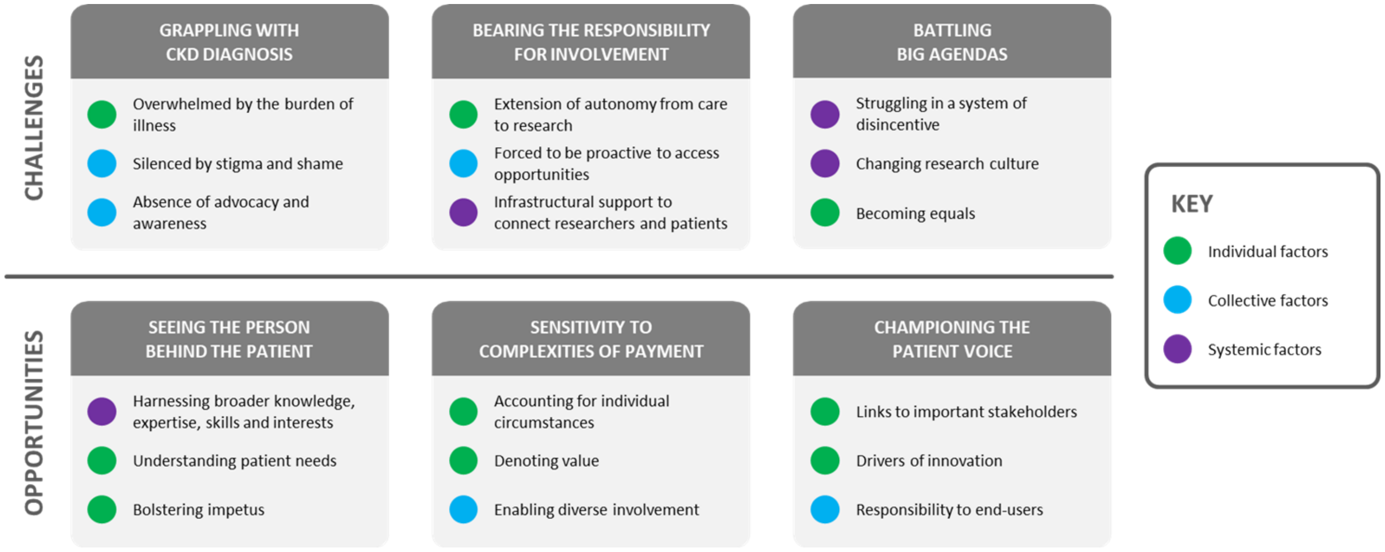

Figure 4.1 Thematic schema

<u>Individual factors:</u> changed from individual to individual depending on their circumstances

<u>Collective factors:</u> shared by participants through being identified as a CKD patient, part of the CKD patient community

<u>Systemic factors:</u> relied on broader infrastructure, and research culture and community

Participants faced challenges in being involved in research as individuals, struggling to cope with their identity as patient and partner. Collectively they encountered stigma and shame around their CKD diagnosis, and were forced to search out opportunities for involvement. They felt systemically discouraged and disempowered as they lacked support and resources needed to tackle health systems and research culture historically set up to silence the patient and recognize the clinician/researcher as the expert. However, they identified opportunities to improve involvement by addressing individual needs and circumstances, leveraging collective motivations to advocate the patient voice, and becoming recognized for their ability to contribute to research beyond their lived experience of CKD.

Table 4.1 Participant Characteristics n=23

Characteristic	n (%)
Patient	18 (78)
Caregiver	5 (22)
Female	13 (57)
Age (years)	
<30	1 (4)
31-40	5 (22)
41-50	4 (17)
51-60	8 (35)
61-70	4 (17)
>70	1 (4)
Country of residence	
Australia	10 (43)
United States	10 (43)
United Kingdom	2 (9)
Denmark	1 (4)
Ethnicity	
White	19 (83)
Indian	2 (9)
African American	1 (4)
Chinese	1 (4)
Education level (highest attained)	
Postgraduate degree	10 (43)
Undergraduate degree	10 (43)
Professional certificate	1 (4)
High School	2 (9)
Employment status	
Full time	9 (39)
Part time	9 (39)
Retired	2 (9)
Student	1 (4)
Disabled	1 (4)
Contract	1 (4)
Involvement in research - stage	
Identifying/prioritising	20 (87)
Designing	9 (39)
Recruiting	14 (61)
Conducting	8 (35)
Reviewing	18 (78)
Disseminating	7 (30)
Implementing	6 (26)
Evaluating	5 (22)
Involvement in research - role	
Advisory board	11 (48)
Steering committee	14 (61)
Expert working group	7 (30)
Workshop participant	16 (70)
Involvement in research – hours per week	
<1	7 (30)
1-2	9 (39)
3-4	2 (9)
5-10	3 (13)
>10	1 (4)
Interview setting	
In person	11 (48)
Online	12 (52)

Table 4.2 Illustrative quotations

Theme	Illustrative Quotations
Grappling with CKD diagnosis	
Overwhelmed by the burden of illness	It’s time consuming. It’s scary… all of a sudden, it’s permeating their whole life. I’m supposed to have time to get involved in research? I don’t have time to do anything except take care of myself…. They want to distance themselves from an all-pervasive disease, I think, in many instances. *(Caregiver, United States)* There were times that I could not even fathom, don’t talk to me about anything, just keep her alive right now. *(Caregiver, United States)* It's been hard for me the last few weeks because I haven't been well ... So, I think that would be very dependent on how well they are, which is a shame because that's probably when we need them to be involved…you're experiencing CKD at perhaps it's worst and you might have valuable input for researchers about this is what I'm experiencing. *(Patient, Australia)*
Silenced by stigma and shame	People get a bad rap with dialysis because they go three times a week, they’re fluid overloaded, they [dialysis staff] blame them. Patients get blamed a lot of the time for having CKD ... A lot of people think it’s hereditary or it’s something that you do. I can’t tell you how many times I’ve told people I’ve been on dialysis and they were like ‘oh, you did drugs and you used alcohol?’ and I’m like no, that’s not it at all…It has nothing to do with anything that I did. I think people shut down, because who wants to get blamed, and who wants to be told that you’re not doing the right thing? It’s hard enough as it is. *(Patient, United States)* There’s also kind of a stigma, unfortunately, with kidney disease. Some people think it’s communicable. *(Patient, United States)*
Absence of advocacy and awareness	Kidney disease isn’t sexy. It’s not the attractive thing. Being a woman, I walk in and I just saw some cereal, it had a pink banner across the top. My immediate thing was recognition of breast cancer. We do not have a high recognition. *(Caregiver, United States)* I just like getting the word out, because it’s the only way things are going to get done. I was like, it was kind of nice that Selena Gomez came out. She didn’t have to, that was pretty cool…. I hate to say this, but sometimes you need a big marquee kind of figure. *(Caregiver, United States)* Not many people know how bad kidney diseases are…whereas if you have a cancer, I'm not downsizing cancer's important, but the importance that it gets rather than the kidney disease, because if you are diabetic, you're prone to kidney disease, if you're obese, you're prone to kidney disease. If have a hereditary condition, you're prone to kidney disease. So there are a lot of chances to get a kidney disease than to get a cancer, but the people take it so easily when you have a kidney disease and the lack of awareness, and even preventing the progression of getting kidney diseases. *(Patient, Australia)*

Bearing the responsibility for involvement	
Autonomy in their own care	Especially here dialysis is so paternalistic, and we're not taught that we want to advocate for ourselves. *(Patient, United States)* There are a lot of people who are getting more engaged in their own treatment, and I think that's the first step before they start getting involved in other things. *(Patient, United States)* Obviously the clinic is your number one captive audience, I mean you've got ninety percent of the patients in the American population sitting in clinic. Of those, let's just say forty are not even capable, but you've got still fifty percent…. What I call it when I'm speaking is you check your independence at the door. Ninety percent of the patients that choose in-centre walk in, check out, sit in a chair and they're done. *(Patient, United States)*
Forced to be proactive to access opportunities	I kept telling them I wanted to be involved… there's not a lot of opportunities to be involved in research unless you're doing drug research. *(Patient, United States)* I'm cheeky, I don't mind reaching out to someone. If I know they know something, I'll just write them and introduce and usually we know some of the same people and we refer to them or whatever… personally I just have the drive. *(Patient, Denmark)* If you make it hard for patients to get involved in something, you may just lose them forever. It may be their only chance to interact in a different way with the organization or with the community. *(Patient, United States)*
Infrastructural support to connect researchers and patients	I think the biggest failing is re-engaging the people who already are interested, just relying upon them to see an EOI or see something that one group puts out somewhere, I just think there's a lot of missed opportunity there for recruitment. *(Patient, Australia)* We're trying to come up with that, the National Kidney Foundation, like a board on the web that patients can go to and look at research opportunities…The Juvenile Diabetes Foundation, they have a great way of doing that. What they do is you can go in and you can register to be part of their research…it gives me options on what I'm willing to do. Am I willing to go into hospital? What am I willing to do? And where am I willing to go to? Is it locally? Am I willing to go out of state? And then based on those results when research comes up that fits my demographic of whatever, they will kick that opportunity to me via email. *(Patient, United States)*
Battling big agendas	
Struggling in a system of disincentive	There are people now running large hospital systems like University of Southern California, like UCLA, that are bean counters, that are basically accountants. They're trying to stretch that dollar as far as they can, and therefore they dictate how much time you have with each patient. *(Patient, United States)* In many cases, the system is designed to be paternal. It pacifies the patient. It says come 3 times a week, sit down, shut up. *(Patient, United States)* 'I don't know how to educate my patients. I don't have time.' The system doesn't reward you. We're dealing with is a dysfunctional system. *(Patient, United States)*

	Clinics don’t let researchers in, because they only want research that favourably impacts their business…because our entire dialysis is a business model and not a health model, that’s an issue. *(Patient, United States)*
Changing research culture	They’re very good doctors, and they’re younger. Most of them are way younger than I am. It’s very interesting, it’s a real give and take, it’s a real two-way street. We need more of that. We need more of that conversation to go on. *(Patient, United States)* I think that I’ve seen a change in the last two to three years, where instead of a paternalistic type thing, where the doctors or the nurses know best attitude. That’s really gone away, it’s going away, let’s put it that way. Where we realise that it is a partnership between the patients. *(Caregiver, United States)* I think a good example is the American Society of Nephrology, what they’re doing. The American Society of Nephrology is making a sincere effort to change the culture because they realise they have to… the Clinical Journal of American Society of Nephrology now has patient editors on their editorial board. I think those are very tangible actions that are not words. I think there’s a lot of words, but think the ASN is actually leading with some actions. *(Patient, United States)* Sometimes it's like a bit of a tick-the-box. Do we have the consumer voice? But I think more emphasis is being put on that, so that's good…This consumer involvement is quite new so it's a learning curve for everybody. I think it's changing as people are becoming more aware of how to involve consumers better. It might be starting like ticking the box but it is growing to be more than that. *(Patient, Australia)*
Becoming equals	But personally, for me, there is a tendency to sit back and not cut in with something that I may think is important to say or contribution I have because I'm thinking of myself as the patient, and still on a much lower level to the professors, and the doctors, and the surgeons, and whoever else might be in the room…it's taken for granted by other people that they would speak up, when that's not natural or normal for the patient who is used to laying in a hospital bed, receiving instructions about how to care for themselves. It's like role confusion. Over time you can come to expect, ‘Okay, my opinion and input here is wanted and warranted,’ and either time is going to be made for that or you feel confident enough at a certain point to put your hand up and say, ‘What about this?’ *(Patient, Australia)* I think it has been a good experience, and I feel a valued contributor. I don't feel just like the patient at the end of the line, ‘Oh, what do you think, by the way?’ And I feel like a valued member of the team and it felt all very collaborative and very professional. Again, it wasn't a case of, ‘Well, there's the clinicians and here's the patients.’ It was very together, which I think is really nice and really important. *(Patient, United Kingdom)* On a lot of the steering committees and scientific committees, because I've been doing this now for a few years, it feels like we now are only starting to get traction with some equity in respects mostly. This is where I've almost ended up in conflict resolution type processes because with some people in the profession from the research side, there's been a real pushback against treating the patient as an equitable partner and an expert in their field. *(Patient, Australia)*

Seeing the person behind the patient	
Utilizing broader knowledge, expertise, skills and interests	One of the other problems you have of getting patients involved is patients are only looked at on the basis of what's on their medical charts. Not who they are. Not the fact that they might have been a CEO of a company, or they might have been a university professor. *(Patient, United States)* People want to treat consumers like they can just bring their lived experience and that's it. They can just quote their emotional stories and move on. No, no, no. I want the research methodology to be strong. I want the research dissemination to be strong. *(Patient, Australia)* Another example is I see a lot of is 'Oh, we've done this research and now we want to make an infographic.' Someone in the office who doesn't have training and experience in how to create infographics, 'We'll put it together because we don't have a budget.' I go, 'Well, what if you went out to your consumers and said, 'Does anybody work in this field? Is anyone a graphic designer and would like to donate their time pro bono to create this professional infographic for us?'' Harness the ability of these people because they're here. Why do you guys only look at us as someone to ask questions about tolvaptan? *(Patient, Australia)*
Understanding patient needs	One of the research groups I'm on did an introductory video that explained things. Trying to limit the amount of reading, I think. And having more diagrams and really short, a few groups are pulling out lots of short research summaries. *(Patient, Australia)* There are so many clever people where they're involved with the research and who know so much. And occasionally I think it's forgotten that I am only a consumer. And even though I'm a nurse and have that background, there's still a lot that goes over my head. *(Patient, Australia)* Instead of saying renal, say kidney to me. So those simpler words, switching the words where consumers can understand. *(Patient, Australia)* What I ask my researchers to do is develop a simple one-page summary of your research. And then next to it have how long I need to spend on that research. For example, how many meetings I need to do if I'm going to do three months, or if I'm involved in this research for the next one year, how many meetings do you want me, and how long those meetings are? Breaking that into simpler steps, because I'm living with that chronic condition and have dialysis and I have a treatment to go through. So instead of you just coming, saying, '[Patient], are you available?' You just have to set the agenda, or set the tone, right from the start saying 'We expect you have to spend 20 minutes for a meeting, for the next three months in three meetings.' And what skillset do you require from me. *(Patient, Australia)*
Bolstering impetus for involvement	I was always on the lookout for changing things, making things better. I just feel I've got so much experience, so much knowledge, and so much sadness, I have to turn it into something positive. I need something good to come out of this. So that's always been my driving force. I have to use it somehow. *(Caregiver, Australia)* I started to talk to other patients and I took a leadership role, started using some of the skills I had from my career and put them into my work with the kidney groups, and started feeling a sense of purpose again. *(Patient, United States)* I was looking to add things to my professional resume… it was professional development, but also if anything I thought I would be able to help more patients and if I were going to go to work on digital related things, I could use those for later jobs. *(Patient, United States)*

	Research is a way to stimulate my brain and my thinking… opportunities to meet people, make connections, learn more stuff, and that all helps me. In a way, it helps me cope. It helps me cope with sometimes the realities and the crap part of having renal disease, which is not always a great deal of fun. *(Patient, Australia)*
Sensitivity to complexities of payment	
Accounting for individual circumstances	With the way my disability's set up, I'm not able to accept too much without losing some benefits, like my medical, so it's very important. I find it more rewarding, to volunteer, because then I don't have an agenda. It allows me to get invited to sit at the table with people that otherwise maybe I couldn't have these conversations with… especially at a government level, where they know that I'm not there representing a corporation or anything like that. *(Patient, United States)* One friend of mine, she can't be compensated, nor does she want to be compensated because in the United States, she's on something called Social Security Disability Income… if she gets too much, then the government sees her as working or capable, and they're like, 'Well you don't get access to that anymore, we're taking that away,' and she's afraid to lose that. So she does more of, I want you to pay all my expenses to come to this meeting. On the other side of it, I know a patient, he does so much work, he would absolutely in a heartbeat say, 'Yes, I should be compensated for this.' The most challenging piece is that it's different for each person. *(Patient, United States)*
Denoting value	I don't get paid. Every time I raise that issue, I say we're not getting paid for this. And there's silence. It's like, 'these suckers think we're actually going to pay them?' *(Patient, United States)* So if you're an academic journalist, or a senior editor or reviewer for some group and you're asking a patient to review, whatever that person gets paid to review, the patient should get the same… equal to the other people participating. *(Patient, United States)* I know there's a lot of people with CKD who are also researchers who are doing an awful lot, basically, for free. I think that's almost really taken for granted because it's like, 'Oh, you're a consumer', but to me, it's just going too far. It's really disrespectful. *(Patient, Australia)*
Enabling diverse involvement	If there's financial support, your foods are taken care of and things like that, I think that gives patients opportunities to come down and be involved. I think sometimes it's just being aware that some people won't have the money there to even catch the train. *(Patient, Australia)* Because it's not always put into the budgets. Some of the universities insist on adding overheads, so if you've got an expensive researcher and then you're left with patient involvement, the tendency is to go for the person who's geographically closer, can walk into your building. I think cost is a barrier, but that's because it's not included early enough in the process. Maybe they should mandate a particular percentage [for consumer involvement]. *(Patient, United Kingdom)* It's very heavily dependent on a few highly motivated individuals who give up a lot of their own personal time, at personal cost to get involved. *(Patient, United Kingdom)*

Championing the patient voice	
Links to important stakeholders	We meet with Medicare, CMS Medicare, to talk about reimbursement issues and also the quick measures, the quality measures, because those have to be patient meaningful… we were invited by the Government Accounting Office, to come and give input to them when they were mandated by Congress to look into the barriers for home dialysis. *(Caregiver, United States)* I'm networked to other patients out there and I know people's strong points and skills. *(Patient, Australia)* I do a lot of advocacy on Capitol Hill, again because of my proximity, being an hour away there's no excuse not to. *(Caregiver, United States)* I chair that [clinical study group] and rally people together, and encourage patient participation in all the studies that people are planning to do…through being the CEO of the UK group, I can then reach thousands of patients to take part. *(Patient, United Kingdom)*
Drivers of innovation	A lot of them would be healthy, relatively, but they wouldn't have the courage to go back to work. There wasn't so much information out there on the web. And I felt recipients do need that. They need to, when they are down, when they're sick, when they're looking for information, they need to find something that they can read and be informed before they go in for their surgery, while they are going through the experience, and even after. So that's what I did. I put together this repository of information online for recipients across all sorts of organs. *(Caregiver, Australia)* I proposed a research project on a device to detect your optimum fluid in your body. We went through their process, they looked at it through what's called the Kidney Health Initiative and decided to support that idea. It's the first patient-approved project that they've ever done, so it's pretty exciting. *(Patient, United States)* I recruited 8 patients who have quite significant experiences of pain, and we had a day and a half workshop. Afterwards, the primary researcher said that there were some things that he'd never expected that came out of these two days. Since then KDIGO always had a patient carer peer group within their meetings. I do think we were pioneering in that respect, to making sure that our voice was heard as equally as the geneticists' voice and the nephrologists' voice. *(Patient, United Kingdom)*
Responsibility to end-users	I think that the patient voice is really important. Even if we don't necessarily speak up all the time, we remind people why they're doing this research There is an opportunity for research gains and possibly policy gains or treatment gains to be made from every person contributing what they can. With my stake being a little more unique, there is a bit of an obligation that I want to fulfill in contributing what I can. *(Patient, Australia)* It's a pretty huge responsibility to represent the paediatric [voice], I'm the only one in this particular, and it's going to expand in time but right now we serve a certain term, so I'm always on the lookout for someone to supersede me when I go. I do not want to lose a paediatric voice. *(Caregiver, United States)*

Table 4.3 Patient and caregiver recommendations for involving them in research

Theme	Suggestions and considerations from the participant perspective
Grappling with CKD diagnosis	• Involve patients early in their journey so they are inducted and equipped to be involved and can contribute even in periods of illness. Consider the support that may be required to facilitate this • Use caution and care when approaching someone who is unwell to become involved for the first time. Consider passive approaches such as posters or flyers in this instance • Encourage and support patients/caregivers to build confidence to be involved • Consider that since patients with CKD have a lifelong illness “there are lots of people who are available to provide input” and they are potentially able to be involved “from the start through to the finish of any projects because they live with that experience for long periods of time” • Provide education and increase awareness of CKD in the broader population to discourage discrimination and stigma
Bearing the responsibility for involvement	• Encourage patients/caregivers to become active participants in their own care through strategies such as shared decision-making • Cultivate relationships in clinical care to start the conversation about becoming involved in research • Educate clinicians on how to reach and communicate with patients/caregivers • Demystify research – educate patients/caregivers about research, its benefits for them and opportunities to become involved • Use multiple channels to reach patients/caregivers and advertise opportunities for involvement (e.g. social media, flyers, email) • Develop relationships with patients/caregivers groups and provide information and education sessions explaining your research and how patients/caregivers can become involved • Develop a database of interested patients/caregivers that can be contacted and re-engaged for new projects • Invite patients/caregivers to participate in scientific meetings
Battling big agendas	• Aim to involve patients/caregivers as equal members of a collaborative research team (e.g. include in all meetings and correspondence) • Educate clinicians in medical school about how to work with patients/caregivers • Consider communication training for clinicians • Ensure simple language is used • Develop a “patient-centred research standard” • Enlist patients/caregivers to assist in developing involvement strategies and avoid tokenistic involvement • Open access to scientific meetings to patients/caregivers including patient/caregiver speakers in equal roles and sessions specifically designed for patients/caregivers
Seeing the person behind the patient	• Develop a database of patients/caregivers and include profiles that contain their interests, skills and expertise as well as their lived experience • Consider relevant experience when selecting or matching patients/caregivers with research projects • Develop a terms of reference outlining what is expected of patients/caregivers including their role/tasks, number of meetings, expected workload in hours, and timeframe of the research as well as privacy and confidentiality policies • Take time to talk to patients/caregivers about any special needs they may have, including physical and cognitive challenges, technological or logistical support • Consider providing education or training for patients/caregivers researchers on basic research methodology and subject matter background

	• Provide support/assistance to help patients/caregivers voice their perspectives, particularly in settings where they are working with clinicians/researchers • Include more than one patient/caregiver in any group • When scheduling meetings ask patients/caregivers for their availability • Consider having a dedicated patient/caregiver liaison so that patients/caregivers know who to contact to raise and concerns or questions • Be sensitive to patient/caregiver experiences including taking time to understand their individual journey, and use care when talking about potentially challenging topics – e.g. mortality • Communicate often throughout the process, including explaining any delays • Report back to patients/caregivers at the end of the process to explain what has come out of the research, and how they contributed • Consider using an ‘interest-based’ approach when selecting patients/caregivers to work with • Highlight the potential benefits to patients/caregivers individually (e.g. education, purpose) and more broadly (e.g. improving future patient outcomes)
Sensitivity to complexities of payment	• Include patient/caregiver involvement in the budget at grant stage – view this as an investment similar to any other budget items that contribute to research quality • Use a cost calculator or budgeting tool such as the one from INVOLVE to understand rates of compensation • Be upfront with discussions about payment/compensation • Always ask the patient/caregiver for their preferences, or if payment may impact benefits they receive • Be transparent about what other team members are receiving for their involvement, and align patient/caregiver compensations with this, ensure they feel valued as an equal team member • Cover any expenses incurred in advance – patients/caregivers may not have access to funds or credit cards to cover expenses up front
Championing the patient voice	• Utilize patient/caregiver networks and connections to expand involvement • Utilize patient/caregiver networks and connections to gain support from policymakers, government, patient organizations and the public • Ask for input, ideas and solutions from patients/caregivers from their lived experience • Build capacity to support patient/caregiver led research through mentorship and training

Table 4.4 Illustrative examples of participant contributions to research

Stage/s	Activity
Priority setting	• Contributed to grant proposals and applications • Identified research priorities • Lobbied government for funding • Conducted community consultations • Contributed to developing a national strategic action plan and submitted to government for funding
Research design	• Designed and developed surveys • Designed a flyer for recruitment • Provided feedback on patient-reported outcomes • Simplified enrolment processes for a trial • Contributed to device design (risk/benefit, tolerance etc…) • Contributed to trial design • Provided lived experience to inform intervention design • Reviewed patient facing documents for readability • Edited research protocol with patient friendly language
Data collection	• Conducted survey • Conducted interviews • Conducted focus groups • Recruited participants for a trial • Recruited participants for workshops • Recruited participant for focus groups • Facilitated workshops • Provided access to participant groups/databases/networks
Data analysis	• Reviewed data • Analysed qualitative data (interview transcripts)
Dissemination	• Wrote research papers/reports • Presented the research at universities to medical students • Edited scientific journals (with an emphasis on patient/caregiver involvement) • Moderated panels • Publish research reports to patient/caregiver audiences • Presenting and sharing research findings with other patients/caregivers • Attended and presented at scientific meetings • Held webinars for patients/caregivers • Developed plain language summaries and infographics for patients/caregivers • Created informational videos for patients/caregivers
Implementation	• Changed policy on home dialysis without a caregiver • Wrote a submission to regulator to support drug approval after survey study
Evaluation	NA
All stages	• Steering committee member • Advised on patient/caregiver involvement strategies • Co-chaired project steering committee • Developed an international patient registry • Led research work groups • Patients/caregiver organizations partnered with research groups • Involved across all stages of developing an internationally validated pain score for PKD • Sought funding and conducted focus groups to develop patient decision-aid

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Chapter 5: Principles and strategies for involving patients in research in chronic kidney disease: Report of National Workshops

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This chapter is structured as per the journal article.

5.1 Abstract

Background: There is widespread recognition that research will be more impactful if it arises from partnership between patients and researchers, but evidence on best practice for achieving this remains limited.

Methods: We convened workshops in three Australian cities involving 105 patients/caregivers and 43 clinicians/researchers. In facilitated breakout groups, participants discussed principles and strategies for for effective patient involvement in chronic kidney disease research. Transcripts were analysed thematically

Results: Five major themes emerged. *Respecting consumer expertise and commitment* involved valuing unique and diverse experiential knowledge, clarifying expectations and responsibilities, equipping for meaningful involvement, and keeping patients ‘in the loop’. *Attuning to individual context* required a preference based multipronged approach to engagement, reducing the burden of involvement, and being sensitive to the patient journey. *Harnessing existing relationships and infrastructure* meant partnering with trusted clinicians, increasing research exposure in clinical settings, mentoring patient-to-patient, and extending reach through established networks. *Developing a coordinated approach* enabled power in the collective and united voice, a systematic approach for equitable inclusion, and streamlining access to opportunities and trustworthy information. *Fostering a patient-centred culture* encompassed building a community, facilitating knowledge exchange and translation, empowering health ownership, providing an opportunity to give back, and cultivating trust through transparency.

Conclusions: Partnering with patients in research requires respect and recognition of their unique, diverse and complementary experiential expertise. Establishing a supportive, respectful research culture, responding to their individual context, coordinating existing infrastructure and centralising the flow of information may facilitate patient involvement as active partners in research.

5.2 Introduction

Involving patients in research has emerged as a policy imperative to align research with patient priorities, and thereby maximise the impact of research on practice and policy[1,2]. This has spurred widespread efforts by global organisations to involve patients as active partners contributing across the research process from setting the research agenda through to design, dissemination and implementation[3-7].

Patient involvement can improve the relevance, quality and translation of research for improved outcomes[1,5,8-11]. For example, patient involvement in the design of a clinical trial in cancer increased recruitment and retention rates[12-14]. Patient involvement in the Outcome Measures in Rheumatology (OMERACT) initiative for the past two decades has led to increased focus and reporting of patient-centred outcomes such as fatigue and sleep[15,16]. In nephrology, patient perspectives have ensured that the scope of clinical practice guidelines for polycystic kidney disease, renal biopsy and infectious diseases in patients on haemodialysis cover patient-relevant topics such as education and psychosocial support[17-19].

Patient involvement in research is particularly relevant in chronic kidney disease (CKD) as patients face a life-long condition with mortality rates up to 100 times higher than the general population, and impaired quality of life [20-22]. Yet there remains a substantial mismatch between the priorities of patients and research[10,11]. A recent review indicated that 80% of clinical research did not address the top 10 research priorities identified by patients on or nearing dialysis[23,24], and outcomes considered critically important to patients, such as fatigue and cardiovascular disease, are infrequently reported in trials[22,25,26].

Major steps have been taken towards empowering patients as partners in research[27,28]. The Patient-Centered Outcomes Research Institute (PCORI) in the United States and INVOLVE in the United Kingdom were established to support patient-centred research and patient involvement across the research cycle [14,29,30]. The Kidney Health Initiative (a partnership between the American Society of Nephrology and the US FDA) has formed a Patient and Family Partnership Council for the patient voice to influence the development of drugs, treatments, devices and clinical trials[31]. Recently, the Standardised Outcomes in Nephrology (SONG) initiative was launched to bring together patients, caregivers and health professional to establish consensus-based core outcome sets for clinical trials[32].

However, there are barriers to involving patients in research[5,33]. Patients may feel disempowered, uncertain about their role and overwhelmed by the burden of their disease and treatment [5,34]. In the research community, there is scepticism about the value and impact of patient involvement and concerns about diminishing scientific rigor and the 'representativeness' of patient partners[1,3,35,36]. Time and funding have also been identified as barriers[3,4,35]. Thus, researchers remain uncertain about how to effectively communicate opportunities for involvement, provide necessary training and support or plan for meaningful involvement[35].

Systematic reviews[2,28,37-41] of methods for patient involvement focus on the stages (e.g. designing a trial) and roles (e.g. steering committee member) of patient involvement

[5,11,27,28,33,34,36,42,43], yet sparse data exist to inform best practice on how to engage and involve patients in research. While frameworks for involving patients in research exist[3,6,44-46], it can be difficult to translate these into tangible recommendations for practice. Further compounding this, and despite minimal evaluation of patient involvement initiatives[11], it is becoming known that practices previously viewed as empowering (e.g. one patient representative on a panel) are now viewed as disempowering and tokenistic by patients[35,45].

Improved reporting and evaluation of effective methods for engagement and involvement, in conjunction with more data from the patient perspective, are needed to understand the potential for patients to impact and shape the future of health research[2,3]. As part of the Better Evidence And Translation in Chronic Kidney Disease (BEAT-CKD) research program, we aimed to understand and describe patient and health professional perspectives about patient involvement in research in CKD, and to identify practical solutions to engage and involve patients across the spectrum of BEAT-CKD research initiatives. In this report, we use the term ‘patient’ to refer to all persons with lived experience of CKD unless otherwise specified. Further definitions can be found in Box 5.1.

Box 5.1 Definition of patient, involvement and engagement

Patient/consumer: refers to a patient, caregiver or family member with lived experience of chronic kidney disease.

Involvement: refers to a sustained and meaningful contribution to the research process as more than a research subject or participant and can range from consultation to partnership.

Engagement: refers to the establishment of a relationship between patients and researchers and is the first step of involvement.

5.3 Workshops

Context and overview

BEAT-CKD is a collaborative research program that aims to improve the lives of people with CKD by generating high-quality research evidence to inform healthcare decisions made by patients, health professionals and policy makers[47]. The program supports research and translation platforms – Caring for Australasians with Renal Impairment (CARI) clinical practice guidelines[48], Australasian Kidney Trials Network (AKTN)[49], Australian and New Zealand Dialysis and Transplant (ANZDATA) registry[50], and Cochrane Kidney and Transplant[51] - and the scope of the research activity is global. To inform a patient involvement strategy, we convened three workshops with patients and health professionals about principles and strategies for patient involvement in research in kidney disease.

Attendees and contributors

The BEAT-CKD Management Committee and Kidney Health Australia invited patients and health professionals to attend through their networks and social media. In total, 105 people with lived experience, including 70 patients (CKD Stage 1-5 and non-dialysis dependent, on dialysis, with a kidney transplant), 28 caregivers (caregivers, parents and family members) and seven who did not indicate their role, and 43 health professionals (nephrologists, researchers, dieticians, nurses) attended the workshop.

Workshops

We conducted one workshop in Sydney, Adelaide and Brisbane, from August to December 2017. Following presentations about research, attendees participated in one-hour breakout discussions with six to ten patients and health professionals per group to encourage exchange of diverse perspectives (total 17 groups). Each group had a facilitator. Question guides were informed by literature on patient involvement in research (Appendix D.1), to elicit perspectives on why and how patients want to be involved in research[2,7,14,29,40,44,45]. The discussions were audio-recorded and transcribed verbatim. TG coded the transcripts using HyperRESEARCH software for qualitative data management[52]. Concepts relating to participants' perspectives on patient involvement in research were identified and grouped into themes, which were reviewed by the facilitators and investigators to ensure all data were captured in the themes. All attendees and contributors were invited to provide feedback on the draft workshop report by email, within a two-week timeframe. Comments and feedback were addressed and integrated into the final manuscript.

5.4 Results

Summary of the Discussions

The discussions across all workshops were synthesised into five themes: respecting patient expertise and commitment, attuning to individual context, harnessing existing relationships and infrastructure, developing a coordinated approach, and fostering a patient-centered culture. The respective subthemes are described in the following section with reference to the patient or health professional group where relevant. Selected illustrative quotations are provided in Table 5.1. Figure 5.1 depicts a summary of the themes. Suggestions for practical approaches to involve patients in research are shown in Table 5.2.

Respecting patient expertise and commitment

"The power of people's lived experience is immense"

Health professional

Valuing unique and diverse experiential knowledge: Health professionals acknowledged that patients could identify important outcomes for research such as sleep and quality of life, and that having patients actively involved "puts a human focus" on research and reminded them of the "reason you're doing it all." Patients emphasised that all lived experience should be valued, and to include caregivers who were "a goldmine of information." Some patients were unsure about how they could contribute to research or thought that their opinion "doesn't count" because they did not have a medical background. Others believed their experience provided complementary expertise – "You do bring in knowledge that [health professionals] don't have…the reason people ask the wrong question is because they don't have the experience of living with it."

Clarifying expectations and responsibilities: Research could be "kind of mystical" for patients, and health professionals recognised that "one of the things that happens poorly is the explanation [of what is required] upfront". They suggested to clarify the patient's role, expected commitments and potential impact of their involvement to encourage patient research partners.

Equipping for meaningful involvement: Patients wanted health professionals to use "plain simple English", to provide adequate education and training, and to avoid putting patients "on the spot" by giving them time and resources to prepare before meetings and opportunities to respond afterwards. Patients thought involving one patient on a steering group of health professionals was tokenistic and overwhelming for the patient and suggested “at least three” patients were needed to give them confidence in voicing their opinions, and to convey diverse perspectives. They suggested that involving patients based on relevant expertise would enable meaningful contribution, and that financial reimbursement may allow a more diverse group of patients to be involved.

<u>Keeping patients ‘in the loop’:</u> To maintain interest, motivation, and engagement in research, patients explained that they wanted to be included in regular communications and information “that you don’t need a medical degree to understand.” Contributing to research was “a two-way street” and patients believed they should receive updates on projects they had been involved in even if the results were not yet available. Health professionals agreed that efforts to maintain communication with patients would serve to sustain relationships for longer term and meaningful involvement in research.

Attuning to individual context

“Once the patient is happy and healthy, then they’ll be able to go on and do research”

Patient

<u>Preference based multipronged approach to engagement:</u> Considering patients’ preferences for communication, and tailoring the approach accordingly was suggested. Some older patients asked to be contacted by telephone or letter, while some younger patients preferred social media. Patients thought that innovative visual methods of communication such as infographics and YouTube videos would be an effective way to deliver potentially complicated research information.

Reducing the burden of involvement: For patients, it was necessary to have flexibility given their health and life priorities. One patient could not commit to research activities that required him to take annual leave as he was saving this for his transplant, while another remarked that dialysis has “already chewed up enough of your day, you’ve already lost enough of your life”.

Being sensitive to the patient journey: Patients urged researchers to “not hit you up for it [involvement] when you’re at your worst” and to demonstrate sensitivity to each individual’s CKD journey. Some patients were “overwhelmed” at diagnosis and not in the “right frame of mind” to be involved in research. For patients on dialysis, the treatment burden was “all-consuming”, and it would be difficult to be involved in research when commencing dialysis. Fatigue and poor concentration could also constrain the patient’s ability to be involved.

Harnessing existing relationships and infrastructure

> *“If we could piggyback onto diabetes research, that might raise public awareness”*
>
> Patient

Partnering with trusted clinicians: Nephrologists and dialysis nurses that already had established relationships with patients were thought to be best placed to induct patients into research opportunities, and health professionals agreed that their networks of clinician colleagues could be further utilised to reach more patients.

Increasing research exposure in clinical settings: Patients “had time to kill” and suggested showing videos about research opportunities whilst waiting for appointments in clinic. Patients thought “posters are really good” to share opportunities for involvement in research.

Mentoring patient-to-patient: Some believed that patients with previous research experience would be the best person to engage new patients "because they've been through it. The more you hear about the process, the more you start to think 'I could probably cope with that'." Patients thought they were uniquely placed to reach new patients in their own networks, including those residing in rural/remote areas. Some health professionals thought moderating peer interactions may be needed.

Extending reach through established networks: Patients were frustrated by the lack of public awareness about kidney disease, compared with cancer and diabetes for example. They urged for more "marketing", "promotion" and "education" to "raise the profile" of kidney disease in the community and not just in hospital settings. One patient noted, "[The kidney information booth] was set up in the [hospital] foyer for kidney health week, saying we'll take your blood pressure... People who come to the hospital already know they've got high blood pressure. You should be outside Bunnings [hardware store]."

Developing a coordinated approach

> *"If you all get together, you're there for one common cause, but being separate, it's not really helping anyone"*
>
> Patient

Power in the collective and united voice: One patient explained that researchers are "diluting the power of their communication" as "there doesn't appear to be one umbrella organisation that is driving communication [about research] to the consumer." They agreed that a collective message would be more effective in reaching patients to get them involved in research.

Systematic approach for equitable inclusion: Some patients were concerned that many patients may never be approached, while others become stuck on "this treadmill of being on all these different things." Patients were confused and frustrated with privacy laws that prevented researchers contacting them through existing registries (e.g. ANZDATA) as they felt this denied them the opportunity to be involved. They suggested patients be presented with an opportunity to be involved in research through an opt-out registry for involvement in research for all patients with kidney disease – "Why can't we just have a kidney disease database [for researchers]?".

Streamlining access to opportunities for involvement and trustworthy information: A comprehensive and reliable information platform covering all nephrology research in Australia, "a one stop shop", such as an online "hub" or "portal" would give patients access to opportunities for involvement and connect researchers to patients who want to be involved.

Fostering a patient-centred culture

"It's about spinning it around and not talking about the clinical stuff. It's talking about the patient's reasons of why it's important, what's in it for the patient"

Health professional

Building a community: Patients, particularly those on home dialysis, often felt isolated and viewed involvement in research as an opportunity to connect with other patients and the research community through shared goals and experiences. Engaging patients in research could be improved if researchers facilitated the development of a community for research involvement and "took research to the patients" on their own terms in a convenient, informal, and unintimidating environment (e.g. patient support groups). This could provide opportunities for social interactions and educational discussions about kidney disease and research and establish connections for future relationships between patients and researchers.

Facilitating knowledge exchange and translation: Patients believed they would be motivated to become involved in research if they knew about current research in nephrology and understood how research and their involvement could improve care. They often felt that they lacked knowledge and information about their illness, and valued opportunities to learn more about their disease from health professionals. Dissemination of research findings to patients could be an effective method of engagement in research.

Empowering health ownership: Patients faced uncertainties with their prognoses and treatments, and therefore viewed their contributions to shaping and directing research as gaining ownership over their health. Whilst they may not glean benefits directly from being involved, they wanted opportunities to assert their priorities and preferences - “I believe in the power we have as consumers to change what’s important to us.”

Providing an opportunity to give back: Helping future generations, paying forward the benefit they have received from others, and giving something back to the community of health professionals who have cared for them motivated patients to become involved in research.

Cultivating trust through transparency: A culture of communication and transparency could address mistrust of the research community among patients, particularly regarding distribution and prioritisation of funding and researchers’ motivations. Patients identified a lack of involvement in grants and funding decisions resulting in resources being directed to research “contrary to what the patient might have picked”. Some believed researchers were “in there to get a degree” or were “led kicking and screaming” by mandates to involve patients. Being involved in all stages of the research cycle from applying for grants to disseminating findings and evaluating programs would serve to allay these fears and allow for trust to develop between the research and patient communities, to encourage more involvement in research.

5.5 Discussion

Patients and health professionals believed that involving patients as research partners required respect and recognition of the unique and complementary expertise of patients, consideration for the individual context, leveraging clinical and community networks for increased research exposure, and streamlined, systematic approaches that unify research silos to foster a patient-centered culture for research. To enact these, practical approaches were suggested which covered selection and engagement; training, support and education; empowering the patient voice; and connection and community (Table 5.2).

These workshops support previous findings suggesting the engagement and selection of patients for involvement across the research cycle requires a plan for diversity [1,5,36]. Attendees suggested that strategies should support the inclusion of a broad range of experiences through involving multiple patient partners in different capacities and different stages (i.e. advisory group, co-investigator). However, the notion that patients need to be “representative” to be able to make a meaningful contribution to research was challenged. It was argued that the intrinsic value of the unique expertise and lived experience that patients bring as research partners can serve to ground research in real world applications, resulting in research that is relevant and important to all stakeholders, supporting both the moralistic and consequentialist arguments previously raised for involving patients in research [3,35,53].

Consistent with existing patient involvement frameworks from organisations such as PCORI and INVOLVE, patient partners can be better supported to make meaningful contributions through the provision of education and training in research methods to help contextualise their role and strengthen their capacity to contribute [6,7,46,54]. Novel to these workshops was the recognition of the need to consider the individual context of the patient and the impact this might have on their capacity for involvement. Awareness and sensitivity to the patient's competing personal, professional and health-related priorities was expected to support respectful engagement by avoiding adding to overwhelming burdens at difficult times in the patient journey.

Empowering patients to contribute meaningfully is well recognised with increasing understanding that approaches such as having one patient on a steering committee can be tokenistic and disempowering[16]. A multipronged approach may be necessary to enable patients to confidently voice their needs and concerns, and detailed suggestions are available in Table 5.2.

Also distinct to these workshops was the concept of establishing a nation-wide community of patients and health professionals for research partnership. Patients identified an opportunity to combat isolation and build trust with the research community by connecting patients and researchers in informal and social environments where they can learn and gain from each other. Of note, attendees acknowledged the need for a systematic way to engage patients in research. They viewed current ethics laws preventing researchers from using existing databases to contact patients with opportunities for involvement as a hindrance, denying them opportunities to shape the future of kidney research and therefore their own health outcomes. This highlights opportunities to draw from principles of participant-centred initiatives that leverage advances in technology and use social media to address consent and privacy concerns, while establishing long-term interactive partnerships[55].

Whilst we were as inclusive as was feasible to obtain a broad range of perspectives, we note it is not possible to include a completely “representative” group of patients (or health professionals) as they are contributing based upon their individual experience and knowledge[56,57]. We acknowledge that the transferability of the findings to other settings is uncertain.

Partnering with patients in research requires respect and recognition of their unique, diverse and complementary experiential expertise. Multiple and flexible approaches are necessary to consider the patient circumstances, stage in their CKD journey, and existing burdens of their illness and treatment. Harnessing relationships with trusted clinicians and community organisations may help to broaden patient involvement and raise awareness of research in kidney disease, while innovative infrastructure to streamline the way patients engage with the research community can be further explored. Robust tools and frameworks to evaluate patient involvement in research are also needed[10,11,36].

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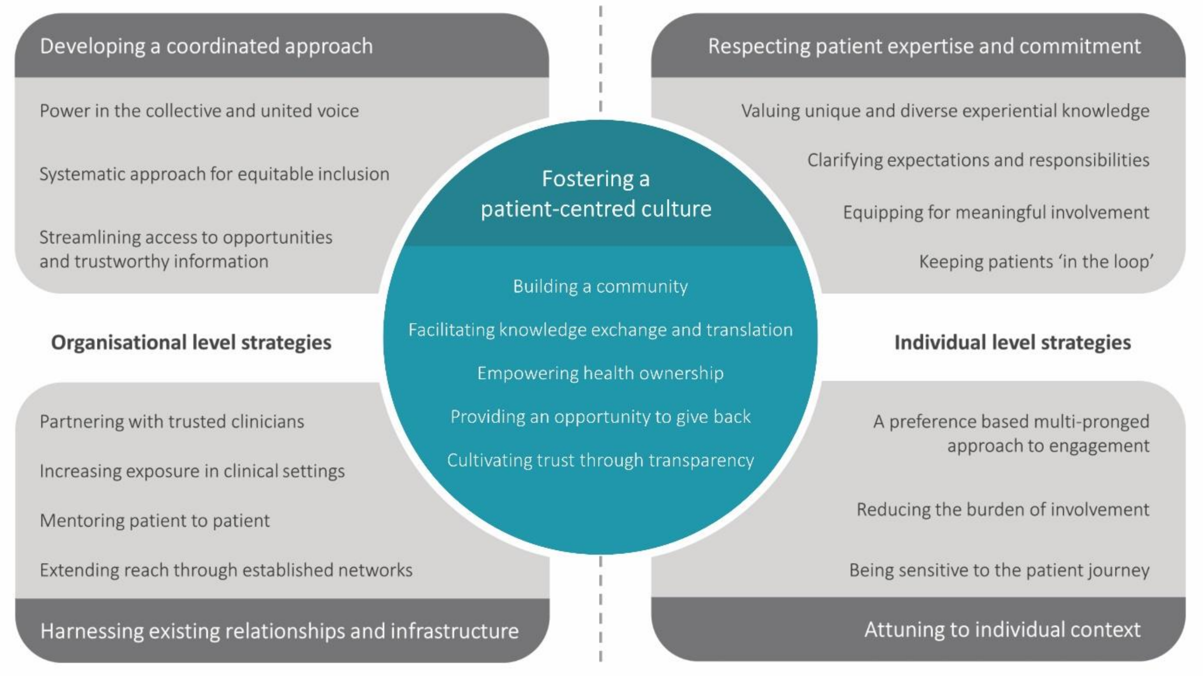

Figure 5.1 Strategies and principles for patient involvement in research

Workshop attendees identified strategies for patient involvement that could be addressed at an individual level (i.e. respecting patient expertise and commitment and attuning to individual context). However, some strategies may require collaborative efforts at organisational levels to instigate meaningful change (i.e. harnessing existing relationships and infrastructure and developing a coordinated approach). Underpinning these strategies were principles grounded in fostering a patient-centred culture and included building a community of patients and researchers for research partnership where both parties can learn and benefit from one another.

Table 5.1 Illustrative quotations

Theme	Illustrative Quotations
Respecting patient expertise and commitment	
Valuing unique and diverse experiential knowledge	Things like sleep or whether someone can go to work or someone's quality of life or their mental well-being we're just starting to measure. Patients probably could have told us 50 years ago that we should be measuring them, but they weren't asked. (Female, Sydney, Health Professional) You do bring in knowledge that they don't have. The reason people ask the wrong question is because they don't have the experience of living with it. (Male, Sydney, Patient) It puts a human focus on it. It's important to know who you're working for, not just a group or a consumer but actual people and faces and lives and stories. (Female, Sydney, Health Professional)
Clarifying expectations and responsibilities	One of the things that happens poorly is the explanation upfront as to why you're doing what you're doing, and what the potential benefits are, just the clarity of that. Because research can be kind of mystical, but it can also be really hands on and grounded if you explain it correctly. (Female, Adelaide, Health Professional) Maybe in the communication it could be about what it would mean in terms of the time that it would take to be involved? What the impact to our lives as a patient would be. (Female, Brisbane, Patient)
Equipping for meaningful involvement	I was horrified to hear that gentleman say they had a consumer involved in one of the studies, and they pulled out because it was too technical. Was that a matter of we won't replace the consumer because it's too hard for consumers or was it just the wrong consumer? You simplify or train, or if it is going to be at that level then you select those consumers. Either way, don't just go "it didn't work". (Female, Sydney, Patient) They're talking about a separate group that would inform the other groups, just a consumer group, because you don't want to get the ratio wrong either where you just have a whole bunch of doctors and then maybe one or two [patients]. (Female, Sydney, Patient) When you get more time to think about it, rather than closed in a room and put on the spot. (Female, Sydney, Patient)
Keeping patients 'in the loop'	We've got to tell you about research that's going on generally in the world. We probably have the duty to relay that. We currently don't do that at all. And the duty to tell people about what's going on, studies you can join, and then communicating the results of the work that you were engaged in as a person. I think we haven't done that well. We've done it a bit through the KHA website. You probably don't even know that. There should be a consumer summary. (Female, Brisbane, Health Professional)

	If I was going to contribute to things, I'd like there to be some way where you got to hear an end result or just progress. That's a frustration for me. If there was some way to be connected to some sort of feedback that you don't need a medical degree to understand. (Male, Sydney, Patient)
Attuning to individual context	
Preference based multipronged approach to engagement	I love the idea of a document that's patient-written for patients about what the research is about. What's going to work is a movie and one sheet of paper that summarises what the benefit to me or the benefit to the kidney community in general is. (Female, Brisbane, Patient) It makes complete sense that this gentleman here would like to have a booklet or a pamphlet, whereas my daughter who's 18 and just been diagnosed with a polycystic kidney disease is going to look at everything online, when she's ready or when she's interested. She's not interested in the piece of paper. Being able to deliver that useful information in different forms- (Female, Adelaide, Patient)
Reducing the burden of involvement	I used an annual leave day today. I don't care, but if it was a regular, well I'm saving my annual leave for my transplant. (Male, Sydney, Patient) It's the flexibility that will be the key. People need to be able to choose the level of engagement that they want to do. (Male, Sydney, Patient)
Being sensitive to the patient journey	The sicker I became, the more vague I became. Your attention span is really affected. You can't concentrate at all, focus. Whether that can be addressed in keeping notes very basic or something like that, rather than complicating. (Male, Sydney, Patient) They've got to have some sort of knowledge of when they hit you up for it [involvement]. Not when you're at your worst. Also, if you're on dialysis, you want to do it at home. You don't want to sit there for another hour. You've chewed up enough of your day, you've lost enough of your life. (Female, Brisbane, Patient)
Harnessing existing relationships and infrastructure	
Partnering with trusted clinicians	Every kidney patient goes and sees a nephrologist or a kidney doctor, the kidney specialist can play a bigger role than they do about talking to the patient about research. (Male, Brisbane, Patient) It needs to come from the nephrologist first, even just to say there's a website out there, or we've got this organisation if you're interested in research, because I think a lot of people probably don't even think that there's research going on. The idea needs to be put into their head first, that there is research going on in the area, and then they can pursue it if they are interested. (Female, Adelaide, Patient)
Increasing research exposure in clinical settings	When I go to a hospital, you normally see a poster wall about the papers. I think there's a volume of material already prepared, ready to be told to an audience, and you probably need to have some downscaling of the language in them to make them more suitable to the patients, and then you could put that in a newsletter. (Male, Brisbane, Patient) There's a TV in the waiting area. Instead of running commercials, you just run your ads for research. (Male, Brisbane, Patient)

Mentoring patient-to-patient	I found [the patient's] talk very helpful, because he's been through it. And he was able to talk to me about how it works. The more you hear about the process, the more you start to think, yeah, I could probably cope with that. (Male, Sydney, Patient) It's just that if you do the patient to patient interactions though, you'll have to mediate it really carefully. My experience with a lot of them is that they turn into misery pits. (Female, Sydney, Patient)
Extending reach through established networks	If we could piggyback onto diabetes research, I think that might raise a lot of public awareness. (Female, Adelaide, Patient) I went to the hospital, they were set up in the foyer, kidney health week, we'll take your blood pressure. I said people, you're in the wrong spot. People who come into the hospital already know they've got high blood pressure. You should be outside at Bunnings [Hardware Store]. That's exactly right. they said that's not a bad idea. Where do you reckon they'll be in March this year? Sitting in the hospital. (Male, Brisbane, Patient)
Developing a coordinated approach	
Power in the collective and united voice	There are so many not for profit organisations or bodies, but there doesn't seem to be one umbrella organisation that's driving all the communications to the consumer, to the patient, so everyone knows where to look. I think everyone's trying to do the right thing. There are so many of them that it's diluting the power of the communication. (Male, Adelaide, Patient) If it's not central or under one banner, you get competing priorities between organisations. (Male, Sydney, Health Professional) If you all get together, you're all there for one common cause, but being separate, it's not really helping anyone. (Female, Sydney, Patient)
Systematic approach for equitable inclusion	You're not increasing your pool if you keep on asking the same people over and over again, there are new patients being added all the time. There's nothing upfront, a survey or whatever, to say are you interested, do you want to participate in this sort of thing? (Male, Brisbane, Patient) I know they have difficulty finding people to put themselves forward. In some cases it's always the same person who puts themselves forward, and they may not necessarily be the right person, but because they're on this treadmill of being on all these different things, then the true reflection of somebody with a passion to be on it doesn't get on it. (Female, Adelaide, Patient) Why not? Why can't we just have a kidney disease database? (Male, Brisbane, Patient) We really need to get a database of people and see whether they actually want to be contacted. (Male, Brisbane, Patient)
Streamlining access to opportunities and trustworthy information	If you had a one stop shop. If there could be a tailored site where kidney-specific things could be put – perhaps research results, or trials or whatever. Something that ordinary people can understand, because we're not all medically inclined. (Female, Brisbane, Patient) Almost like a dating site. You know what I mean. A Tinder trial site. (Female, Adelaide, Patient) There should be a central hub where you can find results, you can find organisations you can join, you can find research you can be involved in. (Male, Sydney, Patient)

Fostering a patient-centred culture	
Building a community	Our approach to communication becomes sanitised. The coffee group idea gives a sense of a relaxed atmosphere. Just having a group like that, where it’s just a general friendship group, and you’re not there for a purpose, you’re there just because you got commonalities. (Male, Adelaide, Health Professional) It’s about building a community first, is that right? (Female, Adelaide, Health Professional) Take the questions to the people where you know your groups are on the day, instead of you trying to say can you listen to us? (Female, Brisbane, Patient)
Facilitating knowledge exchange and translation	Gaining information empowers you and that helps you to make decisions about how you can involve or improve your care. Gaining information gives you insight as to what type of research you might want to be interested in or involved in, and what capacity that you want to be. (Female, Brisbane, Health Professional) That’s really what we want [researchers to speak informally at patient meetings]. We had a transplant lady come and talk to us all about what they do with transplants. We would all come around, that was just fantastic. (Female, Adelaide, Patient)
Empowering health ownership	We need to explain what’s in it for the patient, what’s the opportunity that it might present, what’s the potential benefit for the future, and why is it important for patients to get involved. If we make the messaging all speared towards why the consumer’s part of this is so important, we’ll get much better uptake. It’s spinning it around and not talking about the clinical stuff. It’s talking about the patient’s reasons of why it’s important. (Female, Brisbane, Health Professional) I believe in the power that we have as consumers to change what’s important to us, and make sure that we understand what’s going on in the research community. (Female, Brisbane, Patient)
Providing an opportunity to give back	The way that I feel about it is that I got so much out from all the people, the doctors and the nurses who looked after me over the years. It’s been fantastic, really. I feel that anything I can contribute, I’d be very happy to have an opportunity to do anything. (Male, Sydney, Patient) Why would someone want to participate, it’s because they want to help other people. (Male, Adelaide, Patient)
Cultivating trust through transparency	I just thought research was a waste of money up until today, because you never heard of an outcome. (Male, Adelaide, Patient) There’s sort of a them and us mentality. Most of these researchers, they’re in there as good guys. They have some bad habits, we all do, but they’re in there to help us, and we have to respect that as part of the process. Some of them are [there to get a degree], but it’s actually good to engage younger researchers and develop the culture of research as we move on in time. (Male, Sydney, Patient)

Table 5.2 Suggestions for effective engagement and involvement

Domain	Considerations and suggestions from workshop participants for patient/caregiver involvement
Engagement and selection	• Plan and budget for patient/caregiver involvement at the earliest stage of the research project. • Clearly define role (i.e. level of involvement, stage of research, specific responsibilities), required commitment (i.e. time, financial, emotional), and expected impact (i.e. directly on a specific project, on the wider kidney community agenda, or expected benefits for future generations) with ‘terms of reference’ document, allow time for reflection and questions. • Roles may vary by project and/or research stage and can range from consultation to partnership. • Consider the potential benefits for the patient/caregiver and clearly explain these (e.g. developing new skills, greater understanding of disease, contributing to research for future generations). • Consider the cross-section of patients/caregivers involved (e.g. urban vs. rural, experienced vs. new, older vs. younger, well vs. unwell) and structure projects to enable broad participation (e.g. join from remote locations, schedule meetings outside of business hours). • Select patients/caregivers for whom your project is most relevant, and whose expertise is best suited (e.g. target peritoneal dialysis patients for projects/interventions designed for peritoneal dialysis patients). • Use engagement methods preferred by target population (e.g. phone/letter for older patients, social media for younger patients, in clinic/center for hemodialysis patients). • Advertise opportunities in waiting rooms (e.g. videos, pamphlets, posters). • Work with physicians and nurses to engage patients/caregivers. • Ask patients to suggest new patients to be involved, consider having patient mentors to induct new patients into research projects.
Training, support and education	• Provide education for patient/caregiver partners (e.g. current research activities, kidney disease). • Provide training in research methods (e.g. study design, academic writing, critical appraisal, recruitment). • Consider psychological, mental and physical demands of involvement: ○ Ensure environment is accessible and can accommodate for patient needs (e.g. place to do peritoneal dialysis, elevator access). ○ Use communication strategies and simple language to reduce cognitive burden of involvement on patients. ○ Have referrals available in case of distress (e.g. social worker). ○ Offer flexible meeting options (e.g. videoconferencing, regular breaks, time for questions). ○ Where possible, embed opportunities into routine care (e.g. transplant clinics). • Financial reimbursement/aid may assist patients/caregiver to become involved: ○ Consider financial burden/sacrifice of involvement on patients/caregivers, and at a minimum, cover costs incurred (e.g. travel expenses, time off work). ○ Consider implementing a ‘jury duty’ system to reimburse patients/caregivers for their time. ○ Consider budgeting for paid patient/caregiver research partner position/s.

Empowering the patient voice	• Appoint more than one patient representative on a steering committee/working group. • Consider forming a diverse patient advisory group to oversee research activities. • Produce induction packets with relevant materials (e.g. glossary of terms/acronyms, background reading, helpful resources, videos). • Use lay language where possible and explain technical/medical terminology when necessary. • Allow time before or after meetings for patients/caregivers to consider their response/opinion (e.g. send meeting agenda with topics to be discussed, follow up post-meeting). • Involve across all stages of the research, however early involvement, before grant stage, allows patient/caregiver contributions to be more easily integrated. • Partner patient/caregiver with experienced research ‘buddy’ whom they can reach out to with any problems, questions or suggestions.
Connection and community	• Establish and expand a database to contact patients. • Consider building a patient dedicated research showcase portal/hub with lay language summaries of research and impacts, and opportunities for involvement to connect patients with researchers with potential for patients/caregivers to customise their profiles (interests, experience, CKD stage etc…). • Keep communications open with regular updates via newsletters, emails, texts, and/or social media, even if no formal results are available. • Collaborate with patient/caregiver and community organisations (e.g. present at patient support groups) to raise awareness of kidney disease, encourage knowledge exchange and advertise opportunities for involvement. • Establish relationships with other research organisations to pool resources for patient/caregiver engagement to mitigate competing priorities, leverage collective goals and streamline communications to patients/caregivers. • Disseminate research findings in plain language and informal settings to patients/caregivers to educate them about the outcomes and impact of research (including the impact of their involvement), build trust with the research community and encourage future involvement.

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Chapter 6: “Can I go to Glasgow?” Learnings from patient involvement at the 17th Congress of the International Society for Peritoneal Dialysis (ISPD)

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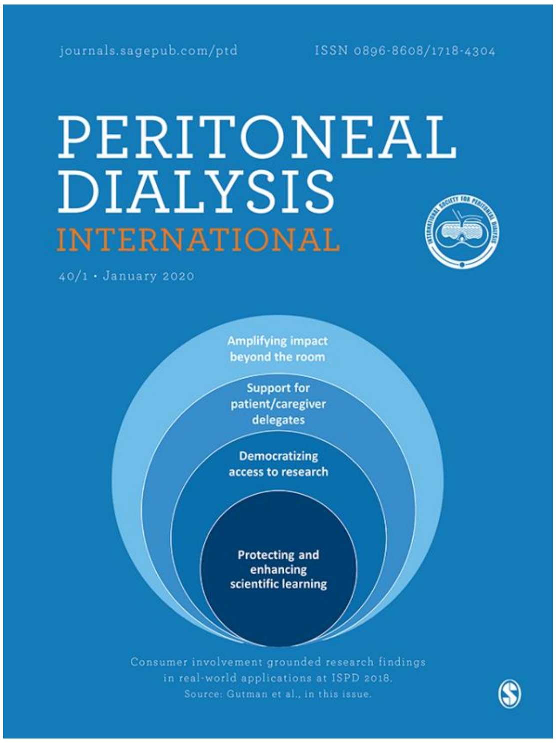

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Amplifying impact beyond the room

Support for patient/caregiver delegates

Democratizing access to research

Protecting and enhancing scientific learning

Consumer involvement grounded research findings
in real-world applications at ISPD 2018.
Source: Gutman et al., in this issue.

This chapter is structured as per the journal article.

6.1 Abstract

Background: Recognition of the discrepancy between the research priorities of patients and health professionals has prompted efforts to involve patients as active contributors in research activities, including scientific conferences. However, there is limited evidence about the experience, challenges, and impacts of patient involvement to inform best practice. This study aims to describe patient and health professional perspectives on patient involvement at the Congress of the International Society for Peritoneal Dialysis (ISPD).

Methods: Semi-structured interviews were conducted with 14 patients/caregivers and 15 health professionals from six countries who attended ISPD. Interviews were recorded and transcribed verbatim, and transcripts were analyzed thematically.

Results: We identified four themes: protecting and enhancing scientific learning (grounding science in stories, sharing and inspiring new perspectives, distilling the key messages of research presentations, striking a balance between accommodating patients and presenting the science); democratizing access to research (redistributing power, challenging the traditional ownership of knowledge, cultivating self-management through demystifying research); inadequate support for patient/caregiver delegates (lacking purposeful inclusion, challenges in interpreting research findings, soliciting medical advice, difficulty negotiating venue and program, limited financial assistance in attending); and amplifying impact beyond the room (sparking innovation in practice, giving patients and families hope for the future).

Conclusions: Patient involvement at the ISPD Congress clarified the applicability of research to patient care and self-management, democratized science, and strengthened the potential impact of research. More structured support for patients to help them purposefully articulate

their experience in relation to session objectives may enhance their contribution and their own learning experience.

6.2 Introduction

Growing recognition of the discrepancies between the priorities of patients and health professionals has prompted widespread efforts to involve patients and their families as active contributors in research activities, including scientific conferences[1-5]. In 1992, the Eighth International AIDS Conference catalyzed the “patients included” phenomenon when they invited patients to attend[6]. Since then, scientific meetings such as the Outcome Measures in Rheumatology (OMERACT) and Medicine 2.0 have patients comprising 10% of delegates. These patient delegates are involved in designing the conference program, presenting, and chairing sessions. More recently, patients have been included in nephrology conferences. In 2018, for example, patients convened and presented a session on “Developing Therapies That Matter Most to Patients: A Patient-Centered Approach to Innovation” at the American Society of Nephrology 2018 Kidney Week in San Diego[7]. At the Australian and New Zealand Society of Nephrology Annual Scientific Meeting (ANZSN ASM) a patient gave a plenary presentation, and special educational sessions were developed and co-chaired for and by patients and caregivers and live-streamed globablly[8,9].

Patient attendance and contributions at conferences have led to innovation and widening of research agendas through meaningful discussions, alignment of research priorities with patient needs, diversifying opportunities for dissemination, and establishing researcher-patient collaborations[2,3,8-12]. The Patients Included Charter for conferences[10,13,14] provides a framework for organizations to demonstrate “their commitment to incorporating the experience and insight of patients"[13] by having patients "on the program, on the stage and in the audience"[15,16]. In July 2016, the Accreditation Council for Continuing Medical Education (CME) announced new criteria for accreditation with commendation that includes engaging patients/public in the planning and delivery of CMEs as planners and faculty in the accredited conference or program[2,17]. Despite this, there has been little evaluation of patient involvement in scientific conferences, and concerns remain regarding tokenism, the power dynamic between patients and clinicians, dilution of scientific rigor and financial burdens associated with patient/caregiver attendees[18,19]. Further insights from patients and health professionals are needed to strengthen strategies and actions for involving patients in medical conferences to ensure that they are empowered to make meaningful contributions and to mitigate any risks of involvement.

As a home-based therapy, peritoneal dialysis (PD) success is dependent on patient/caregiver knowledge and self-management and is a poignant example of the potential for patient-clinician partnership in education. In support of the conference theme "patient-centered care", the International Society for Peritoneal Dialysis (ISPD) Scientific Committee opened registration to the 17th Congress to all patients and their families. We aimed to describe patient, caregiver and health professional experiences and perspectives on the process and impact of patient/caregiver involvement at the Congress of the ISPD to inform future efforts to involve patients in conferences, including in Glasgow for ISPD 2020.

6.3 Methods

We used the Consolidated Criteria for Reporting Qualitative Studies (COREQ) to report this study[20].

Context

For the first time, patients and their families were invited to attend and/or to speak at the international biennial ISPD Congress hosted in Vancouver, Canada, from the 6th to 8th May 2018, by their nephrologists and through hospitals and dialysis clinics. Registration fees were waived for patients/caregivers. In parallel to other pre-Congress workshops, a patient and family education day was held on Saturday 5th May 2018 targeted at patient attendees with patient-important topics, however all delegates could choose to attend any of the parallel sessions. Patients were included as speakers, panelists and/or co-chairs in most sessions during the main conference. Patients were identified as ‘delegates’ on their registration badges, and not distinguishable from other attendees. In total 1465 delegates registered to attend part or all of the conference including physicians (n=734), nurses (n=218), patients/caregivers (n=128), nephrology fellows (n=82), pharmacists (n=30), dieticians (n=22) and other (n=251). Eighty-three patients/caregivers registered to attend the congress and the pre-congress workshop, and 45 registered for the congress only. To support patients on PD in attending, a patient lounge with nursing staff was available for patients to rest or perform a bag exchange. The Congress program was available via a mobile app, which was also used in sessions for live polling and questions.

Participant selection

Health professionals and patients with any stage of chronic kidney disease (CKD) and their family members who were delegates of the Congress, and were English-speaking and aged 18 years or over, were eligible to participate. We initially aimed to recruit 30 participants and applied a purposive sampling strategy to capture a diverse range of demographic (i.e. age, gender, country, profession) and clinical characteristics, and roles at the conference (i.e.

delegate, speaker, chair). Participants were identified from the program, mobile application, or through face-to-face meetings at the conference and invited via email/message to participate. Informed consent was obtained from all participants. Ethics approval was granted by The University of Sydney (2018/136).

Data collection

TG conducted semi-structured interviews with participants, either face-to-face when possible or via video-conference from May to June 2018. The interview guide was developed based on a review of the literature[1,3,6,10,12,13,18,21] and discussion with investigators (Appendix E.1). All interviews were audio recorded and transcribed verbatim. We conducted interviews until we reached data saturation within each stakeholder group (i.e. patients/caregivers and health professionals).

Analysis

Using thematic analysis and principles from grounded theory[22], TG read the transcripts line-by-line and inductively identified preliminary themes relating to participants perspectives on patient involvement in the conference. These themes were discussed and revised by AT, KM and AB, who had also independently read the transcripts, to ensure the coding structure captured the full range and breadth of the data before being entered into HyperRESEARCH software for coding and thematic analysis[23]. The preliminary findings were sent to participants to provide feedback within a two-week timeframe, and additional concepts were integrated into the final analysis (i.e. member-checking)[24].

6.4 Results

We contacted 42 participants: 11, did not respond, 31 agreed to be interviewed, and 2 interviews could not be completed (no follow up response). Of the 29 participants, nine (31%) were patients and five (17%) were caregivers from Canada (n=13) and United States (n=1), and 15 (52%) were health professionals (nephrologists, physicians, nurses, pharmacists and dietitians) from Australia (n=2), Brazil (n=1), Canada (n=7), Sweden (n=1), United Kingdom (n=2), United States (n=2). The participant characteristics are shown in Table 6.1. The duration of interviews ranged from 15 to 50 minutes (mean 25 minutes). Nineteen (66%) were conducted in person and the remainder were conducted via video-conference.

We identified four themes: protecting and enhancing scientific learning; democratizing access to research; inadequate support for patient/caregiver delegates; and amplifying impact beyond the room. The following section describes the respective subthemes and the description reflects the perspectives from all groups (patients/caregivers and health professionals) unless otherwise specified. Selected quotations to support each theme are available in Table 6.2, and suggestions and considerations for patient/caregiver involvement in conferences are provided in Table 6.3. Figure 6.1 shows the relationships between themes and subthemes.

Protecting and enhancing scientific learning

Grounding science in stories: At the conference, patients/caregivers discussed how PD allowed flexibility in their lifestyle, and expressed their fears about technique failure, which served to “testify” to health professionals’ presentations, validate research outcomes and make the results more applicable to the day-to-day work of clinicians. Hearing directly from patients/caregivers was “humanizing” and gave the presentations more “depth” and “authenticity”, reminding health professionals “why we are here”, which they acknowledged “often gets lost in esoteric academic discussions [at conferences]”. Some noted that having a patient/caregiver tell their story at the beginning of a session “put a face on the science”, “reshaped the way people are listening to the information that follows” and opened the door to challenging conversations on patient-centered topics, for example caregiver burden and bereavement.

Sharing and inspiring new perspectives: Given the dearth of “hard evidence” in nephrology, “insider perspectives” from patients/caregivers “shed a new light” on topics such as symptom burden and equity of access. Patients/caregivers discussed the impacts of PD on outcomes such as their ability to work and travel. Seeing the research “through another lens” highlighted “how sometimes health care professionals are out of touch with patient and caregiver needs”, which impacted the way some health professionals interpreted findings. This also emphasized the need to include patient-reported outcomes (e.g. fatigue) in clinical trials. Discussions between patients/caregivers and health professionals allowed for deliberation about “what the paper says should happen versus what reality shows when you add in the patient factor.” Particularly for nurses, “connecting knowledge and research to how the patient is actually going to deal with and process that information” enabled them to understand and integrate competing priorities, for example being able to swim while avoiding catheter complications and infections.

Distilling the key messages of research presentations: Allied health professionals and younger nephrologists noted that presentations were more “digestible” for all attendees because patients/caregivers were present. They noted that speakers emphasized the key points of the research, relevance to clinical practice, impact on patient outcomes, and avoided jargon

– “[speaker] did an amazing job of making the topic fun, interesting, filtering enough scientific evidence to prove his point but keeping it at level that was easily interpreted, whether you’re a patient or a PhD times five.”

Striking a balance between accommodating patients and presenting the science:
Patients/caregivers did not expect speakers to alter the content of presentations as they did not want to detract from professional learning. Whilst some more experienced health professionals, particularly speakers, were concerned that the quantity of content was reduced to cover the context more clearly for patients/caregivers, others stressed that the purpose of the conference was to share new ideas and that details (i.e. description of methodology) could be disseminated through other platforms such as journal articles. Health professionals believed that having parallel sessions for patients/caregivers provided options for delegates without compromising the delivery of scientific content. Both groups valued the ability to attend either “track”.

Democratizing access to research

Redistributing power: Patients/caregivers were empowered by having their voices heard by health professionals in an international forum and believed their involvement challenged the traditional patient-clinician relationship. One patient explained that it is “not the medical procedure that affects us most, it’s the whole relationship that has to be improved”. Health professionals explained that “none of us is of higher value than anyone else” and “we want patients involved and educated so they can challenge us” to ensure their needs are addressed. Some nephrologists stressed the importance of learning from patients in an external conference setting because in the context of their clinical practice the “patient is the hostage and we are the captor. It takes a really confident patient to be able to tell you what they really think, because they're actually in a dependent situation."

Challenging the traditional ownership of knowledge: Health professionals explained that "the point of research is not only for researchers." Inviting patients/caregivers to be "part of the conversation" challenged the traditional hierarchy of knowledge ownership by health professionals and gave patients/caregivers direct access to research findings.

Cultivating self-management through demystifying research: Attending research presentations allowed patients/caregivers to access "deeper information" rather than the "bits and pieces" they received during clinical appointments. The conference presentations gave them further insight related to the advice they received from their clinicians. One patient explained that by attending the "less is more in PD" session they understood why their nephrologist was not worried that they missed an exchange while they were traveling. Health professionals explained that PD patients were an "engaged" and "motivated" cohort with greater responsibility for their own care than most patients and should have access to information to strengthen their capacity for self-management, for example to prevent peritonitis.

Inadequate support for patient/caregiver delegates

Lacking purposeful inclusion: While patients/caregivers appreciated the opportunity to speak, some felt telling their story was "not enough". Some were unsure about the purpose of their talk, did not relate to the topic they were asked to speak about, or felt they had inadequate guidance in preparing to integrate their perspectives with the other speakers. Some health professionals noted that patient/caregiver talks could have been more "specific" and "purposeful", articulating the parts of their journey that aligned with the learning objectives of the session. For example, health professionals wanted to know what patients were struggling with regarding peritonitis in the session on PD infection.

Challenges in interpreting research findings: While patients/caregivers did not expect to understand everything some health professionals worried that patients "may hear something that wasn't said" and were concerned that patients would relay "misinformation" back to their nephrologist. Others were concerned about the "blunt" and insensitive way some health professionals presented data such as mortality risk, which may cause unnecessary distress for patients/caregivers. No patients/caregivers reported any distress from what they had heard.

Soliciting medical advice: Some nephrologists were uncomfortable with patients asking for specific individual medical advice. One nephrologist was concerned that patients "hijacked" the sessions by asking about "their own personal concerns". Another nephrologist recalled a patient asking about her blood pressure during a session and noted that the chair gave a general answer and suggested that she should consult with her nephrologist.

Difficulty negotiating venue and program: Some older patients/caregivers struggled with walking long distances and navigating the conference with no paper program. It was challenging for them to assess which sessions would be most beneficial for them to attend because they had difficulty understanding the language in the program. For example, the term 'basic science' was misunderstood to be a basic outline of the research - "It was called basics, and they were basics to the medical profession but not to the patient. That was a little bit misleading in that title."

<u>Limited financial assistance in attending:</u> While registration was free and many patients/caregivers had their parking or travel costs covered by the British Columbia Renal Agency, some patients/caregivers were frustrated that they did not receive financial assistance for accommodation or honoraria to speak. For some, this meant that they were only able to attend the conference for one day as accommodation and time off work were too costly. Both health professionals and patients/caregivers commented that they knew of patients who wanted to attend but did not due to the expense.

Amplifying impact beyond the room

<u>Sparking innovation in practice:</u> After hearing the patient/caregiver perspective on topics such as transitioning from PD to hemodialysis, health professionals discussed changing the way they explained these topics to their patients to ensure they were understood and addressed. The patient and caregiver presentation on PD and intimacy inspired some health professionals to be proactive in developing education programs for staff and patients. Some health professionals, particularly nurses, began to think more broadly than symptom management, to the impacts on quality of life such as being able to swim or travel. Health professionals reported that interacting with patients/caregivers helped them to see the value in consumer involvement in other areas, such as prioritizing topics or identifying patient-centered outcomes, which encouraged them to consider more meaningful partnerships in future projects rather than viewing it as a “tickbox exercise”.

<u>Giving patients and families hope for the future:</u> Patients/caregivers felt they were able “see behind the curtain” and understood that “[clinicians] do not have all the answers, but we’re looking”, which gave patients/caregivers reassurance that progress is being made and instilled confidence in their clinical team. It was “uplifting” for caregivers to see patients engaging

with health professionals as equals in panel discussions. The opportunity to meet and learn from other patients with different stages of CKD and treatment provided comfort in shared experiences and hope for their future.

6.5 Discussion

Patient involvement in the 17th Congress of the ISPD allowed health professionals to consume research through a different lens, where patient priorities and care were at the forefront. It also empowered patients/caregivers to join the conversation as equals, contributing their expertise and experiences to shared learning, and gaining knowledge to take ownership of their health. However, health professionals and patients/caregivers suggested that patient/caregiver contributions should be more targeted and relevant to conference learning objectives, and support should be offered to patients/caregivers to enable them to be active delegates. The limited financial support for patients/caregivers may have prevented more extensive and substantive contribution. Despite the challenges identified, most health professionals believed there was inherent value in having patients/caregivers included, and some were motivated to change their practice because of what they had learned from patients/caregivers. Patients/caregivers gained insight into research and how it informs their care and treatment, giving them comfort and strengthening trust in their health care team.

Allied health professionals and younger nephrologists in particular perceived the patient/caregiver voice as adding to their professional learning as the presenters drew direct links between the research and patient care and experience. Some clinicians advocated to simplify the language and refine core messages of research as this would benefit not only patients/caregivers, but also themselves. However, some nephrologists who had been practicing for a longer period felt they already had extensive experience working closely with their own patients.

In previous studies, concerns have been raised regarding the power dynamic between patients and their clinicians which may hinder patients from sharing their true perspectives[18,25]. Some health professionals argued that it was important to hear from patients/caregivers who were not directly under their care, as their own patients may feel disempowered in clinical settings to express their views. However, in this conference, patients felt supported and encouraged when their own nephrologist was present and did not feel that it inhibited their contribution. Moreover, patients/caregivers believed having their voice heard by international health professionals alongside the science empowered them to impact the way health professionals interpreted findings based on their experiences and priorities. It has been recognized previously that partnership with patients reflects a “a fundamental shift in the power structure in health care”[19], reinforcing the concept of “relational empowerment”[3]. Equal participation rights for patients/caregivers in this conference and direct access to information about their disease and treatment challenged the traditional hierarchy in the patient-clinician relationship, serving to equalize the power distribution.

Our findings identified similar challenges with, facilitators for and impacts of involving patients in scientific conferences as have been reported in previous studies[6,8,12,18,21]. Responsive evaluations by the OMERACT group also found that some patients who attended conferences felt unprepared and unsure of their role and what was expected of them[21]. Support organized by a patient-led liaison group, including the development of introduction packs, glossaries, information modules and a training day, facilitated better inclusion, which was also suggested by some of the participants in this study (Table 6.3)[12]. Patient involvement in the OMERACT conferences has resulted in the study of important patient-reported outcomes such as well-being, flares and sleep disturbances, and the inclusion of fatigue in the rheumatoid arthritis core outcome set[12,18]. Likewise, patient involvement at the ISPD Congress brought patient-important outcomes such as fatigue, intimacy, travel and physical activity to the forefront of scientific discussions, emphasizing the need to address these in research.

A survey conducted with health professionals from the UK who attended the AIDS conference in Amsterdam in 1992 found that 50% and 57% reported increased motivation for their clinical work and research respectively[6]. Our study echoes these findings of impact beyond the scope of the conference, with health professionals motivated to make changes to their practice and research agendas to make them more patient-centered. More recently, patients were involved in co-designing two patient research sessions for the 54th ANZSN ASM[8]. Members of the Better Evidence and Translation in Chronic Kidney Disease (BEAT-CKD) Consumer Advisory Board were responsible for determining the aims, topics and formats of the patient programs, and hosted two sessions with participants attending in person and via free live streaming. Recommendations from this evaluation to advertise to patient delegates early, waive registration fees, include patients as co-designers, provide briefing and support for patient speakers in advance, encourage inclusiveness through multiple attendance modes, and provide supplementary materials including slides and recorded presentations after the conference also aligned with our findings (Table 6.3)[8]. Participants also suggested that all patients/caregivers should be named delegates in the program (with permission), acknowledged, be financially supported to attend, given a clear brief to speak on, and have the opportunity to shape the agenda of the conference and specific sessions, which have previously been suggested by patient partners[1,11,15,26].

Patients have advocated to be seen as “more than [their] stories”[3,9,11], and in our study both health professionals and patients/caregivers acknowledged that inviting patients to share their stories was a powerful first step, however it was “not enough” on its own, and that for their involvement to have a greater impact they needed support to tailor the presentation of their experiences to the learning objectives of the meeting. We also found that patients wanted to harness the knowledge gained about their disease and treatment to improve their capacity for self-management and take more ownership of their health, which could ultimately improve outcomes[3,12].

Recently, frameworks have been developed to support successful involvement of patients in conferences. The ‘Stanford Framework for Patient Partnership in conferences’ encompasses four pillars of accommodation, codesign, engagement and education[10]. The ‘Patients Included Charter’ mandates that patients be involved in the design and planning of the event, be included in the program as speakers and delegates, have their accommodation and travel expenses paid for in full in advance, have any disability requirements accommodated for, and be provided free remote access through video streaming where possible[13]. Our study highlights specific areas to strengthen and improve patient involvement including goal-directed presentations that align with conference/session learning objectives, teamed presentations between researchers and patients/caregivers, reducing the administrative and organizational burden for patients through arranging travel and accommodation on their behalf, developing and distributing induction packets prior to and summary packets post conference to maximize patient/caregiver learning, assigning a contact person for patients/caregivers during the conference, allocating time to patient questions, providing opportunities for patient networking early in the conference and debriefing toward the end of the conference, and keeping patients/caregivers connected and informed about future opportunities (Table 6.3).

Involving patients in conferences has the potential to enhance education for health professionals and patients/caregivers. We suggest that the roles for patients should be clearly defined, and support provided to enable patients to deliver purposeful presentations that are synergistic with the other speakers and align with the learning objectives of the meeting/session. Education and training programs to support these efforts may be considered for patient speakers at medical conferences. For example Can-SOLVE CKD Network has developed a flexible online workshop titled “Storytelling for Impact” that provides patient partners with coaching and tools “to support compelling and impactful storytelling”, helping patients to articulate the parts of their journey that are most relevant in the context of the research[27]. Addressing topics that are of interest and importance to patients, in formats that are preferred by them, and at times and modes that are accessible, are also suggested.

We applied a purposive sampling strategy to elicit a broad range of views, and investigator triangulation and member checking ensured that thematic analysis captured the full range and depth of the data. However, there are some potential limitations. All participants were English-speaking, and of the patients/caregivers only one was not Canadian, which may limit the transferability of the findings to other regions or populations.

Patient/caregiver involvement at the ISPD Congress clarified the applicability of research to patient care and self-management, democratized science, and strengthened the potential impact of research. Perspectives from and interactions with patients/caregivers enriched health professionals’ learning by centering the research on patient/caregiver priorities. Patients/caregivers were empowered to be active delegates with equal participation rights, giving them direct access to knowledge and insights about their treatment and care which they believed would lead to improved self-management vital for successful PD. Having well-defined and co-produced objectives and more structured financial and academic support for patients/caregivers may enhance their involvement, contribution, and learning experience at scientific conferences.

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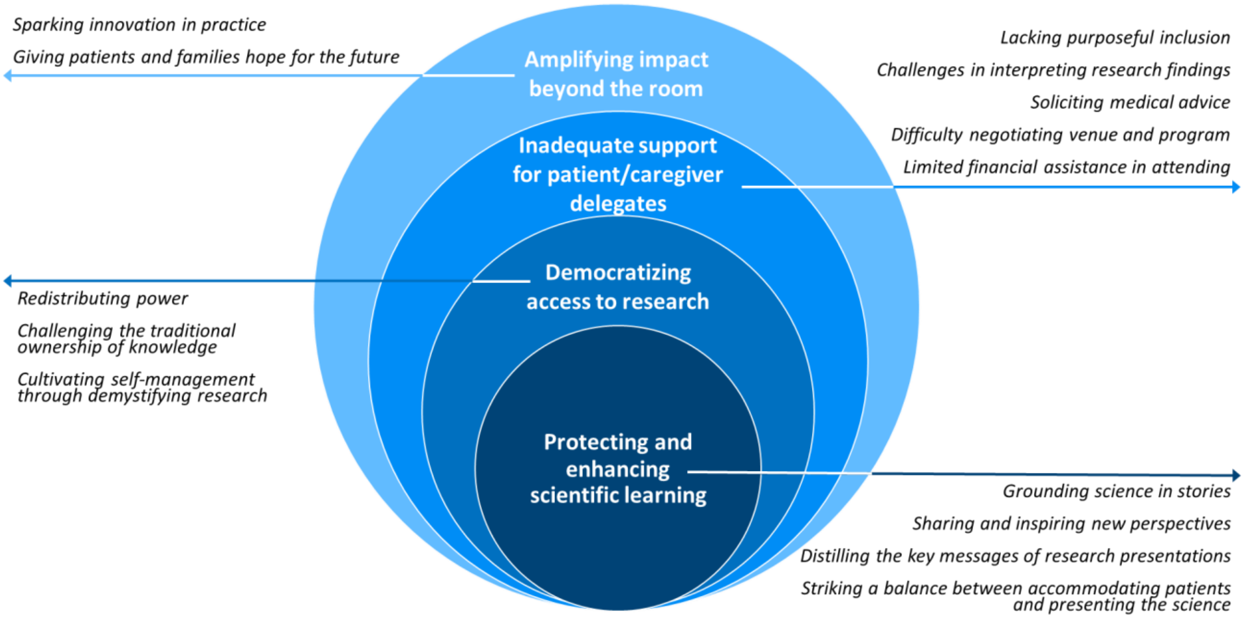

Figure 6.1 Thematic schema

At the core of conferences is the scientific learning that takes place. Inviting and involving patients and caregivers in conferences enhances this learning through linking research to the real world and highlighting priorities for clinical settings. Distilling key messages of research presentations to accommodate lay audiences can benefit all delegates, particularly patients/caregivers and less experienced health professionals. Involving patients/caregivers challenges the traditional ownership of scientific knowledge thereby democratizing access to information that can be used to improve self-management and build trust between patients and clinicians, despite concerns from health professionals about patients’ capacity to interpret this information. However, inadequate support for patient delegates can limit their potential to both contribute to and gain from the conference. Purposeful involvement that weaves patient perspectives together with session learning objectives and ensures patients understand their role and capacity to contribute can inspire changes in practice (both clinical and research), lead to future research partnerships, and give patients and their families hope for the future, while serving to enhance scientific learning through impactful storytelling and confidence in voicing innovative perspectives.

Table 6.1 Participant characteristics n=29

Characteristic	**Patients/caregivers (n=14) n (%)**	**Health professionals (n=15) n (%)**	**All n (%)**
Role			
Patient	9 (64)	-	9 (31)
Caregiver	5 (36)	-	5 (17)
Nephrologist	-	7 (47)	7 (24)
Nurse	-	5 (33)	5 (17)
Other (Pharmacist, dietitian, Internist)	-	3 (20)	3 (10)
Female	8 (57)	9 (60)	17 (59)
Age (years)^			
<30	1 (7)	-	1 (3)
31-40	-	6 (40)	6 (21)
41-50	-	2 (13)	2 (7)
51-60	2 (14)	3 (20)	5 (17)
61-70	6 (43)	3 (20)	9 (31)
>70	4 (29)	-	4 (14)
Country of residence			
Canada	13 (93)	7 (47)	20 (69)
United States	1 (7)	2 (13)	3 (10)
Other*	-	6 (40)	6 (21)
Country of birth			
Canada	8 (57)	4 (27)	12 (41)
United States	2 (14)	2 (13)	4 (14)
United Kingdom	-	3 (20)	3 (10)
Not reported	1 (7)	1 (7)	3 (10)
Other**	2 (14)	5 (33)	7 (24)
Education level			
10th Grade	2 (14)	-	2 (7)
12th Grade	2 (14)	-	2 (7)
Professional certificate	5 (36)	-	5 (17)
Undergraduate degree	2 (14)	5 (33)	7 (24)
Postgraduate degree	2 (14)	10 (67)	12 (41)
Not reported	1 (7)	-	1 (3)
Employment status			
Full time	2 (14)	12 (80)	14 (48)
Part time	-	1 (7)	1 (3)
Retired	7 (50)	1 (7)	8 (28)
Other***	4 (29)	-	4 (14)
Not reported	1 (7)	1 (7)	2 (7)

^missing n=2; * including Australia, Brazil, Sweden, United Kingdom; ** including Australia, Brazil, El Salvador, Hong Kong, Jamaica, Slovakia, Sweden; ***including part time employed and studying, part time employed and caregiving, claiming disability

Table 6.2 Illustrative quotations

Theme	Illustrative Quotations
Protecting and enhancing scientific learning	
Grounding science in stories	You see the expert academics presenting, and then a patient would stand up and give their perspective, and it was validating some of the characteristics that the academic had, but also humanizing. Making it more about what we do, so when we see patients, how that translates. They humanized it. They put a face on the science." (Female, Nurse, Canada) I think they make it real, they make it authentic. We get brought back to the focus that this is about patients, sometimes we forget. Sometimes we get lost in our various esoteric academic discussions. I think it does refocus us, I think that's really important. I think that we should start almost every session, or every day with being realigned to the patient story to make sure that we're on message. (Male, Nephrologist, United Kingdom) It was a patient that went there and talked about how it was for him to start dialysis, and how it was for him to go back to his activities after being on dialysis. Then you start talking about outcomes in trials and things like that. It makes a lot of sense, it puts things into context. I think that example reshapes the way that people are listening to the information that follows that. (Male, Nephrologist, Brazil)
Sharing and inspiring new perspectives	That one [session on caregiver burden and bereavement] hit home because her mum was in our program. To hear her side for me, of living the experience on that side, and knowing what I saw on my end and what we intervened with and did, and where the bright spots were, the shortfalls were, hit home a lot harder for me than almost anything an academic person could have told me. (Female, Nurse, Canada) Hearing patients speak at ISPD, hearing patients speak at the events that we have done, I just find that it sheds a new light. Sometimes I find that we can get so focused on checking off the things that we need to cover that we might miss things. (Female, Dietitian, Canada) We have to understand totally what their [patients] situation is to improve it. That's the bottom line. I think meeting them in a different situation where you discuss these things, because really in the clinic there is rarely time to discuss these things properly. The patient will learn and listen to our perspective, which is of course different to their perspective. We will learn of their perspective also. Give and take. (Male, Nephrologist, Sweden)
Distilling the key messages of research presentations	We should be able to get messages across in a lay-language. Any research, any scientific content, you should be able to. There's certain science at these sort of conferences, some of the cellular type, lab-based science, but you still should be able to explain that. Why is that important, why are we doing this, and what's it relevant to the patient at the end of it. I think it's quite good. Even for a health professional. I think sometimes the hard sciences, sometimes there is a lost in translation, shall we say. What relevance is this, what impact is this going to have, because all research should have a patient benefit in the end. (Female, Nurse, United Kingdom) Even as a registered nurse, there's been a lot of content where I've been thinking wow, I didn't realise they were such high-level scientific information. However, this morning in the 'modality' session there was a speaker. I think he did an amazing job of making the topic fun,

	interesting, filtering enough scientific evidence to prove his point but keeping it at level that was easily interpreted, whether you're a patient or a PhD times five. (Female, Nurse, Canada) It definitely led to sometimes avoiding jargon and trying to simplify messages, which is probably not a bad thing. Really, I find the talks, they don't need to be in-depth or scientific. They just need to be able to convey the theme or the idea. Because it's really new ideas or themes that I'm looking for, that I can take back either to my clinical practice or research. I don't need all the super detailed, that someone might be looking for. If I want the detail, I'll go and approach them later. (Male, Nephrologist, Australia)
Striking a balance between accommodating patients and presenting the science	I find that some of the sessions are pretty simplified, and it's been easier to understand. But I guess as patients, you kind of learn the language. I think for some things, it would be good for people to have that support or even a simplified session. But that's the thing, we don't want to make it so boring for healthcare professionals. Trying to find a happy medium with both. (Female, Patient, Canada) I don't speak research talk, and I appreciate the information, but I don't always follow it, the same as patients would, so I felt like everyone met in the middle. It was good to see that. (Female, Nurse, Canada)
Democratizing access to research	
Redistributing power	By having us there, it's challenging doctors to take the risk of coming from behind the curtain and meet with me at a lunch table. I sat at a lunch table with a whole group of them... What was really neat was that when I did identify myself as a caregiver, instead of there being a sense of them withdrawing from me, the doctors at the table seemed delighted and asked me what I had been speaking on. When I told them, they were more engaged. One of the nurses said oh, you must find it odd that we've been joking around so loosely. One doctor said yeah, because when usually you see us professionally, you see just one side of us, you don't get to see this other side. I said, but people are people. (Female, Caregiver, Canada) What I wanted to share in that conference and in that panel was that it's not the medical procedure that affects us most. To us, it's the whole relationship, authority, and doctors and patients' relationship is the one I think that has to be improved. (Male, Patient, Canada) We need to recognize people for who they are, valued for who they are. None of us is of higher value than anybody else, but the issue is that power arrangement. (Male, Nephrologist, United Kingdom)
Challenging the traditional ownership of knowledge	We're not trying to hide anything from patients. This is the age of open disclosure and information. Patients have a right to know what their mortality is. I suppose it goes back more – it's not about telling them information, it's more about helping them interpret what that information means for them. (Female, Nephrologist, Australia) The point of research is not only for researchers. (Male, Nephrologist, Sweden)
Cultivating self-management through demystifying research	I was so impressed listening to the less is more one, because when we go travelling, we use the two bag system, but at night he uses a cycler. When we use the two bag system you're supposed to have four exchanges, but once in a while we'll take a tour that lasts all day so we miss one of them. Dr. X knows we're doing that, and he says it's okay. Your results are still really good. But when I listened [to the 'less is more' session] I thought, oh, okay, that's why he's okay with that. (Female, Caregiver, Canada)

	It’s so, so important for patients to learn these things, because the doctors often can’t, don’t, won’t. If you don’t take care of your own healthcare and learn, like I learned about incremental. If I hadn’t done that, I wouldn’t be able to travel the way I do. I’d be restricted with my treatments much more. I have freedom because of that, and I didn’t learn it from the doctors, I learned it from being at these conferences. It’s made a huge, huge difference in my health. (Female, Patient, United States) I think it’s a good thing to allow patients to get information from more places...They’re doing home therapy...I actually think they’re a group of patients that generally want to be involved in decision-making, and they want to be involved in preserving their ability to be at home. If you have that group of motivated type of patients, then you’ll do them a disservice if you’re not allowing them to get more information. (Female, Nephrologist, Australia)
Inadequate support for patient/caregiver delegates	
Lacking purposeful inclusion	To be honest with you, I didn’t really like it because it was like to tell your story. For me, that’s not enough. To tell the story… I don’t want to say political, but it should be some kind of empowerment from the part of the patients that I don’t see. Even in that conference. (Male, Patient, Canada) I wasn’t sure why I was on the panel because it was short, and I didn’t get to put in as much as I wanted to. (Male, Patient, Canada) I feel like it could be more structured. For example peritonitis. We have all these very knowledgeable and prestige speakers on stage. Can we include a patient with peritonitis talking about their experience, what are they struggling with, what would increase the rate of peritonitis from their perspective? I think it’s so nice to include that in their discussion, especially in the renal world there is not a lot of evidence, necessarily, studies, so we rely on that patient perspective a lot and what matters to patients. (Female, Pharmacist, Canada) That was very informal, I thought. Just go there and tell your story, that was my perception that was what the patient was instructed to do. If you plan a session and then you ask the patient to speak about specific things, I think it’ll be much better than it was here. It is a good start, you feel the potential, but we’re not there yet. Every time I saw a patient there I felt that they really didn’t know what was expected from there. That’s very different from what was requested from the speakers, right? (Male, Nephrologist, Brazil)
Challenges in interpreting research findings	I do sometimes worry that if some of the patients might be sitting in sessions and hear information and not understand whether it was significant, not significant… What I wouldn’t want is for a patient to hear information about all these awful complications like peritonitis and EPS and all this stuff and to come away thinking oh my god, everything terrible is going to happen to me because these doctors talk about it all the time, and maybe not understand the circumstances about that. (Female, Nephrologist, Australia) They may hear something that wasn’t said. They may go back to their physician and say ‘I was at the ISPD meeting and a doctor said such and such’, when he didn’t. (Male, Nephrologist, United States)
Soliciting medical advice	I don’t want to be approached by patients and being asked, what shall I do about my problem, a bit like this patient was asking a very personal question for her and asking an opinion directly of the nephrologists on the panel. But I think they handled that well in the sense that, it’s a fair question for her to ask, and to relay back that this is a fair question, but it’s question that needs to be answered by the person that knows you, which is your nephrologist… it’s really down to the individual nephrologists attending to be able to field that. (Female, Nephrologist, Australia)

	I do feel that it is inappropriate to ask medical questions, personal medical questions at a conference. I thought there might be quite a lot of that, but pleasingly and somewhat surprisingly there was less than I anticipated. (Male, Nephrologist, Australia) At the general assembly meeting the day after the dinner, some ladies walked in, turns out they were patients. They… turned the discussion to things that had nothing to do with the business of the society and dealt with their own personal concerns. (Male, Nephrologist, United States)
Difficulty negotiating venue and program	To our detriment, I have to admit, I did not look closer at the program. Because I truly didn't think it involved us. That was my feeling from even the registration. I guess we didn't immerse ourselves, and I don't think we got as much out of it as we could have. (Female, Caregiver, Canada) I think it's a great idea going green, but they should have had some stuff for people who don't have [smartphones]. (Male, Patient, Canada) I think that the professionals need to have their sessions. I think that's quite fine, and that's great. But there was another patient couple beside us and they were in the same situation. It was called basics, and they were basics to the medical profession but not to the patient. That was a little bit misleading in that title. (Female, Patient, Canada) I think just make it more patient-friendly. This conference is really good at using the app on the phone, but a lot of our patients don't even have a smartphone. I remember one of our panellists on Saturday, her and her husband went to a different room for the morning because they didn't know how to get here clearly, and then they needed more directions. It's just simple logistics, you can have a designated person to help them navigate these things before the conference and on the day, that would be perfect. (Female, Pharmacist, Canada)
Limited financial assistance in attending	I think that the financial thing is a huge part of it, and I think that's why a lot of patients probably don't attend these things when I think they should. If it was more resources that are covered, I think patients would love to come more often if they can, medically. (Female, Patient, Canada) There was a restriction on how much financial support patients could receive for making their own trip here, whether that was airfare, mileage, parking costs, or accommodations. That was a barrier to quite a few patients. They didn't have the finances to be able to get to the conference even though they were really wanting to. It wasn't financially possible, and as we mentioned, budget is tight to begin with. (Female, Nurse, Canada)
Amplifying impact beyond the room	
Sparking innovation in practice	I always think that hearing the patient voice goes a long, long way. People taking it home, from that clinical trial session that we set up, I think it would've been a much drier session without a patient there saying this matters. I've learned, it's not so much about the data, it's about the heart. People who go away from a meeting thinking yeah, that spoke to me, I'm going to try and do something about that because of the way that it came through to me. I think you're communicating on various levels, the cerebral level but also the why-we're-here level, what do we get up for in the morning kind of level. That sort of thing is important too. I think it does go beyond the conference. (Male, Nephrologist, United Kingdom)

	We’ve since started putting out some info in a newsletter, so it’s changed our practice, my practice anyway, because I try to bring up intimacy more than I ever would have before. (Female, Nurse, Canada)
Giving patients and families hope for the future	Yeah, so the thing is, the points that he brought up when he had his five minutes – in his defence – were very good, and every doctor on the panel that stood, except for one, said ‘what [patient] said, what [patient] said’. That was very uplifting for myself to see, that they listened to what he said even in his five minutes. That was awesome. That part of it, I think was great. (Female, Caregiver, Canada) It is not all known, it is not fully understood, they do not have the answers, but here we’re looking. (Male, Patient, Canada) They [patients] did find reassurance in the fact that progress was being made in a number of areas. (Male, Nephrologist, Australia

Table 6.3 Suggestions and considerations for involving patients in future scientific conferences

Domain	Considerations and suggestions from participants for ISPD 2020 in Glasgow
Before the conference	• Include patients and caregivers in the organizing and scientific committees, enlist their help in selecting topics and designing the conference agenda/program • Develop a recruitment strategy and advertise early, ideally at the same time as advertising to all delegates • Plan and budget for patient attendees, including: o Waiving of registration fees o Bursaries to cover travel, accommodation and meal expenses; developing partnerships with local business to arrange discounted/donated goods/services • Reduce the organizational burden on patients by o Arranging for group transportation to assist patients/caregivers in attending o Booking all transport/accommodation on behalf of the patient and their caregiver o Partnering with Industry to accommodate their needs e.g. Baxter to provide supplies for bag exchanges • Ensure patient speakers are given adequate support ahead of time, including: o Specific guidance for patient speakers to prepare their talks in alignment with other speakers, session learning objectives and within the allocated timeframe o Opportunities for practice and feedback o Consideration for the time allocated to patients compared to other speakers in the session for meaningful contributions o Potential to utilize/develop flexible training modules to assist patient speakers in developing a purposeful talk e.g. Can-SOLVE “Storytelling for Impact” • Develop a conference induction packet for patient/caregiver delegates, including: o All logistical information o A detailed, lay language version of the conference program including suggested sessions that may be interesting for patients (if through mobile application – include a patient resource section) o A glossary with common medical terms and acronyms likely to be used in the conference o Advice on how to ask a question o Tips to get the most out of the conference • Consider incorporating patient workshops prior to the conference to discuss conference topics with a bigger group of patients, with opportunities for patient representatives to feedback the perspectives of the group in the main conference • Encourage teamed presentations with health professionals and patients presenting together • Consider including the perspectives of patients unable to attend via pre-recorded video presentations to capture more diverse perspectives

During the conference	• Ensure patients are welcomed and receive appropriate guidance and information from the start of the conference, including: ○ A patient/caregiver check-in desk ○ A personal contact number for any problems/queries during the conference e.g. buddy system, patient and caregiver coordinator ○ The same courtesies extended to other delegates (e.g. speaker ribbon on registration badge for patient speakers) • Host a patient networking event early in the conference to enable patients to meet one another • Utilize concurrent sessions to offer some summary/simplified sessions targeted at patients/caregivers • Accommodate patient needs including: ○ Providing a quiet place to rest or administer therapies (e.g. patient lounge for PD bag exchanges) ○ Access to assistance for impairments including hearing, visual and physical (e.g. wheelchair, preferential seating) ○ Dietary requirements (e.g. low sodium diet) ○ Paper/hard copy information (e.g. program – patients may not have or may not be able to use a smart phone) • Allocate time explicitly for patient questions • Designate “meet-up” times for informal interactions between patients/caregiver and health professional delegates • Provide an opportunity for patients/caregivers to debrief/address any questions/concerns raised from what they have learned at the conference
After the conference	• Provide slides, summary notes or recorded presentations online for future reference • Consider a specific patient/caregiver evaluation and pilot with patient/caregivers • Provide an opportunity for patient/caregiver delegates to stay informed/included in future correspondence/events

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Chapter 7: Identifying and integrating consumer perspectives in clinical practice guideline development for renal biopsy: Consumer workshop report

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This chapter is structured as per the journal article.

7.1 Abstract

Aims: Percutaneous renal biopsy is often essential for providing reliable diagnostic and prognostic information for people with suspected kidney disease, however the procedure can lead to complications and concerns among patients. This study aims to identify and integrate patient priorities and perspectives into the Kidney Health Australia – Caring for Australasians with Renal Impairment clinical practice guidelines for renal biopsy, to ensure patient-relevance.

Methods: We convened a workshop, consisting of three simultaneous focus groups and a plenary session, with ten patients who had undergone a renal biopsy and seven caregivers. Topics and outcomes prioritised by patients and their caregivers were compared to those identified by the guideline working group, which was comprised of seven nephrologists. Transcripts and flipcharts were analysed thematically to identify the reasons for participants' choices.

Results: In total, 34 topics/outcomes were identified, 14 of which were common to the list of 28 previously identified by the guideline working group. Most of the new topics identified by patients/caregivers were related to communication and education, psychosocial support, and self-management. We identified five themes underpinning the reasons for topic and outcome selection: alleviating anxiety and unnecessary distress, minimising discomfort and disruption, supporting family and caregivers, enabling self-management, and protecting their kidney. A new topic on patient care and education was added to the guideline as a result.

Conclusions: Patient and caregiver involvement in developing guidelines on renal biopsy ensured that their concerns and needs for education, psychosocial support, and self-management were explicitly addressed; enabling a patient-centred approach to renal biopsies.

7.2 Introduction

"It's this one size fits all, widget in a factory line, rather than a patient in a context."

Caregiver, female, group 1

Percutaneous renal biopsy is widely used as a diagnostic tool to inform the management of patients with kidney disease [1,2]. The procedure is generally efficacious and safe, with more than 99% of biopsies on average providing adequate tissue for diagnosis, and life-threatening complications such as major bleeding occurring in less than 0.1% [3,4]. However, the procedure is invasive, requires day-stay care, analgesia, may involve sedation, may interfere with current therapy (e.g. antihypertensives), and can provoke fear, anxiety, and stress in patients and their families[5,6]. There are still uncertainties regarding the procedure including the cessation of medications such as antiplatelet and anticoagulant agents, optimal imaging technique, needle type and size, site of biopsy, observation period post-biopsy, definition of sample adequacy and the benefits of surveillance biopsies[1,7,8]. Scant evidence exists on the patient experience, priorities, and psychosocial impacts[5,6,9].

In 2017, work began by Kidney Health Australia – Caring for Australasians with Renal Impairment (KHA-CARI) to develop a guideline for renal biopsy following standard processes for evidence-based guideline formulation[10,11]. As is routine for KHA-CARI guidelines, patient and caregiver involvement is structured though a parallel process to ensure that the guideline scope and recommendations explicitly address patients' priorities[12-15]. This study aims to identify topics and outcomes relating to renal biopsy that are important to patients with kidney disease and caregivers to be included in the KHA-CARI guideline; to describe the reasons for their choices; to compare topics and outcomes identified by patients/caregivers with those selected by the expert working group; and to discuss the process of integrating patient and caregiver feedback into the guideline.

7.3 Methods

We used the Consolidated Criteria for Reporting Qualitative Studies (COREQ) to report this study[16].

Context and overview

KHA-CARI facilitates the development and implementation of clinical practice guidelines for kidney disease, dialysis and transplantation[10]. In March 2017, a working group of seven nephrologists was convened to discuss the development of a clinical practice guideline for percutaneous renal biopsy for all indications in patients with native and transplanted kidneys. In the initial meeting, the working group identified preliminary topics and prioritized eight topics (determined by feasibility) for inclusion in the guideline using the Population, Intervention, Comparator, Outcomes, Methodology (PICOM) framework[10]. To ensure the relevance of the guideline for patients/caregivers, a half-day workshop was convened in October 2017 to elicit patient and caregiver perspectives about topics and outcomes for inclusion.

Participants and recruitment

Participants were eligible if they were aged 18 years or over, spoke English, and had undergone at least one renal biopsy, or were a caregiver of a patient who had undergone a renal biopsy. Participants were recruited from a major Australian university and teaching hospital. We applied a purposive sampling strategy to obtain a wide range of demographic and clinical characteristics, which can help to ensure that a broad range of knowledge, perspectives and experiences are included. Recruitment ceased when a maximum of 25 participants were confirmed as determined by the resources available for this study. Participants were reimbursed $50 to cover travel expenses. Ethics approval was granted by the site's ethics committee.

Data Collection

The workshop program, structure, and question guide are shown in Appendix F.1. Three simultaneous focus groups were conducted by trained facilitators (TG, KEM, PLV), while one facilitator took notes. None of the facilitators were involved in the clinical care of the patients. The question guide was developed based on previous KHA-CARI workshops [12,14,15] and discussion with the research team. Participants were asked to discuss their experiences with percutaneous renal biopsy, and to suggest topics and outcomes they considered important to include in the new guideline. A plenary session was convened and moderated by KEM whereby a spokesperson from each group presented a summary of their discussion. Participants had the opportunity to provide comments, feedback and ask questions, and were asked to clarify and explain their responses. All discussions were audio-recorded and transcribed verbatim.

Data Analysis

Transcripts and flipcharts were entered into HyperRESEARCH to assist with qualitative data management and analysis. PLV extracted all topics/outcomes and categorised these into an existing or new guideline subtopic. TG inductively coded the transcripts line-by-line, to extract data relating to the reasons behind the participants' topic and outcome selection. Similar concepts were grouped into overarching themes. Preliminary themes were discussed and revised with AT, KEM and PLV, who had independently read the transcripts, to ensure the themes captured the range and breadth of the data.

7.4 Results

Participant characteristics

Of the 25 participants that agreed to take part, 17 (68%) attended the workshop, including ten (59%) patients and seven (41%) caregivers. Reasons for non-attendance were due to illness or unknown. Participant characteristics are shown in Table 7.1. Participant age ranged from 37-77 (mean 54.1) years, nine (53%) were male, and nine of the ten patients had received a kidney transplant.

Guideline topics and outcomes

Table 7.2 shows the topics and outcomes identified by the workshop participants and the guideline working group, and highlights the discordance between patients/caregivers and health professionals. In total, 48 topics/outcomes were identified; 34 (70%) by patients/caregivers and 28 (58%) by the guideline working group. Only 14 (29%)

topics/outcomes were identified by both groups. Most of the topics identified by the patient/caregiver group related to communication and education, psychosocial support and self-management. Of note, some patient identified topics including patient education and clinician expertise were initially discussed by the working group, but were not prioritized for inclusion as available resources determined a finite number of topics for evidence review.

Themes

Five themes underpinning topic and outcome selection were identified: alleviating anxiety and unnecessary distress, minimising discomfort and disruption, supporting family and caregivers, enabling self-management, and protecting their kidney. Figure 7.1 shows the relationships among themes and topics/outcomes identified. Illustrative quotations for each theme are provided in Table 7.3.

Alleviating anxiety and avoiding unnecessary distress

> *"Just come and talk to us, ask us what we're feeling or how anxious we are."*
>
> Patient, male, group 3

Long waiting times and perceived lack of communication resulted in patients and their families suffering anxiety due to "not knowing" the reasons for delayed appointments, procedures or delivery of results. Participants believed they were often left waiting before the procedure without explanation, were misinformed about their expected discharge time, or were not attended to in a timely manner after the procedure.

Some patients were distressed because they were not introduced to the doctor performing the biopsy "before being wheeled in" for the procedure. Patients were confused and anxious that a support person was not allowed to be present during the procedure, particularly as they knew it was not performed in a sterile operating theatre. Afterwards, some patients were taken to the radiology department to recover which they found stressful and chaotic. One patient remarked that it was “the scariest thing ever” because he “thought [another patient] was going to die right in front of [him]”. Other patients felt “well informed” and “appreciated” when hospital staff took the time to explain things to them, and when they were allowed to have a support person with them during the procedure.

Patients and caregivers wanted clinicians to tailor the way they delivered information about the biopsy to their individual needs as patients, including the way they conducted the consent process, particularly for those who were “in the storm of transplant recovery”. For some, anxiety clouded their ability to fully comprehend the information they were given and they felt overwhelmed by too much information and “medical jargon”. Some felt they were given too much detail in the consent process about the potential negative consequences, and not enough information about the benefits of biopsy. Others felt they were not given enough information, so they felt vulnerable and sometimes pressured into consenting to a procedure they did not completely understand. Participants wanted clinicians to anticipate these information needs and preferences.

Delayed delivery of results increased anxiety levels as patients feared the worst. They were frustrated with the protocol of scheduling their clinic appointment two weeks after the procedure when they believed the results would be available before then.

Minimising discomfort and disruption

"They said 'don't move', and I'm shaking. They said 'you're nervous.' I said 'no, it's just that cold in here.'"

Patient, male, group 2

Patients expressed feeling dehumanised, "like guinea pigs," and felt the staff and hospital policies didn't consider their time, needs or comfort. Being told to arrive many hours before they were scheduled to be seen by a nurse or doctor was particularly disruptive for patients as it meant unnecessary time away from work and family. Some patients remarked that the procedure room was so cold they had difficulty keeping still for the duration of the biopsy, which they believed put them at greater risk of bleeding.

Many patients described the boredom and discomfort they experienced in recovery while waiting to be discharged, which was exacerbated by the sometimes conflicting information they received post-procedure regarding the duration and position of recovery. Some patients discussed the need to simplify discharge processes, as they were sometimes forced to extend their hospital stay because doctors were not always available to see them immediately. One patient explained how she needed four doctors from different units to approve her discharge before she could leave. Another patient stated that he was forced to stay in hospital overnight because there was not a doctor present that could discharge him, causing him to have to urgently organise care for his children.

Supporting family and caregivers

"It's that context of it's a whole person and a whole family"

Caregiver, female, group 1

Participants were concerned that there was "no caregiver support". Some caregivers were alarmed that they were not contacted when the patient was unwell post-procedure or were not informed when they were ready to be discharged. They also noted that they did not have an appropriate area they could wait in until their family member was taken to recovery.

Patients also believed there was no flexibility with scheduling of biopsies to accommodate family commitments and wanted the hospital to be "more family friendly". Participants explained they sometimes had to "drop everything" and arrange care for their children to be able to attend their biopsy appointment.

Enabling self-management

"We had examples of two different sets of information for post-care we'd been given"

Patient, male, group 2

The perceived lack of clear information and effective communication left patients feeling disempowered. Patients wanted the risks and benefits of the procedure communicated to them clearly so they could make an informed choice about whether to undergo the biopsy. However, some believed "you can't say no" and that doctors "make you feel guilty" when they were reluctant to consent.

Participants wanted clear, comprehensive and consistent information communicated at times when they were not unduly stressed. They wanted written materials explaining how to manage their diet and medications before and after the procedure, as well as a breakdown of the timing from admission to discharge. Some patients were confused about what they were permitted to do post-procedure, specifically about whether they were allowed to drive home. Patients stated they should have been notified before the day of the procedure of the potential need to arrange for alternative transport home or accommodation close to the hospital. There was also concern that non-English speaking patients were not adequately supported, resulting in their inability to self-manage. One caregiver witnessed a patient get up immediately after his procedure because he did not understand the nurse’s instructions or the recovery procedure, which was reported to have resulted in him haemorrhaging.

Many felt that clinicians should explain that biopsies were often conducted in the context of post-transplant protocol and wanted an itemised “visit plan” so they could be prepared for procedures and minimise schedule conflicts. Some patients were frustrated as they were not permitted to have pre- or post-biopsy blood tests at a local pathologist to allow the results to be ready in time for their clinic appointments.

Protecting their kidney

“How much damage does biopsy do to my new kidney that I’ve waited so long for?”

Patient, male, group 2

Patients were concerned that biopsies could damage their kidney, and wanted to avoid unnecessary procedures that might “jeopardise” their already “delicate” kidney. Some questioned the hospital protocol biopsies post-transplant, particularly when their treating nephrologist advised them against it "when everything is going perfectly." Some participants felt doctors were a bit too "willy nilly" in recommending the procedure, and did not understand the necessity of a biopsy if blood and urine tests did not indicate any signs of rejection. Some patients were sceptical when told by their doctors that multiple biopsies wouldn't damage their kidney – "I don't know whether to believe him or not…you can't keep hacking at a tree and think it's going to keep growing." Some patients were reluctant to have an inexperienced doctor or a registrar perform their biopsy, particularly if they had distressing previous experiences, or felt this increased the risk of complications. Some patients avoided pain medication post-procedure because they feared it may damage their kidney.

Impact on guideline development

A summary of the workshop, including a comparison matrix of topics identified by both groups, was sent to the guideline working group for review and discussion. A new topic on patient care and education was added to the guideline to address patient and caregiver concerns, particularly opportunities to improve communication and reduce anxiety (Figure 7.2). Prior to publication, patients and caregivers will be given the opportunity to review the guideline to ensure the patient voice has been represented and integrated. Also, a patient-version of the guideline will be developed to ensure the guideline is accessible to patients and their caregivers/families. This will be written in plain language and reviewed by patients/caregivers before publication.

7.5 Discussion

For patients and caregivers, the topics and outcomes identified for guidelines on renal biopsy focused on patient-provider communication and education, psychosocial impact of the procedure, and self-management before, during, and after renal biopsy. Some of the priorities were raised in the context of feeling anxious, distressed and “lost in the system”, and to alleviate discomfort and minimise disruption to their usual activities, including for family members and caregivers. Patients wanted to be empowered for self-management and shared decision-making, particularly in terms of whether to undergo a biopsy, managing diet and medications prior to and after renal biopsy, being better prepared for making transport or other arrangements, and having immediate access to their results. Patients were also highly concerned about protecting their kidney and thus needed to better understand the need for, and the process of renal biopsy.

The waiting time on the day of the renal biopsy was viewed by patients as extensive and unnecessary, and interfered with their work, family, and other personal commitments. This was reflected in topics relating to the discharge process, and high prioritisation of outcomes that caused disruption and discomfort such as waiting time, recovery position and location, and pain management. Patients and caregivers emphasised the need to support patients with families due to the burden of extra expenses incurred, such as child-care, or distress caused by their absence. Therefore, impact on family and length of stay were considered important outcomes for inclusion, as were the provision of adequate facilities and access to psychosocial support for caregivers.

Participants wanted more information to help them self-manage medications, recover after the procedure, and arrange logistics such as getting home from the hospital. As a result, they prioritised topics such as understanding the consent process and the transplant biopsy protocol, and outcomes such as withholding anti-platelet agents, knowledge and awareness, and return to usual activities. While some patients understood that undergoing the biopsy was for their benefit, many were concerned about the damage the procedure would cause to their transplanted kidney. Number of passes, rate of failure, procedure time, complication rate, loss of kidney/allograft, ICU admission, bleeding, renal survival, and total number of biopsies were all outcomes identified by patients/caregivers as important, which reflected their concerns of protecting their kidney.

There were notable differences in the scope and focus of the topics/outcomes between patients/caregivers and the working group of nephrologists. The work group members prioritized topics and outcomes focused on clinical procedures and outcomes related to minimising the risk of complications, decreasing recovery time, and maximising sample yield. The topics covered anti-coagulation pre-procedure, needle and imaging types, patient position for procedure, bleeding complications and post-operative care; and the outcomes were all procedural or clinical, including hypotension, haemoglobin, number of cores/samples, procedure time, haematuria, haematoma, embolization, nephrectomy, need for blood products and death. In comparison, patients and caregivers were focused on self-management, psychosocial impact, education, patient-provider communication, and impact on family.

Studies in patients undergoing biopsy in other clinical settings have shown similar findings. Women undergoing breast biopsy wanted health professionals to involve their family members, take an individualised approach, provide written educational materials, and decrease the waiting time for results[17]. Patients requiring prostate biopsy felt they lacked information and were ill-prepared, and the disparity between expectations of the prostate biopsy and what occurred in reality heightened their anxiety[18]. Our findings build on the limited evidence from previous small-scale studies that identified information needs of patients before, during and after renal biopsy, by addressing the reasons and preferences for these informational needs, and potential opportunities for improved communication[9].

We elicited a range of patient and caregiver priorities for topics/outcomes to include in guidelines on renal biopsy. However, our study has some potential limitations. This study reports the perspectives and beliefs of patient and caregivers, and may or may not reflect what happens in standard clinical practice. Due to limited resources, patients were recruited from a single centre and were all English-speaking, thus the transferability of findings is uncertain, however some participants did describe their experiences at other sites. Whilst we sought to include patients with both native and transplanted kidneys, most patients who attended had received a kidney transplant. This may be because biopsies are regularly performed as part of routine transplant surveillance, and rarely performed in dialysis patients acutely presenting with renal failure, or in patients with early stage chronic kidney disease. While we did not collect data on the number of biopsies each patient had undergone, and we do not know how many patients had undergone native kidney biopsies prior to having their kidney transplant, some patients did discuss their experiences of renal biopsy prior to kidney transplant.

Our findings reveal opportunities to improve care for patients undergoing renal biopsy. We suggest that a communication strategy between health professionals and patients/caregivers be developed that aligns with the patient journey of renal biopsy, a re-evaluation of protocols for procedures that cause unnecessary anxiety and discomfort, and provision of rapid access to biopsy results. Providing a schedule of upcoming visits, delivering concrete, practical, and consistent pre and post-operative information, clearly explaining the consent process, and keeping patients well-informed of hospital procedures and delays from admission to discharge could mitigate the fear, anxiety, frustration and inconvenience experienced by patients[19-22]. Figure 7.2 outlines the points of contact where patient anxiety can be addressed. Ensuring patients and caregivers feel calm, reassured, and comfortable may be achieved by meeting the clinician prior to the procedure, allowing a support person to be present, making sure the patient is warm enough, having a waiting area for caregivers, allowing patients to recover in a quiet place, and providing supervision for less experienced operators. Existing procedures should be reviewed and if necessary modified to address these key aspects. Aligning clinic appointment schedules with the availability of results to ensure that patients receive their results as soon as possible may help to relieve anxiety and better enable self-management. These issues may be relevant to broader patient cohorts undergoing day procedures both within the context of renal care (e.g. dialysis line insertion, peritoneal dialysis catheter insertion, arteriovenous fistula construction) and across other disciplines. The findings of the workshop may be used to design a survey to be conducted among a larger sample of patients, to identify and assess the frequency of opinion on renal biopsy across a wider and more diverse population.

Patient and caregiver involvement in this study has expanded the scope of the renal biopsy guideline and its recommendations to explicitly address patient/caregiver concerns and needs for education, psychosocial support, and self-management; ensuring a patient-centred approach to renal biopsy. Implementation of the guideline is likely to improve quality of care, patient satisfaction, and outcomes in patients undergoing renal biopsy.

Acknowledgment

We thank all the patients and caregivers who participated in this study.

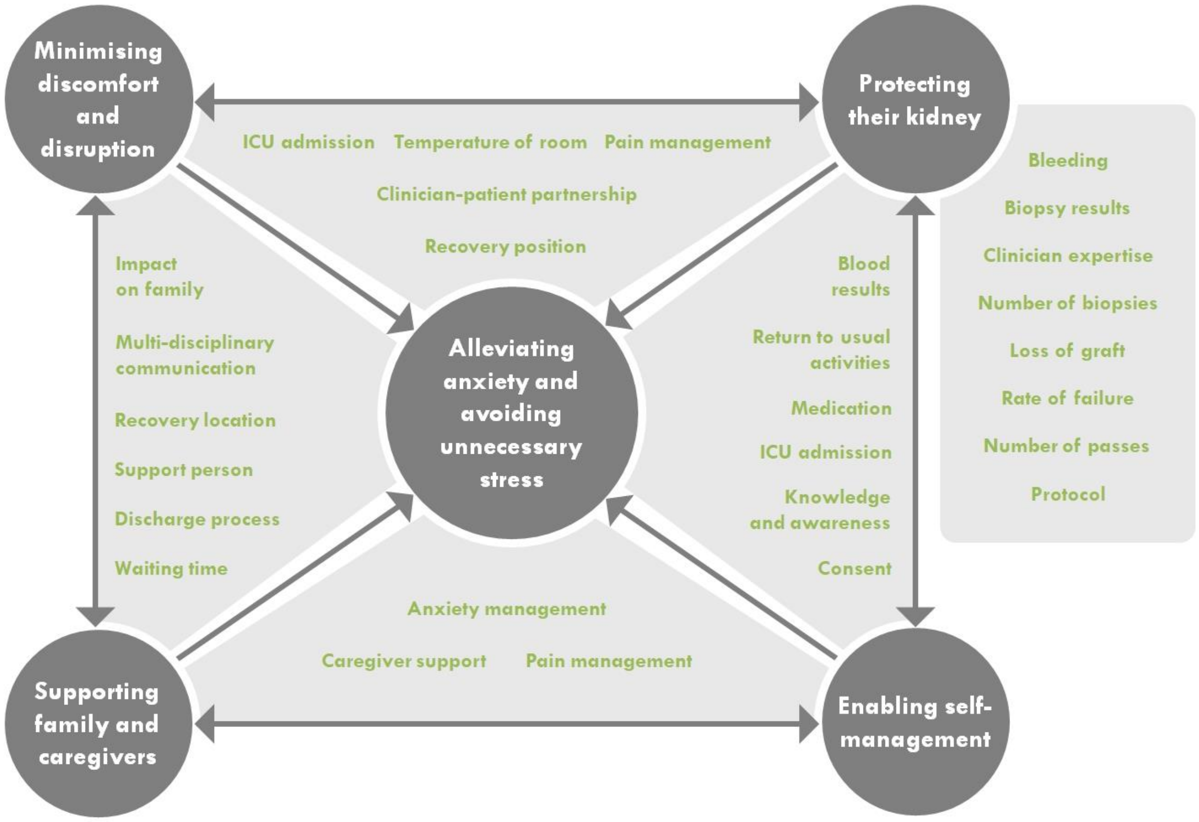
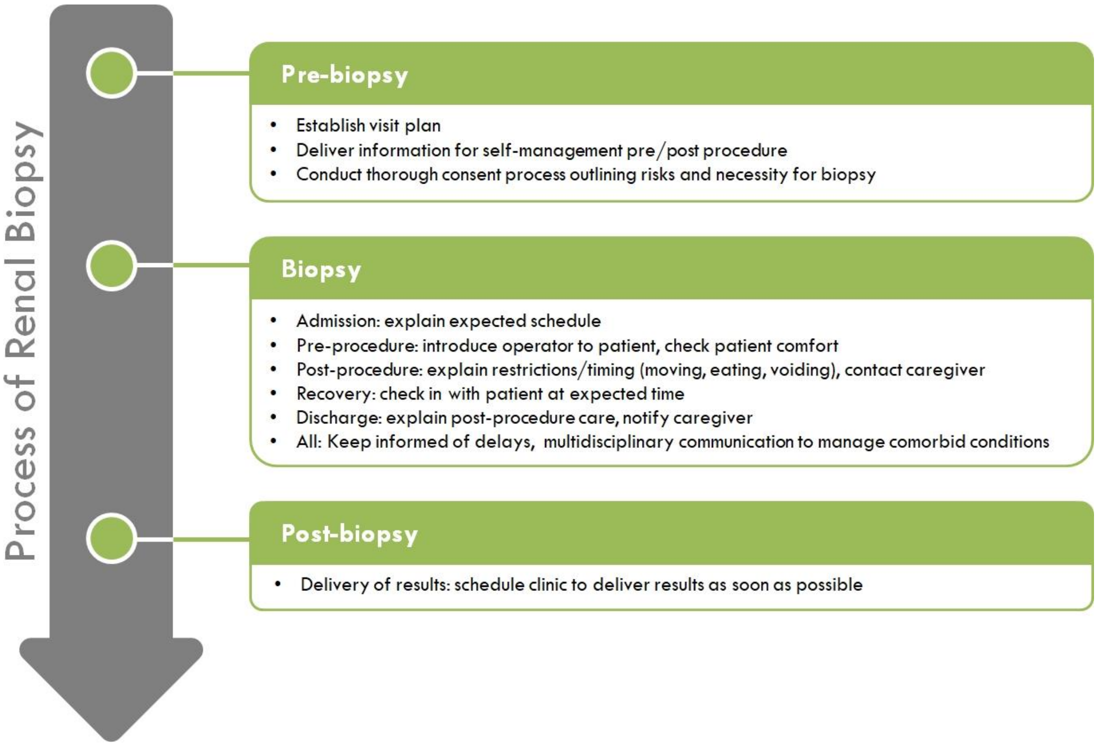


Figure 7.1 Thematic schema and related outcomes

The topics/outcomes identified by patients/caregivers are shown in green. The grey squares show the themes that reflect the reasons for the prioritisation of the topics/outcomes. All of the topics identified either directly or indirectly caused anxiety in patients. Some of the topics/outcomes were specifically related to protecting their kidney.



Pre-biopsy

- Establish visit plan
- Deliver information for self-management pre/post procedure
- Conduct thorough consent process outlining risks and necessity for biopsy

Biopsy

- Admission: explain expected schedule
- Pre-procedure: introduce operator to patient, check patient comfort
- Post-procedure: explain restrictions/timing (moving, eating, voiding), contact caregiver
- Recovery: check in with patient at expected time
- Discharge: explain post-procedure care, notify caregiver
- All: Keep informed of delays, multidisciplinary communication to manage comorbid conditions

Post-biopsy

- Delivery of results: schedule clinic to deliver results as soon as possible

Figure 7.2 Communication throughout the patient journey

Table 7.1 Patient and Caregiver characteristics (n=17)

Characteristic	n	%
Participant status		
Patient	10	59
Caregiver	7	41
Sex		
Male	9	53
Female	8	47
Age		
30-39	2	12
40-49	5	29
50-59	4	24
60-69	4	24
70-79	2	12
Education†		
Primary school	1	6
School certificate	4	24
TAFE	7	41
University	4	24
Employment†		
Full time	5	29
Part time or casual	4	24
Unemployed	5	29
Retired	2	12
Marital status†		
Married	13	76
Single	3	18
Reason for biopsy (n=10 patients)		
Kidney transplant	9	90
Kidney function (native kidney)	1	10

†Missing n=1

Table 7.2 Guideline topics and outcomes for renal biopsy identified by patients/caregivers and the working group

Guideline Subtopic	Intervention/Outcome	Identified by patients/caregivers	Identified by working group
Pre-biopsy			
Medication – coagulants	DDAVP – change in haemoglobin, hypotension, bleeding		✓
	Withholding anti-platelets - change in haemoglobin, hypotension, bleeding	✓	✓
Patient education and care	Patient and caregiver information/knowledge and awareness†	✓	
	Transplantation protocol	✓	
	Understanding consent†	✓	
	Delivery of blood results	✓	
	Impact on family	✓	
	Clinician/patient partnership	✓	
	Multidisciplinary support and communication	✓	
	Anxiety management	✓	
	Caregiver support	✓	
	Waiting time	✓	
Biopsy			
Methods	Needle type/size/guidance		✓
	Imaging type (CT, Ultrasound, blind, real-time, colour/flow Doppler)		✓
	Patient position		✓
	Clinician expertise†	✓	
	Number of passes	✓	✓
	Number of cores		✓
	Adequacy of sample		✓
	Rate of failure (non-diagnostic, abandoned)	✓	✓
	Procedure time	✓	✓
	Patient comfort during procedure (temperature, support person)	✓	
Complications	Complication rate	✓	✓
	Death		✓
	Embolization		✓
	Need for blood products		✓
	Loss of kidney/allograft	✓	✓
	ICU admission	✓	✓
	Wound infection		✓
	Immediate bleeding	✓	✓

Post-Biopsy			
Post-operative care	Bed rest/recovery position	✓	✓
	Recovery location	✓	
	Length of stay/discharge process	✓	✓
	Frequency of observations		✓
	Radiological embolization		✓
	Medical monitoring	✓	
	Pain management	✓	✓
	Anxiety management	✓	
	Multidisciplinary communication	✓	
	Return to normal activities	✓	
	Knowledge and awareness for post-biopsy self-management	✓	
	Delivery of biopsy results	✓	
Bleeding	Rate of bleeding		✓
	Secondary haemorrhage	✓	✓
	Late bleeding	✓	✓
	Renal survival	✓	✓
	Need for blood products		✓
Other			
Protocol	Total number of biopsies	✓	
Total:	48	34	28

†Topics initially identified by working group, but excluded due to feasibility

Table 7.3 Illustrative quotations for each theme

Theme	Illustrative quotations
Alleviating anxiety and unnecessary distress	Just come and talk to us. Ask us what we're feeling or how anxious we are or nervous. *(Patient, male, group 3)* As a patient, not knowing what's going on with you is the worst thing, and even if you do know something's bad, then it's like okay, how do we fix it, what's the next steps to getting better or sorting this out? *(Patient, male, group 3)* Some people found themselves being told initially that they were there for four hours, and then eight hours later they're still there and no one's come to communicate as to what the issue is, and then the anxiety levels go up even further because you know, is there something wrong? *(Caregiver, female, group 3)* The main thing is just getting information as soon as it's available. Don't just leave me in the dark. *(Patient, male, group 3)* You're already very overwhelmed both as patient and caregiver, when you're in the storm of transplant recovery, we're just not taking in the information in the same way, and we want an opportunity in the guidelines to recognise that. *(Caregiver, female, group 1)* Not putting a student doctor on someone who's got high anxiety. *(Caregiver, male, group 1)* They say they can't come in because of the sterile environment, but it's not a sterile environment at all. It's just a room in the radiology department…so I don't understand why. *(Patient, female, group 1)* I found that very reassuring that he allowed me to be with her, because I would have been more stressed if I couldn't see what was going on. I really appreciated that. I felt really well informed as to how it was all going to go, and he explained it all as he was doing it all. *(Caregiver, female, group 3)* To meet the doctor before the crunch time, you know, maybe half an hour before, just on the day. I understand they're busy, but it would be nice to meet him or her before, well, just before the needle's supposed to go in. *(Patient, male, group 3)*
Minimising discomfort and disruption	The long wait before the biopsy really annoyed me. *(Patient, male, group 1)* When you're booked in for eleven, and you're waiting, waiting, waiting. That's the worst thing. *(Patient, female, group 1)* Especially when you know you're going to have a three or four hour wait afterward. *(Caregiver, male, group 1)* She was in emergency, and I felt sick because I thought she was going to die in front of me, that's what it felt like. She was breathing one minute then stopped breathing. That was the scariest thing ever.' And I stayed there for five hours, listening to all this stuff all day. *(Patient, male, group 2)* You're seeing all these really sick people coming in and out, and you've got all this stuff happening next to you, I just think it's not the place for me to be at that time, I need to be away from all that chaos. *(Patient, male, group 3)* Probably the biggest thing as a patient is the waiting and communication. *(Patient, male, group 3)* We were always told to be somewhere at least an hour before we really need to be there. The biopsy procedure, you've got to be there at seven in the morning, and nothing would happen until lunch time. It's this one size fits all widget in a factory line, rather than a patient in a context. It's that context of it's a whole person and a whole family, not we, the whole hospital, want to have everyone in their beds by

	7:30AM waiting for the procedures and in their chairs waiting. Don't care how long you're sitting there. *(Caregiver, female, group 1)* The place was that cold, it was freezing. They said don't move, and I'm shaking. They said you know what, you're nervous. I said no, it's just that cold in here. *(Patient, male, group 2)* Minimising the discharge process and simplifying authority for discharge. *(Caregiver, female, group 3)* When I come in I'm under four different doctors, and all four got to let me go home. I've got diabetes, I've got renal, vascular, and dietary. *(Patient, female, group 3)*
Supporting family and caregivers	I wasn't kept informed at all by the hospital staff. *(Caregiver, female, group 1)* I thought that if I supplied [patient's] name and number, they would call if something went wrong. I came in to pick him up, he'd been sick, lying here for four hours. Really bad. *(Caregiver, female, group 1)* I've got a young family. I have to decide where they're going, if they're going to their nan's or my father in law's so he can get them to school and to their sporting events. If I'm stuck here and you're not telling me nothing, there's people ringing you, how are you, what's going on, can I pick up the kids at school, well I don't know what's going on so I can't tell them. *(Patient, male, group 3)* The whole environment, there's nowhere for caregivers to actually be able to chill down. There is something to be factored that you're actually dealing with people that are already in some ways, in trauma. *(Caregiver, female, group 1)* I've been told when I go for a biopsy, it's urgent. Get in here quickly, just drop everything and go. There's kids there that don't know what to do. *(Patient, male, group 3)*
Enabling self-management	People get told different things, you don't get managed properly. *(Patient, female, group 1)* Give patients a choice, I think it should be giving patients a choice. The doctor should say, look, these are the risks, do you want to know more? *(Patient, female, group 1)* No flow chart, no diagram of what's going to happen or anything. Any documentation with people. Especially if they don't feel comfortable about what's happening. Any documentation's going to help them feel more comfortable. *(Patient, male, group 1)* I think there's one at six months or twelve months. I'm not sure. See, that's the other thing. They don't give you your visit plan. You don't know what's happening in the future. *(Patient, male, group 1)* And it's almost like you can't say no, like they pretty much insist they have to do it. *(Patient, female, group 1)* Post-op information. We're aware that we've got to sit still, we're aware that that's going to be over a period of time, but it's the uncertainty of whether you're going to stay, you're allowed to go, you're allowed to – not drive, I know that, but can you be a passenger? I think it's just purely the clarification is am I in for the day, or am I in for the night? *(Patient, male, group 2)* We had examples of two different sets of information for post-care we'd been given, one in the radiology department and one from the kidney coordinator in terms of what you can and can't do immediately following, so there's some inconsistencies around how the treatment occurs. *(Caregiver, female, group 1)*

	That’s definitely not on any of the sheets. I actually rang up and said well, do I take all my medications? Then they said are you on aspirin or Clexane? They said don’t do the aspirin. By that stage I was on Clexane as well. I had to ask that. *(Patient, male, group 2)*
Protecting their kidney	I understand some registrars need to learn, but learn on someone else, please. *(Patient, female, group 1)* I said to him, how much pain and cutting can the kidney take? And he goes the kidney is pretty strong, it can take as much as we can give it. I don’t know whether to believe him or not… because you can’t just keep hacking at a tree and think it’s going to keep growing. Same with your kidney. You can’t just keep hacking at it. *(Patient, male, group 3)* They don’t tell you, they just tell you there’s scarring and there’s damage there. What damage? What has damage there? *(Patient, male, group 3)* Is the kidney going to be damaged by having this biopsy done? That’s for the people who have a kidney from natural, their own body. The concern for the transplanted person is – we wait so long for this great gift, is it going to be something that’s going to be damaged just for the sake of having a look to see if everything’s okay. *(Patient, male, group 2)* Quantifying or making clear the amount of damage when the results do come through, to make it clear as to whether it’s just a little bit of damage or like, be specific. *(Caregiver, female, group 3)* How much damage does biopsy do to my new kidney that I’ve waited so long for? (Patient, male, group 2) We all know how delicate this is – you’ve got a new kidney that you want to work, and someone’s in there who could jeopardise it, there’s no going back. *(Patient, male, group 2)* You’re only allowed to take a certain amount of painkillers because you don’t want to damage the kidney. *(Patient, male, group 2)* Yeah, so I’m coming up to my twelve month one, and everything is going perfectly, and now they’re talking about doing a twelve month one, and my nephrologist doesn’t think that it’s necessary to have one when everything is going perfectly, because every biopsy you do does a bit of damage to the kidney. So why is it, when everything is running perfect, that you have to have a biopsy at twelve months just because it is protocol? I understand if there’s something wrong, to have one, but when everything’s right, why damage my kidney for, when it’s going great? *(Patient, male, group 3)*

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Chapter 8: Child and parental perspectives on communication and decision-making in pediatric CKD: a focus group study

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This chapter is structured as per the journal article.

8.1 Abstract

Background and Objectives: Effective communication and shared decision-making improves quality of care and patient outcomes, but can be particularly challenging in pediatric chronic disease as children depend on their parents and clinicians to manage complex healthcare and developmental needs. We aimed to describe the perspectives of children with chronic kidney disease (CKD) and their parents with regard to communication and decision-making.

Study design: Qualitative study

Setting and Participants: Children with CKD (n=34) and parents (n=62) from six centers across six cities in Australia, Canada and the United States participated in 16 focus groups.

Analytical Approach: Transcripts were analyzed thematically.

Results: We identified four themes: (1) disempowered by knowledge imbalance (unprepared and ill-informed, suspicion of censorship, inadequacy as technicians); (2) recognizing own expertise (intuition and instinct unique to parental bond, emerging wisdom and confidence, identifying opportunities for control and inclusion, empowering participation in children); (3) striving to assert own priorities (negotiating broader life impacts, choosing to defer decisional burden, overprotected and overruled, struggling to voice own preferences); and (4) managing child's involvement (respecting child's expertise, attributing 'risky' behaviors to rebellion, protecting children from illness burden).

Limitations: Only English-speaking participants were recruited, which may limit the transferability of the findings. We collected data from child and parent perspectives, however clinician perspectives may provide further understanding of the difficulties of communication and decision-making in pediatrics.

Conclusions: Parents value partnership with clinicians and consider long-term and quality of life implications of their child's illness. Children with CKD want more involvement in treatment decision-making but are limited by vulnerability, fear, and uncertainty. There is a need to support the child to better enable them to become partners in decision-making and prepare them for adulthood. Collaborative and informed decision-making that addresses the priorities and concerns of both children and parents is needed.

Non-technical summary: We conducted a multi-national focus group study to explore child and parent perspectives and preferences about communication and decision-making. Our data revealed children with CKD and their parents perceived a knowledge asymmetry, which for some limited their capacity and confidence to contribute to decisions and communicate their concerns and goals. Experiential learning and gaining familiarity with the clinical setting enabled some parents to trust their 'gut' instincts, empowering them to communicate concerns, however some children felt that their preferences were sometimes ignored or dismissed by their parents and clinicians. Parents were challenged with the tension between allowing their child decisional autonomy and taking responsibility to protect their child from the illness burden. Our study highlights the potential for miscommunication and differing priorities between parents and children and provides opportunities for clinicians to improve communication, partner with patients to empower then to become active decision-makers, and recognize parent and child expertise.

8.2 Introduction

Shared decision-making is a cornerstone of patient-centered care and improves patient knowledge, satisfaction, adherence, and outcomes[1-5]. However, this process is particularly challenging in pediatrics because of the dynamic and complex relationship triad that encompasses the autonomy of the patient, legal authority of the parent, and the beneficence and clinical acumen of the physician[1,2,6-9]. This complexity is compounded by the constantly changing nature of these relationships as the child matures.

Shared decision-making is “an interactive process in which patients (including families) and physicians simultaneously participate in all phases of the decision-making process and together arrive at a treatment plan to be implemented”[2]. However, integrating the often conflicting priorities of the child, parent, and clinician for competing treatment options is not straightforward[2]. Failure to involve patients and families in decision-making can exacerbate disempowerment, fear, decisional conflict, and disengagement from healthcare, which can jeopardize safety, quality of care and outcomes for children[2-5,10-13]. Despite this, evidence on child and parental perspectives on communication and decision-making in pediatrics is sparse[2,14-16].

The challenges in communication and shared decision-making are highly relevant in childhood chronic kidney disease (CKD). Children with CKD have a 30-fold increased risk of mortality compared with the age-matched population, and are at risk of serious comorbidities and impaired quality of life, which can limit their perceived capacity to participate in shared decision-making[3,17]. Limited evidence exists on communicating and shared decision-making with children and families dealing with chronic and complex disorders[15,16,18]. This study aimed to describe the child and parental perspectives on communication and decision-making in CKD to identify opportunities to improve shared decision-making, with an ultimate goal of improved care and better outcomes for children with CKD and their families.

8.3 Methods

This focus group study was conducted as part of the Standardized Outcomes in Nephrology – Children and Adolescents (SONG-Kids) Initiative[19]. We used the Consolidated Criteria for Reporting Qualitative Studies (COREQ) to report this study[20].

Participant selection

Parents of children aged 0-21 years and children aged 8-21 years with stage 1-5 CKD, receiving dialysis, or who had received a kidney transplant were eligible to participate. All participants were English-speaking to minimize disruptions to the dynamic of the focus group discussions and due to the lack of resources for multilingual trained facilitators.

Participants were recruited from three centers in Australia (n=44), two centers in Canada (n=16), and one center in the United States (n=36). The site investigators were asked to apply a purposive sampling strategy when selecting patients and their families from their database to ensure a broad range of demographic (age, gender, socioeconomic status) and clinical (CKD stage, diagnosis) characteristics. The researchers approached participants who gave permission to be contacted to provide the time and venue details to participate in the focus group. Informed consent was obtained from participants aged over 18 years. Parental consent and written assent was obtained for those aged under 18 years. Participants received

$50 reimbursement (in their local currency) to cover travel costs. Ethics approval was provided by the Institutional Review Board of all participating centers (Appendix G.1).

Data Collection

Two-hour focus group discussions were conducted separately for parents and children, externally to their treating hospitals, from June 2016 to August 2017, until data saturation. All groups were audio-recorded and transcribed verbatim. Question guides were developed from the literature and discussion with the investigators (Appendix G.2 and G.3). One investigator (CSH, AT, TG) facilitated the group while a second investigator (AJ, LJ, AT, TG, AR) took field notes.

Analysis

The transcripts were entered into HyperRESEARCH software to facilitate qualitative data analysis. TG inductively coded the transcripts line-by-line using thematic analysis and principles from grounded theory to identify concepts related to participants' perspectives on communication and decision-making[21]. Preliminary themes were discussed and revised with AT, CH and SB who had independently read the transcripts. Investigator triangulation ensured that the analysis captured the full range and breadth of the data. A thematic schema was developed to show the relationships among themes (Figure 8.1).

8.4 Results

In total, 62 parents and 34 children participated in 16 focus groups. Participant characteristics are shown in Tables 8.1 and 8.2. Parents were aged from 24 to 58 years and most were mothers (47, 76%). Twenty-five (40%) parents had children who had CKD stage 1-5, 14 (23%) had children on dialysis, 22 (35%) had children with a kidney transplant and one parent did not report CKD stage of their child. Seven (11%) parents had a child with CKD aged younger than 8 years. Children ranged from 8 to 21 years (including younger children aged 8-12 years, adolescents aged 13-17 years and young adults aged 18- 21 years), 19 (56%) were male, 17 (50%) had CKD stage 1-5, 5 (15%) were on dialysis, and 12 (35%) had received a kidney transplant. Twenty-nine children had at least one parent who also participated in the study.

We identified four themes: disempowered by knowledge imbalance; recognizing own expertise; striving to assert own priorities; and managing child's involvement. The respective subthemes are described in the following section with reference to the relevant participant group (parent, child) and relationship context (within the triad). Selected quotations to support each theme are available in Table 8.3. Figure 8.1 shows the relationships among themes and subthemes.

Disempowered by knowledge imbalance

Unprepared and ill-informed: Uncertainty surrounding their child's prognosis meant some parents felt inadequately warned about their child's need for treatment (e.g. transplant). Some believed they were given "false hope" and "unrealistic" expectations regarding medication side-effects and surgery recovery, while others felt they were "getting railroaded into things" (e.g. biopsies) by clinicians. Parents wanted "more education" and fewer "medical terms" to inform decision-making. Younger children struggled to comprehend information from parents and clinicians (e.g. blood results) and wanted more information "in words that [they] could understand". Some adolescents had unanswered questions about their future, such as how potential treatments may impact fertility and "what happens after" graft failure.

Suspicion of censorship: Some parents speculated that clinicians withheld certain information (e.g. graft rejection) or would not discuss new treatments, such as stem cell therapy or new trials, when they asked about them. Some younger children and adolescents suspected they "didn't get told everything" about their CKD and some thought their parents did not want to disclose the severity of their illness.

Inadequacy as technicians: The perceived knowledge imbalance between clinicians and parents further served to disempower parents as they felt forced to relinquish their caregiver role to clinicians and accept treatments that they did not necessarily prefer. For example, some parents believed they did not have the technical expertise required to choose less disruptive and invasive treatments such as home peritoneal dialysis and were therefore forced to accept in-center hemodialysis. This resulted in feelings of guilt and helplessness in not being able to contribute to their child's care.

Recognizing own expertise

Intuition and instinct unique to parental bond: While clinicians were acknowledged as "the expert", parents learnt to regard their own instinct, a "mum gut", as equally important. Parents could detect signals in their child, for example "dry lips", "heavy breathing", or "not growing", that were ultimately determined to be clinically important. At times, parents believed they had to "convince" or "pressure the doctors" to investigate their child's symptoms and were distressed and frustrated when they felt ignored.

Emerging wisdom and confidence: As they became more familiar with CKD and the clinical setting some parents developed knowledge and skills that enabled them to better manage their child's care and were thus able to opt for preferred treatments options, such as home dialysis. A few became increasingly sceptical about treatment decisions made by clinicians, particularly if they resulted in detrimental outcomes or were perceived as unnecessary.

Identifying opportunities for control and inclusion: As parents developed confidence in their own expertise they pursued opportunities for involvement and control. Some challenged the need for repeating invasive procedures such as biopsies, or argued to change their child's immunosuppression dose after "seeing the side effects." Having to face uncertainty and burden of treatment, some adolescents and young adults sought autonomy in making decisions and managing their health and treatment where possible. They wanted to contribute to decisions about medications, diet, and invasive interventions such as dialysis or surgery. Some younger children felt their parents dominated interactions with clinicians which hindered their ability to communicate with clinicians directly.

Empowering participation in children: Parents appreciated when clinicians encouraged independence and responsibility in their child for self-management. While some children were satisfied with how their clinicians incorporated their preferences in their treatment, some adolescents were "worried" about their lack of involvement and how they would cope with the role of primary decision-maker as an adult – "at the moment it's in your mum and dad's hands, when you get older you've got to take it into your own hands."

Striving to assert own priorities

<u>Negotiating broader life impacts:</u> Parents considered the impact of treatment decisions on their child more “broadly” in terms of the long-term risks and felt clinicians tended to focus on immediate clinical outcomes. They questioned “textbook” decisions that often focused on survival, and instead wanted to draw attention to impact on “quality of life”. For example, one mother refused bladder augmentation to avoid her child having to be “catheterized for life.” Children wanted to “hang out with friends, go out, have fun, be normal kids,” but felt restrictions imposed by their parents and clinicians (e.g. diet and physical activity) and the treatment and symptom burden of CKD limited their freedom of choice.

<u>Choosing to defer decisional burden:</u> While parents valued being involved in decision-making in many instances, they were sometimes grateful for the option to ease their own decisional burden by deferring to physicians who were trusted to “know what they were doing.” As CKD was “scary” and involved complex treatment decisions in sometimes life-threatening situations, younger children often felt they lacked sufficient knowledge to determine the best course of action, and children of all ages looked to clinicians and parents for support and “reassurance”.

<u>Overprotected and overruled:</u> Adolescents and young adults were frustrated when their parents were “super protective” and did not trust them to make the right choice as this limited their involvement in decisions that impacted their lifestyle. Children believed they often had no choice regarding treatments and accepted they “have to” “get needles”, “growth hormones”, “go on dialysis” or “have surgery.” However, they were upset and sometimes objected when told by parents not to “play contact sports,” “eat certain foods,” or “do active

things" with their friends, particularly when they believed these did not pose any additional risk.

Struggling to voice preferences: Some children had trouble voicing their preferences because they believed they were viewed as unimportant, particularly when they conflicted with priorities of parents and clinicians. Some refrained from asking questions to avoid appearing "stupid" or because they felt "too shy" to ask. Some children felt unfairly judged or accused by parents and clinicians, for example one adolescent explained that they could not take medications because it made them "sick", but were made by their parents and physician to see a psychologist for non-adherence.

Managing child's involvement

Respecting child's expertise: Some parents valued their child's experience as the patient (e.g. recognizing symptoms) and said that their child "makes the decision, because it's [their] body," however, they sometimes felt the need to intervene if they perceived their child's preference had unacceptable risks or consequences. One mother overruled her adolescent's preference for an open nephrectomy, and instead elected to have laparoscopic surgery to minimize risk and recovery time.

Attributing 'risky' behaviors to rebellion: Decisions including refusing to take medication or attend appointments were blamed on "teenage rebellion" by some parents. Some were concerned they couldn't "make [their child] understand" that having CKD meant their choices had more severe consequences than their well peers'. Some parents "struggled" with their child's non-adherence, and attributed this behavior to adolescent risk-taking and defiance.

Protecting children from illness burden: As CKD is a lifelong condition that requires invasive interventions, and is associated with serious co-morbidities, parents wanted to carry the emotional burden of the disease. They debated keeping their child “in the dark” about their prognosis and potential treatments. Some struggled to find a balance between over-protecting their child and giving them the freedom “to be a kid” and participate in ‘normal’ activities.

8.5 Discussion

Children with CKD and their parents perceived a knowledge asymmetry, which for some limited their capacity and confidence to contribute to decisions, manage treatment, prepare for potential complications, and communicate their concerns and goals. Some parents believed that clinicians did not communicate comprehensive information about their child’s disease, which meant they were unprepared to manage comorbidities and unable to choose treatment options to minimize the risk of complications and side effects. Some suspected clinicians avoided discussion on specific topics particularly in relation to transplant outcomes such as rejection. As children matured, they wanted more information about their disease communicated in ways they could understand. While some adolescents and young adults had a better clinical understanding and could take on more responsibilities (e.g. managing medications), some younger children also wanted to be involved in their care and decisions, especially when it constrained their social freedoms. Children sensed that their parents did not want to talk to them about the severity of their disease and prognosis.

Experiential learning and gaining familiarity with the clinical setting enabled some parents to trust their ‘gut’ instincts, empowering them to communicate concerns about the broader impact of treatments on their child’s life, even if it conflicted with the clinician’s recommendations. Other parents elected to defer decision-making to their trusted clinician to avoid the pressure of having to make difficult decisions, and were grateful for the comfort and reassurance they provided. The desire for inclusion and control among children with CKD was apparent for decisions that impacted their ability to be “normal kids”. They wanted to know about outcomes beyond childhood, including their ability to start a family. Some children sought to regain control through self-management, but felt that their preferences were sometimes ignored or dismissed by their parents and clinicians. This is highlighted by the absence of data from the child perspective recognizing their inherent expertise as the patient.

Parents were challenged with the tension between allowing their child decisional autonomy, and taking responsibility to protect their child from the burden of illness and consequences of high-risk choices and behavior. While some parents claimed they allowed their child to make the decisions, they also believed they should filter communication between the clinician and child, and make the final decision if they disagreed with their child’s preferences. Our study also revealed potential miscommunication and differing interpretations between parents and children. For example, one mother attributed her child’s non-adherence to “teenage rebellion”, whereas the adolescent explained their refusal to take medications was because it made them feel ill. One father explained that his child “doesn’t bother [being involved in treatment decisions]” because “she doesn’t really have that understanding”, while the younger child explained she “wanted to know more…so [she knows] what is happening in [her] body” but felt “too shy to ask.”

As found in previous studies across other childhood chronic conditions, parents may strive to protect their children from the burden of ‘knowing’ by managing communication about their disease and treatment, however this can mean that children are inadvertently denied opportunities for involvement in their care[7,14,22-26]. Children want to be aware of what is happening in their own bodies, involved in their own care, and empowered to address concerns and goals that are important to them, particularly as they grow up[6,12,15,24,27-30]. They also desire experience in decision-making through incremental involvement to be better prepared for transition into adulthood when they will no longer rely on their parents as proxies for their health care decisions and responsibilities[7,18,23,30,31].

Mismatches between patient and clinician priorities have been recognized since the early 1960s, resulting in the paradigm shift in the approach to healthcare – from paternalism to partnership[15,32-36]. In pediatrics, differences between parent and clinician priorities have been well established[2,15,28,37]. Shared decision-making models have been developed to manage this discordance and while some aspects (e.g. multi-directional information exchange, presenting all options clearly, determining preferences for involvement[10,32-35,38-40]) may be applicable to the pediatric setting, they do not specifically address the power imbalance the child faces in the triad[14,15,32,33,35]. The Typology of Youth Participation and Empowerment (TYPE) Pyramid developed for youth empowerment in health promotion suggests a pluralistic approach to youth participation when adults and youth transactionally share control, allowing young people to leverage “social capital” and experience from adults, while still allowing them to defer to adults if desired[41]. Aspects of this framework may be useful for researchers and clinicians working in pediatric CKD, however it may not adequately account for the uncertain trajectory of CKD and may not address the changing needs of a developing child. There remains a need to bridge the gap from the parent/clinician to the child, to consider the child’s preferences separately from their parents’ and to identify effective strategies or frameworks to elicit and integrate the child’s perspective in decision-making[9,13,18,36,42,43].

This study was multinational and offers in-depth insights gained from perspectives of a diverse group of children with CKD and parental caregivers. We achieved data saturation, and used investigator triangulation in the analysis to ensure the themes reflected the breadth and depth of the data. However, there are some potential limitations. All participants were English-speaking and from high income countries, therefore transferability of the findings to other populations and settings is uncertain, but we note that participants were diverse in terms of country of birth and socioeconomic status. Moreover, communication issues experienced are likely to be exacerbated in contexts where all parties in the triad do not speak the same language. While participants likely experienced different types of care and education across different centers, our findings show themes were consistent and relevant across all centers. We acknowledge that we included only one patient on hemodialysis at the time of the study so the views of prevalent patients on hemodialysis may not have been captured extensively. However, children and parents of children previously on hemodialysis discussed their past experiences with hemodialysis and five parents who participated had a child receiving hemodialysis. Our data reflect the views of only two out of the three people in the decisional triad. Clinician perspectives may help to better understand where and why breakdowns in communication occur.

Our findings reveal opportunities for communication training for clinicians to 1) improve transparency in communication, 2) promote partnership with their pediatric patients, and 3) recognize patient and parental expertise. Ensuring parents and children understand all the treatment options (including potential need for treatment), and how they might impact medical, social participation, and longer-term quality of life outcomes may help to alleviate anxiety over uncertainties and suspicion of censorship, and enable them to prepare for the challenges of living with a chronic disease[2,8,23,28]. Partnering with parents to engage children in their own care would involve providing them with appropriate resources and education, creating opportunities for them to be heard, and advocating for their preferences[2,44]. There is also a need to explicitly acknowledge the expertise that parents and children can bring to the decision-making process and to consider these in relation to their preferences when making decisions[14,15,32,38,44].

The American Academy of Pediatrics and the United Nations advocate to involve children in decisions about their health and treatment[2,7,8,16]. However, the lack of child-centred interventions for decision-making denies children a voice in their own care[16]. Evidence from the adult population suggests decision coaching in conjunction with decision aids may be effective in increasing participation in decision-making, increasing knowledge, improving alignment of decisions with patient values, and decreasing decisional conflict[12,45]. A systematic review of interventions to support decision-making in pediatrics found that decision-coaching (i.e. individualized, facilitated discussion to prepare the patient for upcoming decision-making[12,45]) had modest effects in improving the decision-making process, decision alignment between parents and children and satisfaction regarding being informed about options[12]. Visual aids, rephrasing, turn-taking, and role-playing, may also be effective for improving communication in children with CKD[13,22,46]. Journaling can be an effective method to engage children and enables them to express their private experiences and emotions safely[47,48]. Further research is needed to assess the effectiveness of different strategies and interventions for communication and decision-making in the pediatric CKD population, and across disease stages. We suggest the need for more studies to improve communication and shared decision-making that target all three members of the decisional triad[12,18]. An online portal or mobile phone application journaling tool where children can express their preferences, report their symptoms, access decision supports and describe their feelings may empower children to become more active in their health. Children could determine what information they want to share with their parents and health care professionals through a messaging or permission system that could give parents and clinicians access to selected content. We suggest involving children and parents in the development, implementation and evaluation of future interventions to support communication and decision-making in this context, and that future research include a specific focus on their perspectives on decision-making and relationship with multidisciplinary team members including psychologists, social workers, youth workers, and play therapists[18,30].

A perceived lack of knowledge and poor communication disempowers children and parents from becoming active participants in their care. Parents want comprehensive information in plain language and recognition of their expertise, and are motivated by a strong desire to protect their child. Children want "reassurance" and to understand the long-term implications of their disease and treatment, and involvement in decisions that impact their ability to live a "normal" life, however they struggle to voice their preferences. An absence of data on the child's expertise as the patient highlights the need to invest in building this expertise incrementally and in line with their development to better equip them to become partners in decision-making and prepare them for adulthood. Effective interventions to support communication and shared decision-making are needed. These can lead to better knowledge and understanding of the condition[15,16,18,49] as well as improved health outcomes[5,8,22,39,50], improved decisional quality[2,15,16,28] and improved patient satisfaction[5,14,16,51].

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We are grateful to the families that gave their time to participate in these groups and shared their valuable experience.

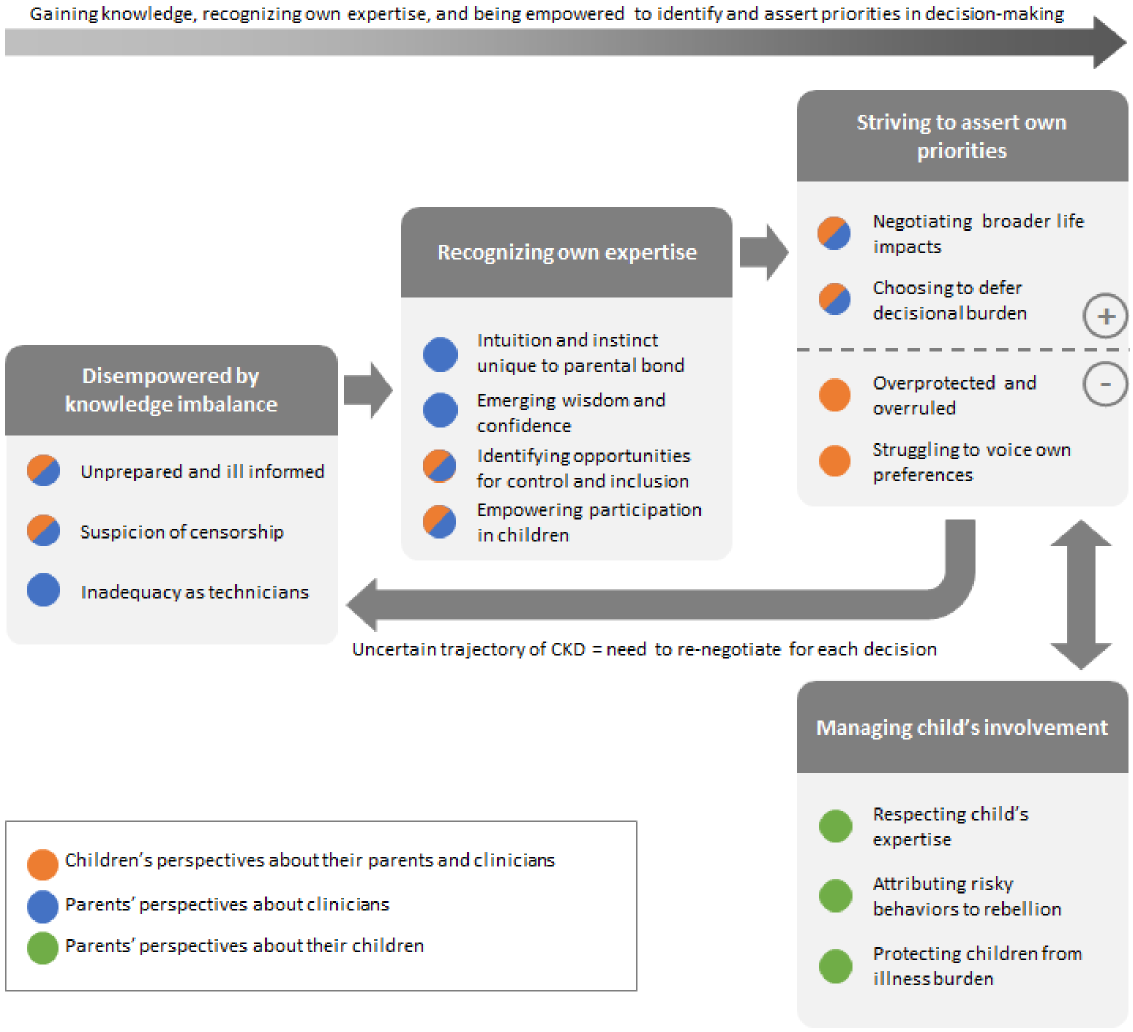

Figure 8.1 Thematic Schema

The uncertainty of prognosis and management of CKD served to compound the already difficult nature of decision-making for parents and children. Some felt they were not given adequate preparation and explanations of treatments (e.g. transplant and immunosuppression), and both parents and children wanted more information about their disease and potential treatments communicated in plain language to inform their decision-making. The lack of transparent information led parents to believe clinicians were withholding information about their child's prognosis. Similarly, children felt their parents were censoring information about their CKD. Parents were further disempowered when they were forced to relinquish part of their caring responsibilities due to their lack of technical expertise (e.g. home dialysis).

Over time parents began to recognize their ‘gut’ instincts added valuable and complementary information to support decision about their child’s treatment, and they developed confidence to challenge clinicians, and subsequently they felt more involved and in control of their child’s health. As children grew older, they too looked for opportunities to be involved in their care and were able to do so when supported by parents and clinicians.

Once parents developed confidence in their own expertise they were able to assert their priorities to ensure clinicians considered the broader impact treatments would have on quality of life. While some remained sceptical from feeling uninformed, others developed trusting relationships with clinicians who they sometimes relied on to ease their decisional burden when decisions were too difficult. While children were also concerned about the impact decisions would have on their quality of life, their view of what was important often differed from their parents (e.g. ability to play sport). Their ability to assert these preferences was limited by their interactions with parents and clinicians which made them feel that their priorities were not important.

Parents were then able to re-establish their role as protectors for their children. However, as their children developed and wanted more control, parents had to balance allowing decisional autonomy and trusting their child’s expertise with the desire to protect their child from the burden of their disease as well as from potential risky behaviors. From the child’s perspective, this limited their involvement and denied them opportunities to practice decision-making in preparation for adulthood.

Table 8.1 Characteristics of the child participants (N=34)

Characteristics	n (%)
Gender	
Male	19 (56)
Female	15 (44)
Country of birth	
Australia	13 (38)
Canada	5 (15)
USA	14 (41)
Other*	2 (6)
Age (years)	
8-12 (young children)	10 (29)
13-17 (adolescents)	20 (59)
18-21 (young adults)	4 (12)
Current CKD** treatment stage	
CKD 1-5	17 (50)
Home peritoneal dialysis	4 (12)
In-center hemodialysis	1(3)
Transplant	12 (35)
Primary kidney disease	
Congenital abnormalities of kidneys/urinary tract	12 (35)
Nephrotic syndrome (cause not specified)	4 (12)
Focal segmental glomerulosclerosis	3 (9)
Polycystic kidney disease	2 (6)
Cystinosis	1 (3)
IgA nephropathy	1 (3)
Not reported or not known	11 (32)

*Other includes: Mexico, New Zealand; **CKD: chronic kidney disease

Table 8.2 Characteristics of the parent participants (N=62)

Characteristics					n (%)
Gender					
	Male				13 (21)
	Female				49 (79)
Carer relationship					
	Mother†				47 (76)
	Father†				13 (21)
	Other (grandmother, aunt)				2 (4)
Country of birth					
	Australia				11 (18)
	USA				9 (15)
	Canada				8 (13)
	Other*				25 (40)
	Not reported				9 (15)
Age (years)					
	21-30				4 (6)
	31-40				10 (16)
	41-50				28 (45)
	51-60				8 (13)
	Not reported				12 (19)
Highest level of education					
	Bachelor degree or higher				19 (31)
	Diploma/certificate/trade				18 (29)
	Secondary school				11 (18)
	Primary school				2 (3)
	Not reported				12 (19)
Household annual income (before tax, local currency)					
		US	CAD	AUD	Total
	$0-39,000	5	1	2	8 (13)
	$40,000 - $59,999	2	1	7	10 (16)
	$60,000 - $84,999	0	6	6	12 (19)
	$85,000-$124,999	7	2	3	12 (19)
	>$125,000	2	1	5	8 (13)
	Not reported	5	0	7	12 (19)
Child's age (years)					
	0-7				7 (11)
	8-12				18 (29)
	13-17				32 (52)
	18-21				3 (5)
	Not reported				2 (3)

Child’s CKD** treatment stage		
	1-5	25 (40)
	Home peritoneal dialysis	8 (13)
	In-center hemodialysis	5 (8)
	Dialysis, non-specified	1 (2)
	Transplant	22 (35)
	Not reported	1 (2)
Child’s primary kidney disease		
	Congenital abnormalities of kidneys/urinary tract	15 (25)
	Nephrotic syndrome (cause not specified)	9 (15)
	Focal segmental glomerulosclerosis	4 (6)
	Polycystic kidney disease	4 (6)
	PUV	4 (6)
	Other^	9 (15)
	No reported or not known	17 (28)

† Twenty (32%) parents were 10 couples with one child with CKD
*Other includes: England, Ethiopia, Fiji, India, Kenya, Mexico, New Zealand, Pakistan, Poland, Somalia, Vietnam; **CKD: chronic kidney disease; ^Other includes: Cystinosis, Eagle Barret Syndrome, IGA Nephropathy, Scleroderma, Neurogenic bladder, bladder obstruction, reflux

Table 8.3 Selected illustrative quotations

Theme	**Illustrative Quotations** ***(participant, child's age, CKD stage, country)***
Disempowered by knowledge imbalance	
Unprepared and ill-informed	They got us bits and pieces of information. It was more medical. Some of it was hard for us to understand. *(Father, 8-10y, transplant, Australia)* I never felt like I had enough information about the procedures. Sometimes I just feel like I'm getting railroaded into things. *(Mother,13-17y, transplant, Australia)* I think parents should be given more education, so that we know what's going on, and we know more about the disease. We lack a lot of outcomes of the studies you do, we should be informed about that. *(Father, 8-10y, CKD, Australia)* It's also a Catch-22 because we want to keep his kidneys as long as we can, keep it healthy for as long as we can. But at the same time, once he reaches 18, he's no longer a child, so finding a kidney is going to be harder. *(Mother, 13-17y, CKD, US)* If you had just told me that was a side effect I'd be prepared. *(Grandmother, 13-17, CKD, Australia)* They were saying things like, oh you won't know yourself, it'll be a magical experience, you'll get to do all these things you've never done, it's going to be wonderful to have this life you've never had, and then it hasn't been. *(Mother, 13-17, transplant, Australia)* So giving false hope, really…instead of a more realistic, instead of painting a fantasy that everything's going to be better. *(Mother, 13-17y, CKD, Australia)* For a few years I thought he only had one kidney and that was it. And then, at one appointment, she just said, "Oh, well when he has his transplant", and I was like, "What are you talking about?" That was kind of when I found out. *(Mother, 13-17y, CKD, US)* I wanted to know if I would get better, but the answer she said didn't actually make any sense. Something printed out in words that I could understand [would be useful]. *(Girl, 8-12y, CKD, Australia)* When you have a kidney transplant they say it can last up to 15 years. But I don't know what happens after that. *(Girl, 13-17y, transplant, US)*
Suspicion of censorship	They say you get kidney rejection episodes which can scar the kidney. I think we've had one, but they haven't told us. *(Mother, 8-12y, transplant, Australia)* [For] his disease [they] are doing cure trials in America starting from this year, but the doctors won't acknowledge it when I bring it up. *(Mother, 13-17y, dialysis, Australia)*

	They didn’t say anything, they just said, “His blood pressure is too high, and you need to go to the Emergency Room.” *(Mother, 18-21y, transplant, US)* The biggest challenge that I see- and the biggest impact to [her daughter’s] life- is her delayed development and her learning disabilities, things that I’m now told go hand-in-hand with transplantation and kidney failure. *(Mother, 13-17y, transplant, Canada)* My mom was crying about it, and I was like, “Why are you crying, what’s going on?” And she was like, “You might have a transplant”. And I was like, “What’s that?” And she wouldn't tell me because I was young. *(Girl,13-17y, transplant, US)* I feel like sometimes like I don’t get told everything. But I don’t know if I am or not. *(Girl, 13-17y, CKD, Australia)*
Inadequacy as technicians	They gave me two options, whether you want to do peritoneal dialysis or haemodialysis, but I chose haemodialysis because I wasn’t confident that I could do the dialysis at home. *(Mother, 13-17y, transplant, Australia)* It is the most helpless feeling. You can’t do nothing. *(Father, 13-17y, transplant, Australia)* They’re like, “and you’re going to have to monitor her blood pressure but don’t worry, we’re going to get you a cuff”. Within a week we were going by ambulance back to the hospital because no one told me how to work the blood pressure cuff. *(Mother, 13-17y, CKD, US)*
Recognizing own expertise	
Intuition and instinct unique to parental bond	I’m following my ‘mum gut’ and I think they respect that. *(Mother, 8-12y, CKD, Australia)* The only reason I brought him into the hospital that day was because he was breathing more heavily than usual. *(Mother, 0-7y, dialysis, Canada)* He had dry lips, and I thought ‘hang on, you drink so much, why are your lips always cracked?’ So I took him to my local GP. Luckily he listened to me, He was down to 29 per cent function, that’s how we found out. The pediatrician said if I’d asked her it would’ve been a straight out no, but I taught her a lesson now that any mum that’s got any queries, that she will listen to. *(Mother, 8-12y, CKD, Australia)* So the struggle with our local MDs, to get us here. She had shown all these signs and symptoms. I went to see pediatricians, I went to see doctors but nothing, they kept saying, “Oh, she’s going to grow, she’s this, she’s that, blah, blah”. *(Mother, 8-12y CKD, Canada)* It took me pressuring the doctors, and pressuring the doctors, to figure out what was going on with her. *(Mother, 13-17y, dialysis, US)*
Emerging wisdom and confidence	I think you sometimes have to put your views [forward]. He [doctor] is the expert. [However] when you are at home you know what is going on. *(Mother, 8-12y, transplant, Australia)*

	You know your child. *(Mother, 13-17y, transplant, Australia)* Initially whatever they would say, we would say okay, but now we question everything Why? What is that going to tell us? Why are you doing that? Is there any risk? Why do you think he needs it? I always ask. *(Mother, 8-12y, transplant, Australia)* I continually now wonder whether it wouldn't have been wiser to transplant her much earlier. And it's the one thing that I would ask, that they look at and study- because it seems to me almost like a black art. How do you know when she needs a transplant? *(Mother, 13-17y, transplant, Canada)* They've been so upfront and honest with it. I'm so thankful that my GP listened to me and didn't question it or didn't blame me. (Mother, 8-12y, CKD, Australia)
Identifying opportunities for control and inclusion	I think now we have become quite vocal in what we want. Especially with immunosuppression. We have seen the effects on [our son], so sometimes it is a bit of a fight with the doctor. *(Mother, 8-12y, transplant, Australia)* He's got another biopsy soon. And I'm thinking why are we having another one? And they're like 'just to see where we are'? And I'm thinking, is this really necessary? Does he really need it? *(Mother, 13-17y, transplant, Australia)* I started doing all my medications, I want it to get to the point where I don't need them to do anything. Not depend on my parents, or fixing the medications I need to take, or the dosages. *(Boy, 18-21y, transplant, USA)* I tend to know a lot in general, like I look into the details of each test, because I know that there's going to be specific ranges which I have to be within. *(Boy, 13-17y, transplant, Australia)* It was very tiring for me to have dialysis and go to school, so I asked my mum to sign me up, so I started homeschool. *(Boy, 18-21y, transplant, USA)* When I ask a question that I really want to know I feel good about it because then I'm aware and I know what is going on with my kidney. *(Girl, 13-17y, CKD, Australia)* They give us choices and keep us well-informed the doctors have been pretty good with their explanations of the medicines. *(Mother, 8-12 y, CKD, Australia*
Empowering participation in children	The doctors did well with making sure she knew what medications she was taking, how much she was taking, what they were for. *(Mother, 13-17y, dialysis, US)* I know my doctor so well, you can say anything and he'll try make it happen. *(Girl, 13-17y, CKD, Australia)* They [doctors] understand. *(Boy, 13-17y, transplant, Australia)* I'm just worried 'cause you're like, at the moment it's in your mum and dad's hands, when you get older you've got to take it into your own hands, got to know about the medications and stuff. *(Boy, 13-17y, transplant, Australia)*

Striving to assert own priorities	
Negotiating broader life impacts	I'm thinking broader, more about their ability to live independently, be an adult. It really worries me how she's going to carry on as she gets older and graduates from high school. What she's going to be able to do, and whether she's going to be able to live independently, or function efficiently. *(Mother, 13-17y, transplant, Canada)* We've spent most of the time in hospital since then, and it's like you know [he is] missing out on his school, his friends. *(Mother, 13-17y, dialysis, Australia)* The transplant surgeons were insisting that he should have his bladder augmented and he'd have to be catheterized for life and I said tell me why we are writing this off before it's had a chance to prove itself? And eventually we sourced an interim solution and subsequently he pees normally. *(Mother, 13-17y, transplant, Australia)* They're only looking at it from a medical scientific outlook, parents care and love, doctors don't care and love, it's a job. *(Mother, 13-17y, CKD, Australia)* Medication can make your kidney last longer. But is that a good longer? How are you surviving that longer? Are you in pain all the time? Are you in discomfort? You have a transplant 2 years early, but you've grown, you've played, you have enjoyed your life. You aren't in hospital all the time… So quality of life, not just how old you are going to get, but are they experiencing life and not just stuck in a hospital bed. *(Mother, 8-12y, transplant, Australia)* Sometimes we want to hang out with friends, go out, have fun, be normal kids. We really can't with medications, we have restrictions. Like, "You can't do this, you might get sick. You can't do this, you might get sick". We have so much we can't. *(Girl, 13-17y, transplant, US)*
Choosing to defer decisional burden	They know what they're doing, I feel really comfortable with that. *(Mother, 13-17y, transplant, Canada)* He's been brilliant, and he's always, ultimately, she's your child, it's her body, and he has said that. But there's times when you don't want the control, you don't want to have to make that decision, and you have to push it back onto them. *(Mother, 8-12y, CKD, Australia)* They were really good at explaining things to us. It was scary, but at least we knew that it could be taken care of. *(Mother, 13-17y, dialysis, US)* Doctors should have that reassurance, like "it'll be okay." *(Boy, 13-17y, CKD, Canada)* I don't talk to doctors a lot. I just sit there and let their mum talk. Well I don't like talking to males to start with. *(Girl, 13-17y, dialysis, Australia)* [I prefer to see doctors with my parents] because they know more stuff than I do. They know more of the terms. *(Girl, 8-12y, transplant, US)*

Overprotected and overruled	There are some sports that I'm not allowed to do at all, for the rest of my life. That I can get. But there's other sports I want to do, like hockey, where the padding is over where the kidney is. I love it. So I'm technically allowed to do it, but my parents won't let me do it. *(Boy, 13-17y, transplant, Australia)* I feel like that can be stressful at home, because your parents are protective - "Hey, have you taken your medication?" "Are you sure?" *(Boy, 13-17y, CKD, US)* My mum and my granny are so like on top of me. Sometimes in a good way sometimes in a bad way because it gets really annoying. *(Girl, 13-17y, CKD, Australia)* My mom is super protective. My mom isn't worried about my meds, but she's worried about the people, the food. *(Girl, 18-21y, transplant, US)*
Struggling to voice own preferences	I had to do something like this for my psychologist because I didn't take my tablets because they made me sick. We had to write a whole list down of what was important and what was not. ..I didn't take my tablets because they made me sick… Oh they are disgusting! I took a sip and I threw up. They said if I didn't drink it I wouldn't be able to eat. I still ate. But I didn't drink it…. *(Girl, 13-17y, dialysis, Australia)* I was very sad, because my parents didn't want me to have a quinceañera because they were worried that when I was having the time of my life, I might get a call, like, "We need you to come on home, we have a kidney for you". *(Girl, 13-17y, transplant, US)* Medical stuff, not social stuff. They [parents] don't understand. *(Girl, 13-17y, CKD, Australia)* I wish I could see my kidney. I don't ask because I don't want to seem stupid. *(Boy, 13-17y, dialysis, US)* Having a child and then passing it on to them. That worries me. I haven't really spoken to people about having children. That can wait. *(Girl, 13-17y, CKD, Australia)* Sometimes I want to ask but I normally don't ask because my parents will speak to them. And I get too shy to ask. *(Girl, 8-12y, CKD, Australia)* Some of the time it's because I'm shy to ask. Or if it is a really big question I'm like worried to ask. *(Girl, 13-17y, CKD, Australia)*
Managing child's involvement	
Respecting child's expertise	We talk about it, and if I don't agree with her I will overrule her, but she is the one that makes the decision, because it's her body. *(Mother, 13-17y, dialysis, Australia)* Two years later [in remission] he got a cold and he knew the achiness of his lower back, he felt his body, you know, how it felt then [at previous diagnosis of nephrotic syndrome], he knew it was the same sort of thing. *(Mother,13-17y, CKD, Australia)*

	She's proud of her scars, she wears them with honour… So it's a battle that she's won, or she's winning. *(Mother, 13-17y, dialysis, Australia)* It's his body, even though it affects the whole family, but he's the one that has to come to terms with what he has for the rest of his life. *(Mother, 13-17y, dialysis, Australia)* [My daughter] gets most of the say. I allow [her] to decide what she wants to do, because she's the one that's gotta go through it, not me. You know, I've gotta be there with her, every step of the way, but I believe the decision's up to [her]. *(Mother, 13-17y, CKD, Australia)* Yeah, with our daughter the big trouble now would be that she's 11 years old, she doesn't bother, she still thinks that she doesn't really have that understanding *(Father, 8-12y, CKD, Australia)*
Attributing 'risky' behaviors to rebellion	*Mother 1*: For me it's her taking her tablets. It's the thing keeping her well, and alive, and it's, it's a struggle when she misses several days, and all you see is the blood pressure go straight back up. *Mother 2*: How old is she? *Mother 1*: Fourteen. *Mother 2*: Fourteen, teenagers! *(Mothers, 13-17y, dialysis/CKD, Australia)* We also get the teenage rebellious I don't want to take my pills today. I don't want to do it anymore. *(Mother,13-17y, transplant, Australia)* We threaten her all the time, she's 15, she's at the age when she's tired of cath and all that. She does everything that all the kids around her do. But she has issues, and that's something we can't make her understand. It's a hard age. *(Father, 13-17y, dialysis, US)* She did sneak off to a party. She did have a drink. We found out through Facebook and we badgered her about her choices…I said you need to know your choices and you need to make the right ones. *(Grandmother, 13-17y, CKD, Australia)*
Protecting the child from illness burden	I went back and forth on that, in terms of letting [my daughter] know how serious it is, or just let her be a kid and me worry about it. Because I tell her all the time "I got this, I'll do all the worrying, you go and be a kid." *(Mother, 13-17y, transplant, US)* I tried to keep my son in the dark for as long as I can. *(Mother, 13-17y, CKD, US)* I'm always saying to my son you can't do football, better not do basketball, can't do this. It's only in the last three months I thought, I better check myself as well, and remind myself well hang on, he's still got to be a child. I'm not going to let him sit there and ponder or worry about what's wrong with him. *(Mother, 8-12y, CKD, Australia)*

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Chapter 9: Discussion

9.1 Summary and synthesis of findings

The overall aim of this thesis was to establish an evidence-based framework for involving consumers in research in CKD. Specifically, this thesis addressed the following objectives:

1. Describe the ways in which consumers have been historically involved in CKD research (Chapters 2,4 and 5)
2. Describe the ways in which consumers have been involved in shared decision-making in CKD treatment and care (Chapter 3)
3. Describe consumer perspectives on involvement in decision-making (Chapter 8)
4. Identify effective methods to engage and involve consumers in research (Chapters 2,4 and 5)
5. Describe the impact of consumer involvement on research output (Chapters 6 and 7)
6. Evaluate consumer involvement projects (e.g. conferences) and consumer contributions to research (Chapters 6 and 7)
7. Develop a practical framework for involving consumers in research (Chapter 9)

I used predominantly qualitative methods to address the objectives above because they were most appropriate and relevant to the research questions as consumer involvement in research in kidney disease is an area with scarce evidence, particularly on the attitudes and experiences of consumers. Chapter 2 was a mixed-methods systematic review using quantitative methods to synthesise the included studies and a qualitative synthesis to scope the current state of consumer involvement in published research in CKD. Chapter 3 included a narrative review to assess the nature of shared decision-making in practice in CKD. A combination of semi-structured interviews and workshops with focus groups were used in the first part of this thesis (Chapters 4 and 5) to address conceptual aspects of the experiences of consumer involvement, and in the second part of this thesis (Chapters 6, 7 and 8) to provide novel insights into the application, implementation and evaluation of consumer involvement in research and decision-making practices in real-world settings. This final chapter provides a synthesis of findings presented as a framework for evidence-based best practice for consumer involvement in research in CKD.

Development of an evidence-based framework for best practice

Part A: Conceptual Development (Chapters 2,3,4 and 5)

Consumer involvement in research (Chapters 2,4, and 5)

<u>How consumers have been involved in the past:</u> Consumers have been involved in research in CKD in varied capacities. Consumers have been co-researchers, members of steering committees with other healthcare professionals, consumer-only boards, and participants in qualitative studies, surveys and workshops to provide input on research (e.g. prioritising outcomes, developing outcome measures[1-3]). They held roles across the spectrum of involvement[4] (Figure 9.1), from being attendees (e.g. conference delegates[3,5]) through to initiating and leading research projects (e.g. proposal for medical device development[6]). Consumers have been involved in every stage of the research cycle from priority setting through to dissemination and implementation[7]. This demonstrates that consumers may be involved at different levels, with various roles, and across the research translation spectrum.

Figure 9.1 The spectrum of levels of involvement and decision-making power

<u>Benefits to researchers and consumers:</u> Some of the reported benefits to researchers were improved recruitment, retention, adherence and communication between the research team and their participants; richer data collection and validation of findings (particularly for qualitative data); improved knowledge translation; better agreement on priorities between stakeholders; and improved access to and relationship with the consumer and broader community. For consumers, being involved in research was empowering – their involvement resulted in more patient-centered research and useful outputs (e.g. plain language summaries), and access to information and education meant that consumers felt better informed and developed confidence to advocate for themselves and other consumers, both in clinical settings and in research. Involvement in research also connected them with other consumers and the consumer community and provided opportunities for personal and professional development.

<u>Challenges:</u> Consumers involved tended to be highly educated, white and from high income countries. Involvement occurred more frequently in the early stages of the research project and most consumers were involved in ‘one-off’ activities with limited decisional power and had limited involvement in later stages to ensure their voices were carried through all phases of the study. Despite evidence showing significant advantages of involving consumers in data collection and analysis in qualitative research, this was rare. There also appeared to be

approaches that reinforced or did not consider power imbalances that may have limited input and contribution, for example by only having one consumer on a steering committee of health professionals.

While consumers were engaged and selected in many ways, often inadequate resources and knowledge meant that a “convenient” approach was used rather than a strategic purposive approach to ensure inclusion of relevant and diverse expertise and experience. As such, consumers from minority groups (including from non-English speaking backgrounds) or those experiencing severe illness for example may have been precluded from involvement. For most, the onus remained on them to look for opportunities to be involved at their own personal cost. Limited diversity was exacerbated by the lack of community and public knowledge about CKD. Generally, consumers lacked the logistical, financial and educational support needed to build confidence in their role as an expert, to navigate the power imbalances they faced, and to manage their illness and treatment burden to be able to contribute optimally.

Reporting and publishing of consumer involvement practices in CKD remains scarce, limiting transparency around consumer contributions and impacts, and the vast majority of published papers did not report using the GRIPP/GRIPP2 checklists designed for reporting consumer involvement in research.

<u>Opportunities:</u> Consumers identified the need for a systematic approach to involvement so as not to dilute the patient voice and to provide opportunities for involvement to engage and involve a more diverse group of patients and caregivers. Raising the profile of CKD in the community could also increase the pool of potential consumers by encouraging earlier diagnosis, and therefore involvement of consumers with earlier stage CKD. Leveraging

consumer motivations for community, knowledge/education, health ownership and altruism were also identified as opportunities to increase and expand involvement in research.

Consumer involvement in clinical decision-making (Chapters 3 and 4)

Power asymmetry and ownership of knowledge: Consumers perceived a power asymmetry whereby clinicians were the owners of health knowledge and decided what to share with them. Despite professional recommendations to use shared decision-making, consumers reported not feeling involved in clinical decisions (e.g. to initiate dialysis[8,9]). Power asymmetry in clinical settings carried over to the research setting, where consumers lacked skills and confidence to participate on the same level as health professionals.

Poor communication and mismatched priorities: Use of jargon, inconsistent, unplanned or poorly timed (e.g. unwell, urgent) delivery of information limited its decisional usefulness for consumers. Prognostic information shared by clinicians for decision-making often focused on clinical outcomes (e.g. mortality) and omitted other outcomes important to patients (e.g. quality of life). Limited time with clinicians to build relationships and trust discouraged future involvement in research.

Building capacity: The SHARE approach[10] (*Seek your patient's participation, Help your patient explore and compare treatment options, Assess your patient's vales and preferences, Reach a decision with your patient, Evaluate your patient's decision*) and the Ask-Tell-Ask communication framework support consumers to become equal contributors in decision making, and develop confidence through education, rapport, trust, and bidirectional information exchange[8]. As well as countering the power imbalance, consumers empowered in their own care were better educated, less overwhelmed by their illness and treatment burden and faced less stigma and shame associated with patient status, thereby improving their capacity for involvement in other contexts, including research.

Part B: Applied Empiric Studies (Chapters 6,7,8)

Consumer involvement in conferences (Chapter 6)

Having consumers as part of the conference validated scientific data through their experiences, helped health professionals (particularly allied health professionals) to apply findings to clinical practice and increased empathy for patients and families. Use of plain language in conference presentations meant research findings were more accessible to all delegates, including consumers. Democratising access to knowledge and information was thought to lead to better self-management, and therefore improved patient outcomes. Successful involvement helped validate the purpose and potential of consumer involvement more broadly for clinicians. However, consumers lacked clear guidance in preparing talks, financial support in attending (e.g. accommodation, honoraria for speaking), and struggled with fatigue with attending long days in a large venue. They also wanted to be involved in capacities other than just telling their story, including contributing to the conference agenda and program. They believed that with additional guidance and planning, their broader skills could be optimised to align their lived experience with conference learning objectives to be more impactful.

Consumer involvement in guideline development (Chapter 7)

Consumer involvement in topic and outcome selection for the CARI biopsy guidelines highlighted discordance between patient and health professional priorities with less than one third of topics/outcomes identified by both groups. Clinicians identified topics/outcomes related to clinical procedures (e.g. bleeding, complications), whereas consumers viewed the biopsy in the context of their broader lives and wanted to be empowered for self-management through better education and communication, and recognition of the impacts on psychosocial outcomes. As a results of patient involvement, a new topic "Biopsy information and education for patients and caregivers" was added to the guideline[11], and a new process was initiated to develop a consumer version of the guidelines, led by a consumer subgroup of the BEAT-CKD advisory committee (Appendix H).

Consumer involvement in clinical decision-making (Chapter 8)

A knowledge imbalance between consumers and clinicians, a lack of information and a perception of censorship on behalf of clinicians limited involvement in clinical decision-making for parents and their children with CKD. Time and experience with the health system allowed them to develop confidence in their own expertise, seek opportunities for inclusion and advocate for wider priorities to be considered by their clinicians.

Applied learnings for consumer involvement in research (Chapters 6,7, and 8)

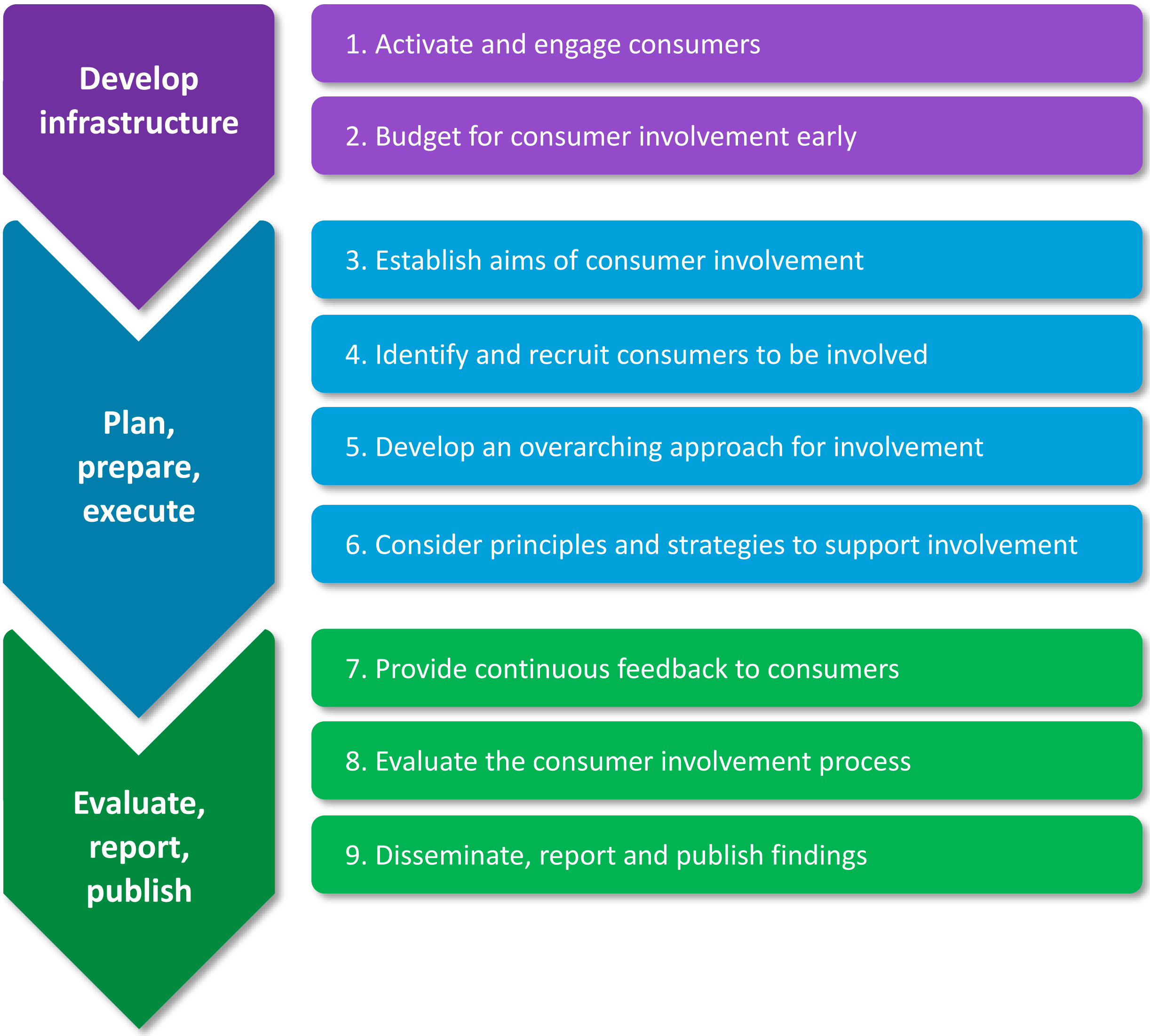
Aligning priorities: Studies of patient involvement in care and research have consistently highlighted discordances in priorities between clinicians/researchers and consumers – clinicians and researchers tended to focus on clinical outcomes (e.g. mortality, cardiovascular disease, hospitalisation) while consumers gave higher priority on symptoms and broader life impacts (e.g. quality of life, ability to work, fatigue). Workshops and focus groups to elicit consumer priorities for decision-making and research helped to align priorities between both groups.

Cultivating confidence in expertise: In both clinical and research settings, consumers needed support in cultivating their expertise, and recognition of the knowledge derived from their lived experience, to be able to build confidence in advocating for their preferences and priorities among health professionals. Together with greater access to information, this served to equalise power asymmetry and better equipped consumers to contribute to research as partners.

Part C: Framework Development (Chapter 9)

From existing work and studies included in my thesis, I have developed a framework for consumer involvement in research in CKD that covers the following domains: 1) develop infrastructure, 2) plan, prepare, execute, and 3) evaluate, report, publish (Figure 9.2). Each domain is described in detail in the following section, with practical and feasible recommendations and examples for researchers based on the evidence generated in my thesis, specific to consumers with lived experience of CKD. At a high level, this framework is designed to encourage consumer involvement broadly, outside of specific research projects by engaging, activating, and communicating with consumer communities. Within specific studies, projects, or programs it aims to provide a roadmap towards successful consumer involvement practice through recommendations for selecting the right consumers, at the right stages, in the right activities to meet the goals and objectives for consumer involvement, underpinned by appropriate principles and strategies.

Figure 9.2. Framework for involving consumers in CKD research



1. Activate and engage

- **Educate consumers and broader community about kidney disease** – raise awareness, decrease misinformation and stigma, increase knowledge of disease, research findings and research methods
- **Involve consumers in shared decision-making in their clinical care** – improve understanding of disease and research, decrease power asymmetry in relationship
- **Educate and train clinicians and researchers in consumer involvement** – planning for consumer involvement, communication, inducting patients into research
- **Understand consumer motivations for involvement**
- **Develop reciprocal relationships with the consumer community and consumer organisations** – provide education/information sessions, engage through social media
- **Develop databases of consumers interested in being involved in research** – elicit experience, skills, and preferences for interests, availability
- **Establish overarching consumer advisory board** – call on this group to oversee/advise on all research projects, consumer involvement strategies
- **Include consumer co-researchers as part of the research team from the beginning** – impact agenda, funding, priority setting, design, involvement strategy
- **Involve consumers in dissemination of research findings** – invite to scientific meetings, share lay summaries through consumer organisations

2. Budget for consumer involvement early

- **Include budget for consumer involvement in grants** – view as essential investment similar to other budget line items
- **Consider payment costs** – daily/hourly rate (US$200 per day recommended by INVOLVE), paid positions
- **Consider reimbursements for expenses** – travel, accommodation, childcare, meals, registrations, licenses
- **Allocate time/funding for consumer liaison**
- **Allocate funding for training** – for consumers and researchers, methods and subject matter

3. Establish aims of consumer involvement

- **Develop clarity on the reasons for involving consumers** – overall aims for consumer involvement in the project/program, what do researchers hope to achieve/gain from involving consumers?
- **Identify where consumer involvement will have the most impact** – specific stage, role or activity
- **Develop specific objectives for consumer involvement** – what are the specific impacts/outcomes? How will these be measured/evaluated?

4. Identify and recruit consumers to be involved

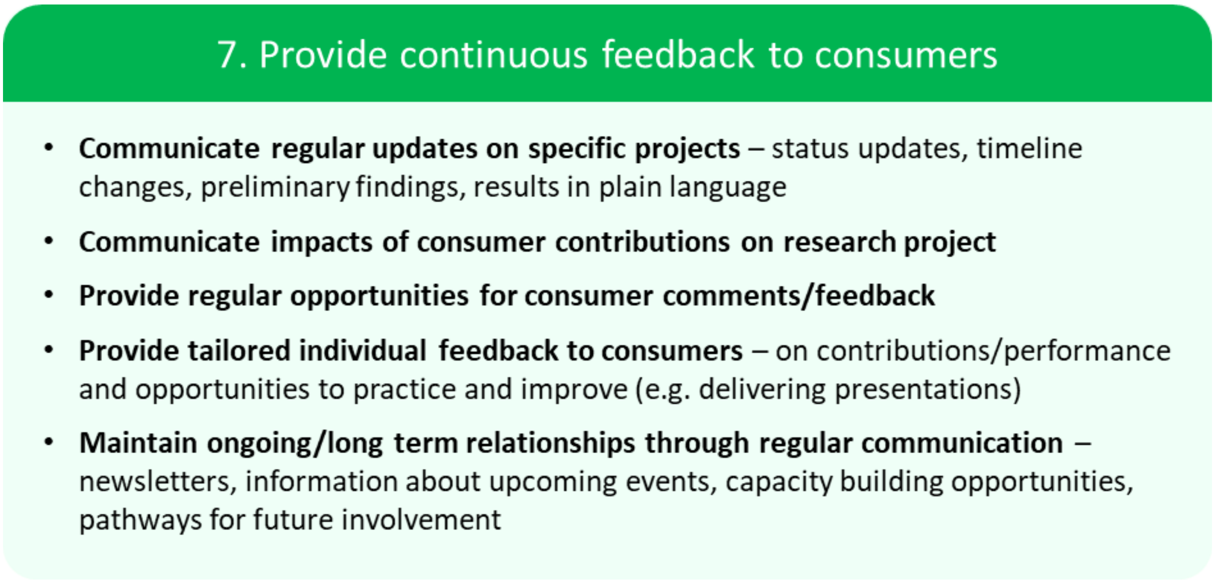
- **Determine who is best placed to achieve the aims and objectives above** – patient vs caregiver, diverse vs specific demographic groups (education, ethnicity),
- **Decide if any specific/special skills or experience are needed** – specific diagnosis/treatment, currently experiencing illness/treatment, Indigenous groups, advocates/representatives, languages spoken, prior experience/connection with research team, special community knowledge
- **Consider broader expertise** – professional background/skills, connections to important stakeholders
- **Avoid convenience sampling** – instead, use purposive sampling, focus on relevant experience, interest
- **Determine the number of consumers you will need** – consider funding, tokenism, accounting for periods of illness
- **Develop strategies to identify and recruit the most appropriate candidates** – consider preferences for the target group, through hospitals/clinics, social media, consumer organisations, posters/flyers, emails, known consumer research partners, clinicians
- **Consider timing of recruiting** – some periods may be too overwhelming for consumers (e.g. diagnosis, dialysis initiation)
- **Discuss motivations with consumers to encourage involvement** – giving back, improving knowledge, developing relationships

5. Develop an overarching approach for involvement

- **Consider how consumers could be involved in each stage** – setting priorities, designing studies, conducting data collection and analysis, disseminating research findings, implementing research findings, evaluating research process/methods
- **Determine which stage/s consumers will be involved to best align with the consumer involvement aims of the project** – single, multiple or all stages
- **Determine what activities consumers will be involved in** – e.g. co-researcher, workshop, survey, reviewing documents
- **Determine the roles consumers will take on** – e.g. partner, advocate, informant
- **Decide the tasks consumers will be responsible for** – e.g. attending meetings, liaising with consumer organisations, recruiting participants
- **Develop a terms of reference clearly outlining roles and responsibilities** – e.g. time required, expected timelines, responsibilities
- **Decide the level of decision-making power consumers will have** – will this be the same as the rest of the research team? Employ mechanisms to support consumers in their roles
- **Ideally involve multiple consumers over the whole project at many points, in different activities/roles** – dependent on aims of consumer involvement and available/allocated resources
- **Design an evaluation plan to assess consumer involvement** – processes, impacts

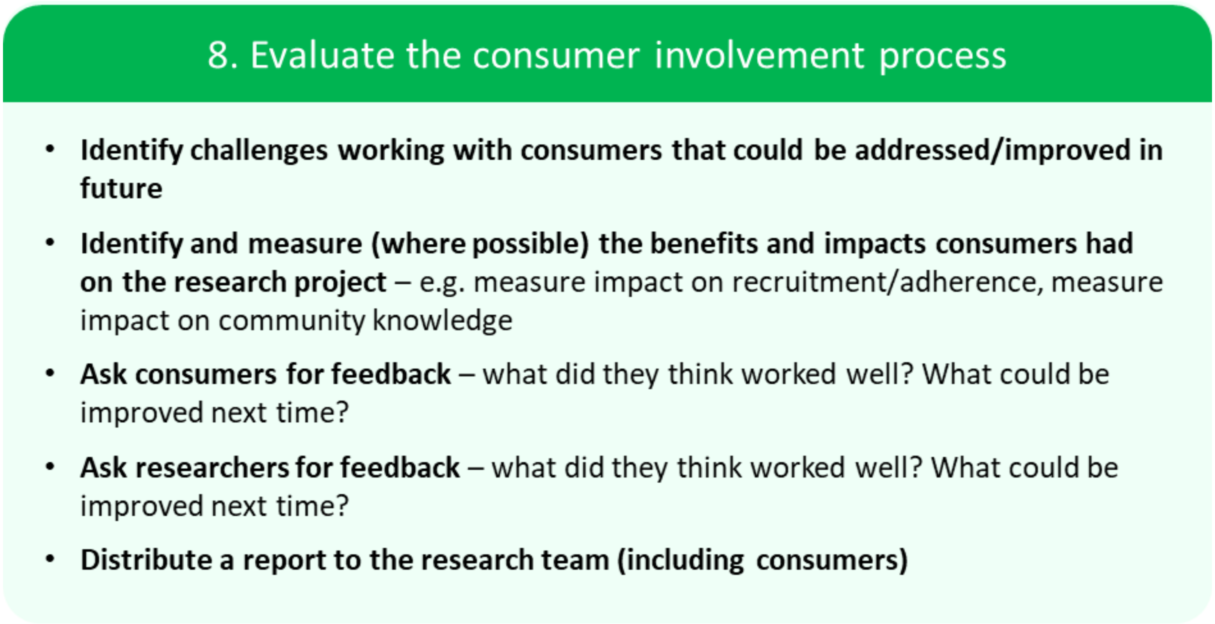
6. Consider principles and strategies to support involvement

- **Respect consumer time** – decrease administrative and logistical burdens, involve in scheduling of meetings, arrange travel/accommodation on their behalf
- **Respect consumer value and expertise** – ensure time/opportunity to be heard, support for involvement from leaders
- **Acknowledge existing power imbalance and employ mechanisms to avoid tokenistic practices** – simple language, inclusivity, induction information packets (glossary/ acronyms, background information, access to resources, contact information,) procedural documents (terms of reference, confidentiality policy, conflict resolution), training for researchers, more than one consumer in any group, develop trusting ongoing relationships, avoid putting consumers on the spot, involve in meaningful activities
- **Account for individual circumstances** – discuss preferences for communication, level of involvement, timelines, payment/reimbursement (e.g. pay expenses in advance)
- **Sensitivity to the patient journey** – discuss special needs with consumers (support in managing illness/impairments/treatment), take care when discussing sensitive topics (e.g. mortality) and have appropriate referrals available.
- **Consider the whole person** – skills and expertise outside of lived experience, commitments outside of research (health, work, family, social)
- **Involve as equals** – same as other team members (e.g. meetings, correspondence, weight on feedback), payment, time allocated, identification/speaker badges (e.g. presentations)
- **Reciprocity** – consider consumer interests/motivations for involvement (sense of purpose, giving back, developing community, professional development)
- **Building capacity** – training and education, long term relationships, regular communication in plain language, consumer peer mentors
- **Diversity** – valuing local/community knowledge, inclusion of vulnerable groups, cultural safety



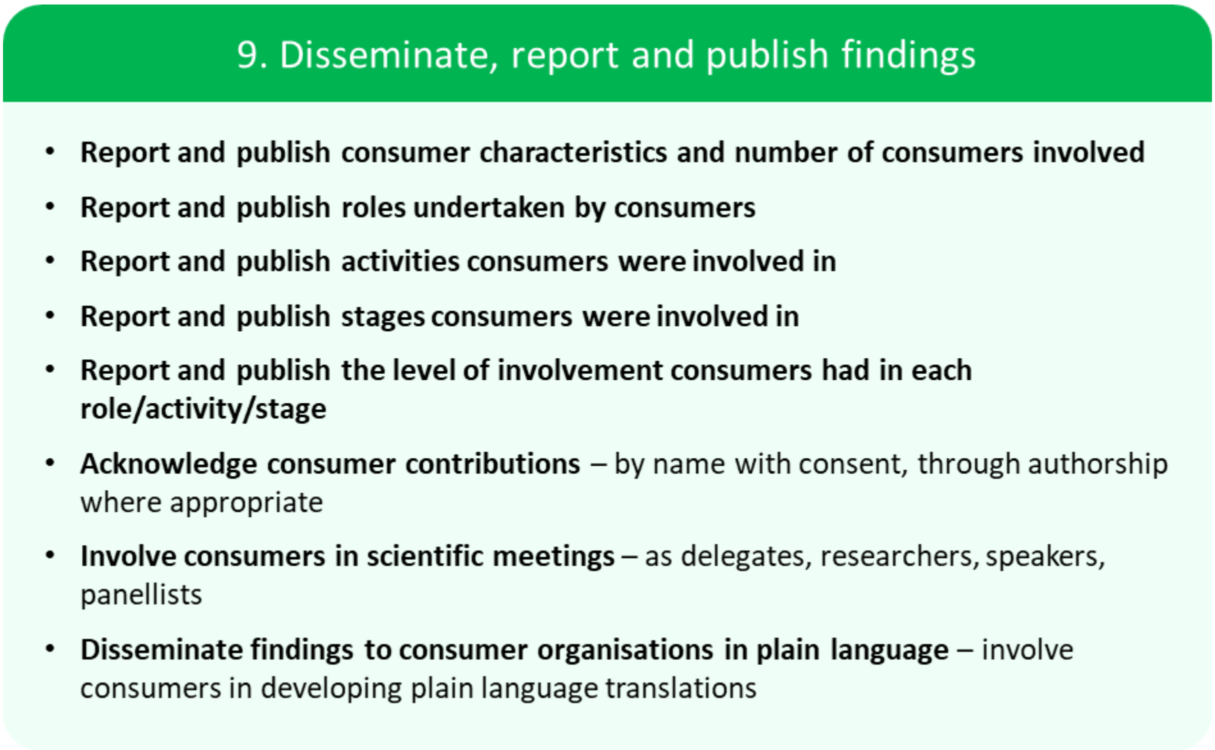
7. Provide continuous feedback to consumers

- **Communicate regular updates on specific projects** – status updates, timeline changes, preliminary findings, results in plain language
- **Communicate impacts of consumer contributions on research project**
- **Provide regular opportunities for consumer comments/feedback**
- **Provide tailored individual feedback to consumers** – on contributions/performance and opportunities to practice and improve (e.g. delivering presentations)
- **Maintain ongoing/long term relationships through regular communication** – newsletters, information about upcoming events, capacity building opportunities, pathways for future involvement



8. Evaluate the consumer involvement process

- **Identify challenges working with consumers that could be addressed/improved in future**
- **Identify and measure (where possible) the benefits and impacts consumers had on the research project** – e.g. measure impact on recruitment/adherence, measure impact on community knowledge
- **Ask consumers for feedback** – what did they think worked well? What could be improved next time?
- **Ask researchers for feedback** – what did they think worked well? What could be improved next time?
- **Distribute a report to the research team (including consumers)**



9. Disseminate, report and publish findings

- **Report and publish consumer characteristics and number of consumers involved**
- **Report and publish roles undertaken by consumers**
- **Report and publish activities consumers were involved in**
- **Report and publish stages consumers were involved in**
- **Report and publish the level of involvement consumers had in each role/activity/stage**
- **Acknowledge consumer contributions** – by name with consent, through authorship where appropriate
- **Involve consumers in scientific meetings** – as delegates, researchers, speakers, panellists
- **Disseminate findings to consumer organisations in plain language** – involve consumers in developing plain language translations

9.2. Consumer involvement in this thesis

As co-investigators and partners in research

During my PhD, I worked closely with Nicole Scholes-Robertson (kidney transplant recipient, previous dialysis patient, and consumer advocate, from regional NSW), who is employed as a research assistant in the research team and is co-chair of the BEAT-CKD Consumer Advisory Board. Nicole has served as a mentor and constant connection to the consumer voice, providing feedback on all aspects of my thesis.

Chapters 2, 4, 5 and 6 all involved consumer research partners in the same role as clinicians and researchers as part of the research team. They contributed to the design of the studies and interview/question guides, to the analysis of qualitative data, and provided comments and feedback on all draft manuscripts. Chapter 2 involved three consumer partners: Nicole

Scholes-Robertson and Chandana Guha (caregiver and parent to a kidney transplant recipient, from Sydney) who are both employed as research assistants with the research team, and David White (kidney transplant recipient, previous dialysis patient, Chair of the Kidney Health Initiative Patient and Family Partnership Council, from then United States). They were involved as co-authors and contributed to the design, analysis and reporting of the findings. Nicole was also involved in the design and screening of the literature search in Chapter 2, and contributed to the study design, analysis and reporting of the findings of Chapters 4 and 6 as part of the research team. All participants in the focus group workshops in Chapter 5, including consumers and health professionals, were included as co-investigators of the paper, contributing their expertise and feedback on the preliminary findings. Three consumers were also involved as co-authors and in the design, analysis and reporting of the study.

Contributing their lived experience (Chapters 4,5,6,7 and 8)

Consumers were involved for their broad expertise and advocacy roles and to provide insights on their experiences being involved in research and were acknowledged for their contributions in the relevant publications (Chapters 4 and 5). In Chapters 6,7 and 8, consumers were involved as experts of their lived experience of specific aspects of conference involvement, guideline development and decision-making and acknowledged as contributors to the research outcomes.

Value and impact

The contributions of the consumers involved in the studies in my thesis strengthened the findings. In addition to contributing to the studies in the same capacity as the other team members, consumers were able to identify themes that were not otherwise identified by researchers and provide novel insights into the reasons underpinning participant perspectives based on their lived experience of a diagnosis or treatment. Having been through health systems and procedures, consumers encouraged discussion among the research team based on their experiences, and highlighted concerns not considered by clinicians/researchers. This provided useful and innovative insights to the implications for research and practice, grounding recommendations in practical applications. Caregivers were able to provide unique information and experiences from their perspective, having navigated the health system alongside the patient without having suffered the symptom or treatment burden of CKD, giving them a unique understanding of the application of research to the clinical setting. They also brought attention to the critical role they play on behalf of their patient as a contributor to their care and outcomes, and how this could be recognised and applied to research findings. Consumers provided suggestions for appropriate and sensitive terminology throughout the study, including in documentation for participants (e.g. interview questions, information, consent and demographic forms), and in the reporting of the findings in the published manuscripts. Finally, they served as a constant connection to the broader consumer community through assisting with or leading the translation of findings and impacts for consumers into plain language formats for dissemination in the consumer community.

9.3 Strengths and limitations

The strengths and limitations of the included studies are reported in detail in the respective chapters. This section will focus on the overall strengths and limitations of the thesis. This thesis used mixed-methods to review and summarise the literature on consumer involvement in research, and qualitative methods to describe consumer and health professional experiences and perspectives on consumer involvement in research and decision-making. All facilitators of focus groups and interviewers were trained and experienced in conducting qualitative research. The four principles of rigour in qualitative research, credibility, dependability, transferability and confirmability, were employed[12]. Credibility was established through investigator triangulation and member checking, whereby multiple investigators were responsible for ensuring the analysis captured the range and depth of the data, and participants were invited to provide feedback and comments on the preliminary findings to ensure the analysis accurately captured their perspectives[13,14]. Credibility was achieved through audio recording and verbatim transcription of all interviews, focus groups and workshops, and the use of qualitative research software for systematic and auditable analysis. Detailed descriptions of context, participants and settings allow for readers to determine the applicability and transferability of the findings to other settings. Use of direct quotations and software to record and track coding ensured findings were derived directly from the data, demonstrating confirmability.

The data from this thesis demonstrate consumer involvement in research on an international scale. The systematic review on consumer involvement placed no restrictions on language, country, date of publication or publication type, ensuring an inclusive approach. Chapter 4 included participants from four countries. Chapter 5 included participants from three cities in Australia. Chapter 6 included participants from six countries. Chapter 8 included participants from six cities in three countries. For interviews, participants were given the choice to conduct them in person (where geographically possible) or online.

However, there are some potential limitations. All qualitative data collection was conducted in English because I did not have the resources required to conduct or translate interviews and focus groups into other languages, therefore all participants had to be English speakers.

Focus groups and workshops were conducted in person at venues determined by the research group. While participants were reimbursed for any costs to attend the meetings, it is possible that perspectives of participants who were unable to travel to attend (e.g. due to sickness, residing in rural/remote areas) were not included in the studies.

9.4 Comparisons with other frameworks

Several toolkits and frameworks have been developed to help the research community better understand how to involve consumers in research in the broader population[7,15-18]. While these frameworks draw from published research, none have conducted systematic reviews of the evidence for their development and there is limited inclusion of recommendations from the consumer perspective.

The PCORI engagement rubric covers theoretical and real-world examples of the potential to involve consumers in the planning, conducting and disseminating of study findings, all of which are echoed in the findings of this thesis[16]. The INVOLVE 'Briefing notes for researchers' provides general reasons for researchers on why to involve consumers (democratic principles, improved quality and relevance, and to satisfy requirements of funding organisations) and on why consumers choose to become involved (to improve quality of care, to have a voice, to give back)[7]. All of these aspects align with the findings from this thesis, however the evidence I have generated takes this further to encourage setting specific aims and objectives for involving consumers. More purposeful involvement of consumers that utilises both lived experience and broader expertise and skills allows for more targeted, meaningful and impactful involvement, and optimal use of resources. This is clearly illustrated in the example of consumer involvement in conferences, whereby goal-directed presentations that align with conference/session learning objectives are more powerful than story sharing alone[5].

The INVOLVE toolkit, the PCORI Engagement Rubric, the NHMRC Statement on Consumer and Community Involvement in Health and Medical Research and the National Framework for Consumer Involvement in Cancer Control support the evidence in this thesis to involve consumers early, provide clarity on roles and responsibilities, and provide access (resources, people, support), payment, training and education to consumers[7,15,16,18]. My thesis develops these aspects further by providing evidence from the consumer perspective on the principles that underpin these recommendations and practical strategies to ensure both consumers and researchers are supported in achieving them, including the need for training for researchers in communication, shared decision-making and consumer involvement methods. It also further defines and establishes consumer roles with concrete examples across the spectrum of decision-making power from attendees to drivers of research (Table 2.2)[3]. These frameworks also recommend to document and record consumer involvement activities. Due to the dearth of published CKD research reporting on consumer involvement, evidence from this thesis emphasises the necessity not only to document and record, but to report, evaluate and disseminate consumer involvement outcomes and impacts to both the research and consumer communities.

The NHMRC Statement on Consumer and Community Involvement in Health and Medical Research provides some practical guidance for researchers on implementing consumer involvement processes, such as developing organisational policies for involvement, building capacity for consumers and researchers through training and mentorship and budgeting for training, honoraria and administrative support, which are also covered in detail in this thesis. While the Can-SOLVE CKD Network toolkit “Engaging Patients in the Research Process”[17] has been developed with the kidney community in mind and poses questions to the researcher about what, who, when and how to involve consumers, it does not provide detailed recommendations for these questions or address specific considerations for consumers with CKD[17]. My thesis and framework (Figure 9.2) provide novel, clear and detailed practical and evidence-based recommendations for researchers about how to involve consumers in CKD research. A focus on diversity over “representation” and the need to include more than one consumer have been addressed in previous frameworks however this thesis highlights specific considerations for selecting and recruiting patients with CKD and their families. The lifelong nature and constantly shifting trajectory of CKD means researchers need to account for individual circumstances, periods of illness and manage consumers’ competing personal, professional and health-related priorities to avoid overwhelming burdens at difficult times in the patient journey.

This thesis adds a further two key elements to previous frameworks and toolkits – the involvement in clinical care and decision-making as a key element to foster future involvement in research, and the need for infrastructure to support ongoing relationships between the research and consumer communities outside of individual research projects. Involvement in clinical decision-making increased consumer knowledge of both their illness and research, and catalysed a shift in power dynamics leading to greater potential for consumers to become involved in research. National advisory boards and online platforms to connect consumers with researchers or research projects would combat isolation, build trust with the research community, and allow for broader inclusion of consumer partners.

9.5 Broader impacts of the studies in this thesis

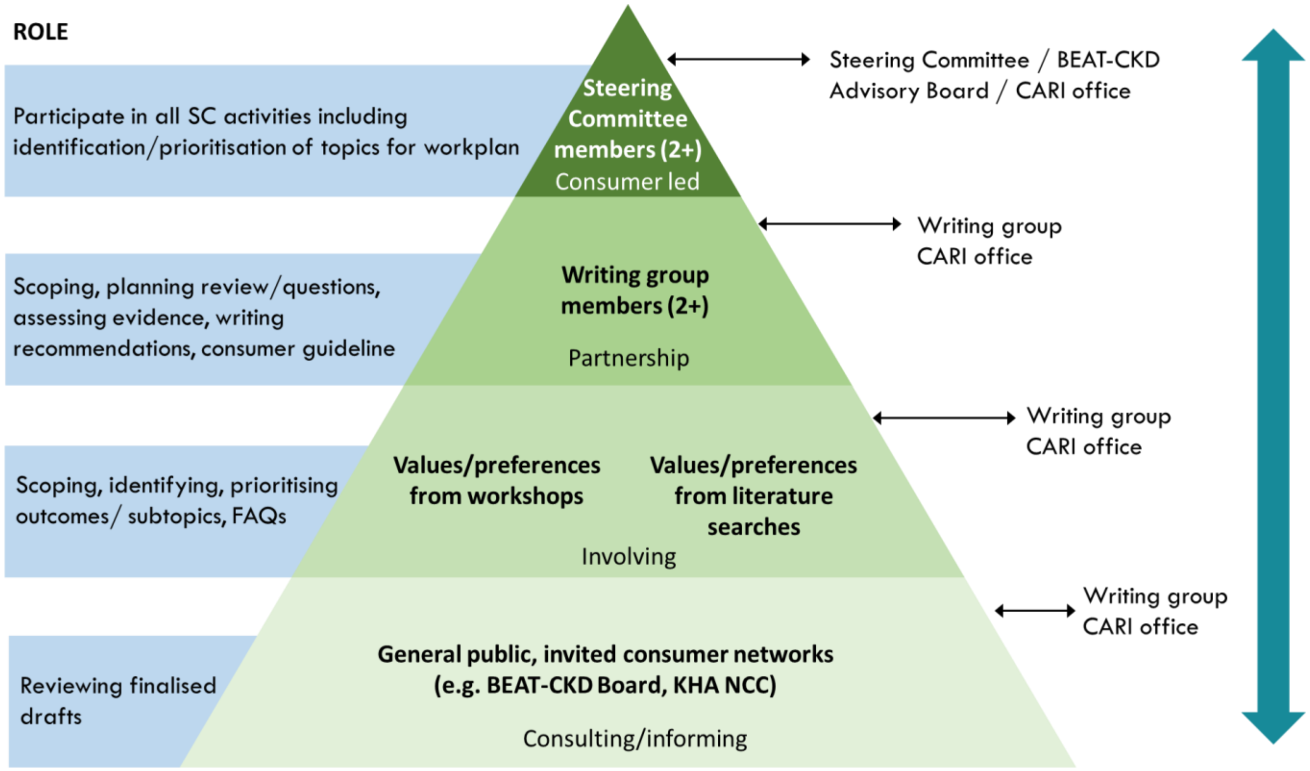
The findings of the studies in this thesis have resulted in broad application and implementation of consumer involvement strategies and practices beyond the scope of the included papers.

Following the focus group workshops (Chapter 5), the BEAT-CKD program established a national Consumer Advisory Board consisting of over 40 members around Australia[19]. This consumer advisory board provides feedback and guidance at a high level on the importance and relevance of CKD research in Australia. On a granular level, members of the advisory board are now involved as co-researchers in every new research project undertaken by AKTN, ANZDATA, Cochrane Kidney and Transplant, and CARI through a process of self-nomination based on relevant skills and interests. BEAT-CKD has also established a YouTube channel posting educational and training videos developed for consumers[20].

Furthermore, and following the findings from Chapter 6, consumers have been involved in the recent Annual Scientific Meetings of Australian and New Zealand Society of Nephrology[21] and the BEAT-CKD annual research meetings. The BEAT-CKD Consumer Advisory Board have designed and delivered consumer focused sessions with both consumers and health professionals, including determining the content of sessions, the mode of attendance (online and face-to-face) and the timing of sessions (length, scheduling) (Appendix A).

Following the findings from Chapters 5 and 7, the CARI guidelines have redeveloped their consumer involvement strategy to include consumers at every stage and level of the guideline development process (Figure 9.3). As well as eliciting consumer preferences for guideline content and developing a consumer version of the guidelines, consumers are now involved as equal partners on the steering committee and writing groups, and lead the development of the consumer versions, supported by research staff.

Figure 9.3 Consumer involvement in CARI guideline development



While the SONG Initiative[22] has always had a significant consumer focus, with the aim of bringing together the shared priorities of all stakeholders, including consumers, more recently and following the findings from Chapter 5, consumers are now involved as members of the executive and steering committees and also as members of the research team.

In conjunction with patient-partner Nicole Scholes-Robertson and based on the findings from Chapters 2, 4, 5, 6, 7 and 8, we have developed a full day training workshop for researchers, delivered through the Patient-Centred Research (PACER) Network[23], which aims to facilitate the development of feasible and evidence-based consumer involvement strategies for all current and future research projects.

9.6 Future research

The findings from this thesis highlight the lack of practical and infrastructural support available to both consumers and researchers for ongoing and meaningful involvement, as well as the dearth of published reports describing and evaluating consumer involvement processes and practice. As part of my postdoctoral research, I would like to validate this framework with multiple stakeholders, develop an automated reporting tool to encourage effortless publication of consumer involvement, conduct training for both researchers/health professionals and consumers and establish a national database for consumer involvement.

Validating and implementing the framework

I would like to conduct a series of international consensus workshops with multiple stakeholders including consumers, researchers, policymakers, funders and journals, as has been done previously through the SONG Initiative and the BEAT-CKD program[24-26]. Parallel to these workshops I would like to conduct a virtual consultation with kidney consumer organisations including Kidney Health Australia, The American Association of Kidney Patients, Kidney Care UK, European Kidney Patients' Federation and the Kidney Foundation of Canada. Once validated, this framework could be actively implemented in future research projects, particularly those under the BEAT-CKD umbrella where significant progress in this area has already occurred. Additionally, the framework presented in this thesis could be converted into an evaluation tool and reporting checklist for CKD researchers.

Developing an automated reporting tool

It remains a challenge to continue to develop the evidence of and best practice for consumer involvement in research while reporting and publishing remains scant. The GRIPP/GRIPP2[27,28] checklists have been developed to improve reporting of consumer involvement in research, however this thesis demonstrated that few studies actually used this guidance in their reporting[3]. While in some instances, where consumer involvement is the focus of the publication, these forms are useful to provide detailed and comprehensive descriptions of consumer involvement, and researchers should be incentivised to complete this reporting, in other circumstances (e.g. reporting a trial), they may be too burdensome for researchers and challenging to fit within with journal requirements (e.g. word limits). As a possible solution for these scenarios I would like to develop an automatically generated figure that provides a comprehensive summary of the types and number of consumers involved, and at what level and stages. Researchers could input these datapoints online through a specific website or directly through the journal submission process and a visual plot could provide an instant illustration for readers about how consumers were involved in the study.

Designing and delivering training for researchers and consumers

Through my role at the University of Sydney and in partnership with consumer co-researcher Nicole Scholes-Robertson, we have developed a training workshop for researchers and health professionals on how to conduct consumer involvement in research. Based on new evidence generated in this thesis I would like to update this workshop and run it regularly to disseminate these findings in a practical way to the research and medical communities. I believe there is scope for this workshop to be converted into a formal unit of study for the University for all public health and medicine students.

Through the BEAT-CKD program and Kidney Health Australia I would like to develop online and face-to-face training modules aimed at consumers to educate them on aspects of kidney disease, current research projects, and research methods.

Designing and developing a national "Involvement Database"

Through established networks of the BEAT-CKD research program, Kidney Health Australia and the Australian and New Zealand Society of Nephrology I would like to establish a national involvement database. Similar to those already established in other health disciplines and countries (e.g. Register 4 research for cancer, Research 4 Me)[29-31] and as requested by the consumer community, this database would connect consumers and researchers based on skills and interests. Consumers could enter their availability, interests, skills and experience and provide a description of how they would like to be involved in research. Researchers could post opportunities to research projects where they are looking for consumer involvement. This database would allow candidates to search for relevant opportunities and even automatically match consumers and researchers based on predetermined criteria. The website of this database could also serve as a valuable resource to engage the consumer community through hosting an information repository for consumers including lay summaries of current research and research methods and provide access to the training modules discussed above.

9.7 Implications for research, policy and practice

The findings of this thesis, and in particular the development of an evidence-based framework with detailed practical recommendations for researchers, will support the research community in the systematic and consistent involvement of consumers in all types of research.

Clinical practice

As shown in Chapters 2, 3 and 8, involvement in clinical decision-making has the potential to empower consumers in health ownership and develop confidence to participate as equals with health professionals. Also, an opportunity to increase the pool of potential consumers to be involved in research by aiming to engage the whole patient population exists. While shared decision-making is recommended by professional organisations including the Renal Physicians Association and the American Society of Nephrology, more emphasis is needed in the training and education of health professionals to support this[9,32]. Additionally, structural changes to reduce time pressures faced by clinicians are necessary to allow for information exchange and relationship building to occur in appointments to enable further involvement.

Research and funding organisations

Consumer involvement in research is widely advocated by funding organisations, however in many cases researchers are uncertain on how to effectively implement it. While this framework summarises the evidence for successful consumer involvement in CKD research, including from the perspective of consumers, it also serves as a blueprint for researchers, providing concrete evidence-based and practical approaches for the planning, conducting, evaluation, and reporting of consumer involvement. It identifies issues specific to CKD consumers for researchers to consider and can be incorporated at any stage of the research project. For funding bodies, it could assist with the development of budgeting tools for consumer involvement to include in grant applications and the necessary technical changes to accommodate consumer chief investigators more easily.

Consumer community and organisations

The collective findings of this thesis serve to demonstrate the significant value and impact consumer contributions have on research. Implementation of the framework will encourage recognition of the broad capabilities that consumers have, and their vast untapped potential to partner with researchers. It will also deepen understanding of individual consumer circumstances and needs and encourage the allocation of more resources to consumer involvement and implementation for formal mechanisms and policies, ensuring consumers are adequately supported and compensated for their contributions.

9.8 Conclusions

In conclusion, the evidence generated by the studies in this thesis informed the development of a practical and feasible framework for the involvement of consumers in research, specific to CKD. These studies highlight the need for more comprehensive and purposeful involvement of consumers in CKD research, as well as the need for more complete and systematic reporting and publishing of consumer involvement processes and practices so that research and consumer communities can better understand and evaluate involvement in published studies and improve it in future. The framework provides a structure for researchers to be able to do this through implementing the evidence-based recommendations underpinned by consumer perspectives. Through my connections with global and national kidney organisations and BEAT-CKD I hope to be able to implement this framework in all CKD research projects in Australia and beyond, develop training tools for consumers and researchers and develop infrastructure to establish a community of consumers and researchers dedicated to consumer involvement in research.

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Appendix A: ‘Knowledge is power’ – A framework for partnering with consumers in developing and delivering a scientific meeting in nephrology

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This appendix is structured as per the journal article.

Abstract

Involving consumers (patients, carers and family members) across all stages of research is gaining momentum in the nephrology community. Scientific meetings present a partnership opportunity with consumers for dissemination of research findings. The Better Evidence and Translation in Chronic Kidney Disease (BEAT-CKD) research collaboration, in partnership with Kidney Health Australia, convened two consumer sessions at the 54th Australian and New Zealand Society of Nephrology Annual Scientific Meeting held in September 2018. The educational objectives, topics and session formats were informed by members of the Better Evidence and Translation-Chronic Kidney Disease Consumer Advisory Board (which at the time comprised 36 consumers from around Australia with varied experience of kidney disease). Patients, health professionals and researchers facilitated and presented at the sessions. In-person and live-streaming attendance options were available, with over 400 total participants across the two sessions. Sessions were also video recorded for dissemination and later viewing. Evaluations demonstrated consumers found the presentations informative, relevant and accessible. Attendees indicated strong interest in participating in similar sessions at future scientific meetings. We propose a framework for partnering with consumers as organisers, facilitators, speakers and attendees at scientific meetings in nephrology.

Introduction

There is a growing movement in nephrology, and other medical specialities, towards meaningful consumer engagement in research. Involving consumers (patients, carers and family members) throughout all stages of the research process can help to align research with consumer priorities, increase participant recruitment and retention, and facilitates dissemination of results, which ultimately enhances the impact of research on health practice and policy.[1–6] Many scientific meetings are convened each year to disseminate current evidence-based practice and new research about kidney disease. As the individuals who participate in research as subjects and for whose ultimate benefit it is conducted, patients and caregivers are being increasingly involved in these events. The 17th Congress of the International Society for Peritoneal Dialysis, held in Vancouver, had a theme of 'Patient-Centred Care' and included a pre-conference day of consumer workshops, consumer speakers, and patient lounges with dialysis facilities, and the entire conference was open to consumers free of charge. A consumer focussed session was included in the 2018 American Society of Nephrology Kidney Week in San Diego, where three patient speakers presented research they had been involved in as investigators. At the 54th Australian and New Zealand Society of Nephrology (ANZSN) Annual Scientific Meeting (ASM) held in Sydney, Australia in September 2018, we convened two research sessions for patients, carers and family members affected by kidney disease, with the sessions open to the public and all conference delegates. Here, we describe the process of partnering with consumers for this event as organisers, facilitators, speakers and attendees, discuss the outcomes and feedback from those who attended, and reflect on lessons learned.

Background and context

The Better Evidence and Translation - Chronic Kidney Disease (BEAT-CKD) research collaboration[7] links four national research and translational platforms; Australia and New Zealand Dialysis and Transplant Registry,[8] Australasian Kidney Trials Network,[9] Kidney Health Australia-Caring for Australasians with Renal Impairment guidelines,[10] and Cochrane Kidney and Transplant.[11] In 2017, BEAT-CKD and Kidney Health Australia (KHA), the peak Australian consumer organisation for kidney disease,[12] held workshops in Sydney, Adelaide and Brisbane with consumers (n = 105) and researchers/health professionals (n = 43) to identify principles for effective consumer engagement in kidney disease research (Gutman et al., 2018, unpublished data). The workshops led to the establishment of the BEAT-CKD Consumer Advisory Board in early 2018, which currently consists of 40 consumers from across Australia ranging from 28 to 76 years of age. The group includes people with chronic kidney disease, people receiving peritoneal dialysis, haemodialysis, carers/family members of dialysis patients, and transplant recipients. Members live in various states of Australia, in both urban and rural areas. The primary objective of this Board is to engage consumers in planning, conducting, disseminating and implementing research across the four BEAT-CKD groups. Themes identified from the 2017 workshops (Gutman et al., 2018, unpublished data) informed strategies for effective consumer engagement in the activities we describe here.

Two consumer research sessions were co-convened by BEAT-CKD, KHA and ANZSN at the 54th ANZSN ASM; a 3 h ‘Research Update for Consumers’ on a Sunday (prior to the main meeting, to accommodate consumer preferences for weekend events) and a 90 min ‘Patient Workshop’ on a Wednesday afternoon (during the main meeting). The ‘Patient Workshop’ followed a closed plenary session in the main meeting titled ‘Patient Perspectives’, which included a consumer speaker [Author N. S-R.].

Organisation of sessions: consumers as conference co-designers

In early 2018, an open and widely advertised application process called for expressions of interest for the BEAT-CKD Consumer Advisory Board. All applicants (n = 37) were invited to attend one of three board meetings in June 2018 held across multiple sites in Australia (including video and teleconference options) and at various days and times (including out of office hours), enabling flexible participation and ease of involvement. The meetings were chaired by the BEAT-CKD investigators and attended by 32 consumers who were provided information through phone call and e- mailed a 'Welcome' document with information about BEAT-CKD and a 'Welcome video' prior. Consumers who attended the meeting in person were provided refreshments and reimbursement for parking fees. The priority action was to obtain consumers' preferences for the aims, topics and formats of the two ANZSN ASM sessions. Consistent with the 2017 workshops, the BEAT-CKD Consumer Advisory Board expressed their desire for knowledge exchange and trustworthy information about trends and recent findings arising from research. The ASM sessions therefore aimed to provide information and 'updates' on current kidney disease research to patients, carers and the public, on topics of importance to consumers in the format of their preference. Board members completed a follow-up survey after the meeting to give them time to reflect and respond, to gauge their interest in attending the sessions (in person or electronically) and to elicit their preferences among 36 research topics collated from the ASM main program and consumers' suggestions. Potential speakers with relevant expertise in the most popular topics were then contacted through the BEAT-CKD investigators' networks, explained the purpose of the initiative, and invited to present. Two consumers (authors A.W. and N. S-R.) were invited to co-facilitate the sessions with a nephrologist and researcher [author S.M.] and deliver a presentation. They were provided information and support prior to and during the event, including feedback on presentations and notes for facilitating the sessions.

Advertising of the sessions, including a registration link, occurred through Twitter, Facebook and e-mails through KHA and BEAT-CKD consumer networks 3 weeks before the meeting.

Registrants were e-mailed the final programs and live-streaming details approximately 1 week prior. All presentations were video recorded. The final session programs can be found on the ANZSN ASM website[13] and reflect consumers' preference for multiple short presentations. Time was allocated for questions and discussion after each in accordance with the Board's suggestion. Free live-streaming of the sessions was available through the BEAT-CKD website (sponsored by Baxter Healthcare via an unrestricted grant to KHA). Remote participation occurred through moderated e-mail.

Attendance

Approximately 25 people attended the 'Research Update for Consumers' session (Sunday) in person, with 240 joining through live-stream. Approximately 15 people attended the 'Patient Workshop' (Wednesday) in person, with an additional 143 through live-stream. Live-streaming data are presented in Fig. 1. While the entire conference was not open to consumers, there was no charge to attend or access these sessions.

Reception and feedback

All attendees were encouraged to complete a paper-based or online evaluation survey. Overall, the formal and informal feedback received was strongly positive and the sessions were rated as valuable and interesting to consumers, and increased their understanding of available evidence. Almost all who completed an evaluation survey agreed the presentation topics were relevant, the lengths appropriate, and the language easy to understand. The presentations on diet and self-advocacy that addressed topics of lifestyle and self-management, continuously prioritised by patients,[14] were identified as most useful. Almost all respondents indicated interest in attending similar events at the next ANZSN ASM, and comments demonstrated appreciation from the community – ‘thanks for including consumers in this way’. Feedback included suggestions to ensure future speakers keep to allocated times.

Reflections

Individuals and families affected by kidney disease have a strong desire to learn about contemporary research that impacts their health. Conferences represent an opportunity for meaningful partnership with consumers to deliver this information and we have identified strategies to ensure such engagement is achievable and worthwhile (Box 1). These closely align with the principles of patient involvement in conferences (accommodation, codesign, engagement, education and mentorship) as recommended in the Stanford Framework for Patient Partnership developed by Chu et al.[15] and suggestions made by consumers.[16]

Box 1 Key strategies for partnering with consumers in scientific meetings in nephrology

- Co-design the aim, themes and format of sessions with consumers
- Convene sessions on days and at times that facilitate attendance
- Offer electronic participation through free live-streaming
- Video record presentations to enable continued dissemination of information
- Provide notice of event details and consumer roles as early as possible
- Use social media prior to, during and after event
- Evaluate various stakeholder opinions of event out- comes and impacts, and feedback to consumer partners
- Early budget planning to reimburse expenses to consumers

Involving consumers in the planning of events as co- designers ensures that presentation topics and session formats reflect their interests and preferences. Consumers with facilitator or speaking roles should be given briefing and offered support well in advance, including from other consumers with relevant experience. Equally important are event logistics, which can be barriers or facilitators to attendance. The greater number of attendees at the Sunday session (both in person and through live-stream) indicate that consumer-directed events should occur on days and at times that enable attendance. Multiple participation methods need to be offered. The overwhelming attendance through the free live-streaming and evaluation responses highlight the technical ease and usability of this as an adjunct to face-to-face sessions. However, we cannot assume that all consumers will be highly engaged with or have access to the technological facilities required for this. Convening ‘meet-up’ events at various satellite locations with live-streaming facilities is one idea that may support involvement of more consumers in these initiatives. These considerations are particularly pertinent for people dependent on dialysis, the elderly and those in rural and remote areas. Video recorded presentations are an investment that also enable continued dissemination of research information and can be brought to consumers, for example, through screens in clinic waiting rooms or dialysis units or online.

Event advertising should occur as early as possible, allowing consumers time to arrange travel, finances and treatment and support if required. Social media is an efficient mechanism for this and enables sharing of related content before, during and after events. Consumers identified clinical encounters as another useful means to inform them of research opportunities through flyers and posters around clinics as well as and direct communication from health professionals. Directly engaging renal units and consumer organisations in advertising these events is one area for development, to support an increased reach to consumers. Finally, keeping consumers ‘in the loop’ about how their contributions have impacted research activity is key to fostering meaningful and sustained engagement (Gutman et al., 2018, unpublished data). The outcomes and follow-on actions from initiatives like ours need to be fed back to consumers in a timely manner, to bring the activity full circle.

Next steps

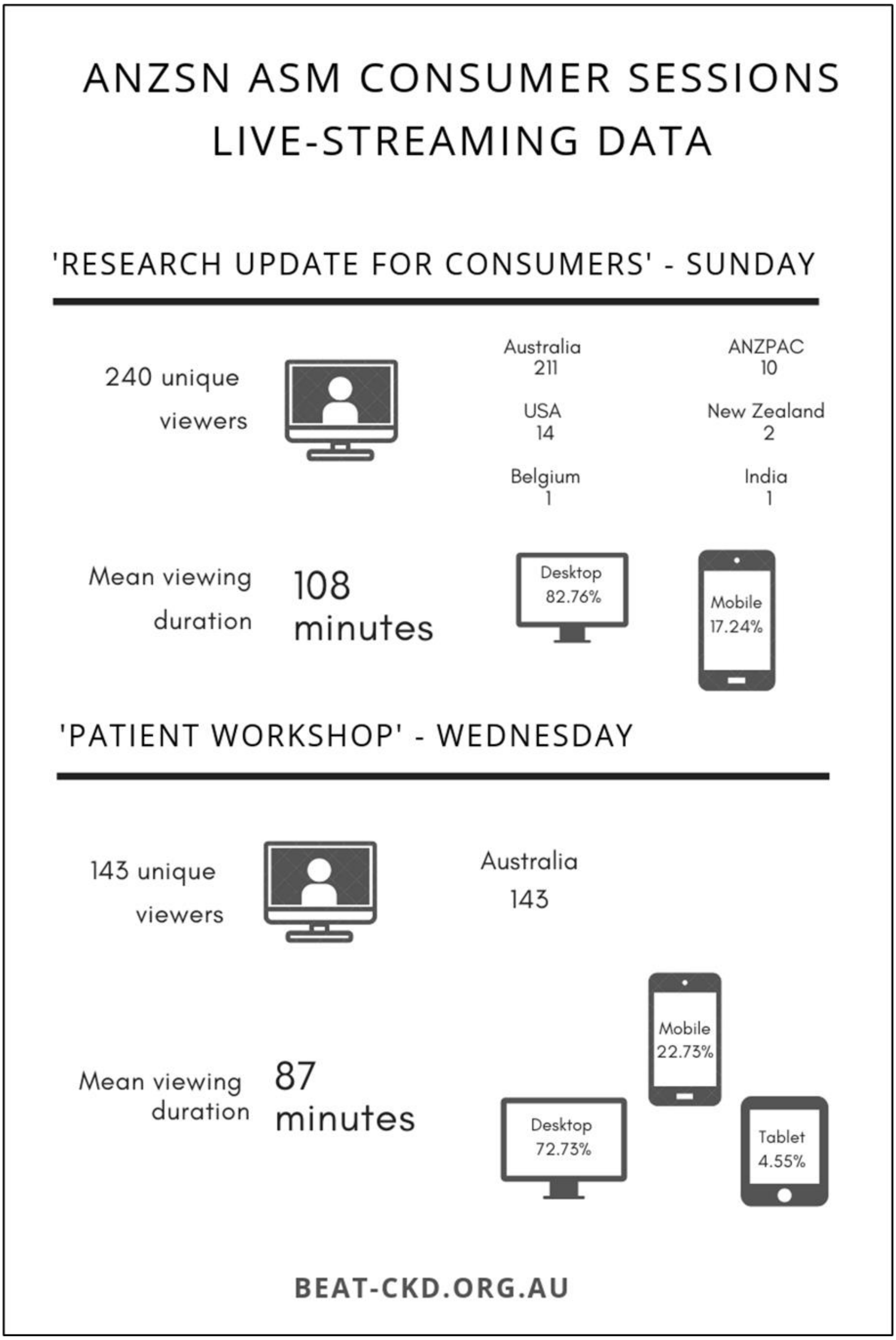
Following the ANZSN ASM, a BEAT-CKD YouTube channel was created and now hosts video recordings of the session presentations which are publicly available.[17] A summary report of this initiative was provided to the BEAT-CKD Consumer Advisory Board and reviewed at their next meeting to gauge interest in convening similar sessions at upcoming conferences. As requested, attendees were also provided copies of the presentation slides soon after the meeting (with speakers’ permission). A summary timeline of the BEAT-CKD consumer engagement activities is presented in Fig. 2.

There are advantages to incorporating consumer sessions into major meetings including taking advantage of speakers’ availability and shared advertising and publicity. Ideally, scientific meetings would include both consumer-intended and academic-level presentations to accommodate a variety of audiences, all of which would be accessible to consumers. Travel and accommodation costs and arranging dialysis therapy make in-person attendance ‘off-putting’ for some. Early consideration is needed to waive conference fees for consumers and arrange dialysis availability. Consistent with the expression, ‘Nothing about us without us’ adopted by disability rights activists[15,18] and other consumer advocacy groups, is a comment from a member of the BEAT-CKD Consumer Advisory Board – ‘I think consumers should have unlimited access to these information sessions, as they are why these meetings come about'.

In conclusion, there is a clear demand amongst consumers for trustworthy information on current nephrological research, spanning topics of prevention, treatment and psychosocial issues from both the academic and patient perspective. The timing and web-availability of the events that communicate this information are critical. True patient partnership will be achieved when consumers are involved in these initiatives as delegates with equal participation rights, and multiple approaches are included to reach a diversity of and as many consumers as possible. We hope this strongly encourages the nephrology community to include consumer-directed sessions at future scientific meetings, in order to contribute to the broader shift towards meaningful consumer engagement in kidney disease research.

Acknowledgements

We would like to acknowledge all members of the BEAT-CKD Consumer Advisory Board who contributed their time, energy and ideas as part of this initiative, and thank them for their ongoing partnership.



ANZSN ASM CONSUMER SESSIONS
LIVE-STREAMING DATA

'RESEARCH UPDATE FOR CONSUMERS' - SUNDAY

240 unique viewers

Australia 211	ANZPAC 10
USA 14	New Zealand 2
Belgium 1	India 1

Mean viewing duration 108 minutes

Desktop 82.76%

Mobile 17.24%

'PATIENT WORKSHOP' - WEDNESDAY

143 unique viewers

Australia 143

Mean viewing duration 87 minutes

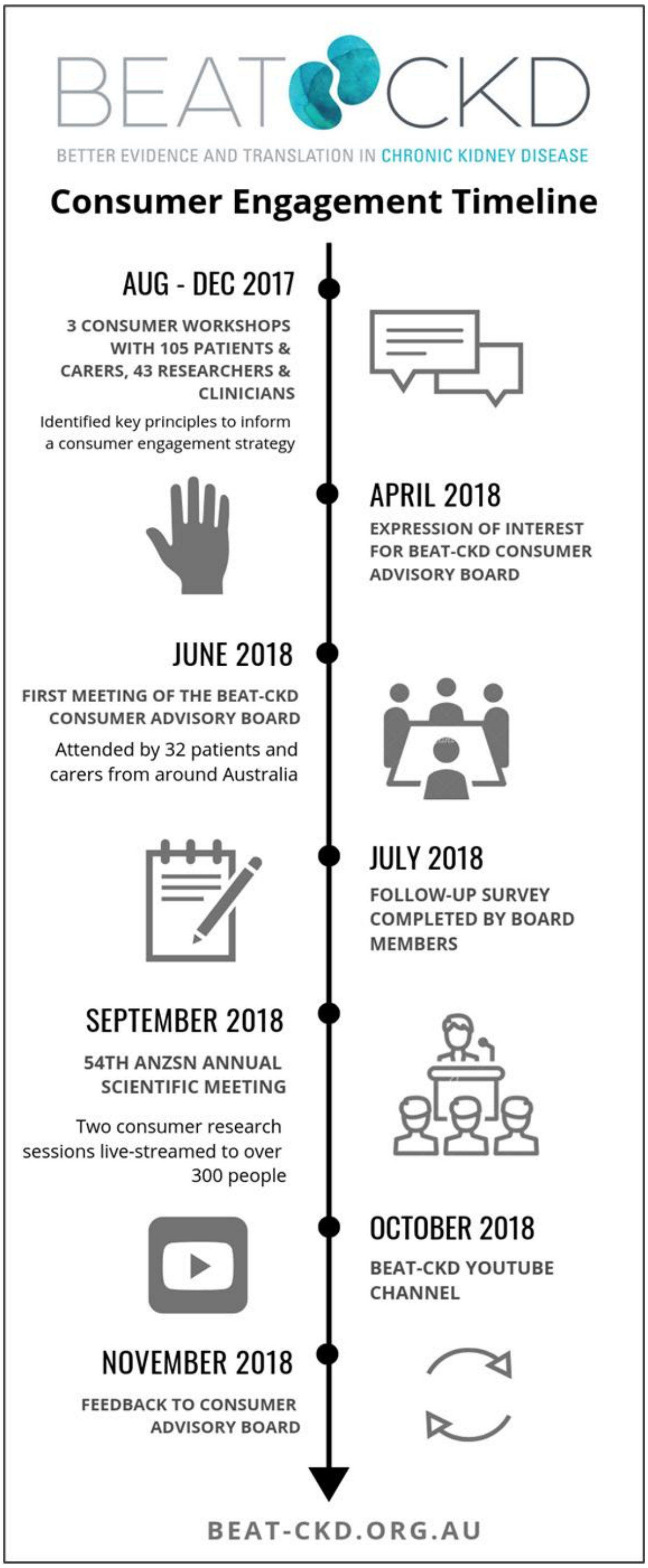
Mobile 22.73%

Desktop 72.73%

Tablet 4.55%

BEAT-CKD.ORG.AU

Fig. 1 Live-streaming data from 54th Australian and New Zealand Society of Nephrology Annual Scientific Meeting consumer research sessions.



BEAT CKD

BETTER EVIDENCE AND TRANSLATION IN CHRONIC KIDNEY DISEASE

Consumer Engagement Timeline

AUG - DEC 2017

3 CONSUMER WORKSHOPS WITH 105 PATIENTS & CARERS, 43 RESEARCHERS & CLINICIANS

Identified key principles to inform a consumer engagement strategy

APRIL 2018

EXPRESSION OF INTEREST FOR BEAT-CKD CONSUMER ADVISORY BOARD

JUNE 2018

FIRST MEETING OF THE BEAT-CKD CONSUMER ADVISORY BOARD

Attended by 32 patients and carers from around Australia

JULY 2018

FOLLOW-UP SURVEY COMPLETED BY BOARD MEMBERS

SEPTEMBER 2018

54TH ANZSN ANNUAL SCIENTIFIC MEETING

Two consumer research sessions live-streamed to over 300 people

OCTOBER 2018

BEAT-CKD YOUTUBE CHANNEL

NOVEMBER 2018

FEEDBACK TO CONSUMER ADVISORY BOARD

BEAT-CKD.ORG.AU

Fig. 2 Timeline of Better Evidence and Translation-Chronic Kidney Disease Consumer Engagement Activities.

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Appendix B: Supporting documentation for Chapter 2

B.1 Search strategy

Search terms for consumer involvement were derived from the 2016 Roger's Search Filter for Patient and Public Involvement[17]

EMBASE 1947 to 30th October 2019: n=836

#	Searches	Results
1	exp Kidney Failure, Chronic/	89653
2	Chronic kidney disease.mp or exp Renal Insufficiency, chronic/	153069
3	exp Peritoneal Dialysis/ or exp Dialysis/ or exp Renal Dialysis/ or exp Peritoneal Dialysis, Continuous Ambulatory/ or dialysis.mp	235634
4	hemodialysis/	106302
5	exp Nephrology/	17402
6	exp Kidney Transplantation or kidney transplant.mp	154559
7	**1 or 2 or 3 or 4 or 5 or 6**	**470702**
8	consumer participation/	49169
9	Patient participation/	26238
10	8 or 9	74980
11	(patient* or public or lay or people or consumer* or user* or citizen or parent or parents or child*).ti	3710480
12	(paticipat* or involve* or engag* or consult* or collaborat* or conducting or conducted or contrib*).ti	477782
13	(questionnaire* or interview* or focus group* or workshop*or peer led or research* or self-report* or qualitative or patient led or public led or self rating or self rated or development).ti	898107
14	11 and 12 and 13	4991
15	((health or research) and (partners or partnership)).ti,ab	54182
16	**10 or 14 or 15**	**131447**
17	**7 and 16**	**836**

Ovid MEDLINE(R) 1946 to 30th October 2019: n=538

#	Searches	Results
1	exp Kidney Failure, Chronic/	91453
2	Chronic kidney disease.mp or exp Renal Insufficiency, chronic/	134711
3	exp Peritoneal Dialysis/ or exp Dialysis/ or exp Renal Dialysis/ or exp Peritoneal Dialysis, Continuous Ambulatory/ or dialysis.mp	175697
4	hemodialysis/	88937
5	exp Kidney Transplantation/ or kidney transplantation.mp	96223
6	**1 or 2 or 3 or 4 or 5**	**313313**
7	consumer participation/	16687
8	Patient participation/	24682
9	7 or 8	41022
10	(patient* or public or lay or people or consumer* or user* or citizen or parent or parents or child*).ti	2719442
11	(paticipat* or involve* or engag* or consult* or collaborat* or conducting or conducted or contrib*).ti	416634
12	(questionnaire* or interview* or focus group* or workshop*or peer led or research* or self-report* or qualitative or patient led or public led or self rating or self rated or development).ti	767582
13	10 and 11 and 12	4122
14	((health or research) and (partners or partnership)).ti,ab	39429
15	**9 or 13 or 14**	**82148**
16	**6 and 15**	**542**

PsychINFO 1806 to 30th October 2019: n=12

#	Searches	Results
1	exp Dialysis/ or exp Kidney Diseases or exp Hemodialysis/ or Kidney Failure.mp	3351
2	Chronic kidney disease.mp. or exp hemodialysis/	1883
3	Renal failure.mp.	1050
4	Nephrology.mp.	246
5	Kidney transplant.mp. or exp Organ Transplantation/	4623
6	**1 or 2 or 3 or 4 or 5**	**8553**
7	consumer participation/	0
8	Patient participation/	2040
9	(patient* or public or lay or people or consumer* or user* or citizen or parent or parents or child*).ti	617211
10	(paticipat* or involve* or engag* or consult* or collaborat* or conducting or conducted or contrib*).ti	122018
11	(questionnaire* or interview* or focus group* or workshop*or peer led or research* or self-report* or qualitative or patient led or public led or self rating or self rated or development).ti	284498
12	9 and 10 and 11	2553
13	((health or research) and (partners or partnership)).ti,ab	27174
14	**8 or 12 or 13**	**31414**
15	**6 and 14**	**12**

Grey Literature Search

<u>Journals</u>

1. Research Involvement and Engagement (n=15 screened, n=2 unique included)
2. The Patient (n=82 screened, n=0 unique included)
3. Health Expectations (n=5 screened, n=0 unique included)

<u>Website search</u>

1. INVOLVE - https://www.invo.org.uk/ (n=6 screened, n=0 unique included)
2. International Society of Nephrology - https://www.theisn.org/ (n=1 screened, n=1 unique included)
3. Kidney Diseases Improving Global Outcomes - https://kdigo.org/ (n=1 screened, n=0 unique included)
4. Can-SOLVE CKD Network - https://www.cansolveckd.ca/ (n=1 screened, n=0 unique included)
5. American Society of Nephrology - https://www.asn-online.org/ (n=0 screened, n=0 unique included)
6. Kidney Research UK - https://kidneyresearchuk.org/ (n=3 screened, n=0 unique included)
7. Kidney Health Australia - https://kidney.org.au/ (n=6 screened, n=3 unique included)

Citing GRIPP/GRIPP2

n=6 screened

n=4 unique included

B.2 Detailed characteristics of included articles

Author	Type of article/study	Activity	N (%)*	Country	Concept
Archdeacon (2013)	Special feature; Project protocol	Board of directors (voting) Patient advocacy groups	2 (10) ^	US	KHI, improve safety, foster innovation
Barnieh (2014)	JLA PSP	Steering Committee Online survey Workshop	5 (45%) ^ 16 (53%)	CA	Research priorities for patients on or nearing dialysis
Bernstein (2019)	Narrative review/summary	NA	NA	CA, US, UK	Patient involvement in nephrology nursing research
Bishop (2017)	Trial Protocol, feasibility study	Patient Involvement group with team lead	^	UK	Acceptability of exercise for transplant recipients
Blackburn (2018)	Mixed methods study (surveys, document analysis, workshop)	5/200 projects reported involvement with renal-urogenital patients	NA	UK	Extent, quality and impact of patient and public involvement in primary care research
Blomqvist (2010)	Participatory Action Research; focus groups	Co-researching group	4 (100)	SE	Supporting daily life of patients with CKD (in clinic and outside)
Bonventre (2019)	Feature; Project protocol	Patient Advisory Committee Patient Advisory Group National Kidney Foundation	8 (100) 3 (100) ^	US	KHI Technology Roadmap for innovative approaches to KRT
Cho (2019)	Focus group study	Steering Committee Focus Group	3[76] 154 (100)	AU, KR, FR	Identification and prioritisation of core outcomes for research in ADPKD
Clemens (2019)	Qualitative study (focus groups, interviews)	Patient research partners	2 (29%)	UK, CA	Patient-centred care for diabetes and advanced CKD
Conway (2019)	Blog post	American Association of Kidney Patients (AAKP)	NA	US	Integrating patient input into kidney disease research - examples from AAKP
Cukor (2016)	Review	Patient advisory groups/councils Patient advocacy/research groups Steering committee members	NA	US	Patient engagement in PCORI funded studies in kidney disease
Demian (2017)	Narrative Review	Consumer co-researcher/author	1 (20)	CA	Opportunities for engaging patients in kidney disease research

Doyle (2019)	Pilot single group pre- and post-test intervention study	Focus group Patient information meeting Patient representative group	8 (100) 65 (100) ^	IE	Consumers involved in design of intervention - focus group, meeting/workshop
Duff (2018)	Protocol for community consultation	Focus groups	^	AU	Engaging Indigenous peoples in guideline development
Duncanson (2019)	Brief communication/ report	Consumer advisory board Consumer co-researchers/authors Consumer conference attendees: in person online	40 (100) 2 (14) 40 (^) 383 (^)	AU	Partnering with consumers in conferences
Elliott (2018)	Interview study (follow up - JLA PSP)	Consumer co-researcher Interview participants (previously involved in steering committee, workshop and/or online Wiki-platform)	1 (14) 12 (52)	CA	Long term views on JLA priority setting partnership for CKD research priorities
Elliott (2018-2)					Perceived significance of engagement (Patients, caregivers, HCPs) in research PSP – impact on participants
Finderup (2019)	Evaluation study	Interviews Advisory board	29 2 (25%)	DK	Engaging patients in evaluating a shared decision-making intervention for dialysis choice
Fowler (2017)	Commentary	KHI Patient and Family Partnership Council	≤10 (100)	US	Strategic guidance to KHI – including patients, families, caregivers in KHI activities
Gutman (2019)	Workshop report	Consumer co-researchers Workshop co-investigators	3 (18) 105 (71)	AU	Principles and strategies for involving patients and their families in CKD research
Hurst (2017)	Workshop report	Project workgroup Workshop	3 (21) 60 (55)	US	Involving consumers in the development of medical devices - KHI
International Society of Nephrology	Website	NA	NA	INT	Guidance on how to involve consumers throughout the research process, particularly for clinical trials
Kelly (2018)	Report	SA Aboriginal Community Reference Group Workshop participants	3 (100) 19 (100)	AU	Indigenous community involvement, guideline development, priority setting/scoping
Knight (2016)	JLA PSP	Steering group Patient organisations Initial survey Prioritisation survey Workshop	^ ^ 113 (62) 117 (46) 11 (55)	UK	Defining shared priorities for future research in Kidney Transplantation

Levin (2018)	JLA PSP, Program Report	Steering Committee: 2012-2013 2014 Survey: 2012-2013 2014 Workshops: 2012-2013 2014 Patient co-researchers (training)	5 (50) 6 (60) 210 (100) 309 (100) 16 (47) 18 (72) 56	CA	Canadians Seeking Solutions and Innovations to Overcome Chronic Kidney Disease (Can-SOLVE CKD): Form and Function – accelerating knowledge translation to clinical research and practice, top 10 research priorities for CKD
Lopez-Vargas (2018)	Workshop report	Workshop	35 (47)	AU	Research priority setting for childhood chronic conditions (including kidney disease)
Loud (2013)	Project report	Consumer co-researcher Advisory group	1 (33) 6 (100)	UK	Involving a consumer advisory group in improving variation in primary for patients with CKD (Quality improvement project)
Mader (2018)	Commentary	Patient led research network	^	UK	Patient led research hub
Manera (2019)	Delphi Survey	Steering committee Focus groups: 126 Survey participants	2 (14) 126 (100) 207 (24)	INT	Developing a core outcome set for patients receiving peritoneal dialysis
Manns (2014)	JLA PSP	Steering committee Survey respondents Workshop	5 (45) 210 (66) 16 (47)	CA	Research priorities for patients on/nearing dialysis
Marks (2018)	Co-researcher involvement	Co-researcher Public Involvement Research group Reference group Advisory group	1 (20) 15 (100) 4 (57) 4 (36)	UK	Roles of the co-researcher as part of the research team
Mick-Ramsay (2019)	Report	Co-researcher Consultation participants Top End Renal Patient Advisory & Advocacy Committee, KHA	1 (11) ^ ^	AU	Indigenous community involvement, guideline development, priority setting/scoping
Miller (2017)	Workshop report	Workshop	11 (100)	AU	Priority topics/ outcomes for infectious diseases in HD patients
Molnar (2017)	Narrative review	Patient advisory committees	NA	CA	Opportunities and challenges for patient engagement in kidney research
Navaneethan (2015)	Editorial	NA	NA	US	Patients as stakeholders in setting priorities for kidney disease research
Nielsen (2019)	Participatory design study	Workshops	8 (28)	DK	User involvement in development of telehealth intervention to improve transplant

Nierse (2012)	Responsive methodology	Co-researchers	2 (33)	NL	Collaboration and co-ownership in research
		Focus groups	^		
		Interviews	27		
Odgers (2017)	Systematic review	2/83 studies → CKD		INT	Research priority setting in childhood chronic disease
		1. Chesney – None	0		
		2. Langman – Workshop	^		
Paterson (2010)	Community based research project	Co-researcher	1[76]	CA	Toolkit for Aboriginal people on HD
		Community Advisory Committee	8 (73)		
Patzer (2014)	CBPR for RCT in dialysis clinics	Steering committee	^	US	Reducing disparities in access to Tx
Rosaasen (2010)	Intervention development	Co-researcher	1 (11)	CA	Patient-oriented research project to improve patient education
		Consultants	8 (40)		
Schipper (2011)	Qualitative study	Interviews	20 (100)	NL	Priorities for social science research for patients on dialysis
		Focus groups	54 (100)		
		Patient advocacy groups	^		
Tong (2012)	Workshops report	Consumer advisor	1	AU	Consumer involvement in topic and outcome selection for guidelines
		Workshops	23 (100)		
Tong (2015)	Workshop report	Workshop	30 (52)	AU	Research priorities in CKD
Tong (2015-2)	Systematic review	Of 16 studies identified, 4 explicitly involved patients:			Research priority setting
		1. Manns	**		
		2. Schipper	**		
		3. Rys-Sikora - online public consultation/voting	^ (7)	US	
		4. Tong - focus groups	63 (100)	AU	
Tong (2016)	Workshop report	3 focus groups	18 (100)	AU	Consumer priorities for guidelines ADPKD
Tong (2018)	Workshop report	Workshop	6 (7)	INT	Implementing core outcomes in kidney disease
Vargas (2008	CPPR (community partnered participatory research)	Workgroup	^	US	Awareness, prevention, early intervention
		Delphi survey	^		
		Conference	25 (10)		
		Survey	^		
Ward (2018)	Commentary	Consumer advisory councils	^	UK	Patient/ caregiver involvement in RCT
		Co-researcher	1		

* % of group made up of consumer; **Reported in included study; NA Not applicable; ^ Not reported; JLA PSP=James Lind Alliance – Priority Setting Partnership, P=patient, US=United States, CA=Canada, UK=United Kingdom, SE=Sweden, AU=Australia, KR=Korea, FR=France, IE=Ireland, DK=Denmark, INT=International, NL=Netherlands

B.3 Definitions of terms relating to consumer involvement

Term	Definition
Community-based participatory research	"A collaborative approach to research that equitably involves all partners in the research process and recognizes the unique strengths that each brings."[48,62]
Patient-researcher partnership	"The mode in which patients are engaged in research, which implies that each partner contributes something of equal value to the common enterprise. It encompasses more than having patients engaged as study participants. Furthermore, it is important to make a distinction between patients as research partners and patients as participants in surveys or focus groups. In the latter, patients are participants and their feedback and discussions serve to answer the research question. In patient-research partnerships, patients' input is sought to direct the various phases (preparatory, execution, and translational) of the research project."[35]
Patient engagement	"Patients having a "meaningful and active collaboration in governance, priority setting, conducting research and knowledge translation.""[45] "Collaborative research done by, with, and for patients to inform health care and health research decisions and questions."[35] "The establishment of a relationship between patients and researchers and is the first step of involvement."[41]
Patient/consumer/ public	"A patient, caregiver or family member with lived experience of chronic kidney disease"[41] "Fredriksson and Tritter (2017) [10] make the distinction between patient and public in PPI. Firstly, people with direct experience of health conditions either themselves or through a member of the family and secondly, people who have a more general interest in health and bring a 'public' view to health research."[25]
Involvement	"Research that is carried out "with or by" members of the public."[29] "A sustained and meaningful contribution to the research process as more than a research subject or participant and can range from consultation to partnership."[41] "Research being carried out 'with' or 'by' members of the public rather than 'to', 'about' or 'for' them"[25]
Co-researcher	"Equal partners of the research team and carry out some or all of the research activities alongside or independent of the academic researchers."[25]
Co-production	"The recently published NIHR INVOLVE guidance of research co-production cites the key co-production principles as sharing of power, inclusion of perspectives, respecting the value and knowledge of all contributors, reciprocity, and the building of relationships"[65]

Appendix C: Supporting documentation for Chapter 4

C.1 Participant organisational affiliations

- Kidney Health Initiative Patient and Family Partnership Council (Collaboration between United States Food and Drug Administration and the American Society of Nephrology) - https://khi.asn-online.org/pages/group.aspx?ID=KHI-PFPC
- Home Dialyzors United - https://www.homedialyzorsunited.org/
- Better Evidence and Translation in Chronic Kidney Disease (BEAT-CKD) Consumer Advisory Board - https://beatckd.org/about-us/consumeradvisoryboard/
- PKD Charity UK - https://pkdcharity.org.uk/about-us
- PKD Australia - https://pkdaustralia.org/
- ESRD Network Organizations, CMS - https://www.cms.gov/Medicare/End-Stage-Renal-Disease/ESRDNetworkOrganizations
- Kidney Health Australia - https://kidney.org.au/

Appendix D: Supporting documentation for Chapter 5

D.1 Break out group discussion questions

1. Why involve consumers? What should consumer involvement aim to achieve?
2. To what degree or level do you think consumers should be involved in research – why, and what are the priorities?
3. What would be the most meaningful, and effective ways of involving consumers in research across all aspects?
4. How should we communicate to patients/consumers about kidney research (format for communications, what works best, diversity of styles/methods etc)
5. How should we involve consumers in kidney research? (logistics of getting consumers to the table, timepoints, types of research, modes of communication etc)
6. What are the consumers views on researchers keeping their data / using and sharing their data? (consent, privacy etc)
7. How should the research community feedback the results of research to consumers and the community?

D.2 Workshop summary report distributed to participants

KIDNEY® HEALTH AUSTRALIA

BEAT CKD
BETTER EVIDENCE AND TRANSLATION IN CHRONIC KIDNEY DISEASE

Consumer Engagement Workshops

Overview of workshops across Australia

In 2017, BEAT-CKD and Kidney Health Australia (KHA) hosted 3 Consumer Engagement Workshops across Australia in Sydney, Adelaide and Brisbane.

A total of 148 people attended, including 105 patients and carers.

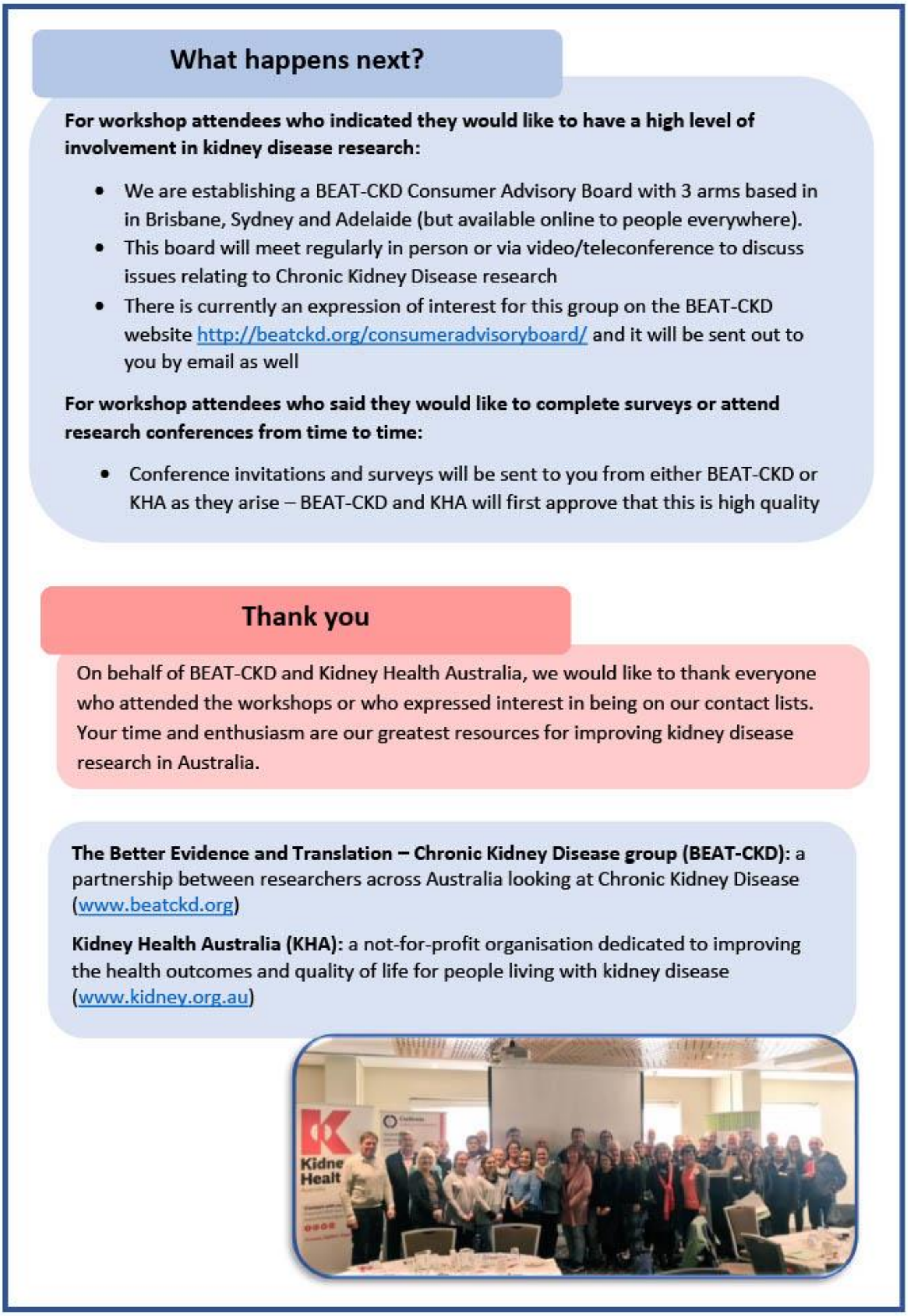
The workshops included:

- Presentations from researchers about what is currently happening in Chronic Kidney Disease research
- Presentations from patients and researchers about their perspectives of consumer involvement in research
- Discussions about important research topics

Why did we host the workshops?

To make research relevant and important to patients, we want to involve them in all stages of the research process. Patient and carer opinions are critical in deciding important topics for kidney research and how research should be conducted.

The purpose of the Consumer Engagement Workshops held in 2017 was to take the first step in this process – to reach out to patients and carers and ask them how they would like to be involved in research and what research they would like to see. The information we collected from the workshops via surveys and focus groups have helped us to understand how consumers would like to be involved and what issues are most important to them.



What happens next?

For workshop attendees who indicated they would like to have a high level of involvement in kidney disease research:

- We are establishing a BEAT-CKD Consumer Advisory Board with 3 arms based in in Brisbane, Sydney and Adelaide (but available online to people everywhere).
- This board will meet regularly in person or via video/teleconference to discuss issues relating to Chronic Kidney Disease research
- There is currently an expression of interest for this group on the BEAT-CKD website http://beatckd.org/consumeradvisoryboard/ and it will be sent out to you by email as well

For workshop attendees who said they would like to complete surveys or attend research conferences from time to time:

- Conference invitations and surveys will be sent to you from either BEAT-CKD or KHA as they arise – BEAT-CKD and KHA will first approve that this is high quality

Thank you

On behalf of BEAT-CKD and Kidney Health Australia, we would like to thank everyone who attended the workshops or who expressed interest in being on our contact lists. Your time and enthusiasm are our greatest resources for improving kidney disease research in Australia.

The Better Evidence and Translation – Chronic Kidney Disease group (BEAT-CKD): a partnership between researchers across Australia looking at Chronic Kidney Disease (www.beatckd.org)

Kidney Health Australia (KHA): a not-for-profit organisation dedicated to improving the health outcomes and quality of life for people living with kidney disease (www.kidney.org.au)

Appendix E: Supporting documentation for Chapter 6

E.1 Interview guide

A) Patient interviews

1. Motivation and attendance
   - How did you become involved/find out about ISPD?
   - Why did you attend ISPD? What were your expectations? What do/did you hope to achieve?
   - Did you receive any support in attending – do you have any thoughts about this? (Financial? Materials? Education?)
   - Were there any challenges you faced in attending? Do you think these (or other potential challenges) may have prevented some patients from attending? What are your thoughts on this?
   - Is your doctor attending? How do you feel about expressing your opinion in front of them?
2. Role and contribution
   - Could you describe your role/contribution at ISPD?
   - Did this have an impact on you personally – why/in what way?
   - What did you like most about being involved in/contributing to the meeting?
   - Were there any challenging aspects about being involved in this – could you please describe these?
     - Accessing scientific content? Need for more lay content sessions?
     - Physical conference environment/facilities?
     - Consideration for illness/symptoms?
   - How did you feel speaking/participating together with health professionals?
   - Do you think you/other patients contributed to the meeting in general – in what way?
3. Improving patient involvement
   - Can you suggest how we could improve the way in which patients are involved at medical conferences (ISPD)?
   - [If they have indicated they have attended other conferences] How did ISPD compare to other conferences you have attended regarding patient involvement?

B) Health professional interviews

1. Can you briefly describe how patients were involved at ISPD?
2. What do you think have been/or do you think there are benefits of patient involvement at ISPD?
    - What did they contribute?
    - How did it change the conference – in what way? (for you personally and in general?)
3. Were there any concerns or challenging aspects about patient involvement – could you please describe these?
    - Accessing scientific content? Need for more lay content sessions?
    - Physical conference environment?
4. Can you suggest ways to improve how patients are involved at medical conferences (ISPD)?
    - [If they have indicated they have attended other conferences] How did ISPD compare to other conferences you have attended regarding patient involvement?

Appendix F: Supporting documentation for Chapter 7

F.1. Detailed workshop run sheet

Time	Duration	Details and question guide
10:30	15 mins	Room set up
10:45	30 mins	Registration – consent form, demographic form
11:15	5 mins	Welcome and introduction • Welcome and thank participants • Introduce KHA-CARI guidelines group • Overview of the day
11:20	15 mins	Introduction to guidelines • PowerPoint presentation
11:35	55 mins	Small group discussion - *Q's projected on screen*
	5 mins	Introduce the task and do a round of introductions • *Before we start can I get everyone to write their name on a piece of A4 paper and fold it like this so that we can all see your name please. A few things before we introduce ourselves:* • *The session will be recorded* • *Everyone will get a chance to speak, please speak one at a time* • *Please respect each other's opinions* • *There are no right or wrong answers* • *The recordings will only be heard by the facilitators* • *No names will be published with the results* • *Observing, field notes, media* • Icebreaker • *OK now, we'll quickly go around the table and in 30 seconds please say your name and the first thing you think of when I say 'kidney biopsy' – who would like to go first? (then go round the table)*
	50 mins	Generating topics *For the next 50 minutes we are going to try and come up with topics that you think are important and should be included in the kidney biopsy guidelines. In the first 15 minutes, we'll have a brief discussion about your experiences of having a renal biopsy.* 1. Experiences of renal biopsy (15 mins) - What is it like to have experienced renal biopsy? - How did you feel in the lead up to your procedure? 2. Guideline questions and topics (35 mins) - In the next 30 mins, we will develop some topics and questions that you think should be included in clinical practice guidelines for kidney biopsy. *One person nominated to write on flip-chart* - What topics do you think should be included in guidelines for renal biopsy – why? *E.g. lifestyle modification, psychosocial support, patient education etc*

		- For each topic, what outcomes do you think are important? What sorts of things should be measured? *E.g. symptoms, quality of life, days in hospital etc* - What do you think health care providers (nephrologists, GP, nurses) need to know about patients' experiences and perspectives regarding renal biopsy? - Is there anything else that you feel is important to add?
12:30	30 mins	Lunch
1:00	30 mins	Small group discussion continued • *In the next 30* mins *we will finalise our topics and try to identify the issues of high importance that should be included in clinical practice guidelines.* *One person nominated to present discussion to wider group* • Ensure all participants have spoken • Refer to the clinician derived topics and outcomes to stimulate further discussion • Summarise discussion • *Observer, field notes, media*
1:30	20 mins	Wider group discussion • Each small group to present groups' discussion and topics • Other groups to comment / feedback
1:50	10 mins	Thank you and evaluation/reimbursement • Thank you for sharing your views and experiences of undergoing renal biopsy and formulating questions you believe are relevant to the care of patients. • Remind participants about the consumer guidelines. If you would like to stay involved in future research projects please tick the box in the continuing research form. • Provide evaluation sheet
2:00		Pack up

Appendix G: Supporting documentation for Chapter 8

G.1 Participating Sites and Institutional Review Boards

British Columbia Children’s Hospital, The University of British Columbia Research Ethics Board, Vancouver, Canada.

Alberta Children’s Hospital, The University of Calgary Research Ethics Board, Calgary, Canada.

Texas Children’s Hospital, The Institutional Review Board for Baylor College of Medicine and Affiliated Hospital, Houston, United States.

The Children’s Hospital at Westmead, The Sydney Children’s Hospital Network Human Research Ethics Committee, Sydney, Australia.

The Royal Children’s Hospital, The Melbourne Children’s Campus Research Ethics and Governance, Melbourne, Australia.

Lady Cilento Hospital, The Children’s Health Queensland Hospital and Health Service Research Governance, Brisbane, Australia.

G.2 Child question guide

We want to know what it's like for you to have kidney problems

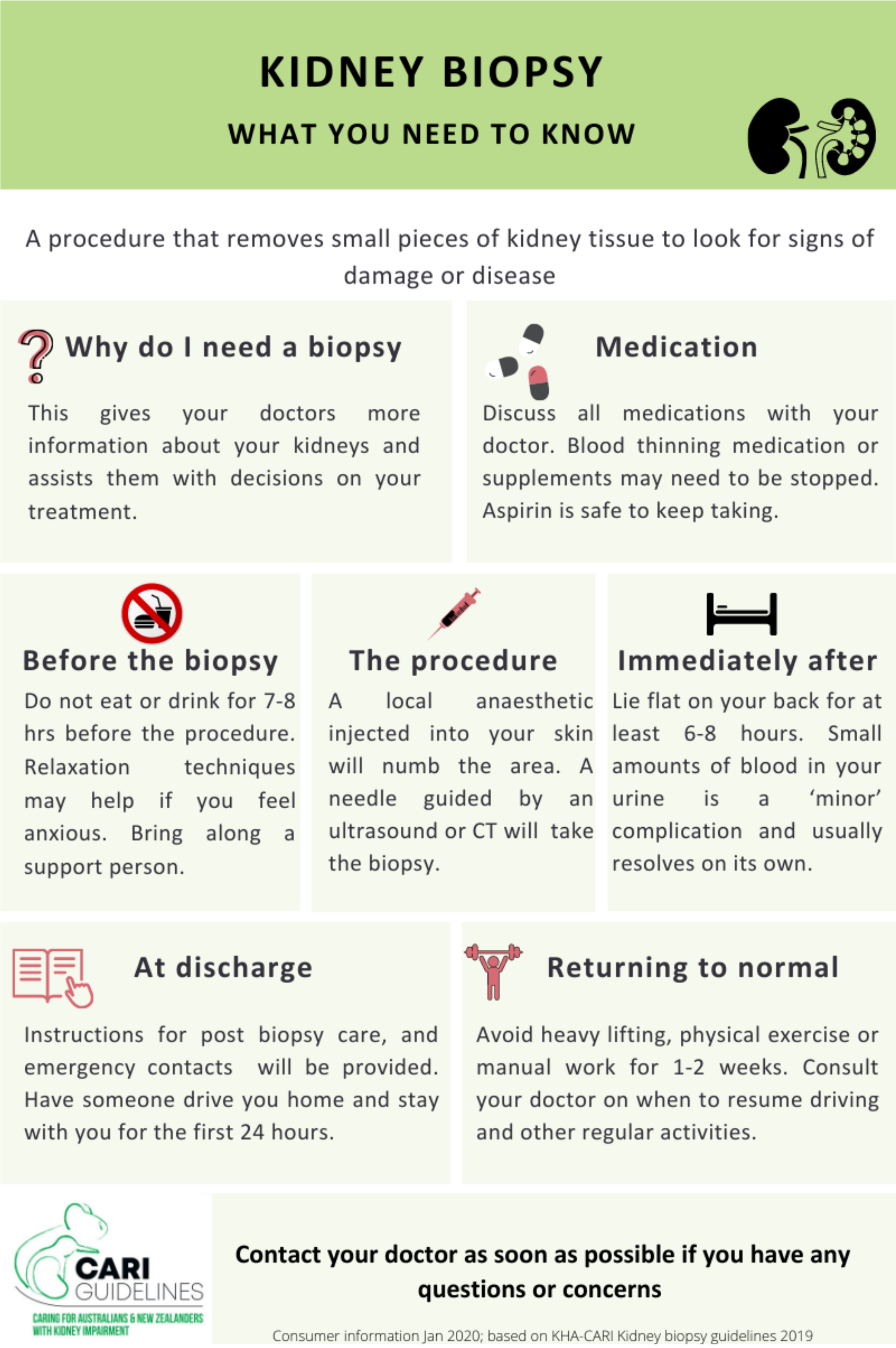
- Can you tell us a little bit about your kidney problems (how long, what treatment you have?)
- What is it like to have kidney problems?
- What are the hardest things about having kidney disease? (school, family, hospital)
    - How could this be better addressed?
    - How well do you think these things are understood by the health care staff?
    - In what ways do they understand? In what ways don't they understand?
- When it comes to making decisions about treatment – who makes the decisions? Do you feel you get to have a say about your treatment – why?
- Can you think of a time when you felt involved or not involved in a decision?
    - How did it go? What did you want to happen?
    - If you spoke up, what happened?
    - Knowing what you now know, how would you make the same decision?

G.3 Parent question guide

- How did you first find out that your child had kidney disease? How did you react/feel?
- How has the kidney disease/dialysis/kidney transplant changed your life and your child's life?
- What is the most challenging thing about caring for a child with kidney disease, and how do you deal with it?
- How well do you think these things are understood by the health care staff? In what ways do they understand/don't they understand?
- What do you think about the information you have about your child's kidney problems or treatment?
- To what degree do you feel you are involved in making decisions about your children's treatment – why?
- When it comes to making a decision about treatment, who makes the decision?
- Has there been a time when you may have not agreed with the doctor – what happened then? Was it resolved – how/why?
- Do you think your child is involved – in what way?
- Can you think of a time when you felt involved or not involved in making a decision about treatment?
- What happens when you child might disagree with what you think is best for them?
- Is there anything else that you think might be important to add about communication or decision-making?

Appendix H: Supporting documentation for Chapter 9

H.1 Kidney Biopsy Consumer Guideline infographic



KIDNEY BIOPSY

WHAT YOU NEED TO KNOW

A procedure that removes small pieces of kidney tissue to look for signs of damage or disease

Why do I need a biopsy

This gives your doctors more information about your kidneys and assists them with decisions on your treatment.

Medication

Discuss all medications with your doctor. Blood thinning medication or supplements may need to be stopped. Aspirin is safe to keep taking.

Before the biopsy

Do not eat or drink for 7-8 hrs before the procedure. Relaxation techniques may help if you feel anxious. Bring along a support person.

The procedure

A local anaesthetic injected into your skin will numb the area. A needle guided by an ultrasound or CT will take the biopsy.

Immediately after

Lie flat on your back for at least 6-8 hours. Small amounts of blood in your urine is a ‘minor’ complication and usually resolves on its own.

At discharge

Instructions for post biopsy care, and emergency contacts will be provided. Have someone drive you home and stay with you for the first 24 hours.

Returning to normal

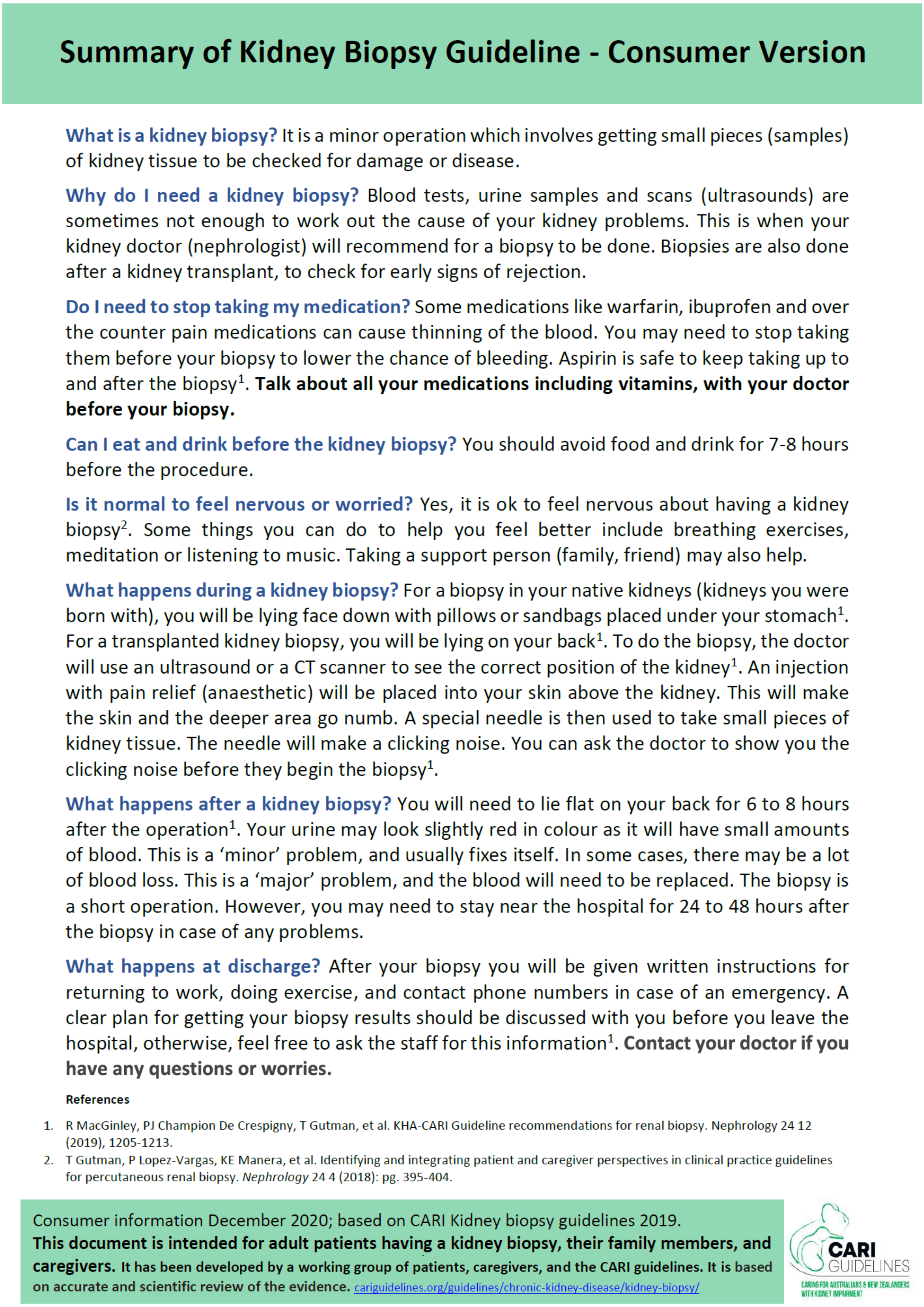
Avoid heavy lifting, physical exercise or manual work for 1-2 weeks. Consult your doctor on when to resume driving and other regular activities.

CARI GUIDELINES
CARING FOR AUSTRALIANS & NEW ZEALANDERS WITH KIDNEY IMPAIRMENT

Contact your doctor as soon as possible if you have any questions or concerns

Consumer information Jan 2020; based on KHA-CARI Kidney biopsy guidelines 2019

H.2 Kidney Biopsy Consumer Guideline one page summary



Summary of Kidney Biopsy Guideline - Consumer Version

What is a kidney biopsy? It is a minor operation which involves getting small pieces (samples) of kidney tissue to be checked for damage or disease.

Why do I need a kidney biopsy? Blood tests, urine samples and scans (ultrasounds) are sometimes not enough to work out the cause of your kidney problems. This is when your kidney doctor (nephrologist) will recommend for a biopsy to be done. Biopsies are also done after a kidney transplant, to check for early signs of rejection.

Do I need to stop taking my medication? Some medications like warfarin, ibuprofen and over the counter pain medications can cause thinning of the blood. You may need to stop taking them before your biopsy to lower the chance of bleeding. Aspirin is safe to keep taking up to and after the biopsy[1]. **Talk about all your medications including vitamins, with your doctor before your biopsy.**

Can I eat and drink before the kidney biopsy? You should avoid food and drink for 7-8 hours before the procedure.

Is it normal to feel nervous or worried? Yes, it is ok to feel nervous about having a kidney biopsy[2]. Some things you can do to help you feel better include breathing exercises, meditation or listening to music. Taking a support person (family, friend) may also help.

What happens during a kidney biopsy? For a biopsy in your native kidneys (kidneys you were born with), you will be lying face down with pillows or sandbags placed under your stomach[1]. For a transplanted kidney biopsy, you will be lying on your back[1]. To do the biopsy, the doctor will use an ultrasound or a CT scanner to see the correct position of the kidney[1]. An injection with pain relief (anaesthetic) will be placed into your skin above the kidney. This will make the skin and the deeper area go numb. A special needle is then used to take small pieces of kidney tissue. The needle will make a clicking noise. You can ask the doctor to show you the clicking noise before they begin the biopsy[1].

What happens after a kidney biopsy? You will need to lie flat on your back for 6 to 8 hours after the operation[1]. Your urine may look slightly red in colour as it will have small amounts of blood. This is a 'minor' problem, and usually fixes itself. In some cases, there may be a lot of blood loss. This is a 'major' problem, and the blood will need to be replaced. The biopsy is a short operation. However, you may need to stay near the hospital for 24 to 48 hours after the biopsy in case of any problems.

What happens at discharge? After your biopsy you will be given written instructions for returning to work, doing exercise, and contact phone numbers in case of an emergency. A clear plan for getting your biopsy results should be discussed with you before you leave the hospital, otherwise, feel free to ask the staff for this information[1]. **Contact your doctor if you have any questions or worries.**

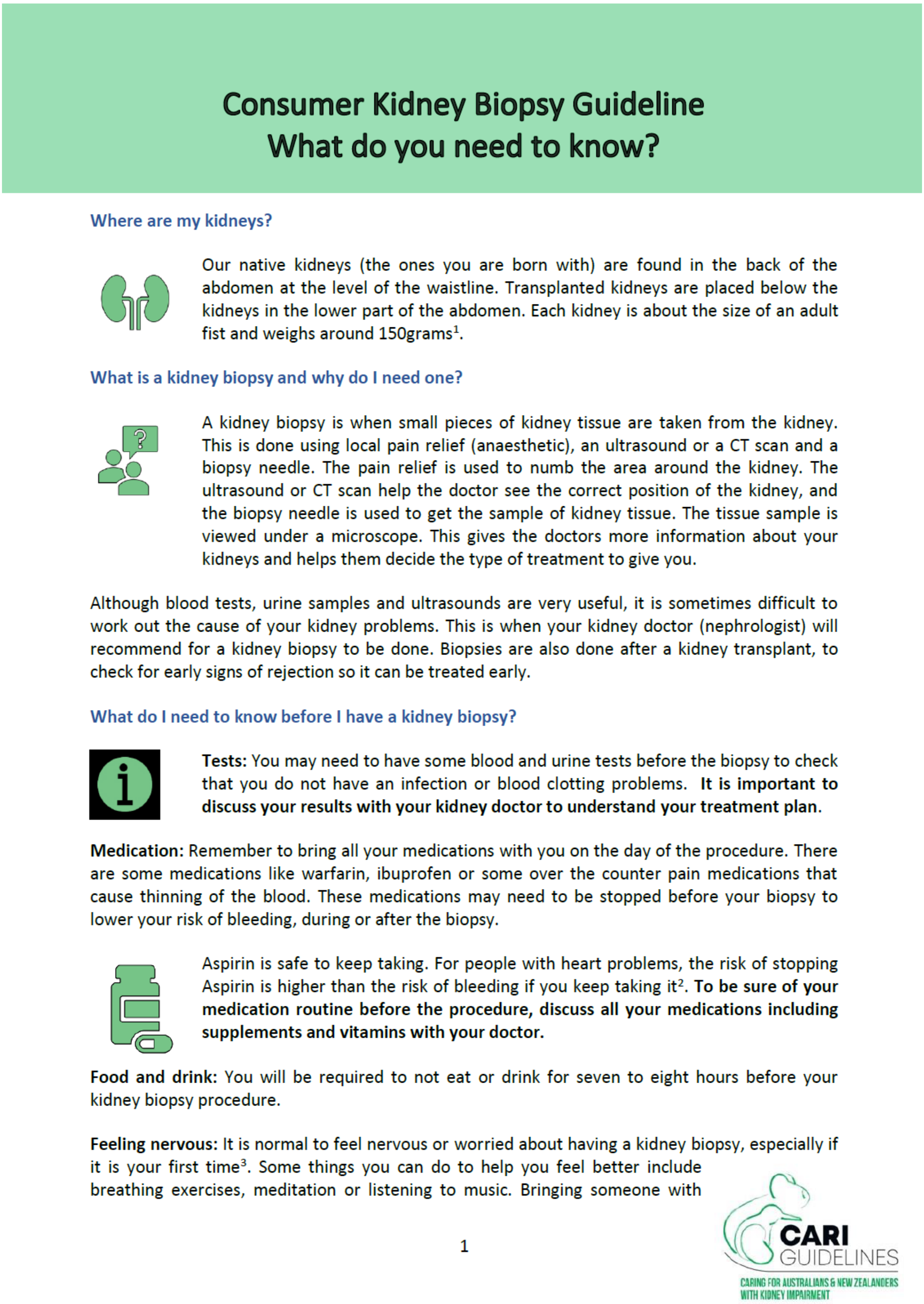
References

1. R MacGinley, PJ Champion De Crespigny, T Gutman, et al. KHA-CARI Guideline recommendations for renal biopsy. Nephrology 24 12 (2019), 1205-1213.
2. T Gutman, P Lopez-Vargas, KE Manera, et al. Identifying and integrating patient and caregiver perspectives in clinical practice guidelines for percutaneous renal biopsy. *Nephrology* 24 4 (2018): pg. 395-404.

Consumer information December 2020; based on CARI Kidney biopsy guidelines 2019.
This document is intended for adult patients having a kidney biopsy, their family members, and caregivers. **It has been developed by a working group of patients, caregivers, and the CARI guidelines. It is based on accurate and scientific review of the evidence.** cariguidelines.org/guidelines/chronic-kidney-disease/kidney-biopsy/

CARI GUIDELINES
CARING FOR AUSTRALIANS & NEW ZEALANDERS WITH KIDNEY IMPAIRMENT

H.3 Kidney Biopsy Consumer Guideline



Consumer Kidney Biopsy Guideline
What do you need to know?

Where are my kidneys?

Our native kidneys (the ones you are born with) are found in the back of the abdomen at the level of the waistline. Transplanted kidneys are placed below the kidneys in the lower part of the abdomen. Each kidney is about the size of an adult fist and weighs around 150grams[1].

What is a kidney biopsy and why do I need one?

A kidney biopsy is when small pieces of kidney tissue are taken from the kidney. This is done using local pain relief (anaesthetic), an ultrasound or a CT scan and a biopsy needle. The pain relief is used to numb the area around the kidney. The ultrasound or CT scan help the doctor see the correct position of the kidney, and the biopsy needle is used to get the sample of kidney tissue. The tissue sample is viewed under a microscope. This gives the doctors more information about your kidneys and helps them decide the type of treatment to give you.

Although blood tests, urine samples and ultrasounds are very useful, it is sometimes difficult to work out the cause of your kidney problems. This is when your kidney doctor (nephrologist) will recommend for a kidney biopsy to be done. Biopsies are also done after a kidney transplant, to check for early signs of rejection so it can be treated early.

What do I need to know before I have a kidney biopsy?

Tests: You may need to have some blood and urine tests before the biopsy to check that you do not have an infection or blood clotting problems. **It is important to discuss your results with your kidney doctor to understand your treatment plan.**

Medication: Remember to bring all your medications with you on the day of the procedure. There are some medications like warfarin, ibuprofen or some over the counter pain medications that cause thinning of the blood. These medications may need to be stopped before your biopsy to lower your risk of bleeding, during or after the biopsy.

Aspirin is safe to keep taking. For people with heart problems, the risk of stopping Aspirin is higher than the risk of bleeding if you keep taking it[2]. **To be sure of your medication routine before the procedure, discuss all your medications including supplements and vitamins with your doctor.**

Food and drink: You will be required to not eat or drink for seven to eight hours before your kidney biopsy procedure.

Feeling nervous: It is normal to feel nervous or worried about having a kidney biopsy, especially if it is your first time[3]. Some things you can do to help you feel better include breathing exercises, meditation or listening to music. Bringing someone with

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you to the hospital may also help. They can be with you before and after the procedure, but not while you are having the biopsy.

What position will you be in during the biopsy?

For a native biopsy, the most common position is lying on your stomach with pillows or sandbags placed under your stomach. For a kidney that has been transplanted the biopsy is normally done with you lying on your back[1].

These positions give your doctor the best access to your kidneys and make sure you are as comfortable as possible. The procedure usually takes 30-45 minutes. If you are not able to be in one of these positions for that long, speak with your doctor to work out what can be done.

What happens during the biopsy?

The biopsy may be taken from the left or right native kidneys. **This procedure involves the use of needles.**

First, an injection with pain relief will be placed into your skin above the kidney and then more deeply, down to the edge of your kidney. This stings a little but will quickly make your skin and the deeper area go numb. Then a needle will be used to take small pieces of kidney tissue. The needle will make a clicking noise when the biopsy is taken. You can ask the doctor to show you the clicking noise before the biopsy, so you know what to expect[2].

To work out exactly where the biopsy needle should be put in, your doctor will use an ultrasound, but sometimes a CT scanner may be used. This will depend on what the unit uses or who is doing the biopsy, a nephrologist (kidney doctor) or radiologist (imaging doctor)[2].

When the biopsy is taken you may be asked to hold your breath for a few seconds to stop any movement around the kidney area. The doctor will then remove the needle containing a small piece of kidney tissue. This is usually done at least two times to make sure there is enough tissue to be tested. When finished, the doctor will put a small bandage over the area where the needle went into your skin. You will not need any stitches. The bandage can be taken off after 24 hours.

What happens immediately after a kidney biopsy?

After the operation you will be asked to lie flat on your back for 6-8 hours[2]. Transplant patients may have an extra weight (often a sandbag) placed over the biopsy site. Strict bed rest is needed to reduce any risk of bleeding. During this time the nurses will take your blood pressure, temperature, monitor your pain levels and check for bleeding at the site. Patients with higher risks for complications may need to be monitored for up to 24 hours.

Before leaving the hospital, you will be asked to pass urine to check for any signs of bleeding. It is normal to have small amounts of blood in the urine after a biopsy – this is a 'minor' problem and almost always settles on its own. In very rare cases there may be a 'major' problem which may cause a lot of blood loss that will need to be replaced. Your doctor will order an ultrasound to check and make sure the bleed is taken care of in the best possible way.

What happens at discharge?

At discharge, you should be given clear written instructions for how to look after yourself including returning to work and exercising. You will be given phone numbers in case of an emergency. A clear plan for getting your biopsy results should also be discussed with you before you leave the hospital. If not, feel free to ask the staff for this information[2]. There can be different wait times for the results of a biopsy depending on the type of test ordered. The best person to let you know how long it will take for the results to be available, is your doctor.

You will not be able drive or go home on public transport after the biopsy, so it is important you arrange for someone to take you home. This will be more comfortable, safer and also quicker for you to return to the hospital, in case you develop any problems on the journey home. If you live far from the hospital, you may need to arrange to stay overnight close to the hospital instead of going home. In some cases, you may stay overnight in the hospital, so it is important to talk with your doctor well before the biopsy.

Call the doctor if you have any of the following:

- More than a teaspoon of bleeding at the biopsy site
- Swelling, redness, ooze, or pus at the biopsy site
- Feeling dizzy, faint or light-headed
- Increasing pain around the biopsy site
- Too much back pain or groin pain
- Blood clots or blood in the urine that is getting worse after 2 days
- Nausea, vomiting or temperature higher than 38 degrees (fever)

Returning to normal activities

This is different for everyone but:

- Avoid heavy physical exercise or manual work for 1-2 weeks. Ask your doctor when you can start driving and doing other regular activities.

- Depending on your type of work the doctor will let you know how much time off you will need. You can get a medical certificate on the day of the biopsy, so please ask if you need one. If your work involves heavy lifting it may take you longer to get back to work.

References

1. https://kidney.org.au/your-kidneys/prevent/what-your-kidneys-do
2. R MacGinley, PJ Champion De Crespigny, T Gutman et al. KHA-CARI Guideline recommendations for renal biopsy. *Nephrology* 24 12 (2019): 1205-1213.
3. T Gutman, P Lopez-Vargas, KE Manera et al. “Identifying and integrating patient and caregiver perspectives in clinical practice guidelines for percutaneous renal biopsy” *Nephrology* 24 4 (2018): 395-404.

Glossary of terms

- **Biopsy-** a procedure to remove a sample of cells or tissue from your body to be analysed through a microscope.
- **Percutaneous-** any medical procedure that is done with a needle through the skin.
- **Nephrology –** is the study of the kidneys and the diseases that affect it.
- **Native kidneys –** the kidneys that you are born with.
- **Guidelines-** Guidelines help doctors understand the best ways to diagnose, treat and even prevent diseases and conditions. Guideline recommendations are based on the strongest available scientific evidence.

If you wish to find out more information about your kidneys and kidney biopsies then here are some reliable websites to visit:

- CARI guidelines - http://www.cari.org.au/CKD/ckd_guidelines.html
- National Kidney Foundation. What is a kidney biopsy? New York: National Kidney Foundation; 2017. Available from: https://www.kidney.org/atoz/kidney-biopsy
- Kidney Health Australia: https://kidney.org.au/your-kidneys/prevent/what-your-kidneys-do
- Kidney Care UK: https://www.kidneycareuk.org/about-kidney-health/

This document is intended for adult patients having a kidney biopsy, their family members and caregivers. It has been developed by a working group of patients and caregivers in partnership with the CARI guidelines and is based on a rigorous and scientific review of the evidence.

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1. C Rowan
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3. A Fenech