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Psychosocial and bioethical challenges and developments for the future of vascularized composite allotransplantations

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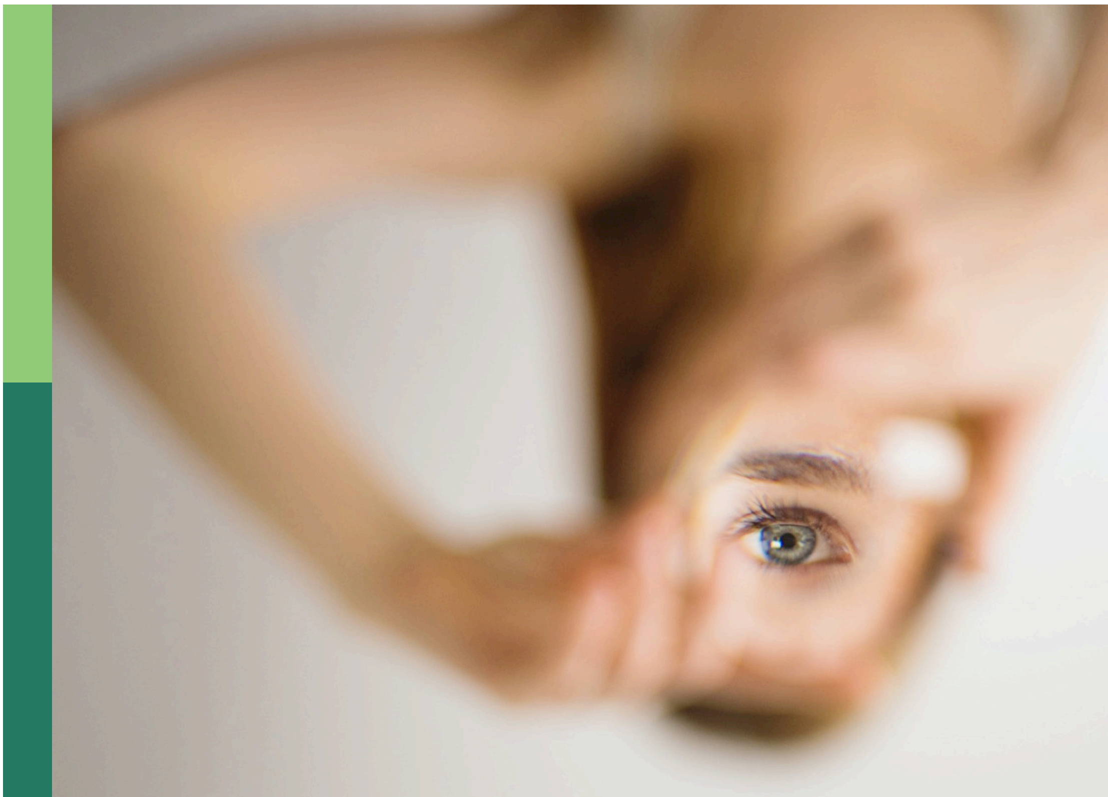

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Psychosocial and bioethical challenges and developments for the future of vascularized composite allotransplantations

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Editorial: Psychosocial and bioethical challenges and developments for the future of vascularized composite allotransplantations

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Editorial on the Research Topic
Psychosocial and bioethical challenges and developments for the future of vascularized composite allotransplantations

Vascularized composite allotransplantation (VCA) remains a relatively new field of medicine and transplantation, with a history of successful transplantation starting fewer than 25 years ago, and including more than 150 transplants to date. The potential to restore lost function and to improve quality of life for individuals with severe impairments propelled early research focused on the technical and immunological aspects of these procedures. As more cases are studied and advances in the field are made, the importance of psychosocial and bioethical considerations has become increasingly relevant (Kinsley et al., 2020, 2022).

The aim of this Research Topic is to create a foundation of high quality articles that address psychosocial and bioethical challenges and developments in the evolving field of VCA with viewpoints driven by research and clinical experience, across the varied types of VCA (e.g., hand, face, and uterus transplantation). Furthermore, this review summarizes recent discussions and conclusions reached by the Chauvet Workgroup, an international multicenter and multidisciplinary platform to bring together expertise and to learn from VCA cases performed worldwide. The Chauvet Workgroup's mission is to improve understanding of psychosocial and bioethical factors in VCA through an open global platform for all interested parties with no defined ownership and broad collaboration in research and clinical practice. The Chauvet Workgroup holds great value and potential because of its core principle of collaborative international research and clinical approach to the psychosocial evaluation in VCA. The Chauvet Workgroup has been endorsed by both the International Society for VAC (ISVCA) and American Society for Reconstructive Transplantation (ASRT) with regular presentations at their meetings to summarize activities of the biennial workshops (Jowsey-Gregoire et al., 2016; Kumnig and Järvholm, 2022).

Psychosocial aspects of VCA span the lifecycle of VCA. Initial evaluation and assessment of candidates participants, optimization of psychosocial variables, peri-operative support, and preparation and management of post-transplantation changes and adjustment are just some of the areas of research and interest (Kinsley et al., 2021). Success in VCA requires a match between the operation and patient. We often consider this "appropriate patient selection" (Kumnig et al., 2012, 2014). However, psychosocial research also allows, perhaps more importantly, optimization of potential candidates to ensure adequate support and interventions to expand candidacy. Thus, psychosocial evaluation should include both a comprehensive evaluation combined with follow-up protocols and supportive interventions before (considering risk-benefit issues) and after VCA (for example, dealing with the graft in daily life, etc.) (Jowsey-Gregoire and Kumnig, 2016; Kumnig and Jowsey-Gregoire, 2016).

In addition to the psychosocial aspects that span the lifecycle of VCA discussed herein, bioethical concerns need to be considered as fundamental elements of patient evaluation and follow-up protocols that provide the foundation for successful VCA (Gordon and Siemionow, 2009; Gordon et al., 2011). For example, a variation in clinical practices pertaining to candidate evaluation and informed consent processes mean that some candidates my receive less rigorous evaluation or information from others, which may undermine their consideration for candidacy or shared decision making. Thus, VCA organizations providing bioethical and policy oversight of the organ transplant system should establish standards to ensure that all VCA candidates/recipients are treated equitably (Gacki-Smith et al.).

While VCA raises bioethical concerns pertaining to all principles, VCA challenges some principles more than others. Specifically, respecting patients' self-determination (autonomy) is particularly challenging given the limitations of data on long-term psychosocial and clinical outcomes. Given the relatively small number of patients receiving VCA organs, the VCA field faces challenges in amassing sufficient information about the risks and benefits that VCA recipients commonly experience. Accordingly, VCA candidates may find it difficult to make meaningful informed treatment decisions and provide informed consent to pursue VCA.

This Frontiers Research Topic demonstrates the need for more collaborative, multicenter research in order to optimize these highly complex medical procedures, and to bring together expertise and to learn from as many of the VCA cases performed as possible. This Research Topic builds on the work of many research and clinical teams and aims to provide a unifying framework to evolve research on psychosocial issues. The authors describe the relevant psychosocial and bioethical considerations for the formation of an international research platform and outline its vision and current process, based on international research collaborations including most predominantly that of the Chauvet Workgroup (Kumnig et al., 2022). Additionally, recent efforts of the Organ Procurement and Transplantation Network (OPTN) and the United Network for Organ Sharing (UNOS) support these developments. Finally, this paper collection identifies key issues that should be addressed in future research.

Statistics on this Research Topic: This Research Topic was open between May 2022 and February 2023. After a rigorous and constructive reviewing process of numerous submissions, ultimately 10 articles by 53 authors were accepted. Since its inception, this Research Topic has received over 7,200 views (as of March 2023).

Overview of this Research Topic: To cover these themes, this Frontiers Research Topic consists of the following types of manuscripts: original research, review and mini review, conceptual analysis, brief research report, and opinion. The submission of original articles was particularly appreciated, presenting recent research innovations in this field, including results of qualitative research initiatives to investigate relevant psychosocial outcome predictors.

Author contributions

All authors listed have made a substantial, direct, and intellectual contribution to the work and approved it for publication.

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We are grateful to all contributors to this Research Topic. We thank the reviewers who helped the authors and us to create an innovative and high-quality publication. We would also like to thank all patients and colleagues who have generously shared their experiences within the field. Finally, we hope that readers will enjoy reading this Research Topic as much as we have enjoyed editing it.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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OPINION
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Patient Advocacy in Vascularized Composite Allotransplantation

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Keywords: vascularized composite allotransplantation (VCA), patient advocacy, vulnerability, ethics, special protections

INTRODUCTION

Over the last two and a half decades vascularized composite allotransplantation (VCA) has developed into a promising therapeutic option for persons who have suffered the loss of limbs, major facial disfigurement, substantial damage to the abdominal wall, primary uterine infertility and other conditions.[1] Candidates considering VCA face a difficult choice between continuing to live with their current condition, with its substantial negative impact on their quality of life, or taking on the substantial risks and burdens of a transplant. Furthermore, the outcomes of upper extremity and facial VCA are highly dependent on the capacity and will of the patient to live with an awareness of the risks, cope with the burdens and persist in careful adherence to immunosuppression and physiotherapy. Despite certain advantages of upper extremity and facial VCA in terms of function and cosmesis, the practice has elicited a great deal of ethical concern, including concern that candidates and patients may require special protections because they are especially vulnerable (Hartman, 2007; Perpich, 2010).

This essay aims to argue that upper extremity and facial VCA candidates are rightly considered to be especially vulnerable and then explore how the provision of a patient advocate can provide protections during the screening, selection, decision-making process and post-surgical period. This argument will review the use of the concepts of vulnerability and patient advocacy in health care, describe how patient advocates were involved in some of the earliest upper limb transplantations in the United States and suggest how patient advocates in the context of upper extremity and facial VCA may be able to offset some of the concern about the vulnerability of candidates and patients.

VULNERABILITY

Since its first appearance in the literature of bioethics more than 40 years ago,[2] there has been a steadily growing interest in the concept of vulnerability. Concern for the vulnerability of research subjects and, to a lesser extent patients in clinical practice, has been expressed through the inclusion of the term in various reports, guidelines, declarations and articles.[3] In general, the term has been used to draw attention to circumstances in which subjects or patients may be unable to adequately defend their interests.

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[1] Other types of VCA that have been attempted or seriously considered include transplantation of the esophagus, larynx, knee, penis, and tongue.

[2] For a detailed accounting of the use of the term in bioethics, see ten Have (2016).

[3] Examples include the Belmont Report, the Council for International Organizations of Medical Sciences (CIOMS) Guidelines. The UNESCO Universal Declaration on Bioethics and Human Rights, and the Declaration of Helsinki. Recent articles treating the topic include Monacelli et al. (2016), Amgel and Vatne (2017), and Bracken-Roche et al. (2017).

The broadest definition of vulnerability is susceptibility to harm. This is a universal human condition, as one aspect of our natural state of mutability. Theoretical ethics has suggested our universal human vulnerability is derived from our biological and social dependence upon others.[4] Some have argued against the relevance of a broad definition of vulnerability for bioethics (Wrigley and Dawson, 2016), but it is in fact the foundation for the well-recognized principle of non-maleficence, which is often expressed by the Latin, "*primum non nocere*," or "first, do no harm."

More commonly, however, bioethics is concerned with forms of special vulnerability. Special vulnerability refers to particular ways in which an individual may be more susceptible to harm than most others, due to characteristics of that individual and/or his or her context. These characteristics are thought to diminish the individual's capacity to defend against threats to his or her interests or wellbeing. An obvious example would be unconsciousness. Poverty, lack of education, disability, race and gender are also sometimes identified as sources of special vulnerability.

Vulnerability is relevant to ethics because it draws our attention to potentially avoidable or remediable human suffering. Discussions of vulnerability in health care ethics typically suggest that we have a duty to provide special protections for those who are classed as especially vulnerable (ten Have, 2014).[5] Much of the focus on vulnerability in bioethics has been on issues involving the principle of respect for autonomy, as in cases where subjects or patients are not adequately informed, unable to process the information sufficiently, or under such duress that they are essentially coerced. Yet while limited autonomy is one source of vulnerability, there are certainly other reasons why individuals or groups may not be in a position to protect themselves. Not only may one's decisional capacity be compromised, but so too may be one's ability to carry out one's expressed preferences or desires.

In order to successfully address vulnerability in a subject, patient or population, one must first specify the vulnerability. To what exactly is the person or population vulnerable, for what reasons, and to what degree? In what ways and to what extent might the vulnerability be offset? With regard to candidates for vascularized composite allotransplantation of face or upper extremities, special vulnerability may take a number of forms.

First, they may be understood as medically vulnerable. Medical vulnerability applies to candidates for VCA because it refers to persons who are so seriously ill or injured that they may be attracted to research protocols by unrealistic expectations (Benvenuti et al., 2021). As Nickel points out, "In general, research subjects tend to underestimate the level of risk or impact associated with participation in biomedical research," while overestimating the likelihood of potential benefits. Subjects do so, Nickel claims, because of the effects of "'motivated bias,' i.e., errors based on a desire to believe something is true." (Nickel, 2006) Given that those drawn to VCA tend to be those who have been unable to adapt to their disfigurement or disability, while others of equal or greater injury do adapt, the question is raised whether VCA inadvertently targets the most desperate among the disabled and disfigured who may therefore also be the most vulnerable (Rumsey, 2004; Bradbury, 2012).

Second, candidates for VCA may be understood as socially vulnerable. Social vulnerability applies to candidates for VCA whose condition has led to social isolation. Facial defects often lead to such isolation (Strandmark, 2004; Strong, 2004; Svenaeus, 2012).[6] Upper extremity defects may also cause isolation, as a result of their effect on the individual's self-image or because the functional consequences of the defect exclude the individual from certain activities. Examples would be the loss of the ability to continue one's career, certain activities of daily living or familiar leisure activities. A sense of social isolation may also be created by the increased level of dependency that occurs as a result of a defect.

Loss of independence due to disability has been associated with a decrease in psychological wellbeing and subjective estimates of quality of life, a limitation of employment opportunities, and social stigma or marginalization. Persons with disabilities frequently "report giving up established ways of doing things, and forgoing numerous activities, plans and goals." Various factors have been identified as playing a role in the subjective perception of dependence, including not only an individual's pre-existing coping skills, but the "cultural norms and societal values" to which the individual has been exposed (Gignac and Cott, 1998). When facial defects are of such a nature as to prevent normal eating or even normal breathing, and when upper extremity defects render persons unable to drive, maintain employment, feed themselves, etc., a state of dependency may be created which individuals may be so anxious to escape that they are willing to take far greater than normal risks.

Third, candidates for VCA may be deemed vulnerable due to the complexity of the decision and the limits of imagination (Fischer et al., 2021). In order to make a decision, candidates must attempt to imagine a future in which they will be confronted with major burdens and risks. While they may receive substantial benefits, they are also taking on what amounts to a kind of chronic illness, some aspects of which will diminish their quality of life. They may struggle to cope with the side effects of immunosuppression or the rigors of physiotherapy.[7] Upper extremity recipients will endure a period of time, often several months, during which they will actually be less functional and more dependent than before the surgery. They must try to imagine how the treatment and the transformation thus wrought will affect their relationships with family members, friends, or co-workers. Additionally, all candidates should assume that a time will come when the graft will be lost. In order to make a sound decision to proceed, candidates must imagine life under these

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[4] See Goodin (1985), Nussbaum (1986), and MacKenzie et al. (2014). This idea of dependence as a natural state is traditional, going back in the western tradition at least to the divine declaration in Genesis 2:18. "It is not good for the man to be alone."

[5] The Principle of Respect for Human Vulnerability and Personal Integrity: Report of the International Bioethics Committee. Available online at: http://unesdoc.unesco.org/images/0021/002194/219494E.pdf (accessed January 25, 2014).

[6] Rumsey (2004, p. 22–23) and Bradbury (2012, p. 193–196).

[7] Failures to cope with their post-transplant condition have been the source of complications, including graft loss in some cases. See Tintle et al. (2014) and Kumnig and Jowsey-Gregoire (2016).

conditions and determine whether or not they will be able to rise to the occasion.[8]

In addition to these forms of special vulnerability which apply to VCA candidates and may compromise their ability to provide adequate initial consent, it should be recognized that the treatment itself imposes upon the recipient new forms of vulnerability which must be borne by the patient thereafter. A recipient is susceptible to harm from comorbidities associated with the surgery, post-surgical infections and both acute and chronic graft rejection. The recipient is also susceptible to harms associated with the long-term use of immunosuppression, such as increased infection risk, the development of diabetes, kidney damage, and increased rates of malignancy (O'Neill and Godden, 2009; Hautz et al., 2011; Shores et al., 2011; Pomahac et al., 2012). In order to manage this new vulnerability, the recipient must carefully maintain the schedule of immunosuppression, participate actively in physiotherapy for years, and self-monitor for signs of rejection for the rest of his or her life. The patient must also reckon with the likelihood that the decision he or she has made to have the transplant may contribute directly to an earlier death. Support in coping with the complications and carrying out the responsibilities is one means of offsetting the added vulnerability.

PATIENT ADVOCACY

The terms "patient advocacy" and "patient advocate" appear frequently in the literature of healthcare but lack any singular or settled definition. "Patient advocacy" has been used to describe efforts of patients themselves to obtain access to or improve treatment (Epstein, 1995; Brashers et al., 2000), as a description of a central feature of the nursing ethos (Bu and Jezewski, 2007; Choi, 2015), and as a description of individuals whose primary role is to assist patients in navigating their way through the complexities of modern health care systems.[9] Patient advocacy in all its forms exists to redress conditions which place patients at a disadvantage, particularly power differentials between patients and providers or patients and systems (Erlen, 2006; Reid, 2022).

Patient advocacy in the form of assisting patients as they navigate their way through treatment may assume different foci at different times. It may focus on pursuit of the patient's best interests, protection and promotion of the patient's rights, formal representation of the patient, or empowerment of the patient by providing information, assuring understanding and providing emotional support (Brazg et al., 2016; Abbasinia et al., 2020). Regardless of the particular focus, the primary duty of the patient advocate is to the vulnerable patient. In the words of Bragz et al., "Patient advocacy is one response to patients' experiences of vulnerability, and it can be utilized as a tool to improve patients' participation and engagement in their healthcare."[10]

The value of a patient advocate has already been recognized in research (Cauhan and Eppard, 2004; Katz et al., 2012; Salamone et al., 2018) and in the field of transplantation. In the context of living donation, the Organ Procurement and Transplantation Network (OPTN) requires the involvement of an independent living donor advocate (ILDA). The responsibilities of this particular kind of patient advocate are to "represent, advocate, protect and promote the best interests" of those who express an interest in donating an organ while alive, by providing information about the process and risks, ensuring free, uncoerced and fully informed consent and providing support for those prospective donors who are not allowed to donate.[11] The assumptions behind the requirement for an ILDA is that potential donors may be vulnerable due to a lack of knowledge or failure to appreciate the burdens, risks and possible negative outcomes for both donor and recipient. Potential donors may also be vulnerable to coercion, especially if the person in need of a transplant is a spouse, sibling, parent or child of the donor.

THE LOUISVILLE EXPERIENCE

From 1999[12] to 2011, a team in Louisville, Kentucky, involving the Jewish Hospital, the Christine M. Kleinert Institute for Hand and Microsurgery and the University of Louisville, performed hand transplantation on six patients. Preparation for these transplants began in 1995 when a group of hand surgeons, transplant surgeons, psychiatrists, nephrologists, physical therapists, nurses, tissue typing lab specialists, ethicists and organ procurement organization representatives came together to envision how to create a program. Initial discussions led to a commitment to undertaking a great deal of basic science research and work in large animal (swine) models prior to attempting a transplant on a human patient.

Another major commitment of the Louisville program from the very start was a commitment to ethical reflection, transparency and accountability. The program sought out advice from Dr. Siegler, director of MacLean Center for Clinical Medical Ethics at the University of Chicago. Among Siegler's recommendations was that the team should announce its intentions prior to its first attempt, rather than wait to see whether the procedure would be a success before deciding if it would be publicized. This approach would heighten their

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[8] May captures the profound challenge of illness and treatment for patients. He perceptively reminds us. "[T]o equate the heroic with the aggressiveness of the doctor's technical interventions cruelly overlooks those who bear the true weight of heroism.... the heavy burden of heroism in medicine falls not on the physician but on the patient and the patient's family, as they often face, after the successful rescue, an extraordinarily long and heavy responsibility of chronic care." (William, 1991).

[9] See, for example, the website of the Alliance of Professional Health Advocates. Available online at: https://www.aphadvocates.org/profession-overview/ (accessed September 9, 2017).

[10] Bragz, et al., 177. See also Erlen, 134, who contends that advocacy "may reduce the level of vulnerability."

[11] OPTN has also extended the requirement for an ILDA to VCA, in the event of living donation. At present, uterine transplantation may use living donors; other possibilities for living donation in VCA have been imagined as well. See "VCAs from Living Donors. US Department of Health and Human Services OPTN (Vittone and Crowell, 2021). Available online at https://optn.transplant.hrsa.gov/resources/by-organ/vascular-composite-allograft/vcas-from-living-donors/ (accessed September 9, 2017) (Hays et al., 2015; Vittone and Crowell, 2021).

[12] Information about the Louisville program was obtained through personal communication with Warren Breidenbach, MD, who led the program at Louisville from 1999-2011.

accountability. Another recommendation from Dr. Siegler was that the team consider including a patient advocate for each patient. This recommendation was embraced, and a patient advocate was involved for each of the first six patients at Louisville.

The first meeting with a prospective patient began with introductions to the surgeon and staff. The patient was asked to explain why they wanted a hand transplant, and given an initial introduction to the risks. Psychological and general medical evaluation followed, and the first encounter ended with the candidate being urged to consider the options and risks thoroughly before deciding whether to schedule of second appointment.

At the second appointment, a much more detailed presentation of the procedure and potential complications took place. A potential patient was also informed that a 6-month trial with a prosthesis would be mandatory before the patient could become eligible for a transplant. The patient was also informed of the need for further psychological testing and that an analysis of the family and social support system would have to take place.

After 6 months or more, if the outcomes of the various screenings were acceptable and the patient continued to be interested, the patient was introduced to the concept of a patient advocate. It was explained that the patient was expected to select his or her own advocate, who should be someone who knew them well but not a family member. The role of this advocate was to accompany the patient through the remaining process prior to surgery and help the patient reach a free and informed decision. The ideal patient advocate was someone who had at least some familiarity with medical terms, the ability to identify and weigh burdens, risks and potential benefits, and the ability to construct and communicate a recommendation.

The advocate would have access to all the information about the treatment that was available to the patient, and could ask questions of the treatment team. The transplant team was prohibited from trying to exert influence over the advocate in any way. While the advocate would make a recommendation on whether to proceed, the final decision was up to the patient.

PATIENT ADVOCACY FOR VCA CANDIDATES AND RECIPIENTS

If it is accepted that VCA candidates and recipients are properly regarded as being especially vulnerable, and that patient advocacy is a reasonable way of addressing the needs of vulnerable patients in health care, then the provision of patient advocates for VCA candidates and recipients may be recognized at least as a moral good, and perhaps as an ethical duty. The anticipated role of a patient advocate in VCA would include (but not necessarily be limited to) (1) assistance in the pursuit of information and in the deliberation leading up to the decision of whether or not to give formal consent and (2) continuing support through rehabilitation and adjustment to post-transplant life (Caplan et al., 2019).

Important traits and skills for patient advocacy in VCA include independence from both the health care team and from the patient. Ideally, the patient advocate should be neither an employee of the health care system providing the treatment nor a family member or intimate friend of the patient. Advocates should know the patient well-enough to have a sense of their values and to be aware of their psychosocial strengths and weaknesses, yet not be so close that the advocate would hesitate to challenge the patient's thinking. Independence from both the team and the patient allows the advocate to express himself or herself without excessive concern about how it may be received by either the care team or the patient. Some degree of health care literacy would be important, so that the advocate would not have difficulty understanding information and could potentially "translate" information for the patient as needed. Communication skills are obviously essential, as is psychosocial stability and emotional intelligence.[13]

The involvement of a patient advocate in VCA does not imply that candidates or patients need protection from the transplantation team, *per se*. Rather, it is based on the recognition that even the most caring and careful transplantation team is inherently limited in its ability to address the full range of the patient's vulnerability. Despite best intentions and efforts, it may be impossible for the team to adequately appreciate the strengths, weaknesses and perspectives of the patient and his or her support system.[14] Evidence of the difficulty of doing so is reflected in the frequent assertion that improvement in patient selection is a primary need in the field (Edwards and Mathes, 2011; Kiwanuka et al., 2013; Jowsey-Gregoire et al., 2016; Shores et al., 2017). It may also be impossible for the team to adequately address the post-surgical vulnerability of the patient, particularly when he or she lives at some distance from the center where the team practices.

Likewise, the involvement of a patient advocate does not imply that the patient lacks decisional capacity. The patient advocate does not serve as a surrogate decision maker, but as a trusted counselor. The patient advocate may raise questions, help assure patient comprehension, and offer opinions, but the patient advocate should not be given the authority to override the patient's choice.

The involvement of an independent patient advocate also provides ethical protection for both patients/candidates and transplantation teams who share in the universal predisposition to self-justification and self-deception. The investments of time and money that must be made to establish VCA programs, as well as the potential rewards in terms of the economy of fame,[15] create significant pressures on individuals to justify what they desire or what is necessary for them to achieve a

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[13] Emotional intelligence is defined as "the ability to monitor one's own and other people's emotions, to discriminate between different emotions and label them appropriately, and to use emotional information to guide thinking and behavior." See Oxford University Press (2015).

[14] At least one such program goes so far as to spend several days visiting in the candidate's home community, attempting to gain a better understanding. (Personal communication with Eduardo Rodriguez, of NYU-Langone Medical Center).

[15] For an excellent article on the role of the economy of fame in the development of science, see Franck (2015).

given status. This results in the creation of "confirmation bias" (Haynes and Haynes, 2009; Mendel et al., 2011) in the assessment of candidates. The same pressures may apply to candidates, who may engage in similar practices of self-justification and self-deception. The role of the patient advocate is to be an independent interlocutor, who can raise questions, challenge reasoning, and offer alternative perspectives.

CONCLUSION

Candidates for upper extremity and facial VCA exhibit characteristics associated with special social and medical vulnerability. In addition, the complexity and relative lack of data on these forms of VCA increase their vulnerability. It is an established practice elsewhere in some research and in living donor transplantation to provide a patient advocate to support, advocate for and protect subjects and patients. Furthermore, the use of patient advocates early in the Louisville program demonstrates the feasibility of incorporating patients advocates into VCA practice. It is therefore recommended that serious consideration be given to the recruitment, training and use of patient advocates in upper extremity and facial VCA in the future.

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The author confirms being the sole contributor of this work and has approved it for publication.

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The psychosocial burden of visible disfigurement following traumatic injury

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Hundreds of thousands of individuals experience traumatic injuries each year. Some are mild to moderate in nature and patients experience full functional recovery and little change to their physical appearance. Others result in enduring, if not permanent, changes in physical functioning and appearance. Reconstructive plastic surgical procedures are viable treatments options for many patients who have experienced the spectrum of traumatic injuries. The goal of these procedures is to restore physical functioning and reduce the psychosocial burden of living with an appearance that may be viewed negatively by the patient or by others. Even after receipt of reconstructive procedures, many patients are left with residual disfigurement. In some, disability and disfigurement may be so profound that individuals are candidates for vascularized composite allotransplantation (VCA) procedures, i.e., the transplantation of a vascularized human body part containing multiple tissue types (skin, muscle, bone, nerves, and blood vessels) as an anatomical and/or structural unit. This narrative review paper summarizes the literature on the psychosocial burden experienced by those who have visible disfigurement. While many of these individuals experience stigma and discrimination, relatively few studies have employed a stigma framework to understand the psychosocial sequelea. This paper briefly addresses this framework. Last, particular focus is given to the psychosocial issues of individuals with particularly severe injuries who are potential candidates for VCA procedures.

KEYWORDS

vascularized composite allotransplantation, psychosocial issues, visible disfigurement, stigma, traumatic injuries

Traumatic injury

Traumatic injuries, both those experienced unintentionally through accidents and those that are violence-related, are estimated to constitute approximately 8% of deaths around the world each year (World Health Organization [WHO], 2021). In 2020 in the United States, traumatic injuries accounted for over 270,000 deaths

(Centers for Disease Control and Prevention [CDC], 2022). Over 3 million Americans are estimated to experience non-fatal injuries per year (American Association for the Surgery of Trauma, n.d.). These traumatic injuries are believed to account for an estimated 10% of the life years that an individual lives with a disability (World Health Organization [WHO], 2021). The health care costs associated with acute and chronic treatment of persons who have suffered traumatic injuries is staggering. In 2019, the cost of injuries from fatal and non-fatal injuries was $4.2 trillion (Centers for Disease Control and Prevention [CDC], 2021). This cost includes medical care, work loss, statistical, and quality of life losses (Peterson et al., 2021). The experience of a traumatic injury increases the risk of mental health issues, substance misuse, chronic disease such as cardiovascular disease and type 2 diabetes, as well as poverty, crime, and violence (Pacella et al., 2013; Hughes et al., 2017). There is a dose response between the lifetime experience of traumatic events and the increased odds of developing significant health issues over time (Scott et al., 2013).

The experience of traumatic injuries is particularly high among specific groups of individuals. Severe automobile accidents, injuries from firearms, workplace injuries, fires, and unsuccessful suicide attempts can leave individuals with traumatic injuries. The number of individuals who suffer these injuries around the world each year is difficult to calculate; more reliable numbers are available from selective groups. For example, in 2006, over 8,000 active duty American military personnel suffered bodily injuries resulting in hospitalization (Jones et al., 2010). Between 2003 and 2011, approximately 7,200 American soldiers stationed in Iraq and Afghanistan sustained significant head and neck injuries (Brennan, 2015). During Operation Iraqi Freedom, wrist, hand, and finger injuries accounted for 28.7% of all extremity injuries (Dougherty et al., 2009). Approximately 30% of military personnel who sustained an extremity injury (either upper or lower) also experienced an injury to the face. While the nature of these injuries is often severe, the survival rate from them is high (Dougherty et al., 2009).

The experience of trauma, and the risk of the most profoundly deleterious effects, is highest among those individuals from underserved groups, making traumatic injuries a significant public health issue (Merritt and Benningfield, 2019). As with other major public health issues, social determinants of health increase the risk of both experiencing a traumatic injury and potentially threaten a successful course of physical and/or mental health treatment following the experience.

Medical care of traumatic injuries

The course of recovery from a traumatic injury is influenced by the delivery of high quality health care at all points in the treatment process. Quality emergency care can reduce the risk of fatality, disability, and psychosocial adjustment. High quality rehabilitation services and the promotion of community inclusion by removing barriers to social and economic participation can ensure that people who experience disability following a traumatic injury enjoy the fullest life possible.

Plastic surgeons are often centrally involved in the care of traumatic injuries. The American Society of Plastic Surgeons (2021), for example, reported that approximately 6.9 million reconstructive surgical procedures were performed in 2020. The most common were tumor removal, laceration repair, scar revision, and maxillofacial and hand procedures. In addition to treating functional issues, these procedures have a major goal of restoring physical appearance to an approximation of normal.

Advances in medical and surgical care have improved the survivability and rehabilitation of individuals who have suffered severe, traumatic injuries (Holcomb et al., 2006; Eastridge et al., 2012). However, even the most successful reconstructive surgical procedures leave patients with some degree of residual deformity. For example, an individual who falls off of a bicycle and suffers facial fractures and lacerations may have visible scarring for the rest of her life. In the case of limb loss, some individuals have the option of prosthetics. Advances in prosthetic technology has improved the physical functioning and quality of life of many individuals with disabilities (Magee et al., 2011). Some individuals show tremendous resilience in their ability to adapt to living with profound disability and disfigurement. Others, unfortunately, are unable to benefit from prosthetic arms and hands (Grunert, 2006; DeBerard and Goodson, 2013). The resulting functional limitations often reduce their ability to perform work-related tasks and negatively impact self-esteem, body image, and quality of life. Many of these individuals suffer with concomitant and significant psychosocial burden in conjunction with the residual disability and disfigurement (Grunert, 2006; Magee et al., 2011; DeBerard and Goodson, 2013).

Psychological response to traumatic injury

Individuals can experience a wide range of psychological reactions to a traumatic injury, regardless of whether or not the injury results in permanent changes to one's body and physical appearance. From a psychiatric perspective, a traumatic event, whether it involves a traumatic injury or not, must involve actual or threatened death or serious injury or sexual violence. The individual experiencing the event also must experience intense fear, helplessness, or horror (American Psychiatric Association [APA], 2013).

Two psychiatric diagnoses of relevance to traumatic events are acute stress disorder (ASD) and posttraumatic stress disorder (PTSD). Both ASD and PTSD are diagnosed in persons who experienced (or witnessed) a traumatic event and experience symptoms including re-experiencing the traumatic event in memories, intrusive thoughts, dreams, or flashbacks, avoiding reminders of the event (including medical or mental health treatment visits), and increased feelings of arousal, anxiety, and depression. In ASD, these symptoms begin within 4 weeks of event and last for less than 1 month. PTSD may develop from ASD, but symptoms last for longer than 1 month. In both disorders the symptoms must cause significant distress or impairment.

Not everyone who experiences a traumatic injury experiences ASD and/or PTSD (Resick et al., 2008). While men are exposed to more trauma, women have higher rates of ASD and PTSD (Resick et al., 2008). While it can be difficult to predict why some individuals develop these traumatic stress reactions and others do not, some variables have been identified (Magee et al., 2011). Several social determinants of health (poverty, neighborhood crime), mental health problems in others in the home, and a lack of social support are associated with less positive reactions to trauma (Schroeder et al., 2021). The experience of adverse childhood experiences includes situations of physical, emotional, or sexual abuse, physical and emotional neglect, as well as exposure to household stressors including family member's substance use, mental illness, incarceration, intimate partner violence, divorce/separation, or death also is associated with greater psychological difficulties with later life trauma (Felitti et al., 1998).

Depression is common among survivors of traumatic injuries. For those who have a visible disfigurement from their injuries, social anxiety disorder may also be observed. It is defined as a marked and persistent fear of one or more social or performance situations in which the person is exposed to unfamiliar people or to possible scrutiny by others (American Psychiatric Association [APA], 2013). The individual fears that he or she will act in a way (or show anxiety symptoms) that will be humiliating or embarrassing. Exposure to the feared social situation almost invariably provokes anxiety, which may escalate to a panic attack. The avoidance, anticipation, or distress in the feared situation interferes significantly with the person's typical, daily functioning.

Psychosocial issues in military veterans

There is a sizable literature on the mental health issues of military personnel who have suffered traumatic injuries or witnessed traumatic events (Hoge et al., 2006; Seal et al., 2007; Kessler et al., 2014; Hom et al., 2017; Kraus, 2017). Individuals who serve in the military are found to have high rates of depression, anxiety disorders, substance abuse, and suicidality following discharge (Hoge et al., 2004; Eisen et al., 2012; Gadermann et al., 2012; Sirratt et al., 2012; Fisher, 2015). Approximately 20% of American military personnel who returned from the Iraq war screened positive for mental health disorders, including PTSD, anxiety, or depression (Hoge et al., 2004; Eisen et al., 2012). The rates of these conditions are even higher among those who were engaged in combat as well as those who experienced and/or witnessed a traumatic event that resulted in injury to themselves or fellow soldiers (Clemency Cordes et al., 2016).

Mental health issues may be of particular concern to veterans who have suffered disfiguring injuries. Approximately 40% of individuals exposed to trauma receive a diagnosis of PTSD (O'Donnell et al., 2003). The rate of PTSD among those who suffered debilitating injuries is believed to be even higher (Fisher, 2015). The permanent and visible nature of these disfiguring injuries serves as a daily reminder of their trauma and is believed to be a stimulus to the development and/or maintenance of PTSD (Van Loey et al., 2003).

Depression also is common. For example, nearly 25% of veterans who suffered traumatic limb loss have been diagnosed with a mood disorder (Reiber et al., 2010). Either as a consequence of or concomitant to depression, substance abuse rates among military veterans are high (Seal et al., 2011). Suicidality is a particularly troubling concern; estimates suggest that approximately 20 American military veterans attempt to end their own lives daily (Department of Veterans Affairs Office of Suicide Prevention, 2016). The rates of these issues among those appearance altering disfigurement is likely even greater.

Combat related injuries that profoundly disfigure military personnel likely have the potential to produce a traumatic brain injury (TBI). In the last several years, the occurrence of TBI among military personnel, as well as the general public, has received increased research and clinical attention. Approximately 440,000 military personnel have experienced a TBI since 2000 (Traumatic Brain Injury Center of Excellence [TBICoE], 2021). In contrast, approximately 1 million civilian Americans are believed to experience a TBI annually; 50,000 die from these injuries and approximately 8–10% have chronic symptoms (Centers for Disease Control and Prevention [CDC], 1999).

While some of these injuries are mild and resolve over time, others have long-term effects. Individuals with TBIs may experience neurocognitive deficits, PTSD, depression, anxiety, substance abuse, and suicidality (Hoge et al., 2008; Litz and Schlenger, 2009; Bryant et al., 2010; Carlson et al., 2011; Mallya et al., 2015; Haabauer-Krupa et al., 2017; Swan et al., 2018). Persistent post-concussion symptoms have been reported in as many 85% of veterans who experienced a TBI during wartime service (Morissette et al., 2011). Individuals with long-lasting, unremitting impairments often report vestibular symptoms, such as dizziness, as well as postural and gait disturbance (Akin and Murnane, 2011; Baldassarre et al., 2015; Howell et al., 2015; Leddy et al., 2015). Other visual-vestibular symptoms that persist beyond the acute period include sensitivity to visual motion and deficits in oculomotor function (Ciuffreda et al., 1996; Kapoor and Ciuffreda, 2002; Heitger et al., 2009; Capo-Aponte et al., 2012; Mucha et al., 2014; Wright et al., 2015; McDevitt et al., 2016; Wright et al., 2017; Cheever et al., 2018).

These symptoms also are associated with impairments in psychosocial functioning (Bryant, 2008; Hoge et al., 2008; Wares et al., 2015). As noted above, PTSD is common in military veterans, although the TBI-PTSD relationship remains somewhat unclear (Carlson et al., 2011; Haabauer-Krupa et al., 2017). The prevalence of PTSD in military veterans is commensurate with the rate of TBI, with over 400,000 Veterans being seen for potential or provisional PTSD at Veteran Affairs facilities (Department of Veterans Affairs et al., 2017). TBI and PTSD share several symptoms, including concentration and/or memory loss, depression, irritability/anger, anxiety, as well as dizziness and loss of balance (Kennedy et al., 2007; Walker et al., 2014; Storzbach et al., 2015; Wares et al., 2015). Further complicating the relationship is the observation that TBI often occurs as a result of a traumatic event, a prerequisite for the diagnosis of PTSD (Servatius et al., 2017).

Physical appearance and body image

The experience of a traumatic injury that results in visible disfigurement likely comes with additional psychosocial burden. Over the past 50 years, social psychologists have created a sizable body of research on the role of physical appearance in daily life. This research has repeatedly demonstrated that individuals who are less physically attractive are judged and treated less favorably than those who are more attractive (Eagly et al., 1991; Etcoff, 2000; Langlois et al., 2000; Sarwer and Spitzer, 2012a; Swan et al., 2018). For example, less attractive individuals are rated as being less intelligent, friendly, and kind than those who are more attractive.

Studies of individuals with facial and body disfigurement have replicated these findings (Tobiasen, 1987; Rankin and Borah, 2003; Mojon-Azzi et al., 2008; Masnari et al., 2013; Jamrozik et al., 2019). These results are consistent with the tenants of social-cognitive theory which postulates that portions of an individual's knowledge acquisition, including understanding of the dynamics of social situations, results from observing others in the context of social interactions (Bandura, 1986). The theory states that when people observe an individual performing a behavior, and witness the results of that behavior, they remember the sequence of events and use this information to guide subsequent behaviors.

In the case of visible disfigurement, we learn from watching the behaviors of others that persons who look different should be treated different. In the case of those who are disfigured, that treatment is less favorable. Persons with facial disfigurement are rated as less attractive and assumed to have less positive personality traits as compared to those who are less disfigured or non-disfigured (Jamrozik et al., 2019). This has recently been described as "What is Anomalous is Bad," where those with facial and body disfigurement are believed to have less positive personality traits and are more likely to engage in non-positive behavior (Workman et al., 2021). This is in contrast to the long standing "What is Beautiful is Good" bias (Dion et al., 1972). From a recent study using fMRI, it appears that these negative responses are "hardwired" into the occipito-temporal cortex as well as anterior cingulate cortex of the brain (Hartung et al., 2019), which may explain the particularly negative reactions that facial disfigurement illicits (Stone and Potton, 2019; Rasset et al., 2022). Encouragingly, plastic surgical procedures that minimize disfigurement result in more positive perceptions of individuals with disfigurement (Mazzaferro et al., 2017; Jamrozik et al., 2019).

At the same time, studies have suggested that perceptions of our own appearance, otherwise known as body image, play a significant role in quality of life and self-esteem (Sarwer and Spitzer, 2012a; Sarwer and Polonsky, 2016). In studies of individuals without visible disfigurement, there is either no relationship or a weak relationship between an individual's objective appearance and their subjective body image (Sarwer et al., 2011). In studies of individuals with visible disfigurement, there is a more consistent relationship between the severity of disfigurement and degree of body image dissatisfaction (Rumsey and Harcourt, 2012; Crerand et al., 2017). Dissatisfaction with one's appearance and body image is believed to be the motivational catalyst to pursue plastic surgical procedures to improve appearance (Sarwer and Crerand, 2008; Sarwer and Spitzer, 2012b).

Quality of life is a multifactorial construct that involves an individual's degree of satisfaction and level of functioning in several core domains, including physical functioning, psychological well-being, as well as social and work role performance (Ware and Sherbourne, 1992). Body image is an important aspect of quality of life and is a highly relevant psychosocial construct for individuals who are disfigured. Along with pre-injury factors (such as premorbid psychopathology and social determinants of health), peri-traumatic factors (i.e., TBI, functional limitations, pain), and post-injury factors (i.e., social support), quality of life is believed to play a central role in adaptation to a disfiguring condition (Fauerbach et al., 2006; Block and Sarwer, 2013).

The psychosocial burden of living with a disfigured facial appearance cannot be underestimated. Much current understanding of this burden comes from studies of children born with cleft lip and/or palate or more profound craniofacial anomalies. In brief, children born with these conditions are at risk for significant psychosocial issues including depression, social anxiety, and reduced quality of life (Hunt et al., 2005;

Demir et al., 2011; Broder et al., 2012; Tyler et al., 2013). Psychological functioning is related to individuals' satisfaction with their facial appearance and body image (Moss, 2005; Magee et al., 2011; Crerand et al., 2015; Feragen et al., 2015). Adults with a visible disfigurement also report low self-esteem, body image dissatisfaction, depression, and anxiety (Pallua et al., 2003; Rumsey et al., 2004; Magee et al., 2011; Fingeret et al., 2012; Wisely and Gaskell, 2012; Akyol et al., 2013; Keeling et al., 2020). In a study of 458 adults with a range of visible disfigurements, 48% were judged to have symptoms of an anxiety disorder, and 28% had depression (Rumsey et al., 2004). In a separate study, 56% of patients with facial disfigurement were judged in need of mental health treatment (Strauss and Broder, 1991).

In summary, the current evidence base suggests that having a visible disfigurement may increase vulnerability to psychological distress – including depression, anxiety, and body image dissatisfaction. This distress may in part result from negative interactions with others, and/or the anticipation of unwanted attention due to their appearance. Anecdotal reports and a limited body of research suggest that stigma, if not outright discrimination, are common experiences among those with facial or body disfigurement. However, the resulting psychosocial and physical consequence of these experiences is less fully understood at present.

Stigma and discrimination

Goffman (1963), arguably the world's first scholar of stigma, began articulating categories and types of stigma over a half century ago. More contemporary scholars have defined stigma as the devaluation of social identities based on the recognition of difference based on some distinguishing characteristic (Dovidio et al., 2000). There are different levels of stigmatization. At the intrapersonal level, individuals may self-stigmatize their thoughts and feelings about a physical trait. The literature on the psychosocial burden of visible disfigurement detailed above falls at this level. At the interpersonal level, individuals may experience unwanted attention or treatment from others. At the structural level, individuals may encounter systematic, unfair treatment due to policies and practices that perpetuate stigma and discrimination.

A number of studies have documented the interpersonal stigma experienced by those with visible disfigurement (Rumsey and Harcourt, 2012). There are social consequences to having a disfigured appearance. As noted above, individuals with abnormal facial appearance are judged less positively as compared to those with normal facial characteristics (Tobiasen, 1987; Rankin and Borah, 2003; Mojon-Azzi et al., 2008; Masnari et al., 2013; Jamrozik et al., 2017). Individuals with facial disfigurement have been found to be the target of unwanted staring as well as negative appearance-related comments (i.e., teasing or bullying) from others (Sarwer et al., 1998, 1999; Turner et al., 1998; Strauss et al., 2007; Magin et al., 2008; Nishikura, 2009; Bonanno and Choi, 2010; Feragen and Borge, 2010; Lawrence et al., 2011; Bogart et al., 2012; Griffiths et al., 2012; Bogart, 2015; Sobanko et al., 2015; Halioua et al., 2017; Martin et al., 2017; Holland et al., 2019; Visram et al., 2019). The occurrence of this unwanted attention is associated with increased symptoms of depression and lower self-esteem (Rumsey et al., 2004; Crerand et al., 2017). Interestingly, some research has suggested that persons with facial disfigurement are seen less positively than those with other visible physical disabilities (Stevenage and McKay, 1999; Stone and Wright, 2012; Bogart et al., 2019).

Over the past two decades, research on stigma and discrimination of persons with obesity has blossomed (Puhl et al., 2020). This work can be used to further understand the interpersonal and structural levels of stigma in persons with visible disfigurement. Although obesity differs from visible disfigurement in several ways (e.g., obesity is generally viewed as more controllable than disfigurement and thus elicits more blame), it is similar in respect to being visible and impacting physical appearance. In a classic study, children ages 10–11 years were asked to rank order their liking of six children depicted with differences in appearance: obesity, facial disfigurement, wheelchair, crutches, missing hand, or no disability (Richardson et al., 1961). Across multiple groups, the child with obesity and the child with facial disfigurement were rated as the least preferred peer.

Weight-based stigma and discrimination is common. Data from the National Survey of Midlife Development in the United States indicated that rates of weight-based discrimination increased by 66% from 1995 to 2006 (Andreyeva et al., 2008; Puhl et al., 2008). This discrimination is not benign; it has been associated with mental health consequences. In a study of over 22,000 United States adults with overweight and obesity, over half who reported experiencing weight-based discrimination met criteria for at least one mood, anxiety, or substance use disorder; furthermore, the odds for meeting criteria for three or more comorbid disorders were 2.4 times higher than for individuals who had never experienced weight discrimination. Weight discrimination also has been associated with increased risk for all-cause mortality.

Our cultural fascination with physical beauty, as perpetually reinforced by the mass media and entertainment industries, contributes to the structural stigmatization of those with visible disfigurement (Griffiths and Mullock, 2018; Sarwer and Spitzer, 2021). Physically beautiful faces and bodies dominate the screens of movie theaters, televisions, and hand held devices. Most individuals are susceptible to engaging in social comparison to these images resulting in increased body image dissatisfaction for the viewer (Sperry et al., 2009). When those with visible disfigurement are represented in the media, they are more often or not in the role of the evil enemy—from Disney films to the James Bond movie franchise (Sarwer, 2021). These depictions reinforce the "what is anomalous is bad" stereotype.

Stigma and discrimination against those with visible disfigurement is also common (Swift and Bogart, 2021). Adults with facial or body disfigurement have been found to suffer from stigmatization in social situations, including friendships and romantic relationships, as well as overt discrimination in employment (Porter et al., 1986; Koster and Bergsma, 1990; Stone et al., 1992; Ginsburg and Link, 1993; Sarwer et al., 1998; Thompson and Kent, 2001; Horgan and MacLachlan, 2004; Rumsey and Harcourt, 2004; Tartaglia et al., 2005; Saradjian et al., 2008; Wisely and Gaskell, 2012; Mathias and Harcourt, 2014; Sharratt et al., 2018). Almost two-thirds of adults with a facial disfigurement reported avoidance of social situations and 71% of individuals reported that others had not wanted to become romantically involved with them because of their appearance (Broder et al., 2012). Almost 50% of these adults reported that their appearance had affected whether they had been hired for a job (Wisely and Gaskell, 2012).

Application to the evaluation of candidates for vascularized composite allotransplantation

Vascularized composite allotransplantation (VCA) holds great promise for individuals with profound disfigurement and who have not had an acceptable response to the current generation of reconstructive procedures or prosthetics (Siemionow et al., 2009; Hautz et al., 2011; Pomahac et al., 2011; Khalifian et al., 2014; Dean and Talbot, 2017; Siemionow, 2017). To date, over 100 of these procedures have been performed worldwide. Many results have been quite impressive. Presently, active programs of research are investigating advances in surgical treatment and immunosuppression for persons who undergo these procedures (Siemionow et al., 2009; Pomahac et al., 2011; Siemionow, 2017). Similarly, the Chauvet Group, an international group of mental health and medical professionals, is considering the mental health issues in persons who undergo VCA (Kumnig et al., 2014; Jowsey-Gregoire et al., 2016; Kumnig and Jowsey-Gregoire, 2016).

While there is enthusiasm that VCA holds great potential for individuals who experience severe traumatic injuries, as well as those who have lost limbs secondary to systemic illness such as sepsis or cancer, little is known about the psychosocial issues of individuals who may be potential candidates. Many suffer with PTSD, mood, and anxiety disorders; others may overuse substances to address issues of physical or psychological pain. Issues with self-esteem, quality of life, and body image dissatisfaction are common. The psychosocial burden may be so severe that it contraindicates a VCA procedure. Anecdotal reports suggest that approximately one-third of patients who have approached established VCA programs around the United States have not undergone a procedure because of concerns about their psychiatric status. Other reports suggest that a subset of patients who have undergone VCA have experienced a failure of the procedure secondary to behavioral non-compliance (typically with immunosuppression medications) or have requested amputation of their hands.

Most VCA programs require patients to undergo a mental health evaluation as part of the preoperative evaluation process. While the Chauvet group and others have offered some guidance on the nature and structure of these evaluations, an established standard of care has yet to be established. The mental health professional, as well as all members of the VCA team, must consider not only issues of physical functioning, but the degree of psychosocial stress and likely benefit of a successful procedure (Caplan et al., 2019). Compliance with the postoperative immunosuppression treatments and other elements of care are likely psychologically challenging as well. Psychosocial status and functioning may leave one patient better suited for the postoperative challenges and threats to long-term success of the procedure than another. Resiliency is a likely pre-requisite, but is a notoriously difficulty psychological construct to predict and assess. As written by Butler and colleagues before the first VCA procedure was ever performed:

> . . .it may be that people who have well-developed coping strategies and good social skills cope well with disfigurement, while those who find life generally more challenging, also cope poorly with disfigurement. The concern for us as clinicians. . .is that this group may also cope poorly with face transplantation; thus, the very group who might benefit most are those who are least likely to cope. . .., particularly if the results fall short of their expectations (Butler et al., 2004, p. 17).

Candidates and family members may believe that a successfully performed VCA procedure is critical to a new, post-injury life. Yet, the full extent of interest in and attitudes toward VCA among veterans and active military men and women is still largely unknown. The reality is that many individuals with profound disfigurements lead meaningful lives; some dedicate themselves to improving the treatment of others with these types of injuries.

As is the case for other forms of transplantation, support from family members, health care providers, and close friends is key to successful post-VCA adaptation. Patients' social and health networks can also support and weigh in on the decision to pursue VCA as a treatment option. Additional research is needed to understand the nuances of patient, provider, and caregiver attitudes and perspectives on the VCA decision. Caplan et al. (2019) have suggested that patient advocates can provide an additional layer of protection for VCA candidates struggling with mental health issues and considering the risks and benefits of the procedure. Such advocates also can help VCA recipients challenged by compliance with the intense requirements of postoperative medical care.

Conclusion and future directions

Traumatic injuries leave hundreds of thousands of individuals with permanent changes in physical functioning and appearance annually. Multidisciplinary care and treatment can help improve physical functioning, but many individuals experience permanent changes in physical appearance. Some experience untoward reactions to the traumatic event; others struggle with mood and anxiety. Self-esteem, body image, and quality of life is negatively impacted for many. Those with visible disfigurement are routinely stigmatized in a range of interpersonal situations and some experience outright discrimination from others. The psychosocial burden of living with disfigurement in a society where physical beauty is so idealized simply cannot be overstated.

For some individuals with profound disability and disfigurement, VCA offers a new treatment option. Literature on the surgical treatment and medical management of patients who undergo VCA procedures is growing. Understanding of the psychosocial issues of persons who are potential candidates for VCA is still in its early stages, with a limited number of case reports and expert opinion articles in the literature. The relationship between impairments in physical functioning, the psychosocial status and functioning of potential candidates, and their appropriateness for a VCA procedure remains poorly understood.

Robust, hypothesis-driven research on the psychological comorbidities of individuals who have suffered injuries that would potentially make them appropriate for VCA procedures would provide critically important information that could inform who might best benefit from VCA. This work would allow thought leaders to provide more specific information on the psychosocial factors which affect candidacy for VCA. Such information can be used to refine the psychosocial patient selection criteria and its role in optimizing postoperative outcomes. Subsequent standards of care can then be articulated and ensure that candidates for VCA procedures at programs around the world are being appropriately evaluated and treated as they undergo these profoundly life enhancing procedure, and, ideally, lessen the psychosocial burden of living with a visible disfigurement.

Author contributions

DS wrote the initial draft of the manuscript. LS, HG, and JS wrote and revised sections of the manuscript. All authors subsequently read, revised and approved the submitted version.

Conflict of interest

DS currently has grant funding from the National Institute of Dental and Craniofacial Research (R01 DE026603) and Department of Defense. He has consulting relationships with Ethicon and Novo Nordisk.

The remaining authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Information needs and development of a question prompt sheet for upper extremity vascularized composite allotransplantation: A mixed methods study

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Background: People with upper extremity (UE) amputations report receiving insufficient information about treatment options. Furthermore, patients commonly report not knowing what questions to ask providers. A question prompt sheet (QPS), or list of questions, can support patient-centered care by empowering patients to ask questions important to them, promoting patient-provider communication, and increasing patient knowledge. This study assessed information needs among people with UE amputations about UE vascularized composite allotransplantation (VCA) and developed a UE VCA-QPS.

Methods: This multi-site, cross-sectional, mixed-methods study involved in-depth and semi-structured interviews with people with UE amputations to assess information needs and develop a UE VCA-QPS. Qualitative data were analyzed by thematic analysis; quantitative data were analyzed by descriptive statistics. The initial UE VCA-QPS included 130 items across 18 topics.

Results: Eighty-nine people with UE amputations participated. Most were male (73%), had a mean age of 46 years, and had a unilateral (84%) and below-elbow amputation (56%). Participants desired information about UE VCA eligibility, evaluation process, surgery, risks, rehabilitation, and functional outcomes. After refinement, the final UE VCA-QPS included 35 items, across 9 topics. All items were written at a ≤ 6th grade reading level. Most semi-structured interview participants (86%) reported being 'completely' or 'very' likely to use a UE VCA-QPS.

Conclusion: People with UE amputations have extensive information needs about UE VCA. The UE VCA-QPS aims to address patients' information needs and foster patient-centered care. Future research should assess whether the UE VCA-QPS facilitates patient-provider discussion and informed decision-making for UE VCA.

KEYWORDS

informed consent, patient–clinician communication, ethics, treatment decision making, patient-centered care, upper limb amputation, VCA

Introduction

Upper extremity (UE) vascularized composite allotransplantation (VCA) is a treatment option for people with hand and/or arm amputations that involves "transplantation of non-autologous vascularized tissues including skin, muscle, nerve, tendon and/or bone as a functional unit (e.g., a hand) to replace non-reconstructible tissue defects" (American Society of Transplant Surgeons, 2022). To date, 56 UE VCAs have been performed on 37 patients in the United States (OPTN, 2022), and more than 120 have been performed worldwide (Shores et al., 2017).

Upper extremity VCA is one of several treatment options (e.g., myoelectric and mechanical prostheses) for people with UE amputations. UE VCA is an innovative treatment modality that restores body wholeness and function for patients with complex reconstructive needs (Rose et al., 2019). Evidence suggests that people with UE amputations lack awareness and knowledge of UE VCA. People with UE limb loss report receiving insufficient information about their treatment options and report that healthcare providers do not engage in enough discussion about their condition (Nielsen, 1991; Pedlow et al., 2014; Pasquina et al., 2015; Bennett, 2016). Further, little is known about what information people with UE amputations need to know about UE VCA, which is necessary for optimizing their informed consent.

A patient-centered approach to care involves respecting and responding to patients' needs and preferences, so they can make informed treatment decisions (Institute of Medicine, Committee on Quality of Health Care in America, 2001; Epstein and Street, 2011). Elements of patient-centered care include effective communication and information exchange. Effective communication entails a dialogue between provider and patient, and patient question-asking can increase patient engagement, empowerment, and the quality of provider information-giving (Barnlund, 1970; Shepherd et al., 2011; Barton et al., 2020). However, patients commonly do not know what questions to ask to guide decision making (Lopez-Vargas et al., 2014; Lederer et al., 2016; Schwarze et al., 2020). To date, no educational interventions have been developed to increase knowledge and understanding about UE VCA for people with UE amputations.

Communication tools, such as a question prompt sheet (QPS), can facilitate patient-provider communication and support patient-centered care (Belkora et al., 2009; Gordon and Ison, 2014; Brandes et al., 2015; Sansoni et al., 2015; Satteson et al., 2020). A QPS is a list of questions that can

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Abbreviations: JHU, Johns Hopkins University; NU, Northwestern University; QPS, question prompt sheet; UE, upper extremity; VCA, vascularized composite allotransplantation; WRNMMC, Walter Reed National Military Medical Center.

empower patients to ask questions about topics important to them and promote discussion between patients and their providers. QPSs have been shown to help patients obtain information, increase the number of questions asked, improve patient-provider communication, increase patient knowledge, increase patient satisfaction, and reduce or have no effect on consultation time across clinical settings (e.g., oncology, chronic kidney disease, palliative care) (Brown et al., 2001, 2011; Gaston and Mitchell, 2005; Brandes et al., 2015; Sansoni et al., 2015; Arthur et al., 2017; Miller and Rogers, 2018; Jayasekera et al., 2020). No QPS about UE VCA has been created for people with UE amputations.

This study assessed the information needs of people with UE amputations and developed a UE VCA-specific QPS to foster patient-centered care. Our UE VCA-QPS was designed to help people with UE amputations attain greater information about UE VCA and establish realistic expectations through patient-provider communication to make informed decisions about UE VCA.

Materials and methods

Study design

In this cross-sectional study, we used a mixed-methods concurrent triangulation design (Greene et al., 1989; Creswell and Plano Clark, 2007), involving sequential in-depth and semi-structured interviews to develop and refine the UE VCA-QPS, as part of a larger study on decision making about UE VCA. Mixed-methods enabled elaboration and clarification of findings, increased validity of results, and informed subsequent data collection. We followed QPS development approaches used previously (Eggly et al., 2013; Ahmed et al., 2016; Lederer et al., 2016), and leveraged perspectives of patients and experts to ensure that the UE VCA-QPS is patient-centered.

The study was conducted at Northwestern University Feinberg School of Medicine (NU) in Chicago, IL, United States; Johns Hopkins University School of Medicine (JHU) in Baltimore, MD, United States; and Walter Reed National Military Medical Center (WRNMMC) in Bethesda, MD, United States from January 2020 through March 2022. Shirley Ryan AbilityLab in Chicago, IL, United States and David Rotter Prosthetics, LLC in Joliet, IL, United States supported recruitment for NU. The study was approved by the Institutional Review Boards at: NU (STU00209718), JHU (00225728), and WRNMMC (WRNMMC-EDO-2020-0432). NU served as the Institutional Review Board of record for WRNMMC. We used the Consolidated Criteria for Reporting Qualitative Research for quality reporting of qualitative studies (Tong et al., 2007).

Participants and recruitment

Eligible participants were English-speaking adults age 18–65 years with acquired UE amputations who had not yet pursued UE VCA, UE VCA candidates (i.e., individuals who contacted a transplant center to express interest in pursuing UE VCA), UE VCA participants (i.e., individuals who began UE VCA evaluation), and UE VCA recipients. People who were cognitively impaired, and/or had congenital limb loss were excluded.

Multiple techniques were employed to increase sample size and ensure a representative sample (Patton, 2015). We recruited patients from study sites by mailing and/or emailing eligible individuals a letter describing the study, followed by a phone call a week later to screen for eligibility. Research team members did not have prior established relationships with study participants. We also recruited participants through support groups ($n = 304$) and social media websites (Facebook and Reddit) by emailing or posting study flyers online. Interested individuals contacting the team were screened by phone for eligibility. All participants provided verbal informed consent.

Phase 1: Question prompt sheet item development

Data collection

In Phase 1, we conducted telephone in-depth interviews from July 2020–March 2022 to assess study participants' information needs and questions about UE VCA. We drew upon five open-ended questions from the in-depth interview guide, which assessed: UE VCA information needs ("If you were thinking about getting an upper limb transplant, what would you want to know about it?" and "If you were thinking about getting an upper limb transplant, what information would you need?); UE VCA-related questions ("If you were thinking about getting an upper limb transplant, what questions would you have about it?"); and perceptions about a UE VCA-QPS ("What do you think about the QPS idea?" and "Would [the QPS] be worthwhile?"). The subset of candidates, participants, and recipients were also asked what people seeking UE VCA should be informed about ("What questions should people seeking an upper limb transplant ask about it to become well informed?" and "Could you suggest some things about upper limb transplantation that people with amputations should be informed about?").

Interviews assessed study participants' likelihood of using a UE VCA-QPS (on a 5-point Likert scale anchored by "Not At All," "A Little," "Somewhat," "A Lot," and "Completely"); demographics (e.g., age, gender); clinical background (e.g., date of amputation, amputation level and type); and health literacy ("How often do you need to have someone help you when you read instructions, pamphlets, or other written material from your doctor or pharmacy?" anchored by "Never," "Rarely," "Sometimes," "Often," and "Always"; "Never" and "Rarely" responses reflected adequate health literacy) (Morris et al., 2006).

Interviews were conducted by female and male research team members (BK, KV, MD, MN) trained by the Principal Investigator (EJG), a seasoned qualitative researcher, to ensure consistent and high quality data collection. Telephone cognitive interviews were conducted (by BK) with five participants prior to in-depth interviews (January–March 2020) using standard "think aloud" procedures to ensure interview guide questions were interpreted as intended and improve question wording (Singleton and Straits, 2017). Research team members took field notes during and after interviews. Interviews lasted, on average, 78 (range: 37–140) minutes and were audio-recorded and transcribed. Participants were compensated with a $35 gift card.

Content analysis

To identify potential QPS items, two members of the research team at each study site reviewed each transcription and compiled participants' responses about UE VCA information needs and questions into a single document using content analysis (Bernard and Ryan, 1998). Responses were grouped into topics organized in terms of a patient's progress from initiating evaluation to rehabilitation. All interview transcripts were then re-examined by research team members as individual files (within-case) and as a list of all participant responses to each open-ended question (across-cases) (Ayres et al., 2003). The Principal Investigator then reviewed responses under each topic to derive draft QPS items that synthesized the information needs and questions raised by all participant responses. Thereafter, the research staff reviewed the draft QPS items to: (a) confirm that they fully represented all participants' responses, (b) add or delete repetitive or idiosyncratic items, and (c) revise item wording for clarity. This iterative process ensured comprehensiveness. All items were compiled into a first QPS draft comprising 130 items organized into 18 topics.

Thematic analysis

To identify UE VCA information needs, all transcripts were analyzed for themes emergent from the data using constant comparison, inductive, and deductive coding methods (Lincoln and Guba, 1985; Bradley et al., 2007). The research team established an initial codebook by developing deductive codes based on questions asked in the interview guide (e.g., Information Needs). The team then developed inductive codes based on themes emergent from the data during open coding of six transcripts until reaching thematic saturation (Miles and Huberman, 1994; Giacomini and Cook, 2000). Thereafter, two research team members at each study site (JG-S, BK, MD, KV, MN, ML) independently coded transcripts from their site using the finalized codebook in NVivo (Release 1.6.1, QSR International) until reaching inter-rater reliability (Cohen's Kappa $>$ 0.80) (Guest et al., 2011). Then, all transcripts were re-coded. During this process, the two research team members at each site resolved coding discrepancies through discussion. Finally, research team members reviewed all text segments coded as "Information Needs" to identify patterns and themes in study participants' UE VCA information needs and developed code summaries (Keith et al., 2017).

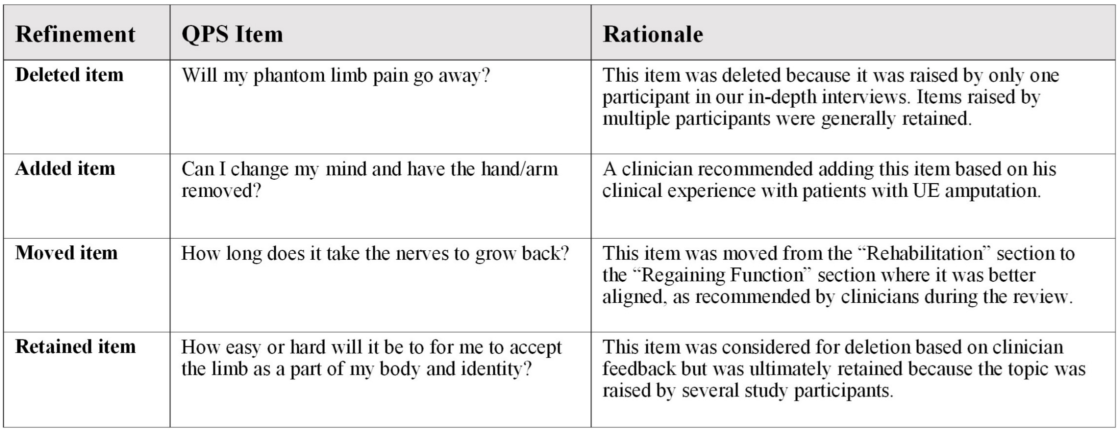
Phase 2: Question prompt sheet item refinement and reduction

Initial item refinement and reduction

Upper extremity VCA-QPS item reduction was performed by three research team members (EJG, BK, JG-S) by identifying redundancy and combining or removing redundant items to retain items that best conveyed the ideas. We assessed the readability of each item using the Flesch-Kincaid grade level formula (Stossel et al., 2012; Centers for Medicare & Medicaid Services [CMS], 2010), and simplified complex words and long sentences in items above a 6th grade reading level (Houts et al., 2006; Brega et al., 2015). Some items remained above a 6th grade reading level because they included terms that could not be restated in a simpler way or included three or more syllables (e.g., transplantation, anti-rejection, recipient). Additionally, we assessed the understandability and actionability of the UE VCA-QPS by applying the Patient Education Materials Assessment Tool (PEMAT) (Shoemaker et al., 2013). Following item refinement and reduction, the draft QPS included 77 items categorized into 16 topics.

Multidisciplinary review

A 7-person multidisciplinary team of study collaborators comprised of researchers, UE VCA clinicians/surgeons, hand reconstructive surgeons, and occupational therapists reviewed the draft UE VCA-QPS and provided feedback on clinical accuracy, relevance to the UE VCA transplant evaluation process, clarity of question wording, and redundancy in question content. Based on the feedback, the research team revised item wording for clarity, moved items to different topic categories, combined items that addressed similar concepts, added items, and deleted items. **Figure 1** includes examples of changes made to items and the rationale, based on multidisciplinary team feedback and research team review. Thereafter,



Refinement	QPS Item	Rationale
Deleted item	Will my phantom limb pain go away?	This item was deleted because it was raised by only one participant in our in-depth interviews. Items raised by multiple participants were generally retained.
Added item	Can I change my mind and have the hand/arm removed?	A clinician recommended adding this item based on his clinical experience with patients with UE amputation.
Moved item	How long does it take the nerves to grow back?	This item was moved from the "Rehabilitation" section to the "Regaining Function" section where it was better aligned, as recommended by clinicians during the review.
Retained item	How easy or hard will it be to for me to accept the limb as a part of my body and identity?	This item was considered for deletion based on clinician feedback but was ultimately retained because the topic was raised by several study participants.

FIGURE 1
Examples of interim UE VCA-QPS refinement based on multidisciplinary feedback and research team review.

the preliminary UE VCA-QPS included 52 items categorized into 12 topics.

Data collection

Semi-structured telephone interviews were conducted from September 2021– March 2022 by female and male research team members (BK, MD, WA, SF), trained by the Principal Investigator (EJG), to refine and reduce the 52-item preliminary UE VCA-QPS. Participants were asked to rate whether each question should be included in the UE VCA-QPS list on a 4-point Likert scale anchored by 4–"Definitely Keep," 3–"Probably Keep," 2–"Probably Cut," and 1–"Definitely Cut." Larger scores indicated a preference for item retention. When rating each question, participants were asked to consider how valuable each question and its answer would be to them if they were interested in the option of UE VCA. Closed-ended questions assessed participants' likelihood of using the UE VCA-QPS if they were considering getting a UE VCA (on a 5-point Likert scale anchored by "Not At All," "A Little," "Somewhat," "Very," and "Completely") and demographic and clinical characteristics (e.g., age, gender, amputation type and level, date of amputation). Open-ended questions assessed perceptions of the clarity of each item and suggestions for improving item wording, preferences between similar items, opinions about item order, and recommendations for adding or deleting questions. Interviews averaged 70 (range: 40–120) minutes and were audio-recorded, while research team members took field notes. Participants were compensated with a $35 gift card.

Mixed-methods analysis

Frequencies and means for each item's Likert score were generated and reviewed. All items with a mean score of 3.00 or smaller were eliminated, resulting in 11 items cut from the list. Remaining items' scores and qualitative responses were analyzed together. Participants' qualitative responses were reviewed by the research team (EJG, JG-S, BK) to identify QPS item changes (e.g., rewording items, combining items, moving items). After refining QPS items, the draft QPS included 38 items categorized into 12 topics.

We then sought feedback on this UE VCA-QPS draft from the study's Scientific Advisory Board (SAB), comprised of four bioethicists, and from the research team's Co-Investigators, who included two UE VCA clinicians/surgeons and two hand surgeons, for further refinement. The SAB and Co-Investigators provided feedback on improving item wording and identifying items to combine, delete, or add. Further, the SAB and Co-Investigators were asked to ensure that items covered perspectives of people with UE amputations, the military, VCA ethics, UE VCA clinical care, hand surgery, rehabilitation, prosthetics, and disability rights. These steps aimed to ensure that the UE VCA-QPS would be relevant and meaningful for patient-provider discussions about UE VCA. Demographics and clinical characteristics of participants in the in-depth and semi-structured interviews were analyzed through descriptive statistics using IBM SPSS Statistics (Version 27).

Results

Demographics and clinical characteristics

Overall, 89 people with UE amputations participated (63.9% participation rate) in in-depth interviews (*n* = 50, 61.7% participation rate) and semi-structured interviews (*n* = 56, 65.9% participation rate) (**Supplementary Figure 1**). Seventeen individuals participated in both the in-depth interview and the semi-structured interview (NU: n = 4, JHU: n = 8, WRNMMC: n = 5). Sixty refused to participate before or after providing consent because they were not interested, too busy, compensation was too low, or they did not show up for their scheduled interview. Study participants included people with UE amputations who had not pursued UE VCA (85%), UE VCA candidates and participants (9%), and UE VCA recipients (6%). Most participants were male (73%), White (74%), on average 46 years of age, and had a unilateral amputation (84%) and had a below elbow amputation (56%) (**Table 1**). Participants were interviewed, on average, 10 years after their amputation. One-third (34%) did not use a prosthesis(es).

Upper extremity vascularized composite allotransplantation information needs

When asked what information they would need if they were to pursue UE VCA, study participants reported different types of information that were organized into five major themes: broad and contextual information, information about the pre-UE VCA period, risks of UE VCA, the UE VCA procedure, and information about the post-UE VCA period. The major themes and subthemes are described below, with illustrative representative quotations presented in **Table 2**.

Broad and contextual information

Participants desired broad and contextual information about UE VCA that included knowing "everything" about it as well as the historical context and present status of UE VCA. Knowing "everything" about UE VCA would enable participants to gain "a more in-depth understanding." Participants desired historical information including "the research that was done," and how the transplant field got "to the point where this is possible." The current status of the UE VCA field pertained to "how many people have had the procedure done" and "the current state of technology." Additionally, only participants at NU wanted to know about the "upfront and lifetime costs" of UE VCA and the insurance coverage and out-of-pocket expenses.

Pre-upper extremity vascularized composite allotransplantation

Several participants wanted information about the processes occurring prior to the UE VCA procedure. This information encompassed the eligibility criteria for UE VCA (e.g., "what would make a good qualified patient…"), the waiting list (e.g., "how long of a wait [would there be] on the waiting list for a set of arms to become available"), and the evaluation process (e.g., what does "the psychological review process… entail"). Participants also desired information about the donor process, such as "how long do you have to typically wait for a donor" and how well can they "match an arm to my body to make it look more realistic for myself." Participants desired learning about the transplant team and clinic, such as the "doctors and clinicians [who have] done it and have had success," "how many procedures they have done," and "where it would be taking place."

Risks of upper extremity vascularized composite allotransplantation

Participants wanted information about the risks of UE VCA, including general risks and the potential affect UE VCA could have on one's life. General UE VCA risks included "the possibilities of what could go wrong" from receiving a UE VCA. Participants desired information on how UE VCA could potentially harm a person's life and lifespan, including the "rate of life-threatening risks," the risk of dying, infection, rejection, and whether recipients would "be more susceptible to cancers or other things like COVID."

Upper extremity vascularized composite allotransplantation procedure

Participants desired information about the UE VCA surgical procedure, including the hospital stay and recovery period. Desired procedure details included "how they attach the bone," the required number of surgeries, and the "length of time the surgery usually takes." Others wanted information about the length of hospital stay post-transplant, and the recovery process: "how long [the UE VCA] is going to take to heal."

Post-upper extremity vascularized composite allotransplantation

Participants expressed interest in learning how UE VCA impacts a recipient's life as it relates to medication and medication side effects, rehabilitation, function, success rate, lifestyle changes, and experiences of UE VCA recipients. Regarding medication and medication side effects, participants desired information on "how many drugs you have to be on for the rest of your life" and the "risk levels of life-time, consistent use of drugs and side effects of the drugs." Study participants also desired information on what "the rehab process [would] be like," how long they would do hand therapy, and "how much time [it would] take out of [their] life."

Participants were interested in learning about the type and extent of functionality a UE VCA would provide, whether recipients would "experience the sensation of touch again," and how long it would take "to get it working." Some referred to their previous "natural hand" and wanted to know if a UE VCA would "work like my old arm worked." Participants

TABLE 1 Participants' demographic and clinical characteristics.

Variable	**Total (*n* = 89) *N* (%)**	**NU (*n* = 31) *n* (%)**	**JHU (*n* = 28) *n* (%)**	**WR (*n* = 30) *n* (%)**
Age, years, mean [SD] (range)	46.2 [10.9] (19–65)	50.0 [9.9] (25–65)	46.6 [10.0] (32–64)	41.9 [11.4] (19–65)
Gender				
Male	65 (73.0)	20 (64.5)	18 (64.3)	27 (90.0)
Female	24 (27.0)	11 (35.5)	10 (35.7)	3 (10.0)
Ethnicity				
Not Hispanic or Latino	80 (89.9)	29 (93.5)	27 (96.4)	24 (80.0)
Hispanic or Latino	9 (10.1)	2 (6.5)	1 (3.3)	6 (20.0)
Race				
White	66 (74.1)	23 (74.2)	23 (82.1)	20 (66.7)
Black or African American	16 (18.0)	5 (16.1)	5 (17.9)	6 (20.0)
Other*	7 (7.9)	3 (9.7)	0 (0.0)	4 (13.3)
Marital Status				
Married/Domestic Partner/Civil Union	57 (64.0)	19 (61.3)	16 (57.1)	22 (73.3)
Never Married/Single	17 (19.1)	5 (16.1)	8 (28.6)	4 (13.3)
Separated or Divorced	14 (15.7)	7 (22.6)	3 (10.7)	4 (13.3)
Widowed	1 (1.1)	0 (0.0)	1 (3.6)	0 (0.0)
Education				
Less than high school graduate	1 (1.1)	0 (0.0)	1 (3.6)	0 (0.0)
High school graduate	17 (19.1)	5 (16.1)	7 (25.0)	5 (16.7)
Some college	26 (29.2)	8 (25.8)	6 (21.4)	12 (40.0)
College graduate	27 (30.3)	12 (38.7)	6 (21.4)	9 (30.0)
Post graduate degree	18 (20.2)	6 (19.4)	8 (28.6)	4 (13.3)
Employment Status†				
Employed Full time	37 (41.6)	11 (35.5)	11 (39.3)	15 (50.0)
Disabled	20 (22.5)	10 (32.3)	8 (28.6)	2 (6.7)
Retired	19 (21.3)	4 (12.9)	4 (14.3)	11 (36.7)
Employed Part time	4 (4.5)	2 (6.5)	2 (7.1)	0 (0.0)
Not Employed	4 (4.5)	3 (9.7)	1 (3.6)	0 (0.0)
Homemaker	3 (3.4)	1 (3.2)	1 (3.6)	1 (3.3)
Student	1 (1.1)	0 (0.0)	0 (0.0)	1 (3.3)
Income				
Less than $15,000	7 (7.9)	5 (16.1)	2 (7.1)	0 (0.0)
Between $15,000 and $34,999	7 (7.9)	1 (3.2)	5 (17.9)	1 (3.3)
Between $35,000 and $54,999	10 (11.2)	5 (16.1)	2 (7.1)	3 (10.0)
Between $55,000 and $74,999	13 (14.6)	5 (16.1)	4 (14.3)	4 (13.3)
Between $75,000 and $94,999	8 (9.0)	2 (6.5)	1 (3.6)	5 (16.7)
More than $95,000	36 (40.4)	12 (38.7)	12 (42.9)	12 (40.0)
Prefer not to answer	8 (9.0)	1 (3.2)	2 (7.1)	5 (16.7)
Primary Health Insurance‡				
Medicaid or Medicare	41 (46.1)	16 (51.6)	14 (50.0)	11 (36.7)
Private	36 (40.4)	14 (45.2)	15 (53.6)	7 (23.3)
Uniformed Services (Tricare)	27 (30.3)	0 (0.0)	4 (14.3)	23 (76.7)
None	1 (1.1)	1 (3.2)	0 (0.0)	0 (0.0)
Other	2 (2.2)	1 (3.2)	1 (3.6)	0 (0.0)
Health Literacy, Adequate				
	80 (89.9)	30 (96.8)	24 (85.7)	26 (86.7)

(Continued)

TABLE 1 (Continued)

Variable	Total (n = 89) N (%)	NU (n = 31) n (%)	JHU (n = 28) n (%)	WR (n = 30) n (%)
Health Status[†]				
Excellent	18 (20.2)	6 (19.4)	6 (21.4)	6 (20.0)
Very good	36 (40.4)	11 (35.5)	14 (50.0)	11 (36.7)
Good	24 (27.0)	10 (32.3)	4 (14.3)	10 (33.3)
Fair	10 (11.2)	4 (12.9)	3 (10.7)	3 (10.0)
Poor	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Dominant Hand Before Amputation[†]				
Right	78 (87.6)	30 (96.8)	23 (82.1)	25 (83.3)
Left	8 (9.0)	1 (3.2)	3 (10.7)	4 (13.3)
Upper Limb Amputated[†]				
Right	43 (48.3)	10 (32.3)	11 (39.3)	22 (73.3)
Left	31 (34.8)	13 (41.9)	12 (42.9)	6 (20.0)
Both	14 (15.7)	8 (25.8)	5 (17.9)	1 (3.3)
Amputation Type				
Unilateral	75 (84.3)	23 (74.2)	23 (82.1)	29 (96.7)
Bilateral	14 (15.7)	8 (25.8)	5 (17.9)	1 93.3)
Amputation Level				
Below elbow	50 (56.2)	19 (61.3)	12 (41.9)	19 (63.3)
Above elbow	37 (41.6)	11 (35.5)	15 (53.6)	11 (36.7)
Both below and above elbow	2 (2.2)	1 (3.2)	1 (3.6)	0 (0.0)
Current Prosthesis Type[‡]				
Myoelectric	39 (43.8)	10 (32.2)	7 (25.0)	22 (73.3)
Mechanic	36 (40.4)	18 (58.0)	1 (3.6)	17 (56.7)
Cosmetic	4 (4.5)	1 (3.2)	1 (3.6)	2 (6.7)
Other	1 (1.1)	0 (0.0)	0 (0.0)	1 (3.3)
None	28 (31.5)	7 (22.6)	19 (67.9)	2 (6.7)
Years Since First[§] Amputation				
<1 year	8 (9.0)	3 (9.7)	3 (10.7)	2 (6.7)
1–2 years	14 (15.7)	5 (16.1)	4 (14.3)	5 (16.7)
3–6 years	20 (22.5)	12 (38.7)	7 (25.0)	1 (3.3)
7–10 years	16 (18.0)	5 (16.1)	5 (17.9)	6 (20.0)
>10 years	31 (34.8)	6 (19.4)	9 (32.1)	16 (53.3)
Data Collection Activity**				
In-Depth Interviews	50 (56.2)	16 (51.6)	17 (60.7)	17 (56.7)
Semi-Structured Interviews	56 (62.9)	19 (61.3)	19 (67.9)	18 (60.0)
Type of Participant				
Person with UE amputation	76 (85.4)	29 (93.5)	17 (60.7)	30 (100.0)
VCA candidate/participant	8 (9.0)	2 (6.5)	6 (21.4)	0 (0.0)
VCA recipient	5 (5.6)	0 (0.0)	5 (17.9)	0 (0.0)

SD, standard deviation; WR, Walter Reed National Military Medical Center.

*"Other" included people who identified as Hispanic or Mexican (n = 4), Asian (n = 1), Native Hawaiian or Other Pacific Islander (n = 1), or multi-racial (n = 1).

[†] Percentages do not add up to 100 because some participants did not respond.

[‡] Percentages add up to greater than 100 due to more than one response from some participants.

[§] Some participants had multiple surgeries for their amputation or multiple amputations.

**Some participants (n = 17) took part in both the in-depth interview and the semi-structured interview.

framed their interest in learning about the success rate in terms of "how many [UE VCAs] failed versus how many succeeded." Others wanted to learn about required lifestyle changes, such as how long recipients might be "out of work" and any restrictions on diet, drinking alcohol, and smoking. Several participants reported interest in learning directly from UE VCA recipients about their experiences: "what kind of things can, and can't they do."

TABLE 2 Representative illustrative quotations about information needs by major theme and subtheme, with code frequency.

Themes/Subthemes	Code Freq. N	Quotations
Broad and Contextual Information about UE VCA		
Everything about it	8	"Well, I think I would need [to know] everything about it, like soup to nuts, like, oh, every aspect" [J008, 43-year-old female with bilateral below-elbow limb loss]
		"I hate to say this, but 'everything'. [Laughs] That everything would include all of the risks, all of the benefits, and the projected recovery time as well as the actual success rates and actual recovery times of other patients." [WR001, 52-year-old male with unilateral below-elbow limb loss]
UE VCA history and current state	13	"I'd like to know the kind of history of it, how did we get to the point where this is possible, how many transplants have been done." [J012, 54-year-old female with unilateral below-elbow limb loss]
		"What I would want to know: the history, the research that was done. . ." [WR017, 57-year-old male with unilateral below-elbow limb loss]
		"The cost. If my insurance covered it. My out-of-pocket procedure costs." [N017, 60-year-old female with unilateral above-elbow limb loss]
Pre-UE VCA		
Eligibility, waiting list, and evaluation process	13	"How one becomes a candidate first of all. . . how do you even get on their radar? And then how, what's the process to find out if I'm a good candidate." [N009, 53-year-old male with bilateral below-elbow limb loss]
		"How long does the process take as far as like, "OK, hey. We received a transplant. We need you here." OK, so how would I get there?" [N015, 37-year-old female with unilateral below-elbow limb loss]
		"If I'm a good candidate, if my case is a good idea for the surgery?" [WR010, 35-year-old male with unilateral above-elbow limb loss]
Donor and matching process	14	"I would be curious about like where it is coming from, like this donor, and so I don't know how much of that information they would share, but if I was getting somebody's limb, I guess I probably would want to know who it was coming from." [J014, 53-year-old female with unilateral above-elbow limb loss]
		"What's the process, as far as choosing aesthetically where the arm comes from? How do they try to match up somebody, how do you get paired with somebody to actually have a transplant from their arm?" [WR008, 39-year-old male with unilateral above-elbow limb loss]
Transplant team and transplant clinic	9	"What is the background of the doctor? What are some of his cases, the number of surgeries, or the hospital staff that are involved and their experience with this? Ideally, I'd want to talk to a patient who's had it done by this doctor. . . . what hospitals are doing it." [N014, 58-year-old male with unilateral below-elbow limb loss]
		"How many has the doctor done? How many years [experience] do they have doing it? What experience do they have?" [WR011, 65-year-old male with unilateral above-elbow limb loss]
Risks of UE VCA		
Risks in general	10	"The risks that would be involved and if that would outweigh the, you know, if the reward would outweigh the risk." [N011, 59-year-old male with unilateral below-elbow limb loss]
		"What are the risks to my health, what are the possibilities of what could go wrong or not happen for me or things like that." [N022, 47-year-old female with unilateral below-elbow limb loss]
Effect on life and lifespan	10	"Would there be the possibility of me dying if I did this. . . if it rejects, what is the possibility of me dying from that. . .?" [J014, 53-year-old female with unilateral above-elbow limb loss]
		"And how does this change my long-term picture? Like, is this going to affect my lifespan? Is this going to shorten my lifespan at all?" [N019, 57-year-old male with unilateral below-elbow limb loss]
Infection and Rejection	7	"What happens if your body rejects the hand. You know, if you find out it wasn't a match or something, do you have to take the hand off?" [WR017, 57-year-old male with unilateral below-elbow limb loss]
UE VCA Procedure		
Surgical procedure	15	"What will be done to my hand, or to my arm. Like, they're going to. what they're going to attach to what." [N006, 47-year-old male with unilateral above-elbow limb loss]
		"I'd want to know all the ins and outs of the procedure itself, first, beforehand, like all the medical risks, the possible things that could happen." [J014, 53-year-old female with unilateral above-elbow limb loss]
		"I would like to know how the surgery would go. How long would the surgery take? How do they attach it to the upper extremity?" [WR009, 62-year-old male with unilateral above-elbow limb loss]

(Continued)

TABLE 2 (Continued)

Themes/Subthemes	Code Freq. N	Quotations
Hospital stay and recovery period	17	"How long would I be incapacitated, like away from my home, away from my family." [N017, 60-year-old female with unilateral above-elbow limb loss]
		"What the recovery process is, how long I would be spending in inpatient?" [WR003, 31-year-old male with unilateral above-elbow limb loss]
Post-UE VCA		
Living with a UE VCA		
Rehabilitation	26	"Where am I going to do my occupational therapy? Do I have to move from my home and live near the hospital because something might go bad. . . have to do that for 6 months?" [N019, 57-year-old male with unilateral below-elbow limb loss]
		"It's like 'How much time does that take out of your life? Would I have to go there and do all my rehab there? How does that work?.' The amount of rehab, how many times a week, how long each time is, where the rehab is and about how long you're doing rehab. . ." [J006, 64-year-old female with unilateral above-elbow limb loss]
		"I guess kind of the timeline as well and how much I would have to commit to physical therapy and occupational therapy. I guess more so just the timeline. Like when would you start seeing results, when should you be able to start moving like the elbow and those types of things." [WR013, 24-year-old male with unilateral above-elbow limb loss]
Medication and medication side effects	17	"How many medications are there? Is this medication something that I have to take for the rest of my life? Is this once a week, twice a week, monthly, daily? . . .if I miss the time, will my body automatically reject the transplant?" [N015, 37-year-old female with unilateral below-elbow limb loss]
		"What does it mean to have some sort of medicine that's in you that's going to hurt you?" [J005, 40-year-old male with unilateral above-elbow limb loss]
		"The first and foremost thing I would want to know is how much medication I would have to take, and for how long." [WR002, 39-year-old male with unilateral above-elbow limb loss]
Lifestyle changes	11	"What do I, do I have to readjust my diet from—way of eating, taking in certain foods? Do I, can I drink alcohol? Can I smoke tobacco?" [N007, 48-year-old male with bilateral, above- and below-elbow limb loss]
		". . . what things might having a transplanted limb make you not able to do in terms of donating blood or in terms of just some ways that that might limit some of your choices going forward." [J012, 54-year-old female with unilateral below-elbow limb loss]
		"I would need to know what . . . the environments that I may be precluded from taking part in, such as swimming, or working outside – how physically active can I be, and what environmental restrictions will that put upon me as to keeping me from doing things that I currently do." [WR001, 52-year-old male with unilateral below-elbow limb loss]
Outcomes of UE VCA		
Functionality, sensation, and other outcomes	34	"You know, am I going to feel like a regular person again with two fully functioning arms? Or am I gonna have one good arm and a half dead arm. Where it's still in the way and not very useful. I just went through all this work and all this procedure and all this surgery for something that I'm not even using still. Which is you know, kind of the problem with prosthetics." [WR003, 31-year-old male with unilateral above-elbow limb loss]
		"I would like to know if, if I will be able to use my hand like my hand was. Will I be able to, as a female, paint the fingers. . .the fingertips? How will I be able to use it functionally, like being able to use a keyboard going back to work? And for me, the biggest thing is, can I use it to do hair?" [WR005, 56-year-old female with unilateral below-elbow limb loss]
		"Is this hand transplant, is it going to replace what I lost? And if not, what percentage will I reacquire, you know? . . . What is my percentage of recovery? That's what I would want to know." [N021, 56-year-old male with bilateral below-elbow limb loss]
		"How is the functionality of an arm transplant? If you have an arm transplant, does it end up being just as functional as your own arm?" [J006, 64-year-old female with unilateral above-elbow limb loss]
Success rate	12	"The surgeries that have been done, . . . how many have turned out wonderful, how many are OK, and how many are not OK." [N018, 63-year-old male with unilateral above-elbow limb loss]
		"I guess the percentages of successful transplant and the non-successful transplants, and the rate of rejection." [WR017, 57-year-old male with unilateral below-elbow limb loss]

(Continued)

TABLE 2 (Continued)

Themes/Subthemes	Code Freq. N	Quotations
Experiences of UE VCA recipients	13	"I'd like to hear what experience people have with it from medical professionals, but also, hopefully or possibly by people who have had transplant of an upper extremity who can at least talk about their experience." [J012, 54-year-old female with unilateral below-elbow limb loss]
		"I think I would want to know the experience of other individuals. . . What is it they weren't able to do with a prosthetic, let's say, but now they can do, or things that were maybe a little bit more difficult with a prosthetic but now they can do it with ease." [N022, 47-year-old female with unilateral below-elbow limb loss]
		"And [I'd] probably like to talk to people [the doctors] have had—you know they have worked on. . . helped with. Because it is a lot easier for the doctors or for people to say well it's supposed to work like that, but the guy that's actually feeling it, the guy that actually has it, he can kind of tell you the real deal, you know." [WR011, 65-year-old male with unilateral above-elbow limb loss]
		"The information I would need is, I'd want to know that someone else had had the procedure and had had success with it. I'd want to talk to that person, hear from that person." [WR007, 41-year-old male with unilateral below-elbow limb loss]

TABLE 3 Likelihood of using a UE VCA-QPS.

Question*	Not at all likely *n* (%)	A little likely *n* (%)	Somewhat likely *n* (%)	A lot/very likely† *n* (%)	Completely likely *n* (%)
In-Depth Interviews If you were considering getting an upper limb transplant, how likely would you be to use a question prompt sheet? $n = 45$‡	1 (2.2)	4 (8.9)	4 (8.9)	6 (13.3)	30 (66.7)
Semi-Structured Interviews If you were considering getting an upper limb transplant, how likely would you be to use a question list about hand or upper limb transplantation in your doctor visit? $N = 56$	4 (7.1)	0 (0.0)	4 (7.1)	18 (32.1)	30 (53.6)

*Some participants (n = 17) took part in both the in-depth interview and the semi-structured interview.
†One anchor in the rating scale differed between the two types of interviews: "A Lot Likely" was used in the in-depth interview and "Very Likely" was used in the semi-structured interview.
‡$n = 5$ participants were not asked the question.

The final question prompt sheet

The final UE VCA-QPS has 35 items, organized into 9 main topics, and fits onto one double-sided page of paper. At the end of the list, patients can note additional questions. The UE VCA-QPS is available upon request.

Mean ratings of QPS items in the semi-structured interviews ranged from 2.70 to 3.93. Items ranked largest, reflecting preference for retention, were primarily about UE VCA risks. Items ranked moderately high were about UE VCA functional outcomes.

Likelihood of using the question prompt sheet

Among in-depth interview participants, most who were asked ($n = 36/45$, 80%) reported being "Completely" or "A Lot" likely to use a QPS (Table 3). Among all semi-structured interview participants, most (86%) reported being "Completely" or "Very" likely to use a QPS.

Discussion

Through mixed-methods research, we developed a 35-item QPS specific to UE VCA to address the information needs of people with UE amputations and facilitate patient-centered care. Our study participants had extensive information needs, focusing on risks, the rehabilitation process, and expectations for functional and other outcomes of UE VCA. The UE VCA-QPS is intended for use in the UE VCA clinical context amongst candidates and participants undergoing UE VCA evaluation. Study participants reported a high likelihood of using the UE VCA-QPS in a clinic visit if they were to pursue UE VCA. The UE VCA-QPS supports patient-centered care by promoting patient-provider communication that addresses patients' unique information needs and fosters information sharing so that patients can make informed treatment decisions.

People with UE amputations are typically healthy people, who generally have little need to know about transplantation. Our study participants had considerable information needs suggesting that they had limited knowledge of transplantation, rejection, and anti-rejection medications, which underscores the need to help people with UE amputations learn about the UE VCA option and establish realistic expectations so that they can make informed treatment decisions. The higher priority placed on risks and functional outcomes by participants in the semi-structured interviews indicates the relative importance that providers should emphasize in their discussions about UE VCA.

Our finding that only NU participants desired information about the costs of UE VCA makes sense considering that: (a) JHU participants included more UE VCA candidates, participants, and recipients and were thus more familiar with the insurance and out-of-pocket costs associated with UE VCA; and (b) all WRNMMC participants were military health system beneficiaries whose healthcare costs are covered by the federal government.

The UE VCA-QPS can be provided to patients in advance of their first visit to the VCA clinic, or it can be provided to them for review while waiting in the clinic to see their provider so that they can become more empowered to communicate with providers. Evidence shows a QPS may be more effective at increasing patient question-asking and provider information-giving when the QPS is provided to patients shortly before they meet with their provider (Sansoni et al., 2015). By reviewing the UE VCA-QPS prior to seeing the provider, the patient can identify questions they find important and become more engaged during their visit. Providers should ask patients for their QPS question list at the beginning of their visit given that other research has shown this provider practice of "endorsement" is effective at increasing the number of questions asked by patients and the amount of information provided by doctors during consultations (Sansoni et al., 2015).

In general, QPSs can vary in format and length. The number of items in other QPSs range from 3 to 169 items, with a mean of 33 items (Kinnersley et al., 2007; Brandes et al., 2015; Sansoni et al., 2015). Our 35-item UE VCA-QPS is comprehensive while also convenient in fitting onto a two-sided single page of paper for easy distribution, or may be viewed as an electronic document on a mobile phone, tablet, or computer.

Future research should assess the effectiveness of the UE VCA-QPS in facilitating communication between patients and providers in the UE VCA clinical context, as well as patients' informed decision-making about UE VCA. Implementation science research should assess the most acceptable, appropriate, and feasible way of delivering and evaluating the UE VCA-QPS.

Strengths of our study include a multi-site study design conducted in geographically diverse locations in the US, and included individuals throughout the US. Our sample included civilian and military participants with unilateral and bilateral amputations that were above and/or below the elbow, which supports the transferability of findings, despite being a challenging population to recruit. Additionally, our sample is representative of the broader population of people with UE amputations in terms of gender, race, and age (Inkellis et al., 2018). Our mixed-methods design facilitated a patient-centered approach to QPS development by involving people with UE amputations in multiple phases of data collection, review, and feedback, and prioritizing their perspectives over other stakeholders' feedback. Further, our multidisciplinary team of study collaborators included UE VCA clinicians/surgeons, hand reconstructive surgeons, and occupational therapists who helped to ensure that the UE VCA-QPS was clinically relevant for the target population. Designing the UE VCA-QPS at a low reading grade level and use of the PEMAT will foster a greater comprehensibility (Shoemaker et al., 2013).

Our study has limitations. Some (19%) participants completed both the in-depth and semi-structured interviews, which may reduce the transferability of study results. Although individuals motivated by the prospect of pursuing UE VCA may have been more inclined to participate in interviews, suggesting a selection bias, study participants' views ranged broadly in their level of interest in pursuing UE VCA. As our study sample included disproportionately fewer Hispanic or Latino individuals compared to the U.S. population of people with UE amputations (10% versus 15%), future research should examine UE VCA information needs among more ethnically diverse participants. We produced the UE VCA-QPS in English; future research should prepare the UE VCA-QPS in other languages.

Conclusion

People with UE amputations desired extensive information about UE VCA, primarily on risks, the rehabilitation process, and functional outcomes. To empower people with UE amputations and foster patient-provider communication about UE VCA, we developed a 35-item UE VCA-QPS. Use of the UE VCA-QPS is designed to address information needs, facilitate patient-centered care, and enhance informed decision making among people with UE amputations undergoing evaluation for UE VCA.

Data availability statement

The original contributions presented in this study are included in the article/**Supplementary material**, further inquiries can be directed to the corresponding author.

Ethics statement

The studies involving human participants were reviewed and approved by the Institutional Review Boards at: Northwestern University (STU00209718), Johns Hopkins University (00225728), and Walter Reed National Military Medical Center (WRNMMC-EDO-2020-0432). Northwestern University served as the Institutional Review Board of record for Walter Reed National Military Medical Center. Written informed consent for participation was not required for this study in accordance with the national legislation and the institutional requirements. All participants provided verbal informed consent.

Author contributions

EJG conceived and designed the research study and participated in performing the research, analyzing and interpreting the data, and writing the manuscript. JG-S and BK participated in performing the research, analyzing and interpreting the data, and writing the manuscript. MD, KV, MN, and ML participated in data collection and analyzing and interpreting the data. TR, WA, and SF participated in data collection. All authors reviewed and approved the final manuscript.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2022.960373/full#supplementary-material

SUPPLEMENTARY FIGURE 1
Consort diagrams for in-depth interviews and semi-structured interviews.

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Using team science in vascularized composite allotransplantation to improve team and patient outcomes

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Reconstructive allografts using Vascularized Composite Allotransplantation (VCA) are providing individuals living with upper limb loss and facial disfigurement with new opportunities for a sensate, esthetically acceptable, and functional alternative to current treatment strategies. Important research attention is being paid to how best to assess and screen candidates for VCA, measure optimal patient outcomes, and support patient adherence to lifelong behaviors and medical regimens. Far less attention, however, has been dedicated to the team science required for these complex VCA teams to form, prepare, and provide the highest quality clinical and psychosocial care to those receiving VCA. VCA teams are unique in that they require specialized team members whose scope of practice may not otherwise overlap. The team also needs to constantly negotiate balancing patient safety with multiple risks throughout the transplant process. This study aimed to elucidate the team science needed for this highly innovative and complex area of medicine. Using in-depth qualitative interviews with 14 VCA team members and observations at team meetings, we found that careful consideration of team composition, team structure, and organizational commitment (e.g., local culture and team values; investment of resources) influences team performance and patient outcomes, but that to be efficient and truly effective, teams need to commit to developing processes that foster collaboration. These processes are action-oriented (e.g., communication, leadership), strategic (e.g., planning, training) and interpersonal (e.g., conflict management, trust building). Dedication and commitment to team science allows teams to manage conflict under stress and exercise ways to leverage strengths to provide optimal performance or patient psychosocial and clinical outcomes. This study can provide insight into quality improvement efforts for VCA teams and guidance for other transplant programs that wish to consider expansion into VCA.

KEYWORDS

vascularized composite allograft (VCA), team science, qualitative study, case study, transplant

Introduction

The first reported hand transplantation was performed in Lyon, France in 1998 (Dubernard et al., 1999). Since then, more than 120 hand transplants in 76 patients and 37 face transplants have been performed, with reasonable functional and esthetic outcomes for optimally selected patients (Thuong et al., 2019). These reconstructive transplants, using Vascularized Composite Allografts (VCA), are providing some persons living with upper limb amputation and facial disfigurement with new opportunities for a sensate, esthetically acceptable, and functional alternative to current treatment strategies (Kaufman et al., 2019). With advances in immunosuppression, surgical and medical techniques, and technology, these transplants while complex and associated with long term risks of immunosuppressive medications, are no longer considered experimental, but feasible. The field, therefore, is at a juncture. These procedures are possible, but they are not yet part of the standard options routinely presented to eligible patients. The psychosocial and ethical challenges associated with VCA and how those challenges are translated into VCA care models and treatment standards are persistent questions. These challenges include how best to identify and select ideal candidates, communicate short- and long-term consequences, support short- and long-term rehabilitation goals, and define and attain optimal clinical and psychosocial outcomes. To optimize patient outcomes and satisfy requirements likely needed for insurance coverage and regulatory needs, clear methods to overcome these challenges are required. Further, these strategies must be considered within the ethical context and assumptions of VCA improving quality of life, not, like other transplants, to save or sustain life (Caplan et al., 2019).

With VCA opportunities still limited to relatively few academic health centers—18 hand and 17 face transplant programs as of 2019—much of the research to fill these gaps is underway with the highly specialized teams that currently have VCA programs (Henderson, 2019). One important area of inquiry with less attention, however, is the team science required to form these complex, transdisciplinary VCA teams and programs that develop and shape models of care and influence the local approach to transplant. Cultural understanding of these highly specialized teams is important, as it is a shaping factor in the ways in which patient-level concerns and challenges noted above are met. Such an understanding can provide insight as other transplant programs consider expansion into VCA and more generally, to advance the understanding of barriers and facilitators required for transdisciplinary teams practicing in highly innovative and complex areas of medicine, areas where the benefits for a patient's quality of life needs to be constantly evaluated with safety and risks.

We interviewed healthcare providers as part of a larger study aimed at understanding the values, attitudes, and expectations of treatment for people with facial disfigurement and upper extremity limb loss. These members of the healthcare team were engaged in discussions about possible treatment options, candidate selection, VCA preparation, surgery, patient recovery and rehabilitation, and in VCA team meetings. One major theme that emerged in the interviews was consistent with definitions of team science: how team members organically formed and came to work collaboratively toward a common goal of establishing a VCA center and providing optimal outcomes for patients. Using a case study approach, we examined this "team science" theme.

Materials and methods

Study case

This study uses a qualitative case study methodology. Yin (2003) defines a case study as an empirical research activity that examines a specific situation within a real-life context. Case studies typically include a limited number of informed individuals with detailed understanding of the case or the context of the case. Qualitative methods for team science have been endorsed because they allow for identification of essential factors, a close proximity to processes (Yin, 2003). When conducted with teams in extreme settings or exceptional circumstances, such as VCA, they provide in-depth findings that can inform other teams in similar circumstances on models of best practice (Solis et al., 2016).

For this case we focused on a VCA team that has successfully completed one VCA transplant, but is actively assessing potential candidates for future procedures.

Study design

From March 2019 to March 2021, we conducted 14 unstructured interviews with the transdisciplinary team that included surgeons, transplant physicians, a psychiatrist, transplant nurse coordinator, social worker, ethicist, and prosthetists. This study was approved as minimal risk by the Institutional Review Board (IRB 18-006889) and oral consent was obtained from all study participants.

Source of participants

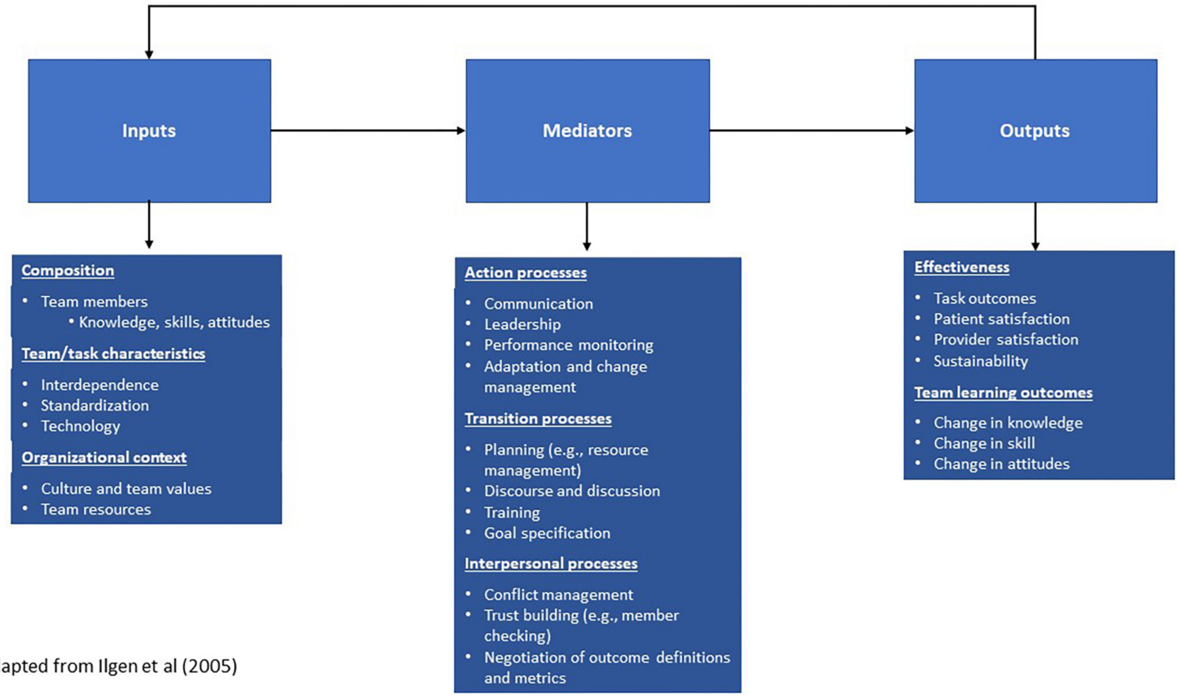
We used a purposeful "maximum variation" sampling strategy to capture differences between providers (Patton, 2002). **Figure 2** shows the composition of the VCA team. Spheres in green represent clinical team members, and spheres in blue represent institutional or administrative team members. Representatives from each of the clinical team spheres were included in interviews.

VCA team leaders representing the surgical team, transplant team and bioethics (indicated by the overlapping green spheres at the center in **Figure 2**), were interviewed. They identified additional key stakeholders for interviews from other spheres. These included physicians and nurses, social workers, physical therapists, transplant coordinators, and prosthetists and protheses fitters involved in the VCA team. Names and contact information for additional key stakeholders were obtained through the VCA team leadership and through snowball sampling. Participants were contacted by either the study coordinator or the principal investigator *via* email to explain the study purpose, consent script, and to request participation. If no response to the initial email was received, one additional follow-up email was sent. A study coordinator contacted interested providers to schedule interviews with one of two interviewers. Both interviewers were experienced Ph.D. trained researchers who had not previously worked with any of the providers. All team members approached ($n = 14$) agreed to be interviewed and completed interviews. Recruitment continued until saturation of information on treatment options, candidate selection, and VCA preparation.

Source of data

The interview guide was initially unstructured, but aimed to understand the team's interactions and perspectives on VCA education, candidate selection, and once a candidate was listed, the pre- and post-VCA preparation and treatment discussions with patients and families. With every subsequent interview, the guide was modified to become semi-structured, and reflected new areas of inquiry or greater concentration on topics that were emerging as critical points for understanding. For example, with little prompting, those interviewed early in the process discussed at length the importance of collaboration and the development of the study team as it related to patient education and outcomes. Probes about team science, therefore, were integrated into the interview guide over time.

All interviews were digitally recorded and transcribed verbatim and then de-identified (removing names and other identifiable features). After each interview, case-based memos were created that captured ideas and compared accounts



*Adapted from Ilgen et al (2005)

FIGURE 1
Input-mediator-output-input framework for VCA team science.

with other participants. Recorded interviews were transcribed verbatim and stored on a secure server for analysis in NVivo (2020).

For team meeting observations, qualitative analysts attended meetings and took notes. Notes were uploaded into Nvivo for analysis. Meetings included 63 invited team members, with attendance varying depending on the case and agenda.

Data analytic approach

The research team used a three-phased thematic content analysis to understand provider attitudes, beliefs, and experiences about the VCA process (Patton, 2002; Bradley et al., 2007). First, three research team members (JG, KS, and CK) independently read the interviews multiple times to become immersed in the data and then began an initial set of codes that captured key concepts from the data (e.g., team collaboration). Second, JG, a health services researcher with qualitative and mixed methods expertise, and CK, a solid organ transplant physician with qualitative research experience, further analyzed the transcripts, met regularly to expand, and refine code development, and create definitions for each code. With the review of each subsequent transcript, codes were refined (e.g., defining roles, coordination, program investment) to reflect a deeper understanding of the team

FIGURE 2
VCA team composition.

science theme. Coding decisions were developed by consensus and documented to provide a clear audit trail on the origin of the codes. After finalizing the code book, JG and CK then coded each transcript and applied codes line by line back to the transcripts. Third, codes were then organized into sets of codes specific to team science and analyzed for relationships across that set of codes.

Results

One major theme across interviews and observation data was how providers worked together to develop and create an environment to promote successful outcomes when stakes are high, and surgeries are infrequent. As shown in **Figure 1**, we coded this theme as "team science" and as we analyzed sub-codes, we recognized the alignment of our subcodes with the constructs from the Input-Mediator-Output-Input framework, the dominant framework in team science that describes dynamic, causal relationships. It details affective, cognitive, and behavioral processes that affect team performance, where inputs and mediators explain variability in team output and viability, and output, in turn, affects the next iteration of inputs (Ilgen et al., 2005). Using this framework assists in helping to better understand the team's experiences and their intentional and unintentional strategies that form the transdisciplinary team and refine team processes for VCA transplants.

Inputs

Inputs included team organizational commitment (e.g., local culture and team values; investment of resources), team structure, and composition, all of which are considered to influence team performance and patient outcomes. Participants described teams by their level of engagement and their unique skills. More often, participants described their role on the "teamlet" (Bodenheimer and Willard-Grace, 2016) or the smaller core teams. There was a deep understanding, however, that the teamlet was part of a larger integrated team and a commitment to that team. Participants considered their organization's cultural values as a key element of how and why the team formed. Institutional and departmental leadership financially invested in developing and sustaining the program, encouraged learning from existing external programs, provided resources that allowed for leaders in transplant and plastic surgery—teams that typically do not share cases—to collaborate and initiate a new model of care. **Figure 2** is a synthesis of team roles and the described levels of team participation, with the inner circle having primary leadership roles, greatest number of responsibilities, and most interdependence.

The VCA team was led by transplant and surgical leaders who drew from existing teams and resources. Notably, the VCA program did not maintain any personnel whose sole employment duties pertained to the VCA program. Salary and benefits were maintained by their primary position and a proportion of their time and expertise was allotted to the VCA program. Further, the VCA team used space in an established solid organ transplant center. Thus, the VCA program did not require infrastructure funds at its inception.

The team members entered the VCA program with established training and expertise in their field and adapted this experience to serve the VCA program. Some of the team members worked in multidisciplinary solid organ transplant teams but many were new to transplantation. Team members without transplant experience reported seeking out workshops or shadowing opportunities elsewhere to build familiarity with transplant practice. Similarly, team members with transplant backgrounds sought opportunities to understand the needs of patients cared for by the reconstructive surgery practices.

Team composition evolved with purpose over time. Leaders chose members for their teamlets based on previous experience, knowledge about transplant or surgical processes, and team members were added as needs emerged. For example, bioethicists were brought on early to help dissect ethical issues of offering non-life saving transplants, navigate candidate selection issues (e.g., ethics of offering bilateral vs. unilateral hand transplantation), and as additional selection and evaluation issues emerged, the bioethicists were integral members of the core VCA team.

> *"The transplant center has an integrated transplant practice, and although all are solid organs or BMT [bone marrow transplant], they are under the same roof, so we should try to merge those [areas of expertise] together'... And if you look at it, you need surgeons to be able to do the procedures. You need physical therapy, like a hand therapist, and we need physiatry because physiatry helps with the rehabilitation. Then you have the amputee clinic because the amputee clinic manages individuals who are amputees, so their expertise was important...So what you're doing there is trying to make sure that, for the normal entities that take care of a particular population, you have representation." (VCA team member, #1)*

> *"[From conversations with another VCA team]...they viewed their team as like a spider web, and there were all these nodal points of information that then would go up through the web. They felt in their team, there was one person who kind of was the spider who caught all the information. And they proposed that with every team, there's one person who kind of functions in that role...in our team, it isn't one single person who does that. I think because of a shared electronic medical record and the [institutional] tradition of us all working in a very close fashion, team meetings, frequent consultations, we're not all separated...."[VCA team member, #2]*

> *". . .I recall that the VCA team began to form as more and more prospects for transplantation began to develop. My primary involvement was to be a part of all of those meetings. I attended all of the VCA meetings and, as a result of that, my council was often solicited with respect to clinically relevant issues related to VCA." (VCA team member, #3)*

Mediators

Mediators included the development of action-oriented (e.g., communication, leadership), strategic (e.g., planning, training) and interpersonal (e.g., conflict management, trust building) processes. These processes shaped how the team considers potential candidates, functioned, and managed conflict under stress, leverageed individual strengths, and prepared for optimal performance. Participants highlighted two key strategies that helped to establish and sustain a highly functional and prepared team. The first strategy was the structure of scheduled meetings for assessment and evaluation of potential VCA candidates. The primary purpose for these ongoing meetings was to review a potential candidate's case by evaluating the medical criteria for transplant, psychological assessments, and a patient's psychosocial well-being and available social support. Team members presented evaluation reports for potential candidates. Although evaluation of medical and transplant criteria were relatively straight-forward, the psychological and psychosocial evaluations were more challenging and ambiguous. Team leaders welcomed differing opinions about the evaluation conclusions from any team member and when concerns were raised, the team had a thorough and deliberate discussion. If deemed appropriate, additional information about that concern was collected or new team members with expertise about that concern were included. Based on participant interview data and observation notes, the unintentional consequence of these meetings was action-oriented and interpersonal processes that built trust, collaboration, and conflict resolution among the team. Team meetings led to frank discussions about selection criteria for candidacy, especially the psychosocial readiness of candidates both before and after listing them for transplant. These meetings continued even when there was not an active candidate listed for transplant or when surgery preparation or performance was imminent, allowing teams to continue to form and develop cohesion. All disciplines involved in VCA were included and participants remarked that leaders flattened hierarchies, thus reinforcing a sense of each discipline's critical contribution to the overall team and trust across disciplines. They emphasized individual expertise and the mutual contribution that each expert brought to the process. After a candidate was listed these thorough and candid discussions promoted a collective commitment and reinforced the team's belief that VCA was a viable, safe, and reasonable option that would benefit the selected candidate.

> *[regarding leadership in team meetings] ". . .taking turns in speaking so that everybody contributes about the same number of minutes to the discussion. It's this very egalitarian way, so everybody feels comfortable enough to say something. There's no top-down leadership style. . .it's easy within medicine for there to be a hierarchy. But this just doesn't feel that way. It really feels like you are truly all equal contributors to a team." (VCA team member, #4)*

> *[team processes regarding selection and evaluation]". . .we began early on, I believe, as a group to formulate a sense of what risk meant to us and what risk meant to those that we assumed were so vulnerable. These terms all began to take on different shades. I think, prior to my involvement [on the VCA team], I would have never had problems articulating what I thought was consistent with the word "risk," but that changed substantially over time, and I believe it did for my colleagues, too. So, we were actively growing together and juggling these very abstract terms, all of which were superimposed on a very intense sense of beneficence and commitment to the welfare of potential recipients." (VCA team member, #2)*

> *[challenges with selection] ". . . if there were a patient who you felt some conflict with or some incompatibility up front that that would be very difficult to work through as a team and to have that patient have a good outcome. (VCA team member, #4)*

> *"[the team process for VCA preparation]. . .it really is like launching a space rocket, that you have to have many, many, many, many, many, many checks and balances. And if the safety light is lighting up, you gotta stop everything, and you've gotta go back and check and not proceed until all systems are go. And if all systems aren't go, then you shouldn't go. I think [the recipient] did get that message, that that's how we would proceed because it's such an enormous responsibility to [the recipient] to do it right. . .. You've gotta have everybody kind of matched and moving in a concerted way together, and all your harnesses have to be buckled up and secured before you can really proceed safely. (VCA team member, #2)*

The second key strategy was VCA team practice sessions in preparation for when a suitable VCA candidate was selected. Team members volunteered their time over 50 Saturdays to create protocols, practice transplant techniques, safety plans, hand-offs, and transitions in the transplant process (Amer et al., 2018). These practice sessions helped form strategic, action-oriented, and interpersonal processes for the team. The team developed a commitment to creating and refining optimal procedures and built trust and cohesion. They formed a collective belief in their skill and efficacy, along with a shared mental model of the VCA process. Practice sessions led to better role clarification, development of contingency plans, and strategies for addressing interpersonal conflicts and administrative barriers. They simulated interpersonal and leadership communication patterns and highlighted areas where additional team coordination was needed. They also informed the need for additional team training, administrative task planning (e.g., blocking operating rooms at short notice), and clarification of necessary procedures outside of the specific VCA procedure (e.g., media relations, communication with and support of donor family). Practice sessions were adapted to the VCA candidate's case once selected.

> *("Interviewer: Were the Saturday sessions everyone just saying "I want to be a part of this" or was it volunteer? How did that happen?) "I mean it was just actually building the team over time. At first it was a small group, two, three people going into the cadaver lab for maybe five or six times or seven times. Trying to decide. . .trying to figure out the basics. And then saying, 'You know what, here's the patient's defect, let's think through how we would do it', and then we thought through that and then once we decided, you know this is a pretty reasonable protocol for this particular defect for our patient. Then we started building the team and rehearsing and of course, still during the rehearsal you are still modifying and improving things." (VCA team member, #5)"*

> *". . .they [the VCA team] practiced and practiced and practiced on cadavers until they almost could do it by memory. They knew every step, every part of step, and it still took, I think he said it took 52 hours, but like an orchestra, everybody came in. I like the analogy of an orchestra. You may be the first violinist but when it's the oboe part, you sit there quietly and let the oboist play. And you can't just say 'Well, I'm the first violinist, you've got to be quiet. I'm going to play.' You play when it's your turn. You come in and do your job and back off and let somebody else do their job. Everybody knows their part. That's what the team's all about. There's nothing special about anyone of us. But as a team we do a pretty good job" (VCA team member, #6)*

> *"I've become interested in how do you build a successful team. . .it is like kind of lightning in a bottle. When you have it, it's great. Trying to recreate it is really difficult. Certainly the anatomy lab sessions help tremendously, spending so many Saturdays together working together on this common project. . .. So I think a huge amount of the credit goes to [the surgical leader] and then, yeah, forming those bonds in the anatomy lab, working towards that common goal. And then it also helps when you have a great outcome, too. It just bonds the team further together." (VCA team member, #4)*

Outputs

Outputs included team learning, patient selection and transplant outcomes, and sustainability of the team and its processes. As noted, team participants had a collective commitment to assuring optimal patient outcomes, regardless of whether a potential candidate progressed to VCA. In addition to the overall success of the transplant, each teamlet had their own individual markers of success for VCA that were associated with their assigned task and area of expertise.

> *". . .afterwards he was able to smile on both sides. Because that's – that's success. Success is getting the smile to – you know it's not perfect but to look like a smile. And he's able to close his mouth." (VCA team member, #5)*

> *". . .as surgeons, we would wanna see function, you know. We would wanna see sensation. (VCA team member, #7)*

> *"If I was to talk to a new team forming,. . .[I] would suggest to them that they get a sense of the rapport-building abilities of the other team members. How do the physician, medical director, and the key physical therapy staff . . .they have to be people who are experienced and known to be able to work well with patients. This is something you've gotta really have some years of clinical experience to do and be vetted by your surgical departments before you move forward. (VCA team member, #2)*

More elusive to the team, however, was building consensus on the optimal approach for determining at each step of the VCA process (assessment/evaluation, procedure, recovery, and rehabilitation) how the expectations and goals of patients are being met. This reflection led to a new cycle of inputs and mediators for the VCA team to consider. These included capturing patient expectations early, assessing alignment of early expectations with procedural realities, and refining tailored educational and rehabilitation efforts to meet those expectations.

> *"[With VCA]. . . I'm definitely thinking about for a future candidate. . .I think that's where some peer mentoring from other patients is so important because there are just so few*

> *people who've ever gone through this." (VCA team member, #2)*

> *"They [VCA recipients] don't want to [only] get their hand back so that they can go back to work, they want their hand back so that they can touch the face of their loved one. So that they can hold the hand of their loved one. . .it's holding hands and feeling the skin of their hands. [The hand] kind of is an intimacy organ, you could say. . . they sustain these primary family relationships. So after I started having these new thoughts [of hands as intimacy organs] I started thinking 'now I really haven't thought this through the right way."' (VCA team member, #8)*

Discussion

The growing recognition that complex problems are often best served by cross-disciplinary expertise and intense collaboration has propelled the evolving field of team science. Transplant surgery, including VCA, relies on transdisciplinary teams, collaboration across specialties, and coordination of processes to identify, assess, and list potential transplant candidates, and prepare for the transplant, surgery, recovery, and rehabilitation (Costanzo et al., 2010; De Pasquale et al., 2014; Cajita et al., 2017). It is surprising, therefore, that little research has been conducted to describe the team science of transplant, and how a transplant program's composition, formation, and interactions might influence the effectiveness of patient outcomes and team effectiveness. In that regard, our study is relatively novel and provides an understanding of the components of effective teams and some of the ongoing challenges in VCA.

In our study we found that VCA teams require specialized team members whose scope of practice may not otherwise overlap. Team members need to negotiate about balancing patient safety, psychosocial well-being, and multiple risks throughout the transplant process. The VCA team in our case study required: (1) a significant investment of institutional, medical and financial resources to form; (2) multiple team members with specific expertise and highly advanced skills; (3) careful and precise protocols to reduce the risk for mistakes/errors when multiple external factors are out of the team's control; and (4) significant practice and simulation prior to these relatively rare surgeries in order to improve precision of technique and to build role definition, trust, and a collective mental model of metrics for success. The team's self-reflection on their care processes identified gaps in how patient expectations were assessed, how they defined success, and how their expectations and definitions of success changed throughout the transplant process.

Our findings are consistent with other studies in team science. Institutional culture and values, for example, especially a culture of cooperation where shared ideas across disciplinary boundaries are communicated and cultivated, is a key indicator to academic innovation (Lee and Jabloner, 2017). As we found in our study, team effectiveness has been shown to hinge on team member familiarity and social cohesiveness, which builds over time and through shared experiences (Stokols et al., 2008). Finally, teams have been shown to perform best when there is cooperation and interdependence of a team's tasks and rewards (Stokols et al., 2008).

Our study does have limitations to consider. First, our examination of team science emerged organically from interview and observation data and was not the original intention of the study. Targeted questions about team science, therefore, were not asked of participants. Future research can build from our findings by developing intentional questions about how teams function and the effect that a team's cohesion has on patient outcomes. Based on our findings we provide potential questions for others to consider when evaluating their own teams in **Supplementary Figure 1**. Second, the VCA team studied had, to date, only completed one VCA surgery. Data from teams that have a longer record of completed transplants may result in different foci or mediators that affect outcomes. Capturing data from an established team with fewer transplants, however, may provide acute insights into the challenges of early formation of a VCA team or for programs considering expansion into VCA. Third, only two sources of data were used: participant interviews and observations. Additional data sources may have resulted in either a deeper understanding of team dynamics or additional characteristics of the team not found in the available data. Future research may benefit from using sensors to capture data, such was done by Rosen et al. (2018) or by using other methods, such as video reflexive ethnography (Carroll et al., 2018) where team members review video of their teamwork in order to reflect on opportunities for improvement. These alternative methods may be especially appropriate for specific stages of VCA, such as surgery and recovery and to inform later phases of team development, reflexivity, and sustainability. Finally, understandably, our data had a greater emphasis on the impact of team science on surgery preparation and performance, and less on how team science could directly affect candidate selection and indirectly, recipient outcomes. In order for teams to reflect on process and system improvement, future research could include ethnographic investigations that include different team approaches from candidate identification through final treatment choices. Future research could also focus on how teams build and maintain cohesion and manage attrition during the less intense stages of candidate identification and assessment or when surgery preparation and performance is not imminent.

In spite of these limitations, findings from this study may be especially helpful for other programs considering how to form a team to expand into VCA or improve existing teams, but they may also be of interest to other highly complex, transdisciplinary teams who perform procedures that are not life-saving, but rather aimed at improving patients' quality of life, such as gender reassignment surgery and bariatric surgery. Based on our findings, these newly forming teams should consider selecting highly experienced, compatible members to participate; an open, methodical, and collaborative approach to leadership; development of team building processes that provides opportunities to practice and build trust; team consensus on definitions of team and patient success; and strategies for self-reflection and evaluation.

Data availability statement

The original contributions presented in this study are included in the article/**Supplementary material**. Further inquiries can be directed to the corresponding author.

Ethics statement

The studies involving human participants were reviewed and approved by the Mayo Clinic Institutional Review Board. Written informed consent for participation was not required for this study in accordance with the national legislation and the institutional requirements.

Author contributions

JG: study conception and design and initial draft of the manuscript. JG and KB: data collection. JG and CK: analysis and interpretation of the data. JG, CK, KB, IH, HA, and SJ-G: critical revisions. All authors reviewed the results, edited early versions of the manuscript, read, and approved the final version of the manuscript.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2022.935507/full#supplementary-material

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Education for patients with limb loss or absence: Aging, overuse concerns, and patient treatment knowledge gaps

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The goals of vascular composite allotransplantation (VCA) for hand are to maximize functional status and psychosocial wellbeing and to improve quality of life. Candidates are carefully vetted by transplant programs through an extensive evaluation process to exclude those patients with contraindications and to select those that are most likely to attain functional or quality of life benefit from transplant. Patient choice for any treatment, however, requires that candidates be able to understand the risks, benefits, and alternatives before choosing to proceed. This study aimed to understand patients' knowledge and perceptions about treatment options for hand loss, including hand transplant. This study will be used to inform a standardized education approach and develop conversation aids for use by clinicians and patients throughout the treatment decision process. Ten individuals who had experienced hand amputation or had congenital limb loss were interviewed to better understand previous and current decisions about treatment, experiences in adjusting to their treatment, and perceptions about hand VCA. From this qualitative interview data, four findings emerged: (1) knowledge and education around VCA as a treatment option; (2) adaptation of individuals with limb loss; (3) fear of risk associated with transplantation; (4) issues of aging and overuse injuries to existing limbs. Results suggests that there is opportunity for expanding education about all treatment options for patients with new loss, long-term loss, and congenital limb loss. Establishing a baseline of knowledge about all options–prosthetics, rehabilitative strategies, and VCA—can help patients evaluate their values and goals of treatment. Issues associated with aging, including overuse and injury, and adaptability over the life course should be included in considerations about treatment choices. Data indicate the need for routinely assessing patient preferences about treatment choice so patients can plan for their future as they adapt and age and as technology for treatments change. To assure that thorough information is provided for current and future decision-making, education about treatment choices and selection procedures for VCA should be standardized.

KEYWORDS

vascularized composite allotransplantation, education, qualitative interviews, standardized tools for evaluation, VCA

Introduction

In 2005, the worldwide prevalence of limb amputations was 1.6 million, and projections suggest that number may double by 2050 (Ziegler-Graham et al., 2008). Treatment options for upper extremity amputations include prosthesis, replantation, and vascular composite allotransplantation (VCA), with prothesis the most common treatment option. VCA is still rare but is becoming a feasible surgical alternative to replantation and prosthesis for some amputees.

The first hand transplant was performed in 1998 (Dubernard et al., 1999). Since then, more than 113 VCA hand procedures have been performed worldwide on 76 patients, some unilateral and some bilateral (Alolabi et al., 2017). Primary goals of hand transplantation are to maximize functional status, emotional status, and quality of life. Patient selection for hand transplant is complex and requires a thorough evaluation to assess the impact, risks, and benefits for each individual and to understand if hand transplantation will improve their quality of life. Developing standardized tools for identifying, evaluating, and assessing potential candidates and standardized approaches for patient education are key to assuring that selected candidates will benefit from transplant (Kumnig et al., 2014; Jowsey-Gregoire and Kumnig, 2016).

As an alternative to replantation and prosthesis, VCA can offer the potential for better sensation outcomes than prosthetics (Pasquina et al., 2006; Schuind et al., 2007). Data from all hand transplant recipients suggests improved protective sensibility of the recipients assessed, with 91% having tactile sensibility, and 82% regaining partial two-point discrimination sensibility. This improved function enabled independent living and, for some, return to full-time occupation (Shores et al., 2017). However, in a study comparing functional and psychological outcomes of hand transplant versus prosthetic users, no significant differences were found in functional outcomes between the two, but transplant patients did have higher scores in psychological areas of "role-physical," "vitality," "role-emotional," and "mental health" (Salminger et al., 2016).

While the option of VCA may provide functional, psychological, social, and even cosmetic benefits for some, there are significant risks associated with lifelong immunosuppression (Dubernard et al., 1999; Hautz et al., 2020). Challenges also exist for VCA candidates around financial burden, missed work, and insurance coverage (Chung et al., 2010; Alolabi et al., 2015, 2017). An economic analysis of hand transplants indicated hand transplant to be significantly more costly than prosthesis and concluded that a unilateral hand transplant would not be considered cost-effective (Chung et al., 2010; MacKay et al., 2014). The decision-making process for treatment options for people with amputation is a balance between weighing risks, burdens, and benefits for all treatments. Seeking to improve functionality is a universal goal across treatment choices, but the broad aspects of quality of life are less frequently assessed. Hand transplant may improve quality of life, but VCA also includes a complex set of life-long risks. Understanding the broad spectrum of quality of life issues is critical for being able to weigh the risks and benefits. Patient satisfaction, for example, is one aspect of quality of life that is significantly related to function (Hautz et al., 2020), but other aspects, such as psychosocial function, wellbeing, and meaning making, are less commonly assessed. Bahler (2019) has commented on factors essential to quality of life that are currently not included in most patient assessments: ". . .our capacity to make meaning, particularly in relation to temporality, embodiment, and intersubjectivity. Without attention to these features of the human condition, assessment of hand transplant recipients' functional capacity and psychological sense of satisfaction will remain incomplete"(Bahler, 2019).

With the expected increase in prevalence of limb loss and the complexity of risks and benefits for treatment options, data are needed to inform the development of standardized tools to assess a broad set of quality of life factors and develop patient education materials that can promote shared decision making. In this study, we aim to fill this gap by utilizing qualitative research methods to better understand the values, attitudes, and expectations of treatment for people with limb loss that may be associated with future quality of life and address patient education needs.

Materials and methods

Study design

To understand factors integral to treatment decision-making and education for potential VCA-eligible individuals (age range = 38–72), we conducted semi-structured interviews between March 2019 and March 2021 with ten individuals with upper extremity loss who were potentially eligible for upper limb transplantation. Interviews were conducted with participants from a variety of backgrounds, and great variation in their path to extremity loss, including congenital loss, active-duty military loss, and loss due to hospital-acquired sepsis.

Source of participants

Purposeful sampling was initially used to identify participants through medical record review of patients with limb absence. Snowball sampling was used for further recruitment as was outreach to amputee advocacy organizations. Participants were contacted *via* recruitment letter or email by the study coordinator to explain the study purpose and request participation. If no response to the initial contact was received, a follow-up contact was made. A study coordinator contacted interested participants to schedule interviews with one of three interviewers (KS, JG, and DF). Interviewers included two experienced Ph.D. trained qualitative researchers and one experienced Master's degree trained qualitative researcher. Recruitment and data collection concluded when thematic saturation was reached.

Source of data

The semi-structured interview guide was created to capture the lived experience of individuals with limb absence, quality of life, and how they approach healthcare decisions. It included questions on how they came to have an upper limb extremity loss, discussed their life without an upper limb(s), their experience or knowledge of prosthetics, and thoughts and interest around hand transplantation. The guide was iteratively adjusted to fit each individual's unique situation and to gather the most detailed and pertinent data from each participant. For example, questions were modified to capture the lived experience of an individual with congenital limb loss and modified again for a person with a military injury resulting in limb loss, as the treatment options and decision-making process may also be different.

All interviews were digitally recorded, transcribed verbatim, and de-identified (removing names and other identifiable features). Recorded interviews were stored on a secure server for analysis in Nvivo (Qi Li Group Pty Ltd, 2020).

Data analytic approach

The qualitative research team (DF and JG) reviewed each transcript as they became available and listened to the interview audios. They independently read the interviews multiple times and listened to the audios to become immersed in the data. Initial key concepts were captured from the data and supported by the interpretive qualitative framework of medical anthropology (Lambert and McKevitt, 2002; Bernard, 2011), emphasizing the lived experience of individuals through the narrative they share about their healthcare journey. Interviews were semi-structured but allowed for flexibility to probe new or emerging issues.

Descriptive coding of interview transcripts ran concurrently with data collection, consistent with iterative thematic analysis (Corbin and Strauss, 2015). After each new interview, DF, a qualitative research analyst, and JG, a health services researcher, with qualitative and mixed methods expertise, independently read and analyzed each transcript and met regularly to reflect on the data and discuss the concepts, themes, and codes. With the review of each transcript and constant comparison across transcripts, a deeper understanding of the themes was reached. Concurrent analysis with data collection allowed for iterative interaction between data and analysis in order to enhance reliability (Morse et al., 2002). Codes and themes were then organized and analyzed through interpretive thematic analysis (Corbin and Strauss, 2008), and each transcript was entered into Nvivo. Data were organized by code and theme.

We concluded data collection after ten interviews because no new themes emerged.

Results

As shown in **Table 1**, three women and seven men, ranging in ages from 38 to 72 were interviewed. Participants were individuals with varying amounts of time since initial extremity loss, from 1 to 64 years. Five had unilateral limb loss and five had bilateral limb loss. None were VCA recipients.

As common concepts and ideas emerged through the interview and coding process, four themes were identified: (1) awareness and knowledge of transplantation; (2) aging with amputation and/or VCA and overuse and injuries of existing limbs; (3) fear of risk associated with transplantation; and (4) contentment and adaptability to limb absence in long-term amputees. Themes included factors impacting knowledge about VCA and treatment decision making. Each subsequent interview helped to explore more depth into initial themes.

TABLE 1 Hand absence interview demographics.

	Gender	Age (years)	Years with amputation	Cause of extremity loss
1	F	64	4	Sepsis/bilateral.
2	M	50	22	Work injury/unilateral.
3	M	64	64	Congenital/partial bilateral.
4	M	38	2	Infection/bilateral.
5	M	65	40	Farm accident.
6	M	71	15	Arm shot off in active duty and reattached.
7	M	49	49 (additional wrist amputation 2 years)	Born without hand but had a bit of wrist. Two years ago, had squamous cell cancer and removed wrist.
8	F	72	1	Sepsis–bilateral. Lost all toes.
9	F	67	64	Gunshot accident as a child. Arm amputated about 1” below elbow.
10	M	59	1	Skin cancer. Amputation of both arms. Skin cancer removed from ear, face, back.

Adaptability to limb absence

Participants living successfully with upper extremity loss/absence identified ways they have adapted their jobs, hobbies, and lives, and continue to adapt as they age.

These individuals have been living successfully with various forms of prosthetics and have adapted their routines. Participants living for longer than 15 years ($n = 6$) with an upper extremity absence also noted that they had adapted their lives to incorporate their limb loss. They had completed education and degrees, changed careers and started businesses, incorporated adaptations to their hobbies and outside activities, maintained family and social relationships, and spoke about their resilience and adaptability as assets. Interestingly, none of the participants interviewed referred to themselves as disabled or handicapped, and some rejected the terms when used in reference to themselves.

“. . .you know, if you’ve maybe got just a couple of fingers, there have been many times I wish I had just one finger on the end of my arm, just one, to be able to do something. That’s a frustration of mine. They’re saying, ‘Well don’t you want the whole hand?’ Not necessarily. The way I do things now, a lot of times, all I need is that one damn finger.” (male, 64, congenital loss of one arm).

“Yeah. I’ve got stuff. I mean we go kayaking. I got stuff to hold the paddle. I’ve got stuff for the ATV. I got some things that hold guns when I shoot that will hold the forearm. Yeah cause that’s what I do at work. So once in a while, I’ll make something that I can’t buy somewhere, and I’ll just make my own, you know. Like even for like when we go deer - for cleaning deer, I got one with a knife on the end I can hold and skin it, you know, and stuff - just weird, different - I got one of the big vice grips so I can clamp things and heat ‘em with a torch, just different things.” (male, 65, lost hand 40 years prior in farming accident).

“I learned how to put screws in my cutting board, put the chicken on it, and that holds it. I have attachments to open bottles with if I need to. I have a one-handed syringe with the CO2 to open a wine bottle, everything adaptive.” (male, 71, arm shot off in active duty 15 years prior and reattached).

“I do mechanic work my whole life. I was born without my hand originally. So, I guess I’ve learned to adapt very well. I did have a prosthetic when I was younger and grew up through the Shriners. But then, as I got older and started to do more, I found–I was very fortunate I had the left arm all the way down, including the wrist. So, milking cows, driving equipment and stuff, I found that I could do better without the prosthetic just because I had the skin, I had the length, I could reach and do everything. I could balance the milkers better. I could drive the equipment better without the big rigid hook being in the way. I know technology has changed, obviously, quite a bit in 30 years [laughs].” (male, 49, congenital hand, additional wrist amputation 2 years prior due to cancer).

“I’ve gotten so used to living without my arm, my prosthesis almost gets in the way. It almost gets in the way because I’ve learned how to do so much in my armpit and holding my stump down and all kinds of stuff . . . I’ve learned how to do stuff so much without my arm, now my arm is like–I’m learning how to re-use my arm and keep it out of the way. When I need it, I need it. I haven’t needed it for nothing yet.” (male, 59, skin cancer led to arm amputation one year prior).

Lack of awareness or knowledge of vascular composite allotransplantation as an option

Participants’ knowledge about upper limb transplantation was varied. Some were aware of cases of upper extremity transplantation but had little knowledge about details of eligibility. Some had an understanding the risk and benefits but did not consider it an option for themselves due to age or the burden of immunosuppression. Some had no knowledge at all. Some variation in knowledge was attributed to time since amputation. Participants that were either born without a limb or lost a limb as a child ($n = 3$) (current ages 64, 49, 67) and prior to the advent of VCA, for example, considered themselves adapted to their circumstances. Their discussions about transplantation suggested that they had some knowledge about hand transplantation but had not taken time to investigate transplant as an option for themselves because they did not see it as relevant to their situation.

Participants suggested that the challenges associated with insurance coverage for treatment or loss of work due to time needed for rehabilitation and recovery was also a concern with any treatment and may contribute to a lack of seeking information about transplant. Communicating with insurance for coverage of prosthetics was identified as an ongoing challenge and worry about insurance coverage for any treatment was discussed. For individuals that see themselves as productive and capable, the loss of work time and rehabilitation burden on family was also identified as a transplant consideration.

"... it takes me back to when people would come to the bedside to talk to me about, 'Wanna transplant this, wanna transplant that, and we can take your thumb, and we can take your fingers, and we can take ligaments from the other side,' you know all these conversations. And anybody else that came into the room was speaking the same language as they were. And I wanted to hear an advocate for the prosthetics, when that was a possible action, and an advocate for all the other options, not somebody who was presenting [all] the options under the umbrella of the one choice..." (male, 71, arm shot off in active duty 15 years prior and reattached).

"I guess this is all a part of like, making the justification for me to get a better hand or arm, and I guess we just have to keep doing this and going through these processes and adjusting what I have so the insurance will see that we've tried because I - I guess there are arms out there now that you can control grasp. I've seen it online - stuff where they actually do implants where they can implant sensors into your arm so like if you open a hand or close a hand or move your index finger, you could do all of that with a prosthetic. So, like I guess if it were ever to get that advanced, and my insurance would pay for it... I've been denied everything, and it was like jumping through hoops just to get me that myoelectric arm that I have...Everything's been just so hard." (male, 38, bilateral loss to infection 2 years prior).

Aging of patient and caregivers and overuse injury worries

When asked about concerns participants had about transplantation, participants organically shared their worries about their age or about aging. They voiced concerns about their own aging and their caregivers aging. With over half the study participants being over the age of 60, many expressed they felt the complexity of the VCA surgery, the recovery time, and the risks associated with lifelong immunosuppression were too much for them at their current age. Three individuals, however, felt they may have considered transplantation if they were younger. Fears of growing older, overuse injuries, and aging primary caregivers are also prevalent.

"I guess the only thing is the fear of aging. Right now, I'm able to do this stuff, and I have a husband who's gonna do this stuff. But what happens when he can't or isn't around. As I get older, I'm not sure - now I've certainly thought a lot about it and looked at a few different things, and my mother is gonna be 90 next year, and she and I have been visiting some senior care facilities, and that's probably where I'll end up. But it's still very unnerving where really, up until this happened, I wasn't that scared of retiring or even living in a senior facility. But now I'm like well - somebody else is gonna have to dress me and help me and all - do all that stuff." (female, 64, sepsis, bilateral 4 years prior).

"Missing the limb, yep...I'm 50 now, and the thing I gotta think about is I don't wanna wear this one out. My biggest fear is I tear a rotator cuff and then your arm is immobilized for three months, and who knows what the outcome is gonna be. What do I got to use?" (male, 50, accident - lost limb at age 28).

"If they actually get to the point where they are doing transplants, if it's not an issue and insurance covers it, I definitely would get on the list. I'd be interested in doing it. I'd check it out just because thinking down the road, you know, like when I get older and I'm not as active or as strong as I am now, life's probably gonna get a little harder... That's the only thing that I really think of is just when I get older and if I'm gonna be able to do the things that I've learned or that I'm able to do now." (male, 38, bilateral lost to infection 2 years prior).

"It's just it eventually comes up–"Okay, I'm getting carpal tunnel." I go, "Gee, imagine that. Only have one hand and you type all day. What do you think you're going to get?" Well, part of it being, you have to look at my age. Do I need something that's that complicated at this stage in my life? So, that's how I looked at it. Now, had I been 30 years old, that would have been a whole different ball game, you know?" (female, 67, gunshot accident as a child).

Perceptions of risks and benefits of vascular composite allotransplantation

Participants described added functional use and dexterity as possible benefits of hand transplantation, but also specified that they would only consider VCA if the function was above and beyond the function gained from their current state or with current prosthetics. Some valued the potential for sensation which can only be gained with VCA, including the greater ability to express emotion and care for others (e.g., hugging their grandchildren). Others described the benefits of social perceptions of having a hand, including looking "normal." Participants also voiced concern about having the transplant rejected and the potential suffering of having to go through amputation again.

"And it's like I can't imagine being able to do most things for myself to go back to being bedridden and needing all that help from everyone until you can actually use your arms" (male, 38, bilateral lost to infection 2 years prior).

"I mean when you've been through the rejections stuff and, basically, the therapy and how long it would take to recover,

that was probably the biggest thing for me, where they're talking, and you could be down for a year just trying to let it heal and rehabilitate. I have a young daughter. I'm not going to be out of anything for a year. That can't happen. I mean I've got to be there for her, and that's the #1 thing I'm going to do, and I can't walk away from my farm" (male, 49, congenital hand, add'l wrist amputation 2 years prior due to cancer).

"Do you want to do this for the rest of your life? And what happens if–I mean I don't know much about this immunosuppressant-type drugs or anything like that; but if you have to take them for the rest of your life, what happens if you can't take them for the rest of your life? Does your hand fall off?" (female, 67, gunshot accident as a child).

Discussion

In this qualitative study of people living with limb loss, participants had adapted to their loss, finding creative ways to modify their activities of daily living and reducing their reliance on others to assist. They discussed their adaptations using prostheses and their knowledge about different treatment options, including VCA. They had variable degrees of seeking out knowledge about VCA and its requirements. We found that some participants had not explored other treatment options, such as VCA, because of concerns about financial burden or lack of insurance coverage. Our findings are consistent with Talbot et al. (2019) who found that adjustment to amputation (limb loss) was inversely related to an interest in transplantation and concluded this may help explain the difficulty in identifying and selecting candidates for VCA. One possible solution for addressing concerns about financial burden or insurance coverage is to have clearer communication early in the evaluation or treatment process about financial burdens and what insurance will and will not cover. This communication should include all options, including VCA and prosthetics, so fully informed decisions can be made.

Our data indicate that there is worry and concern around aging as well as overuse and injury of existing limbs. Other studies have also shown issues related to overuse injury in the population of people with amputation (Jones and Davidson, 1999; Burger and Vidmar, 2016; Cancio et al., 2021), however, our study adds to this literature by addressing how VCA is perceived in relation to issues of aging and overuse. Future research should explore how incorporating issues of aging and overuse into shared decision making about treatment choices and throughout the healthcare journey affects patient satisfaction and quality of life.

This study identifies the need for ongoing education throughout the healthcare journey of individuals with limb absence. Throughout their lives, as needs change with age, injury, and caregiver access, there needs to be changing and adapting education provided to facilitate decision-making. Clinically, healthcare providers can routinely incorporate information about healthcare options and the self-care needed to maintain their treatment choice. Discussing overuse injuries, aging for themselves, and aging of caregivers should begin early, even if the individual is not actively considering transplantation. Included in education should be information for patients that considers adaptations to their limb absence and how those adaptations may be impacted by aging or overuse. Opportunities may also exist for education and mentoring for individuals eligible for upper extremity transplantation. Peer mentorship of new amputees may help individuals see the potential for having productive and capable lives, regardless of treatment option chosen. These conversations may also include topics such as loss of work time and rehabilitation burden on family that some may or may not consider when discussing treatment options.

With increased attention on the importance of standardized evaluation for amputees and VCA recipients, our results also suggest that routine assessment of quality of life for treatment options may be useful. Ability to adapt and contentment of current treatment option are additional factors that may be especially important in this population and may impact decision-making and help to capture concerns about aging and overuse. Furthermore, to assure that thorough information is provided for current and future decision-making, education about treatment choices should be standardized. Routine standardized psychosocial assessment and education should support current and future decision-making about treatment choices, including VCA, for all individuals with limb loss.

Limitations

Noted limitations for this study are its small sample size, which is typical of some qualitative interview studies, especially in unique populations such as this convenience sample of individuals that are difficult to find and recruit from the general population Nonetheless, this sample of ten participants represents unique perspectives from a broad range of amputees that can be considered experts on their own healthcare needs and decision making; and the goal of this study was discovery of new information, which can be achieved through small sample size. Future studies that ethnographically capture the changes and trajectory of quality-of-life issues over time or the impact of educational interventions to reduce ambiguity of treatment decision choices would be important next steps for understanding the interconnectedness of treatment choice, selection criteria, assessment of psychosocial health and quality of life and patient functional outcomes.

Conclusion

Although VCA may be a life-changing option for individuals with loss of an upper extremity, this study highlights individuals who have lived with upper extremity loss for an extended period of time and have adapted their lives around their limb absence as well as individuals who are older and may be less motivated or less interested in limb transplantation. It also indicates the low level of VCA knowledge among these individuals. Lack of awareness and knowledge for this study population indicates an opportunity for education about transplantation earlier. With an increased need to develop evidence-based standardized protocols to evaluate hand transplant candidates, this study suggests the need to include in standardized evaluation: assessment of knowledge base; concerns and insight around aging for amputees; and evaluation of those highly adapted individuals who may be content in their life and may not consider their quality of life improved with a hand transplant. This study identifies the need for ongoing education throughout the healthcare journey of individuals with limb absence. Throughout their lives, as needs change with age, injury, and caregiver access, education should be adapted to support individuals' decision making around VCA and other treatment options.

Data availability statement

The datasets presented in this article are not readily available. Due to the nature of this qualitative study, the participants are recruited from a small population sample. De-identified transcripts may contain contextual information on the participant, their lived experience, and their care that could be inadvertently identifiable. To protect these human subjects, these datasets will not be made available. Requests regarding the datasets should be directed to DW, finnie.dawn@mayo.edu.

Ethics statement

The studies involving human participants were reviewed and approved by Mayo Clinic IRB. The patients/participants provided their written informed consent to participate in this study.

Author contributions

KS, DF, and JG collected the interview data. JG and DF performed the qualitative analysis. DF wrote the first draft of the manuscript. JG wrote sections of the manuscript. All authors contributed to conception and design of the study and manuscript revision, read, and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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In the face of change: Which coping strategies predict better psychosocial outcomes in face transplant recipients?

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Objectives: Face transplantation aims to improve patients' quality of life and psychosocial functioning in patients with a disfiguring injury. With 40 cases worldwide, little is known about coping strategies predicting resilient outcomes.

Design: Six patients followed in Boston, completed the Brief COPE (Carver, 1997) along with validated measures of depression, self-esteem, and quality of life – every 3 months during the first year post-transplant and every 6months thereafter, up to 36 months post-transplant.

Analyses: Due to sample size and distribution of the data, nonparametric tests were used to characterize the relation of coping strategies with psychosocial outcomes.

Results: As expected, active coping strategies were associated with better mental health pre-transplant, while avoidant coping strategies were associated with poorer mental health. Patients using support-based strategies reported better mental health at baseline. Post-transplant, the pattern reversed such that avoidant strategies appeared protective, when looking at mental health trajectories over 18months. Importantly, trends identified during the first 18 months matched the trajectories of all patients with existing data up to 36 months post-transplant, for all outcomes measured.

Conclusion: Different coping strategies support optimal outcomes in the pre-versus post-transplant phases. Pre-transplant data may better inform interventions supporting mental health of transplant candidates than predict post-transplant behavior. Early post-transplant data seems to provide promising insight in long term psychosocial outcomes.

Clinical implications: Our data stresses the need for pre-transplant assessment of coping and post-transplant coping training. Research aiming to optimize post-transplant psychosocial outcomes should consider coping as a promising target for intervention.

KEYWORDS

coping, vascularized composite tissue allotransplantation, quality of life, Selfesteem, depression, psychosocial outcomes, face transplant, outcomes

Introduction

"Some things are in our control and others not. Things in our control are opinion, pursuit, desire, aversion, and, in a word, whatever are our own actions. Things not in our control are body, property, reputation, command, and, in one word, whatever are not our own actions" Epictetus, *The Enchiridion*, 135 ACE.

"When life gives you lemons, make lemonade!" Various origins.

Philosophers and popular wisdom agree: the way we react to adversity can help us come out of it on top. But what is the recipe: what are the best ways to cope? This question is particularly important in the context of experimental procedures, where the psychological and surgical teams advising potential candidates have little to no data available to inform their decisions regarding patient selection and treatment recommendations. Vascularized composite allotransplantation (VCA) comprises surgeries such as face transplant, upper-extremity transplant, abdominal wall transplant, etc. Among them, face transplantation may soon become standard of care and be offered much more widely than it has been so far. Medical teams are looking for guidelines to inform patient selection. This makes it particularly important to identify which coping strategies at baseline are associated with better psychosocial outcomes post-transplant.

Face transplantation is an innovative surgical procedure aiming to restore appearance and function in patients with a history of severe facial trauma. Since the world first case in France in 2005, more than 40 patients have received partial or full face transplants around the world (Oser et al., 2018; Tasigiorgos et al., 2019; Kauke et al., 2021). Yet little is known about the psychological traits fostering successful adjustment post-transplant (Nizzi et al., 2017) and patient reported outcomes about quality of life after drastic physical changes are often received with skepticism (Nizzi, 2021). In this longitudinal study of six face-transplant recipients, we investigated which coping strategies predicted better psychosocial outcomes in terms of self-esteem, depression, and quality of life, within the first 18 months post-transplant, and up to 36 months for the first three recipients.

Several measures of coping have been developed over the past 50 years (Billings and Moos, 1981; Folkman and Lazarus, 1985; Endler and Parker, 1990). We used the Brief COPE (Carver et al., 1989; Carver, 1997). This 28-item measure evaluates 14 coping strategies – each assessed by 2 items in the Lickert-scale – and has demonstrated good reliability/validity in similar populations, such as hospitalized patients with burn injury (Amoyal et al., 2011). Factor analyses have yielded inconsistent findings. However, one constant seems to be the distinction between active-approach coping and avoidant coping (Lawrence and Fauerbach, 2003; Prado et al., 2004). Strategies related to active coping often include acceptance, positive reframing, and a proactive problem-solving attitude. Strategies related to avoidant coping include denial, self-blame, and behavioral disengagement. New factors added to those identified in the 1989 study by Carver and colleagues include support seeking, self-distraction, venting, humor, and religion (Carver, 1997).

The distinction between active and avoidant coping is supported by an extensive literature, both in adolescents and adults, and across a variety of conditions (Canada et al., 2006; Frydenberg and Lewis, 2009; Montel et al., 2012; Asuzu and Elumelu, 2013; Barendregt et al., 2015). We first hypothesized that active coping strategies – such as acceptance and positive reframing – would be associated with a higher self-esteem, higher quality of life, and lower depression levels. Conversely, we hypothesized that items pertaining to avoidant coping – such as denial, self-blame, substance use and behavioral disengagement – would be associated with higher depression scores, lower self-esteem, and lower quality of life scores. Our second hypothesis was derived from clinical practice. When assessing pre-surgical candidates, it is commonly considered to be a good protective factor when a candidate has a strong social support system to rely on. We predicted that support-based strategies would be associated with positive mental health outcomes.

Given the small sample and worldwide population size, our goal in this study is to provide an initial description of coping strategies in relation to key mental health outcomes in face-transplant recipients pre-and post-transplant. Our approach is that of a pioneer study, where it is very difficult to access more cases. We expect our result to be meaningful, but they should be interpreted with caution and the ambition to shed initial light on this under-researched field rather than predicting or generalizing findings. We expect our findings to contribute to informing the direction for future studies.

Materials and methods

Sample

Six face transplant recipients participated in this study (2 females, 25 to 57 years old at time of transplantation, mean age = 38). All patients were followed at Brigham and Women's Hospital in Boston at the time of data collection. For a detailed presentation of each case in the cohort, please refer to the cohort description (Oser et al., 2018).

Procedure

Patients completed both coping and outcome measures at baseline prior to their facial transplantation. Patients then completed outcome measures every 3 months during the first year, and every 6 months thereafter.

Measures

Coping

The brief form of the COPE Inventory (Carver, 1997) is used to characterize the strategies one uses to cope with their stress. The 28-item Likert scale assesses 14 different coping behaviors:

self-distraction, active coping, denial, substance use, emotional support, instrumental support, behavioral disengagement, venting, positive reframing, planning, humor, acceptance, religion, and self-blame. The 4-point Likert scale assesses frequency, with higher scores indicating that the respondent reported using the coping strategy more frequently. A sum score for each of the 14 scales can be derived by summing the individual scores for each of the two items of the scale. The Brief COPE does not provide an overall score, cluster-scores, or cut-off scores. It is recommended to define adaptive and maladaptive composites based on previous literature in the relevant population or second-order factors derived from each sample, and to analyze each scale separately to characterize its relation to other variables of interest.[1] The psychometric properties of this scale have been evaluated in hospitalized patients with disfiguring injuries and have demonstrated good reliability, construct validity, and factor structure (Amoyal et al., 2011). To date, the Brief-COPE has been translated in several languages, including French, German, Greek, Korean, and Spanish.

Self esteem

Rosenberg's Self Esteem Scale (RSES (Rosenberg, 1965) has received more psychometric validation than any other measure of self-esteem (Robins et al., 2001). Scores on this 10-item Likert scale range from 10 to 40, with a higher score indicating a higher level of self-esteem. Whilst originally developed with adolescent populations, the scale has widely used across adult populations (Robins et al., 2001) and has good internal consistency specifically with patients suffering from disfiguring injuries (Nicolosi et al., 2013).

Depression

The Center for Epidemiologic Studies Depression Scale (CES-D) is a short self-report scale designed to measure depressive symptomatology. It consists of a validated list of 20 symptoms, each rated by responders on a scale of 0 to 3 according to its prevalence during the past week (0 = rarely or none of the time, 1 = some or little of the time, 2 = moderately or much of the time, 3 = most or all the time, Radloff, 1977; Lewinsohn et al., 1997). The total score, ranging from 0 to 60 (clinical cut-off at 16), indicates the sum prevalence of depressive symptoms, and corresponds with high sensitivity and specificity to the overall risk of clinical depression (Lewinsohn et al., 1997).

Health related quality of life

The EQ-5D was administered to assess health-related quality of life using a visual analogue scale to rate current health state along a continuous scale from 0 ("worst imaginable health state") to 100 ("best imaginable health state"). The measure has shown good reliability with a range of clinical populations (Cleemput et al., 2004) and strong construct validity in patients with severe skin injuries (Oster et al., 2009; Shaw et al., 2005).

Statistical analyses

Statistical analyses were completed using the R software, version 3.2.2. Significance level was set at 0.05. Due to sample size and distribution of the data, nonparametric tests were used to calculate correlations first within, then across subjects. Bonferroni correction was applied for multiple comparisons.

Within subjects, we explored the relations between outcome measures, based on each patient's scores over the first 18 months post-transplant, using Spearman correlation coefficients.

Across subjects, we explored the relations between the 14 coping strategies using Spearman correlation coefficients (lines 1–17 in Figure 1). We then explored the relation between each coping strategy and each psychosocial measure, at baseline (lines 18–20).

For post-transplant outcome analyses, we computed each patient's average score for post-transplant outcome measures, then correlated this average score with their baseline coping scores (lines 21–23). In half of our sample, we also collected data from 18 to 36 months post-transplant. For these patients, we fit a linear regression line for each outcome measure to determine their outcome trajectories based on data from baseline to 18 months. Trend projections were then tested in data collected from 18 to 36 months post-transplant, using visual analysis of mental health trajectories as well as a comparison of slopes obtained in the linear regressions.

Results

At the individual level, five of the six patients showed associations in the expected direction between mental health outcomes. Depression was negatively correlated with self-esteem (= −0.14 to-0.94) and with quality of life ($r = -0.68$ to-0.97), while self-esteem was positively correlated with quality of life ($r = 0.11$ to 0.99). One patient showed an inverted pattern of response, such that depression was positively correlated with self-esteem ($r = 0.63$, $p < 0.01$) and had a similar trend for quality of life ($r = 0.16$, ns), while self-esteem trended towards negative correlation with quality of life ($r = -0.20$, ns). Results for the three mental health outcome measures are thus reported separately.

At the group level for baseline (Figure 1), only positive reframing had a significant association with mental health outcomes among active coping strategies, with QoL ($r = 0.66$, $p < 0.001$) and self-esteem ($r = 0.41$, $p = 0.05$). Acceptance trended with higher self-esteem ($r = 0.36$, ns). Among avoidant strategies, denial, disengagement, and self-blame were all strongly associated with higher depression and lower self-esteem (Figure 1). In our sample, planning seems to have behaved like self-distraction: both were also strongly associated with higher depression (correlations ranging from 0.69 to 0.71, $p < 0.001$) and lower self-esteem ($r = 0.90$ to 0.93, $p < 0.001$). Planning had the highest correlation with depression scores ($r = 0.82$, $p < 0.001$). Self-distraction stood out among avoidant strategies for a moderate positive correlation with QoL.

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[1] https://local.psy.miami.edu/faculty/ccarver/sclBrCOPE.phtml

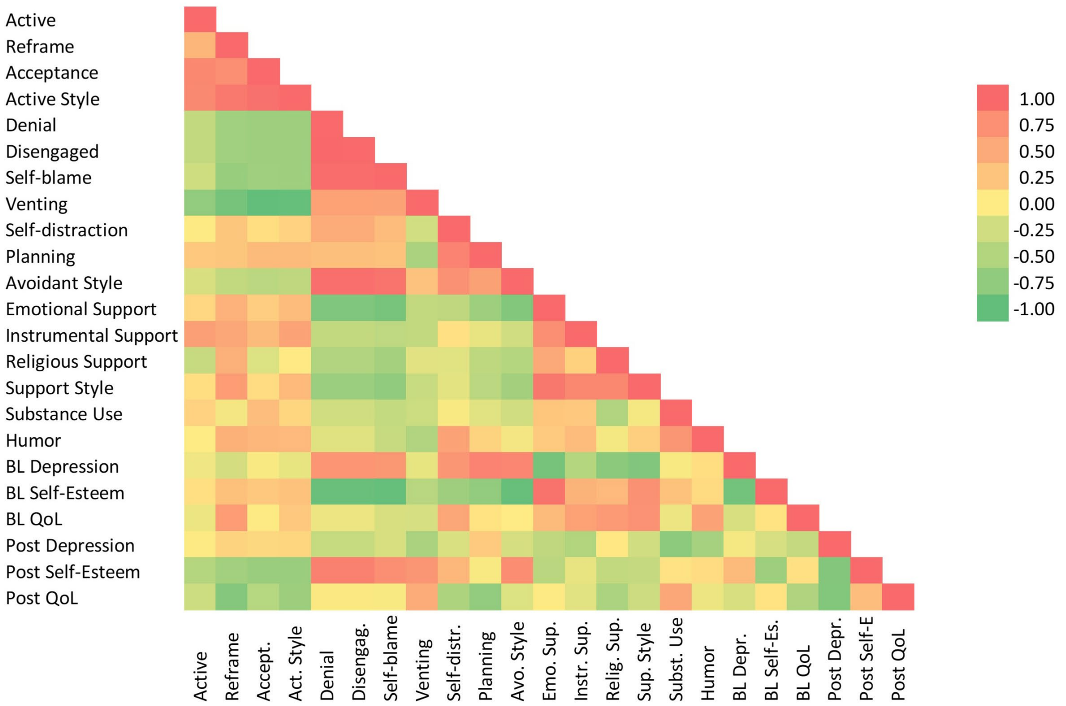

FIGURE 1
Heat map.

All three support-based strategies were associated with lower depression ($r = -0.38$ to-0.81), higher self-esteem ($r = 0.47$ to 0.94), and higher QoL ($r = 0.45$ to 0.67). Receiving emotional support from others was strongly protective, followed by finding support in one's faith. Humor showed a moderate to high positive correlation with QoL ($r = 0.61$, $p < 0.001$).

To look at trends over 18 months post-transplant, a linear regression line was fitted for each patient in each mental health outcome measured. Overall, 2 patients worsened across all outcomes (S1, S5), 2 patients improved across all outcomes (S2, S6), and 2 patients remained stable with mixed patterns (better QoL but worse depression). We found a strong negative correlation both between depression and self-esteem and between depression and QoL, while we found a moderate positive correlation between self-esteem and QoL, all of which went in the expected direction. Comparing pre-and post-outcome measures, depression scores showed a very low positive correlation with each other, while self-esteem and QoL showed moderate negative correlations between baseline assessment and post-transplant trend.

Post-transplant (Figure 1), active coping strategies were associated with worsening self-esteem and QoL trends. Positive reframing and acceptance were associated with decreased self-esteem ($r = -0.51$, $p < 0.05$) and lower QoL ($r = -0.71$, $p < 0.001$). Avoidant strategies like denial, disengagement, and self-blame were all associated with higher self-esteem post-transplant ($r = 0.75$ to 0.84, $p < 0.001$). Venting was also associated with lower depression trend ($r = -0.46$, $p < 0.05$) and higher QoL ($r = 0.55$, $p < 0.01$). Self-distraction and planning continued to be associated with worsening QoL post-transplant. Overall, avoidant strategies correlated with better self-esteem post-transplant.

The three support-based strategies continued to trend towards lower depression but failed to reach significance. Religious support correlated with worsening QoL trends ($r = -0.45$, $p < 0.05$). Humor was associated with lower depression trends ($r = -0.48$, $p < 0.05$).

Finally, outcome data was available to test the predicted trends in three of the patients at 24-, 30-, and 36-months post-transplant. Trends identified in all outcome trajectories were confirmed for all patients (Figures 2–4).

Discussion

Our goal was to describe coping strategies associated with the most resilient mental health outcomes in this sample of face transplant recipients, pre-and post-transplant.

In line with the literature in other clinical populations, active coping strategies were associated with better mental health at baseline, while avoidant strategies showed strong associations with negative mental health. In our sample, poor baseline mental health appeared to be mostly linked to the use of avoidant strategies, such as denial, behavioral disengagement, and self-blame – which were strongly associated with poor outcomes in depression and self-esteem. The main clinical implication of this finding underscores the importance of assessing coping in face transplant candidates, so that interventions targeting ineffective strategies may be implemented to support the mental health of candidates while

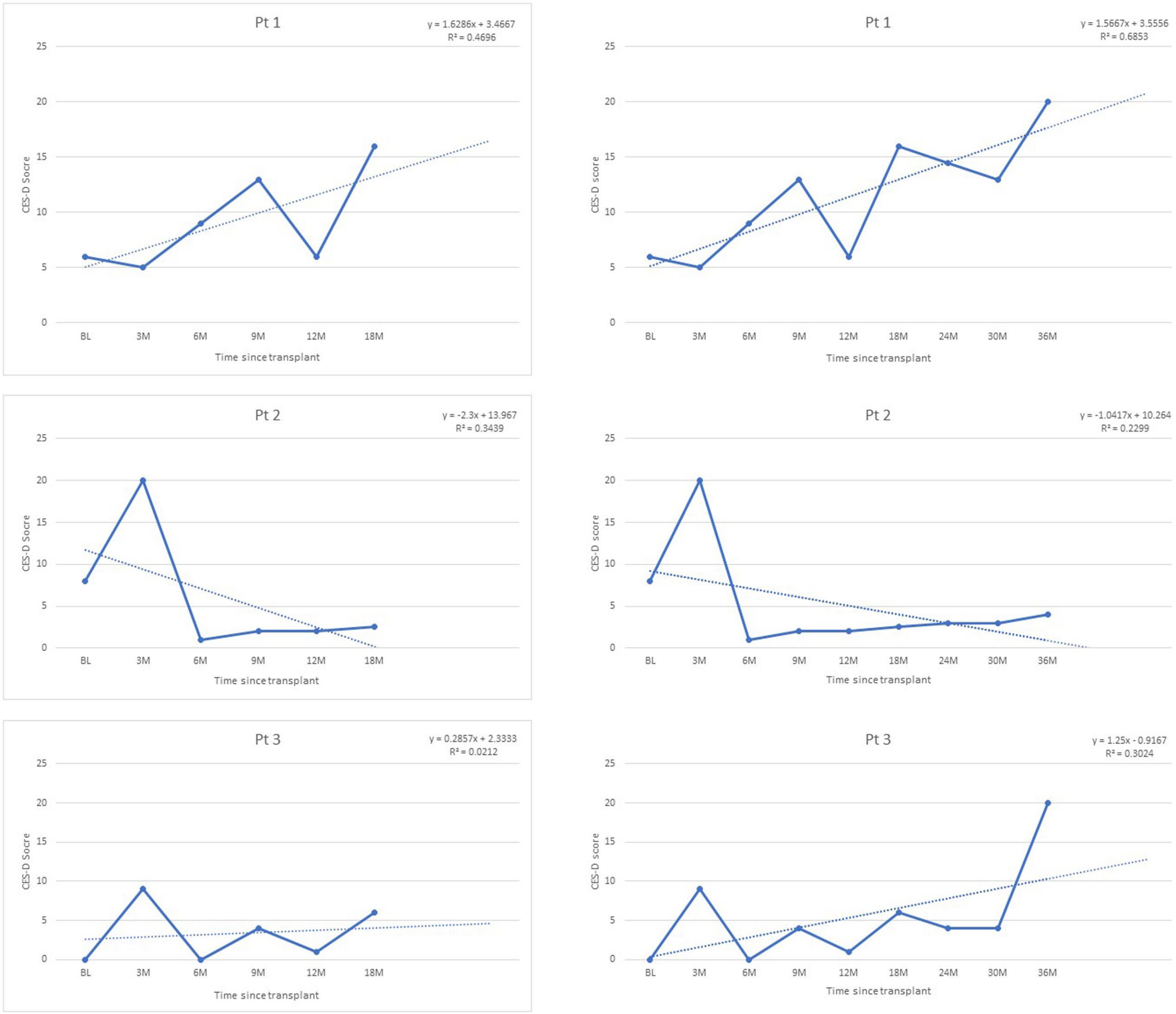

FIGURE 2
Depression trajectories post-transplant, as measured by CES-D scores, range 0–60 (clinical cut-off >16).

placed on a waitlist. Our findings also align with the generally held view that candidates able to leverage support-based strategies due to a strong support network are at an advantage in terms of pre-transplant mental health.

Post-transplant, we found that a different set of coping strategies predicted optimal outcomes. Both active and avoidant coping showed reversed patterns in mental health trends post-transplant compared to pre-transplant. This time, avoidant strategies were associated with better mental health outcomes in the 18 months following the transplant, in particular self-esteem – which may be boosted by ignoring negative experiences during a time of reduced agency in the face of rejection episodes and numerous medical follow-up procedures. Although intuitively surprising, a similar reversal pattern has been described in military studies, where the same coping strategy has been associated with opposite outcomes depending on military status. Namely, help-seeking was associated with negative outcome perception among active-duty Service Members, while it was associated with positive mental health outcomes in Veterans (Blais et al., 2014; Hom et al., 2017; Nazarov et al., 2020). It has been hypothesized that contextual factors, such as the fear of losing a security clearance while on active duty, may explain this reversal. Thus, contextual changes linked to the transition out of active duty similarly reversed the impact of the same coping strategy (Goode and Swift, 2019; Romero et al., 2020). In face transplant recipients, it is possible that the denial detrimental to seeking care pre-transplant played a protective role post-transplant. The loss of protective effect from support-based strategies may reflect a progressive deterioration of the support network of the recipients after years of intensive support, which should be controlled for in future studies when possible.

Although the coping strategies most associated with resilient mental health differed pre-and post-transplant in our sample, it is notable that trends in all mental health outcomes measured

FIGURE 3
Self-esteem trajectories post-transplant, as measured by SES scores, range 10–40.

within the first 18 months post-transplant were confirmed in all patients up to 36 months post-transplant. This suggests that early mental health trajectories potentially offer valuable insight in longer term trends, thus highlighting the importance of an early monitoring of deteriorating mental health to propose supportive interventions.

Clinical implications

Because face transplantation is a life-altering rather than lifesaving surgery, it is critical to select the candidates who stand to benefit most from the transplant in terms of psychosocial outcomes. In this sense, improving psychosocial outcomes is a close second goal to regaining function. The role of the face in our social identity, as much as its importance for both verbal and nonverbal communication through speech and facial expressions, make it one of the most salient physical contributors to our social functioning and interpersonal adjustment. Thus, medical teams have every mandate to improve the empirical markers based on which they can optimize surgical recommendations for face transplantation. Our findings suggest two main implications relevant to candidate selection and recipient care.

First, the discrepancy between coping styles most associated with positive outcomes pre-and post-transplant should offer a measure of caution regarding the protective value of pre-transplant active coping strategies. Rather, the best use of pre-transplant coping data may rest in informing therapeutic interventions at different phases of the transplant journey. When supporting candidates before the surgery, it may be beneficial to train candidates in non-avoidant strategies. Thus, rather than a one-size-fits-all coping recommendation, patients may be informed that optimal coping strategies may vary pre-and

FIGURE 4
Quality of Life trajectories post-transplant, as measured by EQ-Scale scores, range 0–100.

post-transplant, and encouraged to explore new coping strategies if experiencing poorer mental health post-transplant.

Second, and perhaps most promising, the finding that trends in mental health identified during the first 18 months post-transplant were maintained in all patients with available data and for all outcomes measured up to 36 months post-transplant suggests that early post-transplant data may provide reliable information to predict trends for the years following transplant, and valuable context to interpret the unavoidable ups and downs of punctual measurement. We propose that this finding supports the clinical recommendation to use early post-transplant data to guide case conceptualization and treatment planning when supporting the mental health of face transplant recipients post-transplant. To this effect, it may be relevant to increase the frequency of assessment during the first 6 months post-transplant, to propose therapeutic interventions as early as warranted.

Limitations and future studies

As is still the norm in face-transplant studies, the small sample size limits the generalizability of these findings. Therefore, their value lies in both a thorough and novel description of available cases, and in outlining future research directions for the growing field of VCA. We can only encourage the replication of coping studies in other VCA cohorts to complement the descriptive findings reported here.

Another well-documented limitation in similar samples is the presence of a strong bias introduced by social desirability, or the desire to provide responses conforming to the perceived or imagined wishes of the medical team conducting the assessment. This is particularly true pre-transplant, during the candidacy phase, where candidates frequently report inflated self-esteem and underreport problem areas, such as substance use or lack of support. Our findings align with this effect, with a negative correlation between pre-and post-self-esteem scores, along with a strong correlation between avoidant styles (denial, disengagement) and self-esteem post-transplant, suggesting that self-esteem may be maintained by not looking too closely at difficulties.

Finally, it is notable that the three mental health measures did not correlate between pre-and post-transplant assessments. While depression showed only a minimal change, self-esteem and QoL both showed a moderately negative correlation with themselves, which could explain the reversal in pattern found in their association with specific coping strategies pre-and post-transplant. In future studies, assessing coping post-transplant could shed more light on this pattern by determining if patients have altered their coping style. Furthermore, the inclusion of other relevant psychosocial predictors, such as the occurrence of rejection episodes and life stressors, would contribute to a more granular understanding of mental health trajectories post-transplant.

Conclusion

When it comes to helping medical teams with patient selection, only pre-transplant data is available to inform clinical decisions. Because it is the only data available, perhaps too much credit is given to its ability to predict post-transplant outcomes. In this important respect, the pattern of reversal we observed in our sample for the coping strategies most associated with resilient mental health pre-and post-transplant should caution against too strong a confidence in the maintenance of baseline patterns after the transplant. Instead, our findings support that early post-transplant data provides the most promising insight into longer lasting trends in the post-transplant mental health of face transplant recipients.

Data availability statement

The datasets presented in this article are readily available by request to the senior author. Requests to access the datasets should be directed to bohdan.pomahac@yale.edu.

Ethics statement

The studies involving human participants were reviewed and approved by Brigham and Women's Hospital IRB. The patients/participants provided their written informed consent to participate in this study.

Author contributions

M-CN collected the data, conducted the data analysis, and wrote the manuscript. BP provided access to the patients and contributed to the manuscript. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Social support criteria in vascularized composite allotransplantation versus solid organ transplantation: Should the same ethical considerations apply?

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The field of vascularized composite allotransplantation (VCA) is evolving, with some procedures poised to transition from highly experimental research toward standard of care. At present, the use of social support as an eligibility criterion for VCA candidacy is at the discretion of individual VCA programs, allowing VCA teams to consider the unique needs of each potential candidate. Yet this flexibility also creates potential for bias during the evaluation process which may disproportionately impact members of certain communities where social configurations may not resemble the model considered "optimal." We examine the extent to which ethical considerations for social support in solid organ transplantation (SOT) may be applied to or adapted for VCA, and the ethically meaningful ways in which VCA procedures differ from SOT. We conclude that VCA programs must retain some flexibility in determining criteria for candidacy at present; however, considerations of equity will become more pressing as VCA procedures evolve toward standard of care, and further empirical evidence will be needed to demonstrate the association between social support and post-operative success. The field of VCA has an opportunity to proactively address considerations of equity and justice and incorporate fair, inclusive practices into this innovative area of transplantation.

KEYWORDS

social support, vascularized composite allotransplantation, ethics, equity, psychosocial

Introduction

The field of vascularized composite allotransplantation (VCA) has evolved over the last two decades and now includes over 60 active hand, face, uterine and penile transplant programs (Cherikh et al., 2019). Much like in solid organ transplantation (SOT), the use of social support criteria for eligibility is at the discretion of individual VCA programs, providing ample leeway for VCA teams to consider the unique needs of each potential VCA candidate (Jowsey-Gregoire and Kumnig, 2016). Yet, this flexibility also creates the potential for bias to enter the transplant process at multiple touch points, including referral, evaluation, and listing (Ladin et al., 2019a; Mohottige et al., 2021; Reese et al., 2021; Park et al., 2022). Moreover, the very concept of social support in transplant is predicated in part on the presumption that an "optimal" social configuration exists that best positions transplant recipients to fare well (Maldonado, 2019). By extension, communities where social configurations and norms do not resemble the "typical" or "ideal" model may be disproportionately affected by both implicit and explicit biases, thereby exacerbating inequities in access (Maldonado, 2019; Ladin et al., 2019a).

Concerns about the ethics of social support criteria have been raised and debated in the SOT literature (Batra and Rubman, 2019; Berry et al., 2019; Beverley and Reischer, 2019; Fuller, 2019; Goldberg and Foster, 2019; Kelly-Hedrick and Henderson, 2019; Maldonado, 2019; McCauley and Fox, 2019; Parent, 2019; Priest, 2019; Wall, 2019; Ladin et al., 2019a; Mohottige et al., 2021; Reese et al., 2021). In this analysis, we explore the extent to which ethical considerations for social support in SOT may be applied to or adapted for VCA. VCA procedures differ meaningfully from most SOT in a number of ways that are ethically significant. The goal of VCA is to enhance rather than to extend life, and thus competing ethical principles ought to be balanced accordingly. Furthermore, VCA is still generally considered experimental and conducted as research, with implications for weighting ethical priorities that favor greater discretion for individual programs to ensure procedures are safe and effective. And finally, VCA types vary greatly from one another (with a higher degree of variation within each type), and recipients' rehabilitative trajectories differ extensively, again necessitating more nuanced approaches to standards for eligibility criteria.

We consider how the ethical principles of utility and equity should be applied in VCA, and the tensions that arise when they are in conflict. We address the harms associated with bias and discrimination and review several alternatives for providing social support. We then conclude that a one-size-fits-all approach to social support as an eligibility criterion in VCA is unlikely to meet the varying needs of each type of VCA at present. Some discretion must be retained, particularly for lower-volume procedures such as face and penile transplants. That said, the field of VCA has an opportunity to proactively address considerations of equity and justice and can look to SOT for guidance on incorporating inclusive practices into this innovative area of transplantation.

Balancing utility and equity in organ transplantation

The organ transplant system in the United States is guided by the distinct and sometimes competing principles of utility and equity (National Academies of Science, Engineering, and Medicine et al., 2022). Utility seeks to maximize the good that can be derived from available resources, in this case organs available for transplantation (Ethical Principles in the Allocation of Human Organs, 2015; National Academies of Science, Engineering, and Medicine et al., 2022). Given the perennial shortage of organs for transplant, distribution of this scarce resource must take into account where and for whom an organ will confer the most benefit (Clarke, 1995; National Academies of Science, Engineering, and Medicine et al., 2022). The principle of utility informs allocation policy to ensure that organs go to those who will benefit most, balancing need and likelihood of a successful outcome defined by numerous clinical endpoints including overall graft function, graft survival and mortality (Ethical Principles in the Allocation of Human Organs, 2015). At the same time, the transplant system weights these considerations of utility alongside moral obligations to promote equitable distribution of scarce resources and fair access to transplant (Ethical Principles in the Allocation of Human Organs, 2015). Criteria for transplant candidacy and organ allocation reflect the intermingling between considerations of utility and equity, including the use of social support criteria to determine eligibility for transplant.

Social support and the problem of construct validity

Social support first gained attention in the literature as an important element in the relationship between stress and health outcomes (Cobb, 1976). Scholars have addressed social support from a multiplicity of theoretic vantage points, struggling to agree on a consistent definition. Social support can be defined variously as information, as a resource or resources, as availability of helping relationships, and as transactional resource provision, for example. Definitional confusion has resulted in heterogeneity in how the construct is conceptualized, operationalized and measured in the context of health and wellbeing (House, 1981; Chiaburu and Harrison, 2008; Ng and Sorensen, 2008). While existing empirical evidence suggests a link between social support and health outcomes (Cohen and Leonard Syme, 1985; Berkman and Glass, 2000; DiMatteo, 2004; Roth et al., 2005), the relationship between social support, health and wellbeing is not clear, due in part to aforementioned conceptual ambiguity (Kossek et al., 2001; Kim et al., 2005; Roth et al., 2005). Unsurprisingly, research in this area has focused on a variety of outcomes including behavioral, attitudinal, cognitive and/or emotional measures (Cohen and Leonard Syme, 1985; DiMatteo, 2004; Kim et al., 2005; Uchino, 2006, 2009). In the context of organ transplantation, lack of clarity about how to define, operationalize, and measure social support makes it difficult to assess the impact of social support on transplant outcomes, leaving room for bias and discrimination in assessment of eligibility for transplant procedures. This is the case not only in SOT, but also in evaluation for VCA candidacy.

Social support in SOT: Empirical evidence

Approximately 30,000–40,000 organ transplant procedures are performed annually from both living and deceased donors, only 3–20 of which are VCA (Transplant Trends, 2022). To date, no guidelines explicitly define optimal social support in the setting of transplant surgery (Ladin et al., 2019a). In Ladin et al. (2019b) reported different definitions of social support among psychosocial clinicians ranging from informational, emotional, instrumental, motivational, financial, and importance of the patient to others (Ladin et al., 2019b). A study published as recently as 2021 determined statistical significance between social support and medication adherence following SOT but did not assess whether the correlation was due to emotional support or the direct management of medications by loved ones (Huang et al., 2021).

In an article published in the American Journal of Bioethics (AJOB), Berry et al. examined the ethics of social support as a criterion for access to SOT, grounding their analyses in existing empirical evidence addressing the association between social support and transplant outcomes (Berry et al., 2019). They found the empirical evidence linking social support to transplant outcomes insufficient and concluded, therefore, that lack of social support alone should not prevent an individual from accessing life-saving SOT. Furthermore, they suggested the criterion, as presently deployed, inappropriately favors utility and undermines important equity considerations. Specifically, formalized social criteria risk exacerbating the societal disadvantages inherent in marginalized communities, particularly those with reduced health care access and benefits, absent or non-traditional sources of social support, and lower income, thereby not being able to afford aspects of post-operative care such as medications, a live-in caregiver, or transportation to follow up appointments.

However, social support is inextricably linked to patient selection in SOT as surgical outcomes are dependent on the post-operative care period (Ladin et al., 2019a). Despite the importance of post-operative support in SOT, there are no guidelines formally in place in the literature to assist clinicians and transplant programs in establishing standardized approaches to incorporating assessment of social support in the evaluation process for transplant candidacy.

Social support in VCA: Empirical evidence

Current empirical evidence on the impact and validity of social support criteria for VCA is scarce. Although a 2013 study documented statistical significance for patients with adequate documentation of social support system and transplantation failure rate of primarily hand and face, the only definition of social support provided was "suitable resources to sustain medication, adjunctive therapies, and follow-up" (Zhu et al., 2014). In other published VCA research mentioning social support, the criterion is described variably as "strong support from family and community"(Benedict and Magill, 2018), family members who may need to prepare for the media attention that often comes with VCA cases (Kumnig and Jowsey-Gregoire, 2016) or caregivers to aid in tasks of daily living, caring for the patient's children, and providing financial support during recovery (Kumnig and Jowsey-Gregoire, 2016). These heterogeneous considerations further demonstrate the subjective nature of social support as a requirement for VCA eligibility. Finally, the presence of caregivers during the consultation and operative journey as well as mental health screening are discussed in the current VCA literature as important components in establishing psychosocial support (Kumnig and Jowsey-Gregoire, 2016).

Discussion

The ethics discourse sparked by Berry et al.'s target article and accompanying commentaries offers a lens for comparative examination in the context of VCA (Batra and Rubman, 2019; Berry et al., 2019; Beverley and Reischer, 2019; Fuller, 2019; Goldberg and Foster, 2019; Kelly-Hedrick and Henderson, 2019; McCauley and Fox, 2019; Parent, 2019; Priest, 2019; Sharma and Johnson, 2019; Wall, 2019). To adapt ethical arguments about the appropriate use of social support in SOT for application in VCA, we examine ethically meaningful differences between SOT and VCA and ways in which these differences will likely shift over time.

Given the recent evolution of the field of VCA and the small number of procedures that have been performed to date relative to SOT, evidence in the form of long-term outcomes data to support the appropriate role of social support in VCA patient selection is even more scant than in SOT, particularly for lower-volume procedures such as face and penile transplants. And yet, variation in frequency and volume of procedures performed across VCA types means that some higher volume VCAs (hand and uterus) are approaching the transition toward standard of care. For those further along this trajectory, it will become increasingly important to standardize eligibility criteria and shift the focus from the ethical principle of utility toward equity to optimize patient care (Kimberly et al., 2019).

Ethically relevant differences between SOT and VCA

How do these considerations about the appropriate use of social support criteria in SOT translate to the VCA space? To answer this question, we address characteristics of VCA that differentiate this innovative area of transplantation from SOT.

Life saving versus life enhancing

Unlike in SOT which in most cases is considered life-saving, enhancing quality of life is the primary goal in VCA procedures. The ethical considerations of VCA necessitate an alternate lens than SOT, as the risk to benefit ratio for VCA differs and thus requires a different weighting of priorities (Kumnig and Jowsey-Gregoire, 2016). While some benefits of VCA involve significant improvements in function and reduction of pain, other benefits are more psychosocial in nature and include the possibility of social reintegration and considerations for a recipients' sense of identity. This is particularly the case in the context of face transplant, pre-and post-operative body image, and quality of life (Kumnig et al., 2014). Such concerns warrant prioritizing and considering the potential psychosocial harms that come with disfigurement. It might be argued that, for a procedure considered life enhancing but not life-saving, social support could be perceived as carrying more importance as an additional safeguard to protect against potential risk. In the case of a life-saving procedure such as SOT, where the alternative to transplant is death, limited social support might not weigh as heavily against the risk of not proceeding with transplantation. Thus, assurance of robust social support may have a greater role to play at present in patient selection for VCA than in SOT.

Balancing utility and equity in experimental research versus innovative therapy

The clinical research context generates ethical considerations that are distinct from considerations encountered in clinical practice. With respect to patient selection, while research efforts tend to lean more toward utility with the goal of selecting the "optimal" patient to ensure the best possible outcomes and establish proof of concept (Maldonado, 2019), the pendulum will shift toward ensuring fair access once a procedure is well established. Concerns around equity in SOT are at the forefront at present, hence the calls to interrogate the definition and operationalization of social support and its relation to transplant outcomes to ensure that social support criteria are not discriminatory and are grounded in solid evidence (Zhu et al., 2014; Kumnig and Jowsey-Gregoire, 2016; Benedict and Magill, 2018; Ladin et al., 2019a). While some forms of VCA, particularly hand and uterine transplant, are poised to transition toward standard of care, others are still considered highly experimental meaning that ethical considerations of utility still guide approaches to patient selection.

Variation within VCA

VCA types vary greatly by total volume of procedures performed and by rate of performance over time. Moreover, as previously noted, VCA types differ in their status on the developmental trajectory from highly experimental research to innovative therapy approaching standard of care (Diep et al., 2021; Jones et al., 2021; Lake et al., 2022; Wells et al., 2022).

Upper extremity

To date, approximately 148 hand transplants have been performed worldwide (Wells et al., 2022). As a relatively high-volume form of VCA, hand transplant is poised to shift toward standard of care. This procedure has the potential to scale up, and the complexity of the procedure itself is fairly consistent from one case to another, thereby enabling programs to develop expertise.

Face

As compared to hand transplant, face transplant is a resource-intensive, low-volume procedure and is likely to remain so for the foreseeable future. Only 48 face transplants have been documented worldwide (Diep et al., 2021). Each case presents a range of unique technical challenges, and each procedure must be carefully tailored to the specific recipient's needs and anatomical characteristics (including natal characteristics and changes to anatomy as a result of injury or disease) and the anatomy of the deceased donor.

Genitourinary

Uterine – Compared to other forms of VCA, the volume of uterine transplants (UTx) performed annually has increased steeply in a relatively short period of time. To date, data for over 70 UTx have been published (Jones et al., 2021). This is due in part to the nature of UTx, which arguably may be considered more akin to solid organ transplants (Johannesson et al., 2014). Procedures are less variable, allowing for a more rapid development of experience within a UTx program and thus capacity to scale up. The donor-recipient matching process differs from other forms of VCA, without the aesthetic considerations of externally visible hand, face, and penile grafts. UTx is distinct from all other forms of SOT and VCA in that the grafts are intended to be temporary, with removal *via* a second surgery following successful achievement of pregnancy and live birth. Among the various types of VCA, UTx has approached standard of care most rapidly and, in fact, Baylor University in the United States now offers UTx as clinical care outside of a research protocol. However, the procedure is only available to individuals who can pay out of pocket, as commercial insurance has not yet approved reimbursement for all costs associated with UTx.

Penile – At present, only four penile transplants have been performed worldwide and detailed in the literature (Lake et al., 2022). The procedure is still considered highly experimental, and it is unclear whether it may eventually become standard of care. Other reconstructive options are available, although these options have drawbacks in terms of both form and function (Lake et al., 2022).

Important dimensions of social support in VCA

Caregivers in VCA post-operative recovery and rehabilitation

Designated caregivers are considered a vital component of social support for VCA patients to facilitate post-operative recovery (Jowsey-Gregoire and Kumnig, 2016; Kumnig and Jowsey-Gregoire, 2016; Benedict and Magill, 2018). Postoperative VCA monitoring in the years following surgery is critical to successful patient care, and many logistical factors require united efforts on behalf of patients, their caregivers, and clinicians. While specific rehabilitation needs and requirements vary according to VCA type, lengthy rehabilitation is essential in the recovery process for most VCA recipients and may include prosthetic use, adjusting to the visible allograft, and monitoring for rejection. Moreover, further revision surgeries may be needed, particularly in facial transplantation. An established support system to facilitate the extensive logistical demands of postoperative monitoring will likely improve outcomes. The magnitude of postoperative care has prompted discussion about creating useful models for the adjunct care of VCA patients; examples include assessment of quality of life, family support, and psychiatric stability, all of which at present appear to be associated with VCA patient outcomes (Jowsey-Gregoire and Kumnig, 2016).

Social support and mental health in VCA

In addition to establishing the extent of familial or other caregiver support for VCA candidates, preoperative screening and intensive mental health evaluation are important elements of the assessment process for potential VCA candidates. Evaluations are particularly valuable in determining the optimal level of pre-and post-operative mental health support and follow-up (Klapheke et al., 2000; Jowsey-Gregoire and Kumnig, 2016; Kumnig and Jowsey-Gregoire, 2016). Contrary to SOT where the transplanted organ remains inside the peritoneal cavity, most VCA grafts are external. This exterior change in outward appearance such as the face and the hands can present patients with psychological implications related to their sense of self (Kumnig and Jowsey-Gregoire, 2016). Adjunct care to support patients' mental wellness may help ease the adjustment to visible changes to the body, and adequate social support is likely to improve mental health outcomes (Kumnig and Jowsey-Gregoire, 2016; van Pilsum Rasmussen et al., 2020). Currently there is no formalized approach to optimal frequency of mental health follow up in VCA, and it remains largely program dependent. Further examination of adjunct mental health support in the context of social support and its role in VCA recovery would make a valuable contribution to the literature in VCA. While a majority of VCA patients document high levels of social support through their care (van Pilsum Rasmussen et al., 2020), this presence of social support does not necessarily preclude patients from instances of depression or anxiety that may develop during the recovery process (van Pilsum Rasmussen et al., 2020). Understanding the elements of social support that are most closely tied to outcomes during the VCA process will improve approaches to patient selection and patient care.

Social support and adherence to immunosuppression

Adherence to immunosuppressive medication presents another important consideration in VCA. Immunosuppressant adherence is closely tied to successful outcomes in VCA and is crucial in order to prevent major complications, including rejection and graft loss. However, these medications can cause significant side effects, and social support from caregivers has been demonstrated to help recipients maintain adherence and cope with side effects, including mood changes linked to long term immunosuppressive treatment that may make it more difficult for recipients to maintain follow up regimens (Kumnig and Jowsey-Gregoire, 2016).

Moving toward equity in VCA

Important lessons from SOT: Who is harmed by social support criteria?

Berry et al. conclude that the risk of further marginalization associated with the use of social support criteria is greater for individuals of low socioeconomic status, people of color, and individuals with comorbid mental health and substance use disorders (Berry et al., 2019). These demographic groups are disproportionately affected by the implicit biases and contraindications that deny them access to lifesaving procedures (Butler and Wightman, 2021). For instance, black patients are more likely to be uninsured and less likely to be evaluated for transplant (Mohottige et al., 2021). Logistical burdens range from follow-up appointments and access to transportation, to at-home care for those unable to receive support from family. In fact, many patients report the financial stress of covering costs of transportation, medications, procedure, and aftercare outweighed their fears of the transplant itself (Mohottige et al., 2021).

The fact remains that patients of color who are underinsured and who receive less formal education undergo fewer transplants relative to their rates of organ failure, which points to deficits in the current selection process and criteria for eligibility (Reese et al., 2021). In order to address and prevent the perpetuation of these inequities, policies should account for the intersection of race and ethnicity with gender, socioeconomic status, education, and health literacy (Delaney et al., 2021). Neutral transparent evaluations, evidence-based criteria, patient-provider transparency, and revisability in guidelines are some of the factors that are essential in equitable access for transplant patients (Ladin et al., 2019a). Provider confidence and consistency with the definition of social support were found to aid the transparency of waitlist decisions and provision of greater support to the patient (Ladin et al., 2019b). Furthermore, evidence-based initiatives are imperative in establishing criteria that enhance the opportunity for transplant and eliminate the potential for both latent and overt bias within the evaluation process (Berry et al., 2019).

Minimizing bias and subjectivity

As evident from the existing literature, social support as a construct in SOT and VCA is inconsistently defined, and multiple dimensions are often conflated (i.e., instrumental support, informational support, emotional support). Some can be easily supplemented, others less-so. In SOT, the general consensus is that social support has a role to play in transplant evaluation, but that role should be carefully circumscribed, well-substantiated, and standardized across programs (Batra and Rubman, 2019; Berry et al., 2019; Beverley and Reischer, 2019; Fuller, 2019; Goldberg and Foster, 2019; Kelly-Hedrick and Henderson, 2019; McCauley and Fox, 2019; Parent, 2019; Priest, 2019; Sharma and Johnson, 2019; Wall, 2019). Present understanding of the role of social support in transplant outcomes relies on outdated research and highlights the dearth of current research examining the association between social support and transplant outcomes (Sharma and Johnson, 2019). There is also limited information addressing individual biases and motivations of selection committee members, as well as selection committee group dynamics and decision-making processes (McCauley and Fox, 2019). However, patient selection committees would benefit from a consistent definition of social support and understanding of the weight this definition bears in their recommendation to prevent patients from exclusion and further marginalization (Beverley and Reischer, 2019; McCauley and Fox, 2019). Some have even argued from a compensatory justice stance that patients with minimal social support may actually be more deserving of a transplant in order to have more opportunities to develop social support moving forward (Priest, 2019). While these considerations may not be immediately pressing in VCA to the same extent as in SOT, they will likely become relevant in the near future.

Alternative approaches to social support

Several alternatives to the present role of social support in SOT have been proposed. For example, the role of social support could be shifted away from traditional, familial models to a model in which social support is provided by the programs themselves (Parent, 2019). This may be addressed by involving social workers to help patients navigate insurance, financial stressors, and other areas of perioperative support (Goldberg and Foster, 2019). This could reduce the burden of social support being placed on the patient's community and address the disparities across different patient populations who may otherwise be deemed lacking in social support and excluded from receiving the procedure. In addition to the potential for social support as an eligibility criterion to further exclude already marginalized groups, its contribution to gender inequity merits close scrutiny (Fuller, 2019). With the role of caregiver falling more often on women than men, current expectations for social support tend to further exacerbate the gender injustices prevalent in society (Fuller, 2019). This warrants a more equitable system, not only for patients, but for caregivers as well (Fuller, 2019).

As technology evolves, lack of informational, instrumental, and even emotional support may be at least partially mitigated by mobile or social media platforms (Kelly-Hedrick and Henderson, 2019). However, the degree of emotional support that can be provided by a program may be limited and difficult to compare to relationships that have been cultivated over many years and cannot be replaced or standardized (Wall, 2019). Preexisting social support would certainly not be denied or abandoned altogether (Batra and Rubman, 2019; Goldberg and Foster, 2019; Wall, 2019). Different types of social support will need to be examined separately to measure their independent effects on the transplantation and recovery process (Batra and Rubman, 2019).

Strengthening construct validity to improve research design in VCA

Enhanced understanding of the role and specific mechanisms of social support during the pre-and post-transplant experience and its impact on wellbeing can help to identify opportunities to improve policies and procedures, including pre-transplant assessment, preparation for transplant surgery, support during post-transplant hospitalization, discharge planning, and short-and long-term follow up care. The heterogeneous state of the VCA literature (and the transplant literature overall) presents an opportunity for revision and integration of prior theoretical approaches and models, and an effort to more clearly define the construct and its behavioral, affective, cognitive and emotional dimensions would help to inform and prioritize future VCA research design, including the conceptualization, operationalization and measurement of meaningful outcome variables. Collaboration across VCA programs will be essential in generating and assessing the evidence needed to support adoption of a consistent definition of social support.

Future directions for social support in VCA

In the context of clinical research with human participants, a greater degree of discretion for eligibility criteria is generally considered appropriate, as the primary goals of research are to demonstrate safety and efficacy, and therapeutic benefit is not assured. Since VCA procedures vary in their developmental trajectory, individual programs at present have a greater need for flexibility in determining VCA candidacy. However, as VCA procedures eventually shift toward clinical practice, standardizing eligibility criteria, including social support, will become increasingly germane. The field of VCA has an opportunity to incorporate patient-centered, inclusive practices from the outset. By anticipating future ethical shifts from utility toward equity, the field will better support fair access, address calls for greater transparency of the VCA patient selection process and promote a transplant system that is publicly perceived as just.

Author contributions

LK led conception and design of the conceptual analysis, with substantial contributions from OO and ER. LK, OO, and ET drafted sections of the manuscript. All authors contributed to manuscript revision and approved the submitted version.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Psychosocial and bioethical challenges and developments for the future of vascularized composite allotransplantation: A scoping review and viewpoint of recent developments and clinical experiences in the field of vascularized composite allotransplantation

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Vascularized Composite Allotransplantation (VCA) has evolved in recent years, encompassing hand, face, uterus, penile, and lower extremity transplantation. Accordingly, without centralized oversight by United States Organ Procurement and Transplantation Network (OPTN) or European Programs, centers have developed their own practices and procedures that likely vary, and accordingly, present different levels of rigor to the evaluation process, internationally. The importance of psychosocial factors in the selection process and treatment course has been widely recognized, and therefore, several approaches have been developed to standardize and guide care of VCA candidates and recipients. We propose to develop an international multidisciplinary platform for the exchange of expertise that includes clinical, patient, and research perspectives. Patient perspectives would derive from peer education and the assessment of patient-reported outcomes. To establish a foundation for such a platform, future research should review and combine current VCA protocols, to develop the ethical framework for a standardized psychosocial evaluation and follow-up of VCA candidates and recipients. This review presents a comprehensive overview of recent results in the field of VCA, developments in structural aspects of VCA, and provides viewpoints driven from clinical experience.

KEYWORDS

vascularized composite allotransplantation, psychosocial, bioethics, quality-of-life, peer education, patient reported outcomes

1. Introduction

1.1. Current standards and framework development

Vascularized composite allotransplantations (VCA) have moved from a purely experimental option for a small number of patients, to comprising the standard of care of VCA recipients at some institutions internationally (Kumnig and Jowsey-Gregoire, 2016; Hautz et al., 2020). However, this transition has occurred without a detailed, comprehensive, and objective investigation of psychosocial and bioethical factors (Kumnig et al., 2012, 2014a,b; Kumnig and Jowsey-Gregoire, 2016). Ensuring that psychosocial and bioethical implications of VCA transplants are established within the VCA field is highly important; however, standardized protocols for the evaluation and follow-up VCA patients are still evolving (Kumnig et al., 2022). VCA procedures are primarily life-saving, such that quality of life (QOL) comprises central outcomes. The most important development for the VCA field is an emerging recognition that the pre-and post-transplant psychosocial evaluation and treatment is an integral part of any VCA transplant program, and that the identification of at-risk patients and those requiring ongoing counseling is a primary focus of assessment and treatment procedures (Kumnig et al., 2012, 2014a,b; Kumnig and Jowsey-Gregoire, 2016). The psychosocial risk assessment encompasses whether the patient understands the potential surgical complications, the risks of immunosuppression, the potential for rejection and graft loss (Goldade et al., 2011), and the need for adherence with the therapeutic regimen (Matas et al., 2002). Appropriate patient selection is important because of patients' risk of developing a psychiatric disorder or because patients may be struggling with psychosocial burdens before transplantation as well as during the post-transplant period (Rosenberger et al., 2012; Pither et al., 2014).

Psychosocial factors are important elements in the assessment and follow-up care for VCA and require multidisciplinary evaluation protocols. The Chauvet workgroup has been convened with membership from a number of transplant centers to address these issues and to call for ongoing global research collaboration. A multicenter research network, a consortium of collaborating VCA centers, would share similar evaluation approaches so that meaningful research on psychosocial variables could inform the transplant community and patients about psychosocial factors relevant for optimal VCA outcomes (Kumnig et al., 2022).

Because of the extant global diversity in psychosocial evaluation and follow-up routines in VCA, current and future research will need to guide the field regarding this question: What are current standards and what important psychosocial aspects must be considered when implementing or re-structuring a VCA program at a transplant center? Moving one step forward, directly into the psychosocial evaluation and follow-up process, this perspective will particularly address the importance of the assessment and elaboration of 'exit strategies' with candidates planning to undergo VCA or patients who already have been transplanted and their strategies to cope with potential graft loss during the transplant course. This topic is especially important for this field and there is no sufficient explanation as to why this has been neglected for such a long period of time. This important topic is under examination in the kidney-pancreas community of practice *via* American Society of Transplantation (AST), so that future developments in the field of VCA could orientate to such protocols (Alhamad et al., 2022).

1.2. Peer education and consultation concept in vascularized composite allotransplantation

Considering the small number of global VCA centers (about 50 centers worldwide) and VCA recipients (under 200 recipients of different VCA procedures to date, worldwide; Kinsley et al., 2020), international collaborations have sought to bring health professionals and patients together in other clinical contexts (Magill et al., 2019). Therefore, future research will need to address the possibilities of peer education in the field of VCA, educational programs that bring healthcare professionals together as well as connecting patients worldwide (particularly because of the small number of cases). Although international collaboration has been an essential part in the history of VCA, it is still difficult to share experiences and to bring different perspectives together. Thus, the concept of an international platform for experts (e.g., Chauvet Research Group; Kumnig et al., 2022) and patients could lay the foundation to provide this essential global connectivity.

Further, low-threshold consultation concepts for post-transplant VCA patients, for example by routinely assessing patient-reported outcomes (PROs) could guide a targeted expert counseling. That will help to address the specific needs of these patients: e.g., information-related questions, psychosocial issues (e.g., depressive developments, adherence problems). Moreover, such assessment routines could help to identify patients at risk, offering a specific consultation first, evaluate by routinely collecting psychosocial outcomes (PROs), and decide whether additional intensive care may be needed.

2. Important psychosocial issues identified by recent research initiatives

2.1. Psychosocial stability, financial, caregiving, and family responsibility

First, it must be noted that there are financial differences between European countries such as the United Kingdom and

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Abbreviations: AST, American Society of Transplantation; FTx, face transplantation; HRQOL, health-related quality of life; OPTN, Organ Procurement and Transplantation Network; PROs, patient-reported outcomes; QOL, quality of life; UETx, upper extremity transplantation; UTx, uterine transplantation; VCA, vascularized composite allotransplantation.

France, compared to the United States, which may account for differences in what is considered a contraindication for VCA. Finances are generally not considered a significant barrier to healthcare in European countries due to caregiving, hospital, and post-transplant state-sponsored support, like in United States, where the family is commonly expected to provide some financial support to help the patient meet co-payments for care along the continuum of pre-to post-transplant periods (Wainright et al., 2022). While financial issues do not comprise an absolute contraindication to VCA in Europe, such issues could potentially be construed as an absolute contraindication to VCA procedures in the United States. Exemplary in France, there is no financial payment expected from the patient for the operation, hospitalizations, and post-transplant care for the rest of the life of the patient. In the United States, however, insurance companies do not yet cover VCA procedures as they are still considered 'experimental' until more data are collected about patient outcomes. Accordingly, patients may find it challenging to access and cover the financial costs of VCA treatment, immunosuppression, and support themselves in the post-transplant period. Thus, establishing insurance coverage for VCA in the United States could help to expand VCA transplant procedures.

2.2. Coping history, adherence history, and issues of substance abuse

There is early consensus among global VCA centers that active substance use at the time of evaluation is a contraindication to any VCA procedure (Jowsey-Gregoire et al., 2016). However, the question is raised whether a patient who undergoes recommended treatment for addiction can become a candidate in the future. It remains to be determined whether active substance abuse should be a relative and temporary contraindication to VCA and if in that case recommendations for substance use treatment should be made to improve the patient's candidacy.

Recent research suggests that a period of longitudinal follow-up should be part of the protocol, to allow re-assessment and follow-up at multiple time points prior to VCA. The optimal proposed follow-up period was for 1 year from the time of assessment for the potential VCA procedure (Kumnig and Jowsey-Gregoire, 2016).

Particularly in the case of patients with a history of non-adherence with medical recommendations, the evaluation and development of coping and (non-)adherence is important. It is proposed that a psychometric instrument may help with tracking adherence with immunosuppressive medications in the post-transplant period (Jowsey-Gregoire et al., 2016). Recent research also highlights that use of modern technologies (phone apps, digital wrist-worn devices) has the potential to empower the patient and should possibly be considered to assist the patient with adherence to medications. The clinical psychological assessment still remains the most important tool to take care of non-adherence (Kumnig and Jowsey-Gregoire, 2016).

2.3. Psychosocial contraindications to vascularized composite allotransplantation

Most centers consider risk factors in VCA to be relative, and potentially modifiable (Jowsey-Gregoire and Kumnig, 2016; Jowsey-Gregoire et al., 2016; Kumnig and Jowsey-Gregoire, 2016). Certain psychiatric disorders, such as severe personality disorders, active substance abuse (including nicotine), schizophrenia, chronic depression, and bipolar disorder are considered as risk factors for poor outcomes across many VCA centers (Jowsey-Gregoire et al., 2016). Unreasonable expectations, a history of non-adherence, relational difficulties with the team, and lack of social support are also considered risk factors for a poor outcome (Jowsey-Gregoire et al., 2016). The conclusion of past Chauvet meetings that active psychotic illness is an absolute contraindication to any kind of VCA procedure, can be emphasized here as one central absolute contraindication (Jowsey-Gregoire et al., 2016). Substance abuse was felt to be a relative contraindication and it was offered that chemical dependency treatment for those with active substance abuse can improve a patient's candidacy for VCA. Chauvet meeting members considered strong social support for this particularly vulnerable population as equally important in assisting with abstinence from substances. The Chauvet meetings noted that patients with substance use issues who have strong social support do better after solid organ transplantation and that this model can be applied to VCA as well (Jowsey-Gregoire et al., 2016).

In terms of appropriate psychological acceptance of the disfigured body (in case of hand and face transplantation), research found that some patients with bodily disfigurement become social recluses, indicating a possible maladaptive coping mechanism (Sarwer et al., 2022). Therefore, many VCA centers raised the question, whether VCA candidates should be required to demonstrate a period of appropriate social interaction before undergoing the VCA procedure. This requirement may necessitate an understanding of the patient's baseline social behaviors prior to bodily disfigurement (Sarwer and Crerand, 2008; Sarwer and Spitzer, 2012; Sarwer et al., 2022).

Any one risk factor, if severe enough, may constitute an absolute contraindication. The presence of multiple risk factors may also constitute a prohibitive risk. In particular, severe personality disorders, active substance abuse, schizophrenia, and unrealistic expectations would typically be considered risk factors that would be associated with a decision not to approve candidates for VCA transplantation.

2.4. Evaluation for vascularized composite allotransplantation and follow-up after transplantation

The psychosocial assessment is considered the principal means of assessing personality, emotional preparedness, cognitive status, coping style, motivation and expectations, and social support. Psychiatrists, psychologists, and social workers are typically involved in this evaluation process.

Research recommends that candidates undergo the psychosocial evaluation after meeting with the surgical and medical team members (Kumnig et al., 2012, 2014b; Jowsey-Gregoire and Kumnig, 2016). This sequence may allow the psychosocial healthcare professional to assess how well the candidate understands the risks and the benefits of the VCA procedure following a thorough discussion with the medical, surgical, and psychosocial teams.

2.5. Sex and sexuality, esthetics, and occupation

The first point made in investigations was that sex and sexuality in VCA are not openly discussed by the transplant team (Mills et al., 2020). Therefore, little is known about how patients are expressing their sexuality after a VCA procedure. This is particularly important for women considering uterine transplantation and men considering penile transplantation. In the case of hand and face transplantation, embedded in this notion is the fact that they have lived through the stigma of looking different; they may have suffered social isolation. Therefore, transplant teams should consider the role of social identity and how patients perceive their social identity in relation to their sexuality. The question remains: how does one measure esthetic outcomes in VCA? Transplant teams must rely on the subjective standpoint of the patient but we use objective standards to judge this by.

The value of social support for maintaining and restoring good health is well established, and the assessment of social support has been an aspect in the screening of transplant patients for some time, including VCA patients (Ladin et al., 2019a,b). Nonetheless, the concept of social support in transplantation has been subject of several critiques including the lack of a clear definition, the lack of agreement on a method for assessment, and debate over its use as a criterion for exclusion in patient selection. Future research should investigate the role of social support in upper extremity VCA, and evaluate how differences between SOT and VCA may influence the meaning and value of social support for recipients, and suggest ways in which social support may be better assessed pre-transplant and strengthened post-transplant in VCA.

Research also show that assessing the success of VCA functionality and the ability to return to work are important (again, in case of hand and face transplantation), and that it is necessary to consider the pre-morbid occupational function of the patient in order to gain perspective on occupation as an index of success of the transplantation (Smith and Cendales, 2019). The salient consideration is not necessarily whether the patient has gainful employment but rather how occupation is part and parcel of general social functioning. Also playing a critical role in the function of the family, as well as engaging in non-gainful employment such as volunteer work. An equally important consideration for transplant evaluation is whether the patient's occupation changed from pre- to post-transplant; and whether patients were required or underwent job retraining to re-enter the workforce (Bramstedt, 2018). We recommend that patients should indicate prior to transplantation how personally important employment is to them, in order to establish a baseline.

2.6. Special psychosocial issues in uterine transplantation

Uterine transplantation differs from other forms of transplantation (solid organs or VCA) in many ways: (i) it gives women with absolute uterine factor infertility a chance to realize their wish for a biological child; (ii) the clinical outcome is not only relevant for the patient but also for the child conceived; (iii) transplantation is transitory and its endpoint is marked by graft hysterectomy; (iv) the surgical success rate is defined by a technically successful transplantation with a subsequent regular menstrual pattern potentially allowing for pregnancy and live birth of a child to round off surgical success (Brännström et al., 2021); (v) even after successful uterus transplantation pregnancy may still fail; (vi) about two-thirds of donors are live donors due to disadvantages of deceased donation (Kisu and Banno, 2022); (vii) the surgery is even more invasive and complicated for the live donor than for the recipient and for both patients there are high surgical complication rates (Brännström et al., 2021); (viii) additional risks for live donors include possible familial pressure to donate and reduced quality of life due to hysterectomy and sexual dysfunction (Kisu and Banno, 2022); and (ix) thus far, children born as a result of uterus transplantation were born prematurely at a high rate and with an associated high proportion of respiratory distress syndrome (Brännström et al., 2021).

Medically assisted reproduction is a highly complex field not only clinically but also ethically. Some procedures are highly invasive for women and the children they conceive with the technologies applied. This is certainly even more pronounced in the context of assisted reproduction after uterus transplantation. Patients in this scenario are thus transplant patients as well as patients undergoing fertility treatment to fulfil their wish for a biological child. While this wish can be considered "natural," it is also highly shaped by sociocultural context rendering those patients particularly vulnerable.

The new recommended framework of preoperative psychological evaluation has been published (Järvholm et al., 2018; Wainright et al., 2018) and was presented at the first three Chauvet meetings. Representatives of uterine VCA centers suggest addressing the following psychosocial domains prior to transplantation (in addition to the general assess psychosocial aspects in VCA, e.g., psychopathology, adherence, social support, coping skills, substance abuse, knowledge of the procedure, motivations, informed consent, etc.): donors' family planning, coping with childlessness, the couple's relationship to the donor, and motivation for donation. The last three Chauvet meetings raised key questions about critical psychological events after uterus transplantation, including: who should transplant teams favor as a donor, or when is the appropriate time to stop attempts to achieve pregnancy and remove the uterus, and how can transplant teams create supportive strategies that help patients to deal with graft loss ('exit strategies') as well as pregnancy loss?

3. Quality-of-life assessment for vascularized composite allotransplantation

Quality of life (QOL) is considered the most important domain for study both before and after transplantation (Feurer et al., 2004; Jensen et al., 2012; Kumnig et al., 2014b). The Chauvet participants recognized QOL as a relative concept, both within and across cultures, and transplant teams must take into account both of those domains of QOL universally held, and those valued for their uniqueness to a particular environment (Verdugo et al., 2005; Petruzzo and Dubernard, 2011; Prieto et al., 2016). At present, there are no instruments uniquely devoted to evaluating these domains in VCA, but there is wide acceptance that depression and anxiety should be assessed at regular intervals. Recent investigations highlight how important it is to examine QOL from the patient perspective, thus PRO assessment has gained increasing traction in the transplant field. The need for different PROs and specific questions/assessment for different types of grafts (hand, face, and uterine) has already been noted by the United States Department of Defense, which issued a call for proposals on this topic earlier in 2022. QOL assessments should also account for body image adjustments because some patients state they want to feel physically whole, and some report a greater need for a good physical match of the graft. Future research should consider the importance of graft functioning to the patient. For example, some patients are satisfied with a limb that is less functional. Also, the patient's age, gender, race/ethnicity, marital status, education levels, etc., may play an important role in QOL assessment because older persons may differ from younger ones on the importance of different psychosocial domains. In addition, the providers' perspective likely differs from the patients' experiences. The development of a VCA-specific QOL protocol/instrument should consider the following domains: importance of impact of graft outcomes on relationships; in the case of hand transplantation: being able to touch, hold love ones; in case of face transplantation: ability to express emotion; sense of connection with the graft; fear of medical complications; and fear of graft rejection (Smith and Cendales, 2019; Bound Alberti et al., 2022).

4. Informed consent and potential graft loss ('exit-strategies') in the evaluation and follow-up course

As generally known from the field of solid organ transplantation, also in VCA the evaluation and follow-up course are central elements for the success of the procedure. However, in the VCA context, informed consent is of particular importance for several reasons. First, patients need to consider whether the potential benefits outweigh the risks given that the goal of VCA is on restoring functionality and quality of life, rather than on saving life (Cooney et al., 2018). Second, relatively little is known about the psychosocial outcomes of different VCA organ recipients because of the small numbers of patients undergoing VCA. In the United States, for example, VCA transplant programs perform very few VCA transplants per year, making it nearly impossible to collect research data on a sample of recipients large enough to generate meaningful analyzes. Consequently, little is empirically known about patients' experiences of key elements of the informed consent process (i.e., information disclosure, comprehension, voluntary decision-making) that could help potential recipients make more informed treatment decisions about undergoing VCA and help families decide whether to authorize VCA deceased donation of their loved one's organs. One study assessed the availability and quality of VCA public education materials (Van Pilsum Rasmussen et al., 2020), and found that educational materials addressed upper extremity and face transplants more commonly than other VCA types, and that few materials identified patient populations who could benefit from VCA and the requirements for authorizing VCA donation. The study concluded that currently available VCA public education materials did not adequately educate the public (Van Pilsum Rasmussen et al., 2020). Similarly, a focus group study found that members of the public had little knowledge of VCA; reported information needs about who could donate, who needs a VCA, and the success rate; and maintained misunderstandings of VCA (Ferzola et al., 2022). A research study conducting interviews and focus groups among individuals with upper extremity amputations and individuals pursuing or had received an upper extremity VCA about decision-making to pursue VCA found that participants desired extensive information about upper extremity VCA in order to make decisions (Gacki-Smith et al., 2022). Many reported that their decisions in favor of pursuing upper extremity VCA were based on the prospect of regaining functionality and its associated independence, increasing social and physical confidence, and enabling more active parental involvement in childrearing; by contrast, those against pursuing upper extremity VCA reported concerns about their health or limb functioning becoming "worse off," the rigorous rehabilitation process, and having adapted to life without upper limb(s) (Gordon, 2022). Further, individuals maintained various definitions of "success" of upper extremity VCA (Kinsley et al., 2021; Downey et al., 2022). Other research has examined patient's perceptions of the risks and benefits of upper extremity VCA (Jensen et al., 2014). In sum, these studies highlight the need for VCA transplant programs to inform potential upper extremity and other recipients about VCA as part of the informed consent process. Toward that end, a publicly available neutral educational website, *Within Reach*, has been developed to provide patients, families, and healthcare providers with patient-centered information to make informed decisions about upper extremity VCA (Gordon, 2022). While these aforementioned studies focus prospectively on attaining a VCA organ, little attention has been devoted to "exit" strategies for responding to VCA graft loss. Specifically, the informed consent process should address whether recipients will need to undergo the removal of the VCA face or upper extremity or penis, the potential for re-transplantation compared to amputation or prosthetic care as options, as well as strategies and resources available to assist recipients in coping with graft loss (Smith and Cendales, 2019). Particularly because of the life-saving character of face VCA transplantations, these procedures need to be repeated (re-transplantation after graft failure; Kauke et al., 2021). Particularly because of the life-saving character of face VCA transplantations, these procedures need to be repeated (re-transplantation after graft failure). Until now, two patients have had face re-transplantation, one in France and one in the United States (Lantieri et al., 2020; Kauke et al., 2021). One case was notable for significant pain and loss of facial motion prior to removal of the graft. The patient then experienced visual hallucinations due to sensory deprivation after the graft was removed. Following re-transplantation, the patient-reported anxiety but his symptoms gradually improved and he reported reasonable quality of life and was able to resume work on a part time basis 2 years after transplant (Lantieri et al., 2020). A second facial re-transplantation was reported in 2021. Prior to re-transplantation, the patient was reported to have facial tightness, pain, and contraction with functional limitations in eating, drinking, and speaking and was re-transplanted without a period of time in which the donor graft was removed prior to re-transplantation (Kauke et al., 2021). These cases demonstrate both the feasibility and challenges of re-transplantation for face transplant recipients. The very significant loss of function, sensory deprivation if the graft is explanted prior to re-transplantation, and management of pain are notable points. The extensive nature of the allografts in these cases would suggest that other alternatives would not have been feasible. Significant advances in candidate selection, technology, operative technique, post-transplant care, and immunosuppressive management have contributed to the tremendous expansion of the field. Despite these recent achievements, face VCA transplant require complex surgical techniques, excellent immunosuppressive management, and well-established evaluation (limited allograft donor pool) and follow-up protocols as well as continued collaborative and multidisciplinary research efforts (Lantieri et al., 2020; Diep et al., 2021).

5. Bioethical considerations

There are numerous bioethical issues in VCA beyond respecting patients' autonomy by ensuring comprehensive informed consent as outlined above. Specifically, key ethical challenges include: management of the intense doctor-patient relationship, establishing fair patient selection and transparency of outcomes, maintaining donor registries, collecting and sharing data to advance the field of VCA, disparities, and gaining trust and support for the transition of VCA to becoming standard of care covered by insurance. Another issue is VCA procurement as there is no standardization or allocation system in place and it is usually done *ad hoc*. Policies and methods are needed to protect dignity of deceased donors and next of kin in the procurement process (Magill et al., 2019). Determining the needs of particular patients and whether VCA is the right treatment for them or if available alternatives would be more helpful also remains a challenge.

As pointed out by various authors, public awareness about VCA must be raised in order to support public trust (Caplan et al., 2019; Magill et al., 2019; Van Pilsum Rasmussen et al., 2020). While VCAs are legally considered organs in the United States since 2014, in the Eurotransplant member states (Samuel, 2016).

A major bioethical issue of VCA transplantation is the vulnerability of recipients with regard to functional and visible outcome as well as their hopes and expectations for the benefit of the challenging treatment. Other reasons why VCA recipients are considered to be particularly vulnerable is the temporary celebrity status they may acquire (Caplan et al., 2019) as well as potential harms incurred by providing an economy of fame (Magill et al., 2019). Patient advocacy has been identified as an important countermeasure to deal with vulnerability. It may be possible to support recipients' participation in their healthcare and helps to provide "ethical protection for both patients/candidates and transplantation teams who share the universal predisposition to self-justification and self-deception" (Benedict, 2022). Patient advocacy also safeguards living donors (Benedict, 2022). For example, the United States Organ Procurement and Transplantation Network (OPTN) requires the involvement of an independent living donor advocate (ILDA) to protect the best interests of a person who is willing to donate an organ while alive (Benedict, 2022). This is of particular interest also for the VCA context, as living donation for certain VCAs is increasingly practiced (Beederman et al., 2022), and is of particular interest for pediatric VCA (Pomahac et al., 2018).

An important bioethical issue pertains to racial/ethnic disparities in VCA recipients. In the United States, VCA recipients appear to be more White patients than Black or Hispanic patients across VCA organ types. For example, among uterus transplant recipients, 89.2% were White patients, 5.4% were Asian patients, 2.7% were Black, and 2.7% were multiracial (OPTN, 2022). However, the racial/ethnic profile of UE VCA recipients is not clear because many recipients are reported as "unknown" (OPTN, 2022), which precludes the analysis of racial/ethnic disparities in VCA receipt. For example, among all bilateral upper limb recipients in the United States there were $n = 9$ White, $n = 1$ Black, and $n = 9$ unknown race/ethnicity recipients; among all unilateral upper limb recipients in the United States, there were $n = 4$ White, $n = 1$ Hispanic, and $n = 13$ unknown recipients. One pediatric bilateral upper limb transplant recipient was Black. Little is known about factors contributing to racial/ethnic disparities. Future research should assess the role of multilevel factors and social determinants of health in contributing to such disparities. An ongoing challenge entails providing risk information to patients transparently to support their informed decisions, because framing of the information may easily become biased. This issue was recently discussed in detail for face VCA transplantation (Smith and Cendales, 2019) and with regard to implicit bias of the informed consent process. Such bias might influence patients' decisions in a way that makes them too optimistic with regard to their prognosis as well as risks and benefits. Therefore, it is important that doctors are aware of this psychological state of patients that affects their decision as well as of how complex the intertwining of medical ethics and medical practice are (Gilardino et al., 2021, 2022).

5.1. Bioethical considerations with special regard to bioethical considerations

Informed consent is best conceived of as a process. Challenges of this process particularly important in the context of VCA transplantation are information itself (which information should be delivered and how understanding could be assessed), the burdens and long-term commitments of VCA regarding immunosuppression and physiotherapy, and the complexity of how to provide new information on outcomes, particularly those referring to risks and complications in order to improve adherence and care management. An expert group (Brocher bioethics working group) recommended consent as a dynamic covenant in order to promote awareness of the importance of transplant recommendations for both patients and caregivers (Magill et al., 2019).

In order to implement such ethical considerations, Chauvet participants discussed a model of initiating the informed consent process by asking the patient what they know and understand particularly about face transplantation in the last meeting. This serves as a baseline for the physician to appreciate the areas in which the patient requires education about the process. Chauvet participants recognized that there is a spectrum of understanding and that there will be different levels of sophistication in different patients. Yet it remains the physician's responsibility to facilitate informed consent by providing education to meet the level of sophistication of the patient. This process is best achieved by: (1) ensuring that a patient has the capacity to consent (can iterate the risks and benefits and weigh them); (2) conveying the 'nuts and bolts' of the procedure; and (3) a longitudinal process with a proposed time of approximately 1 year from the time of initial evaluation, during which the face transplant team should work to understand the patient's motivations for wanting face transplantation. These recommendations have been discussed at length in the context of face transplantation (Smith and Cendales, 2019; Bound Alberti et al., 2022). But these recommendations present important issues also for the informed consent process in the context of other VCA procedures.

Pediatric VCA is particularly challenging because the effects of the transplant on childhood growth and development are unknown, unlike in the context of kidney, liver, heart, and lung transplantation (Doumit et al., 2014). VCA transplant outcomes may diverge from family and patient expectations and the degree of compliance necessary could become an overwhelming burden (Azoury et al., 2020). Informed consent in the pediatric setting is generally complex (Doumit et al., 2014). Formally, parents or legal representatives have the role of consenting to medical interventions in minors. As there is a general consensus, however, that children should be involved in the consent process according to their decision-making capacity, assessing this capacity at a certain age and in a given situation remains an important area of investigation (Hein et al., 2015). In the VCA context, this assessment can be particularly difficult as treatment options and implications for patient and family are challenging and it might be hard to decide how to best act in the best interest of the child (Azoury et al., 2020). Accordingly, the informed consent procedure needs to be adapted to the pediatric VCA context.

6. Conclusion and future challenges

There are fundamental differences between types of VCA, and we have focused much of this manuscript on common psychological dilemmas and future approaches that exist in VCA the majority of patients. Several VCA procedures (i.e., uterus and upper extremity VCA) have emerged as feasible options to provide a functional restoration following traumatic injuries or of infertility. International experience thus far has shown that successful VCA transplantation requires multi-stage multidisciplinary evaluation and follow-up. Candidate selection and evaluator training regarding assessment and ongoing follow-up to address recipients' post-transplant demoralization, depression, and adherence issues require further refinement to optimize candidate evaluation and follow-up protocols. More broadly, the elaboration of a comprehensive psychosocial framework is needed to provide guidance for individual VCA centers in standardizing their protocols and care procedures. We recommend establishing an international consortium of health care professionals and candidates/recipients (under the auspices of global transplant societies), to facilitate the sharing of experiences and individual perspectives. Use of systematic PROs assessment and follow-up by the recommended interdisciplinary transplant consortium may foster the identification of risk factors and patients' needs outside of routine clinical care, where patients' needs are often not addressed. Ultimately, we envision that such a consortium will greatly improve the exchange and networking among VCA providers and patients through international research that will support the advancement of psychosocial evaluation of VCA.

Author contributions

MK participated in research design, the writing of the paper, performance of the research, and contributed new insights. SJ-G, EG, and GW-F participated in the writing of the paper, performance of the research, and contributed new insights. All

authors contributed to the article and approved the submitted version.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Assessment of quality of life after upper extremity transplantation: Framework for patient-reported outcome scale domains

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Upper extremity transplantation offers the promise of restored function and regained quality of life (QOL) for individuals who have sustained hand or arm amputation. However, a major challenge for this procedure becoming an accessible treatment option for patients is the lack of standard measures to document benefits to QOL. Patient-reported outcomes (PRO) measures are well-suited for this kind of intervention, where the perspective of the patient is central to defining treatment success. To date, qualitative work with experts, clinicians, and patients has been used to identify the most important domains of QOL for PRO item development. Specifically, our group's qualitative work has identified several domains of QOL that are unique to individuals who have received upper extremity transplants, which are distinct from topics covered by existing PRO measures. These include emotional and social aspects of upper extremity transplant, such as Expectations and Perceived Outcomes, Integration and Assimilation of Transplant, Fitting in, and Post-Surgical Challenges and Complications. The broad topic of Satisfaction with Transplant was subdivided into three subtopics: Function, Sensation, and Aesthetics. Satisfaction with Sensation was also identified as a unique domain not evaluated by existing PRO measures. This report operationalizes these eight QOL domains by presenting scoping definitions. This manuscript describes the work that has been completed for domain characterization as an early step toward developing standardized PRO measures to evaluate these important outcomes specific to upper extremity transplantation.

KEYWORDS

quality of life, patient-reported outcomes, upper extremity, amputation, transplant

Introduction

Upper extremity (UE) limb injury and limb loss have been found to affect multiple areas of functioning including emotional well-being, social functioning (Paterson and Burke, 1995; Desmond, 2007; Saradjian et al., 2008; Gallagher et al., 2011; Østlie et al., 2011; Desteli et al., 2014), and physical functioning including activities of daily living, secondary medical conditions, and pain (Madhok and Bhopal, 1992; Davidson et al., 2010; Postema et al., 2012; Passero, 2014). UE limb loss drastically changes multiple aspects of one's quality of life (QOL), thus producing a need for QOL-improving rehabilitative treatments. UE transplant *via* vascularized composite allotransplantation (VCA) offers one such QOL-improving treatment option for those with hand and arm amputation (Elliott et al., 2014; Tintle et al., 2014).

Upper extremity transplantation has been shown to be a surgically and medically feasible treatment option for UE limb loss and is being offered at several surgical centers across the globe (Hollenbeck et al., 2009; Kaufman et al., 2009; Kubiak et al., 2019). However, the QOL outcomes from UE transplant have not yet been systematically or comprehensively documented (Kumnig et al., 2014; Bound Alberti et al., 2022). UE functioning is the primary outcome currently tracked for UE transplant recipients (Petruzzo et al., 2010; Tintle et al., 2014; Shores et al., 2017), for example using the Disabilities of the Arm, Shoulder, and Hand (DASH) measure (Beaton et al., 2001). However, by focusing only on UE functioning, this outcome assessment approach provides a limited picture of the success of UE VCA. While UE functional outcomes are important, they may not be sufficient to justify what is ostensibly a QOL intervention. This is particularly true given the current availability of advanced functional prosthetic arms and hands (Kubiak et al., 2019), which may confer function without necessarily addressing other aspects salient to QOL. Therefore, the full QOL impacts of this intervention remain to be documented objectively. Small samples and single-case studies using limited psychosocial outcomes have been reported (Singh et al., 2015; Salminger et al., 2016). However, the best metrics for evaluating QOL comprehensively for this population remain unknown.

With support from the U.S. Department of Defense's Defense Health Program Congressionally Directed Medical Research Programs, our research team has embarked on a program of study to develop a standardized and comprehensive approach for assessing the QOL outcomes of UE transplant that can be applied across treatment centers to allow for more uniform tracking of outcomes. We are using the methodology for patient-reported outcomes (PRO) measure development outlined and demonstrated by the Patient Reported Outcomes Measurement Information System (PROMIS®) initiative funded by the National Institutes of Health Common Fund (PROMIS, 2013) and the Quality of Life in Neurological Disorders (Neuro-QoL™) measurement system funded by the National Institute of Neurologic Disorders and Stroke (National Institutes of Health, n.d., Fries et al., 2005, Cella et al., 2006, 2007, 2011, Northwestern University, 2021). PROs are lacking for UE transplant, although this format for outcomes assessment is well suited for a QOL-improving intervention like VCA, given that the perspective of the patient is central to defining treatment success. Members of our research team have previously used these methods for developing condition-specific PRO measurement systems for spinal cord injury (Tulsky et al., 2011, 2015b,c; Tulsky and Kisala, 2015) and traumatic brain injury (Tulsky et al., 2016; Tulsky and Kisala, 2019), and we are currently developing item banks for limb injury and amputation (Tyner et al., 2018).

The initial steps in this PRO development process are to obtain substantive stakeholder feedback to guide development of such a system and to review existing measures for content (Bjorner and Ware, 1998; Bjorner et al., 2007; De Walt et al., 2007; Kisala and Tulsky, 2010; PROMIS, 2013). Thus far, our team has documented the domains of QOL impacted by UE transplant by soliciting stakeholder input (Tulsky et al., 2023) using a grounded theory-based qualitative approach (Glaser and Strauss, 1967; Strauss and Corbin, 1998; Kisala and Tulsky, 2010). The findings from this work are described briefly below. Our qualitative work has outlined domain areas specific to stakeholders of UE transplantation and we have used this information to determine where new PROs are needed. The present report describes these newly identified domains and the approach for drafting new PRO items.

Materials and methods

Qualitative stakeholder focus groups and individual interviews

Stakeholder feedback was collected from UE transplant experts and clinicians (i.e., surgeons, nurses, mental health professionals, and physical and occupational therapists) through a series of focus groups, and from patients who have received UE transplant *via* individual telephone interviews. The full methodology and findings have been reported in a separate manuscript (Tulsky et al., 2023) but will be described here briefly.

Three focus groups were conducted at the 2018 meeting of the American Society for Reconstructive Transplantation, and 10 focus groups were held across five UE transplant centers in the United States in 2019–2020. In total, 59 clinicians and other UE transplant experts along with five UE transplant recipients provided input on the most important domains of health-related

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Abbreviations: DASH, Disabilities of the Arm, Shoulder, and Hand; HRQOL, Health-related quality of life; Neuro-QoL™, Quality of Life in Neurological Disorders; PRO, Patient-reported outcome; PROMIS®, Patient Reported Outcomes Measurement Information System; QOL, Quality of life; TMR, Targeted muscle reinnervation; UE, Upper extremity; VCA, Vascularized composite allotransplantation.

QOL (HRQOL) for UE transplant. Trained data collectors and doctoral-level co-investigators conducted all sessions using open-ended semi-structured discussion guides to lead the groups or interviews. Sessions were audio-recorded and transcribed. All participants provided informed consent and data collection was approved by the University of Delaware Institutional Review Board.

This stakeholder input was analyzed qualitatively using a grounded-theory-based (Glaser and Strauss, 1967; Strauss and Corbin, 1998) approach that our team has used in several prior studies to identify important HRQOL domains in other clinical populations (Kisala and Tulsky, 2010; Slavin et al., 2010; Carlozzi et al., 2011; Tulsky et al., 2011, 2015b, 2016). Open, axial, and selective coding were employed to determine the important HRQOL themes (MacQueen et al., 1998; Morgan et al., 1998; Braun and Clarke, 2006; Guest et al., 2012).

The results demonstrated that there are numerous domains relevant to HRQOL after UE transplantation that can be measured by existing PRO systems that cover topics salient to many other clinical populations (e.g., fine motor functioning, anxiety, and pain interference). However, several domains of HRQOL that are unique to UE transplantation were identified as well. This highlighted the need for new item development to ensure HRQOL assessments are comprehensive. The framework defined in this manuscript provides the roadmap for how to draft new PRO items and demonstrates how the qualitative results helped define these new HRQOL domains.

Identifying gaps in HRQOL measurement for UE transplant and defining domains

New HRQOL content domains important to UE transplant recipients were identified in response to the qualitative input from stakeholders (Tulsky et al., 2023). The content coverage of each domain was designed to be directly responsive to the comments from stakeholders. Quotes from the interviews or focus groups were used as source material for new item text. To guide the future development of items, domain definitions were articulated and refined for each of the new domain areas.

Results

Eight new HRQOL content domains were developed based on stakeholder input. The subject, span, and relevant subtopics of each domain were defined. See Table 1 for a brief summary of these eight HRQOL content domains. Figure 1 contains a visual representation of how the domains are related across psychosocial and physical HRQOL. Each section below begins with a summary of the stakeholder input and exemplar quotes from the stakeholders are provided. Information on the subtopics considered for new PRO item drafting are described and examples of items are provided. These draft items will go through several stages of review and refinement before they can be considered ready for use; the draft items are presented here to demonstrate the process of PRO item development and are not intended to be adopted as PRO items for UE transplant HRQOL until the item development process is complete.

TABLE 1 New domains to evaluate HRQOL after UE transplant.

Domain	Content coverage
Expectations and Perceived Outcomes	Satisfaction with results of transplant and overall outcomes as well as accuracy of expectations in retrospect.
Post-Surgical Challenges and Complications	Burdens of post-transplant treatment and therapies; effects on health and personal life.
Integration and Assimilation of the Transplant	Acceptance and identification of the transplant as one's own; feelings of being complete or having something restored.
Fitting In	Comfort in social interactions where other people may view or touch the transplant(s).
Satisfaction with Hand Function	Comfort, confidence, and satisfaction with the functional abilities of the transplant(s) in various daily activities.
Satisfaction with Hand Aesthetics	Satisfaction with physical appearance of the transplant.
Hand Function: Sensation	Ability to perceive sensations with the transplant.
Satisfaction with Sensation	Satisfaction with sensory abilities of the transplant.

Expectations and perceived outcomes

Stakeholders described how it was difficult for UE transplant recipients and their families to truly know what the transplantation and recovery process was going to be like, despite extensive attempts by providers to inform them. Preparing UE transplant recipients for the requirements of recovery—and tempering expectations—was described as a major part of the pre-surgical evaluation process at all sites where experts were interviewed.

> *"Well, I expected to not [feel good] and they told me that, you would not feel good for a while. They were very honest."* – UE transplant recipient
>
> *"I don't know you really ever completely prepare for that.... I had always been healthy."* –UE transplant recipient
>
> *"We aren't selecting patients for hand transplant who walk away saying, 'I want to play the piano and I expect to play the piano.'*

> *So, one of the selection criteria is that they have low expectations and high motivation.… Someone whose need is to look completely normal in society and function as a concert pianist is not going to get a hand transplant from our group.”* –UE transplant expert stakeholder
>
> *“We definitely assess what their expectations are pre-transplant. And…if there's a big mismatch between their pre-transplant expectation and the reality of their outcome then we will see depression. But it's really important to assess whether or not they have realistic expectations and whether they understand what the potential adverse outcomes could be.”* –UE transplant expert stakeholder

To assess how well recipients’ pre-surgical expectations were met, we developed the Expectations and Perceived Outcomes domain. Stakeholder comments coded in this domain included emotional topics, such as feeling happy with results, second-guessing choices, and feeling regret or surprise about the outcome. Experiences and expectations of surgery, rehabilitation, medical side effects, and immediate recovery were also discussed. These emotional and practical topics will be incorporated into items in this domain; for example, *I felt prepared for the risks to my health after hand transplant*.

Integration and assimilation of the transplant

Stakeholders described the process and importance of integrating the UE transplant into one’s physical and mental bodily schema. This included how the recipients experienced the process of accepting the UE transplant as their own limb and also how the UE transplant changed how they felt about their bodies.

> *“So many of those patients, they've tried prosthetics and ultimately, they always see the prosthetic as a foreign object; they're never able to integrate this in their overall body image. This was always a foreign device. Whereas after the hand transplant they referred to the transplant as their own hand, so I think that's something that explains a little bit what it means, this wholeness part. Where with the prosthesis, although they have regained function where you can do things, they never felt whole; they always felt as an amputee. Whereas after transplant they have hands and they feel whole again by using those hands.”* –UE transplant expert stakeholder
>
> *“It makes me feel whole.”* –UE transplant recipient

The Integration and Assimilation of the Transplant domain was conceptualized to assess feelings about how well the UE transplant has been integrated and/or assimilated into the recipient’s life, including somatosensory feelings of bodily integrity (e.g., proprioception and kinesthetic) as well as corresponding psychological experiences, sometimes referred to as a feeling of “wholeness” or being complete. For example, an item like, *My hand transplant makes me feel more complete*, could be representative of this domain. Stakeholder comments coded in this domain included references to acceptance of the limb, such as to what extent the new UE feels like their own. These topics and other salient emotional experiences were targeted for content coverage, such as degree of comfort (or discomfort) with the transplanted limb, feeling like oneself again, and feeling like something lost has been regained.

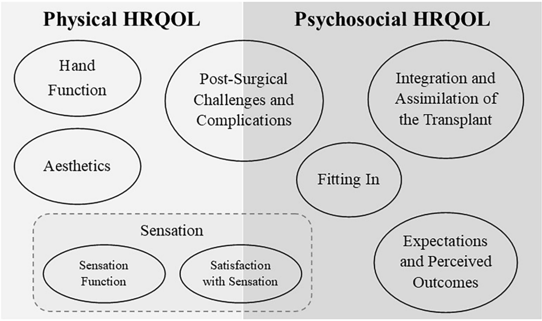

FIGURE 1
The eight new HRQOL content domains are presented above—represented by ovals—grouped into the overarching area of physical and psychosocial HRQOL under which they fall. In the area of physical HRQOL, three domains relevant to satisfaction with transplant were identified: Hand Function, Aesthetics, and Sensation. Sensation was further subdivided by separating Sensation Function from Satisfaction with Sensation. One domain includes aspects of both physical and psychosocial HRQOL: Post-Surgical Challenges and Complications, which includes physical and emotional reactions post-surgically. Psychosocial HRQOL included reflection on past perspectives in light of the completed transplant (Expectations and Perceived Outcomes). The internal reactions to stigmatization or feelings of self-acceptance (Integration and Assimilation of the Transplant) were separated from the external experience of stigmatization/acceptance within the social context (Fitting In).

Fitting in

Stakeholders discussed a number of important social experiences that are unique for UE transplant recipients, both prior to and after transplantation. Descriptions of the experience prior to the transplant were primarily about difficulties fitting in as an amputee, whereas after UE transplant, the comments were both positive and negatively valenced. In many cases recipients felt they were better able to fit in due to the transplanted limbs, although some concerns remained about being noticed as having post-transplant limb differences (for example, mismatches in size or skin tone). Salient emotions included fears of being judged negatively in social settings and relevant behaviors included avoidance of social contact due to concerns about not fitting in.

> *"After getting the hands, I felt like [I] could blend in more."* –UE transplant recipient

> *"…[I]f by doing the transplant they can go out in the world and they're not immediately identified as an amputee; … it's not the first thing that people notice about them."* –UE transplant expert stakeholders

The Fitting In domain was developed to evaluate the recipients' feelings of how well they fit in socially after UE transplant. Stakeholder comments coded in this domain included references to feelings of social comfort or discomfort, feeling judged by others, and potential for embarrassment about the physical appearance or functioning of the transplanted limb. Several comments also referenced the importance of confidence interacting in social settings where the transplanted limb may be observed—for example, decisions about wearing long sleeves or short sleeves and concerns about others staring at their limbs. This was incorporated into an exemplar drafted item, *I feel self-conscious about people seeing my hands*. This domain also captures feelings about being socially accepted and feeling a sense of "normality" with regard to not standing out as different in some way when perceived by others. Topics targeted for content coverage included both positive and negative feelings about fitting in socially, and referenced different social settings, such as being in public or around friends and family.

Post-surgical challenges and complications

Stakeholders described numerous challenges after transplantation, including adverse effects on health from immunosuppressive treatment and requirements for extensive rehabilitation care. There was also discussion of the occurrence of negative emotional outcomes from these new, potentially burdensome experiences. This content domain is multifaceted, covering a wide range of potential post-surgical challenges and complications, spanning aspects of both physical and psychosocial HRQOL (see Figure 1).

> *"You end up living at the hospital essentially for a while. So, you leave your life and you come. What I taught patients is rehabilitation becomes your full-time job from here on out. Or at least, other outside interests that you have, they have to take a back seat if you want to do this. And this has to be what you do every day seven days a week. And it's going to be this way for a year or maybe this way for two years."* –UE transplant expert stakeholder

> *"I do know that the side effects of the steroid that was given to me in such heavy doses caused me to gain a lot of weight."* –UE transplant recipient

> *"After about 5 years of having the hands, the [medication] really started to damage my kidneys."* –UE transplant recipient

The Post-Surgical Challenges and Complications domain was conceptualized as encompassing common post-surgical challenges or problems that may have developed as a result of the transplant as well as associated ongoing physical and emotional burdens of transplantation. Stakeholder comments covered a wide range of topics, including burdens of keeping up with treatment, taking medications, regular checkups, required lifestyle changes, and hand therapy (appointments and at-home exercises). Some of these burdens were described as potentially lifelong. Discussions also described the emotional aspects of complications, including concern about medication side effects, long-term health impacts (e.g., cancer and diabetes), lifespan reduction, rejection episodes, and life-threatening or severe complications. Of note, many of these challenges and complications are faced by individuals who undergo solid organ transplant. Potential items such as, *"My treatment limits my leisure activities"* and *"I feel bothered by medication side effects,"* exemplify this domain. The experiences of recipients with any post-transplant challenges or complications, and the experiences of burden and emotional sequelae from these experiences, were judged to be most pertinent to inform item content coverage in this domain.

Satisfaction with hand function

Stakeholders described that, depending on the recipient, different levels of functioning may be experienced as satisfactory. Some felt that very little UE functioning was a positive outcome, whereas other recipients were seen to have higher requirements for the level of functioning needed to feel satisfied. Discussions of UE transplant function included the movement, strength, and flexibility of the UE as well as feelings of comfort, confidence, and satisfaction with the UE transplant functionality.

> *"My hand functions just as well as a normal hand, to be honest. I can fully grasp, make a fist… you forget that you are an amputee a lot of times."* –UE transplant recipient

> *"I'm very happy even if my hands do not move like normal hands."* –UE transplant recipient

> "*But then there are other patients who have completely different goals, and for them it's really to work out, to do pull-ups, whatnot.*" –UE transplant expert stakeholder

The Satisfaction with Hand Function domain was designed to capture satisfaction with the use and functionality of the transplant, considering the entire UE. Topics covered include general satisfaction and satisfaction with specific functions and uses (e.g., range of motion). Stakeholder comments coded in this domain included both positive and negative statements about the UE transplant functionality as well as descriptions of different activities that could or could not be done with the UE transplant. These different types of functionality and descriptions of emotional reactions to the functional outcomes are important aspects that inform the drafting of items in this domain—for example, *I feel frustrated with how my transplant functions*.

Satisfaction with hand aesthetics

Stakeholders shared that the aesthetic aspects of the UE transplant were important for recipients' evaluation of their overall outcome to a greater or lesser degree depending on the individual's goals in seeking a transplant. For some, the aesthetic aspects were paramount and the procedure was tantamount to aesthetic surgery, while for others, the appearance of the transplant was a secondary or tertiary goal. Stakeholders also explained that the aesthetic aspects of the transplants were expected to change over time, as follow-up procedures (e.g., debulking) could be done to improve aesthetics.

> *"It has been always important for me to have new real hands and not plastic or silicone hands."* –UE transplant recipient
>
> *"They try to match on skin color as well as [donor] sex.. but … there's often a big size discrepancy in the arms …because what's left of [the recipient's] native arm is often very shrunken and small, and then you're transplanting … a normal size forearm."* –UE transplant expert stakeholder clinician

To assess recipients' satisfaction with the external appearance of the transplant, we designed the Satisfaction with Hand Aesthetics domain. Specific subtopics covered in the stakeholder discussions included skin tone of the transplant, size of the transplant, fingernail appearance, forearm bulk, scar appearance, and body hair color. Each item would cover an aspect of only one of these subtopics—for example; *I am satisfied with the skin tone of my transplant*. These subtopics and the need to evaluate the recipient's feelings of satisfaction or dissatisfaction with the aesthetic qualities of the UE transplant were considered to be important considerations when drafting items for this domain.

Hand function: Sensation and satisfaction with sensation

Stakeholders discussed sensation as a major outcome of interest that motivated many recipients to pursue UE transplant. Sensation is one of the functions that cannot be replicated by traditional prosthetics. Sensation has numerous important impacts on daily functioning as well as emotional and social functioning.

Stakeholders described the sensory skills that were missed for UE amputees, and the process of regaining sensation after transplantation. Various sensory skills were discussed as functional abilities, but also for the more personal meaning inherent in these sensory experiences. Sensation was discussed as a socially relevant sense and was closely tied to the desire for improved social functioning after the transplant.

> *"Sensation though is so important. And I can't reinforce that enough as it relates to relationships with those that you love. Your spouse and your children, especially for those that have young children. Hooks don't have any value with young children, and electric hands don't have value with children."* –UE transplant expert stakeholder
>
> *"I can feel what I touch, I can feel if it is hot or if it is cold, if it is soft, or if it is itchy or anything, and … that is something that is very important for me, and it goes with the fact that I can like touch somebody. So, for example, my boyfriend, I can… put my hand on him and I can touch him or feel him or touch his hair or things like that… that really matters for me currently."* –UE transplant recipient

In response to these stakeholder comments, two HRQOL content domains were developed on the topic of sensation. First, the Hand Function: Sensation domain was designed to evaluate recipients' ability to perceive a variety of sensations in the transplanted limb/hand. These included, for example, light pressure, touch, textures, temperature, and pain. The second domain developed was Satisfaction with Sensation. This domain was designed to assess recipients' satisfaction with their ability to perceive sensation with the transplant, including social touch.

These two domains were conceptualized as discrete because stakeholders acknowledged that recipients' degree of satisfaction may not correlate directly with the amount of sensory function they have in the UE transplant. Stakeholders described how acquiring even minimal amounts of protective sensation was experienced as a benefit over prosthetic devices. Hence, Hand Function: Sensation focuses on practical aspects of sensory skills and behaviors as shown in an exemplar item like: *My sense of touch in my hand(s) is good.* Satisfaction with Sensation focuses more on satisfaction and mental/emotional implications of regaining sensory functioning and can be depicted in an item such as: *My hand(s) help(s) me feel closer with people when I touch them.*

Discussion

To understand the HRQOL effects of what is considered to be a QOL-enhancing a procedure, it is critical to systematically assess post-transplant HRQOL from the patient's own perspective. The first steps to implementing routine PRO assessment in a given population are to identify the most important domains of HRQOL to assess, and then to develop PRO items to measure each of these domains. Many of the factors important to understanding the physical and psychosocial HRQOL outcomes of UE transplant are shared with other rehabilitation populations, such as fine motor functioning and ability to conduct self-care activities, pain interference, and emotional difficulties, such as depression, anxiety, and psychological trauma (Cella et al., 2012; Gershon et al., 2012; Kisala et al., 2015a,c; Tulsky et al., 2015a). Additionally, several unique areas relevant to HRQOL after UE transplantation were identified in our recent qualitative work with stakeholders (Tulsky et al., 2023).

After transplant, it is known that recipients face new challenges and risks of complications, and there are opportunities for hindsight and possible regrets, all of which have the potential for condition-specific psychosocial outcomes. These are areas where we determined that new PRO content was necessary, and so we are working to develop the domains of Expectations and Perceived Outcomes and Post-Surgical Challenges and Complications, the latter of which includes aspects of both physical and psychosocial HRQOL.

Psychosocial outcomes such as reduced participation and regaining independence are domains where existing PRO measures can be applied (Cella et al., 2012; Gershon et al., 2012; Heinemann et al., 2015, 2020; Victorson et al., 2015; Kisala et al., 2020). Likewise, measures of depression, anxiety, and traumatization are relevant for UE transplant recipients just as they are for individuals in other rehabilitation populations who have experienced life-altering disabilities (Kisala et al., 2015a,c; Tulsky et al., 2015b). However, the domains of stigmatization and self-acceptance, topics that have been identify as relevant for many other rehabilitation populations (Cella et al., 2012; Gershon et al., 2012; Kalpakjian et al., 2015; Kisala et al., 2015b) are experienced in a particular way by UE transplant recipients. These patients are faced with a unique opportunity to integrate the transplanted limb into their identity (i.e., assimilation), as well as an opportunity to reintegrate socially as a person with ostensibly intact limbs. The concepts of feeling "whole" and "normal"—terms that can evoke negative stereotypes and reflect the type of ableist language typically avoided in rehabilitation research—were repeatedly broached by stakeholders in our qualitative work. Use of these terms in this way implies that if the loss of a limb results in feeling like "something is missing," a transplanted limb is indeed an opportunity to both figuratively and literally feel restored or "whole." These sensitive topics are critical, therefore, for inclusion in assessment of HRQOL outcomes following UE transplant. To respond to this need, we have chosen to develop outcomes domains in the psychosocial HRQOL area of Integration and Assimilation of the Transplant and Fitting In.

From a medical/surgical perspective, satisfaction with the transplant is a broad and vital area for assessment. Based on our stakeholder input and experience with PRO development, we chose to divide the topic of physical HRQOL after UE transplant into three areas: Function, Sensation, and Aesthetics. Functional *abilities* of the transplanted limb can be evaluated similarly to any surgical population, considering, for example, range of motion, grip strength, pain, and the various activities of daily living that require manual motor function and dexterity to complete. Thus, we believe that assessment of functional ability is best left to existing measures, such as SCI-FI Fine Motor, SCI-FI Self-Care, PROMIS Upper Extremity (Jette et al., 2012; Tulsky et al., 2012; Kaat et al., 2019), or Neuro-QoL Upper Extremity-Fine Motor (Cella et al., 2012; Gershon et al., 2012).

In contrast, *satisfaction* with hand functioning as experienced by UE transplant recipients appears to merit a new HRQOL content domain, as difficulties and frustrations with the responsivity and ease of movement of the transplanted limbs/hands are distinct for this population, where capabilities improve gradually with treatment and nerve regrowth—or sometimes not at all. Likewise, the challenges with sensation and aesthetic satisfaction are also unique to UE transplant. Although there are other clinical groups (e.g., spinal cord injury) where UE sensation can be impacted by injury, and the process of developing sensory-motor control for advanced prosthetic limb users has some similarities (Graczyk et al., 2019; Sensinger and Dosen, 2020), the experience of regaining sensory capabilities as donor nerves become reinnervated is distinct in limb transplant. There are similarities for UE transplant recipients to the benefits experienced from targeted muscle reinnervation (TMR) in terms of reduced phantom limb pain and neuroma-related pain (Morgan et al., 2016; Tintle et al., 2016; Dumanian et al., 2019), and similarities with both invasive (e.g., targeted sensory reinnervation) and non-invasive (e.g., armband-based stimulators) technologies for restoring sensory input with prosthetic or bionic devices (Bensmaia et al., 2020), although ideally the outcome of the VCA transplant will go further than any one of these approaches and restore more natural motor control and sensation to the injured limb. Furthermore, the aesthetic and cosmetic aspects are of central concern for many patients (e.g., concerns about mismatched skin tone, body hair color/texture, and musculature/size/bulk of transplanted limb). These issues are layered upon the body image concerns experienced by UE amputees and involve the singular experience of aesthetically integrating donor tissue in a highly visible body location (whereas most solid-organ transplants occur with internal/non-visible tissue). Hence, we have chosen to develop outcomes measures in the physical HRQOL area, including Hand Function, Aesthetics, and Sensation (both Function and Satisfaction with Sensation).

The work in which we are engaged is designed to use stakeholder feedback to identify the appropriate domains for HRQOL assessment and to make the feedback actionable by developing items to measure these important stakeholder-identified areas. The definitions and theoretical structure described throughout this manuscript are critical to the development of items that can measure these areas of function. This marks one of the initial efforts to systematically develop new scales that focus directly on issues of critical importance to those who have undergone UE transplant, but which are absent from existing HRQOL measurement systems.

Conclusion

Standardized, routine, and comprehensive evaluations of UE transplant outcomes are necessary to provide evidence to evaluate this treatment as part of the standard of care for UE injury or limb loss. Based on industry standards for PRO assessments of HRQOL and our completed qualitative research, we recommend that HRQOL assessment for this population includes both existing measures—those that are applicable to many rehabilitation populations, such as measures of depression, anxiety, and pain—as well as measures that are unique to the experience of UE transplant, covering emotional, social, and physical functioning. The eight newly developed PRO domains described herein were designed for this purpose. Future work is needed to finalize the development of new items in these domains and to ensure content validity.

Data availability statement

The datasets presented in this article are not readily available because a data use agreement must be signed prior to release. Requests to access the datasets should be directed to DT, dtulsky@udel.edu.

Ethics statement

The studies involving human participants were reviewed and approved by University of Delaware Institutional Review Board. The patients/participants provided their written informed consent to participate in this study.

Author contributions

All authors contributed to data collection. CT, PK, and DT managed the qualitative analysis. CT, JS, and DT wrote new item domain scoping definitions. CT wrote the first draft of the manuscript. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Psychosocial predictors in upper-extremity vascularized composite allotransplantation: A qualitative study of multidimensional experiences including patients, healthcare professionals, and close relatives

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Background: The impact of patient-specific psychosocial factors on functional outcomes after upper-extremity (UE) vascularized composite allotransplantation (VCA) is poorly understood. The objective of this study was to identify relevant psychosocial predictors for success or failure of UE VCA in an Austrian cohort.

Methods: A qualitative study was undertaken consisting of semi-structured interviews with UE VCA staff, transplanted patients, and close relatives. Participants were asked about their perceptions of factors that either favored or hindered a successful transplant outcome, including functional status before surgery, preparation for transplant, decision-making, rehabilitation and functional outcome after surgery, and family and social support. Interviews were conducted online and recorded with the consent of interviewees.

Results: Four bilateral UE VCA patients, 7 healthcare professionals, and a sister of a patient participated in the study. Thematic analysis revealed the importance of an expert interdisciplinary team with adequate resources for patient selection. Psychosocial aspects of prospective candidates are crucial to evaluate as they contribute to success. Both patients and providers may be impacted by public perceptions of UE VCA. Functional outcomes are optimized with a life-long commitment to rehabilitation as well as close, ongoing provider involvement.

Conclusion: Psychosocial factors are important elements in the assessment and follow-up care for UE VCA. To best capture psychosocial elements of care, protocols must be individualized, patient-centered, and interdisciplinary. Investigating psychosocial predictors and collecting outcomes is, thus, critical to justifying UE VCA as a medical intervention and to providing accurate and salient information to prospective candidates.

KEYWORDS

vascularized composite allotransplantation, qualitative research, psychosocial predictors, psychosocial outcomes, quality of life

Introduction

The primary goal of upper-extremity vascularized composite allotransplantation (UE VCA) is to maximize transplant recipients' functional, emotional, and social quality of life (QoL). Outcomes are much broader than graft survival, range of motion, and other traditional objective endpoints. Accordingly, while immunological, biomechanical, and medical factors are important determinants of transplant outcomes, psychosocial factors also play critical roles. Due to the small number of performed UE VCA cases and the heterogeneous screening and follow-up protocols, few studies of psychosocial predictors of outcomes have been performed worldwide; hence, our understanding of this topic is limited (Kumnig et al., 2012, 2013; Singh et al., 2015, 2016; Jowsey-Gregoire et al., 2016; Heineman et al., 2020; Bound Alberti et al., 2022; Kumnig et al., 2022), although, several transplant centers worldwide have developed specific VCA programs (Kumnig et al., 2013). Recent research (e.g., Heineman et al., 2020; Kinsley et al., 2022) provides a good understanding of functional and sensory, and psychosocial outcomes (Kumnig et al., 2014). Additionally, comprehensive qualitative research initiatives have recently been undertaken to enhance psychosocial outcomes in VCA and discuss the key psychosocial challenges faced in UE VCA today.

As noted above, UE VCA is life enhancing rather than life saving such as in the case in solid organ transplantation (SOT; Dickenson, 1999; Dubernard, 2011). Hence, the risk-to-benefit ratio is quite different than with SOT in which the risks are offset by the lifesaving nature of the procedure (Tobin et al., 2005; Kumnig et al., 2013, 2014). Ideally, candidates will be strongly motivated for transplantation; have demonstrated reliable adherence with medical care in the past; have strong family support networks; and utilize acceptance, flexibility, and problem solving in adapting to the loss of function from the injury/deficit and for future rehabilitation following transplantation (Sears Jr. et al., 1995; Olbrisch, 1996; Leo et al., 2003; Kumnig et al., 2014). A candidate who has been educated about the various risks, benefits, and demands of the transplant experience, and who has been prepared for the psychological stresses of the experience is more likely to have appropriate expectations regarding the risks of immunosuppression and surgical complications, as well as a more realistic understanding of potential functional gains after transplantation (Lanzetta et al., 2001; Sicard, 2011).

In reality, UE VCA candidates may overestimate the benefits of the procedure while underestimating the surgical risks, duration of recovery, demanding post-transplant medication regimen, and intense rehabilitation requirements (Simmons, 2000; Brenner et al., 2002; Baylis, 2004; Brouha et al., 2006; Petruzzo et al., 2010; Sicard, 2011; Kalluri and Hardinger, 2012; Kumnig et al., 2014; Kinsley et al., 2020). Unmet expectations, an inability to incorporate the transplanted hand (s) (Lanzetta et al., 2005, 2007; Petruzzo et al., 2008; Kumnig et al., 2013), and either new or recurring psychiatric conditions have been reported after UE VCA (Schuind et al., 2007), including suicide attempts (Schuind et al., 2006) and request for amputation (Petruzzo et al., 2008; Petruzzo and Dubernard, 2011). Additionally, recipients may be frustrated with the lengthy process of recovery. The loss of ability to perform tasks that were possible preoperatively also leads to initial postoperative decrease in quality of life (Petruzzo et al., 2010; Kumnig et al., 2014).

Fortunately, the majority of recipients have reported successful psychological integration of the hand(s), and improved confidence in appearance and in social situations (Schuind et al., 2006; Jablecki, 2011). Recipients that have assimilated the transplanted hand(s) into their body-/self-image are generally able to develop a sense of "ownership." Additional important outcomes are the observed improvements in QOL and activities of daily living (Kumnig et al., 2014). It has become apparent that patients' coping styles, support from family and friends, and financial factors are important predictors of successful outcomes (Shores, 2011). Recent findings also show that patients' relationships to healthcare providers, as well as to family members and peers, are correlated with satisfaction (Kinsley et al., 2022). Patients may also experience stress due to media attention which has occurred in a number of UE VCA cases (Kumnig et al., 2012); this impacts the decision to undergo a UE VCA procedure and the post-transplant course. Therefore, multidimensional psychosocial evaluation and follow-up protocols should include these additional domains: health literacy regarding transplantation, assessment of pain related to amputation and phantom limb pain, family support, adaptation to prosthesis, financial and family stressors, assessed through multiple interactions with a variety of assessors including psychiatrists, psychologists, social workers, hand therapists, and all team members (Dobbels et al., 2009; Shores, 2011; Kumnig et al., 2014).

This qualitative study has a similar design as the recently published qualitative investigation of Kinsley et al. (2022), which aimed to explore the role of patient-specific factors through a qualitative analysis of interviews with UE VCA recipients.

In the present study, we have adapted and expanded the interview protocol combining the interviews with those of healthcare professionals of the interdisciplinary VCA team and relatives of the UE VCA recipients and contrasting the outcomes of United States VCA recipients with those in Austria. The main goal of this qualitative research was to elucidate relevant psychosocial predictors for success or failure of UE VCA in this European cohort. By understanding these psychosocial factors, we hope to enhance existing heterogeneous screening and follow-up protocols by including identified important psychosocial factors in the evaluation and peri-operative management and optimization of potential candidates for UE VCA.

Materials and methods

Participants

In total five patients underwent reconstructive UE VCA at the Medical University of Innsbruck so far. One of these patients who received unilateral UE VCA in 2009 died due to progressive gastric cancer, leaving a total of four potential patients, which were eligible to be invited to participate in an online interview. Additionally, online interviews with the staff of the Innsbruck VCA team were scheduled to assess the healthcare professionals' overall expertise working in the field of UE VCA. Also, interviews with close relatives of the four transplanted patients have been planned to gather individual experiences of partners/main social daily contacts living with somebody who underwent UE VCA.

Inclusion criteria consisted of all patients and healthcare team members with direct experience undergoing VCA or providing care for VCA patients. These include all patients who have undergone UE VCA at Innsbruck, close family members of patients such as a spouse or primary caregiver, and all core members of the interdisciplinary transplant team, which includes surgeons, bioethicists, rehabilitation

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Abbreviations: UE VCA, upper-extremity vascularized composite allotransplantation; ISVCA, International Society of Vascularized Composite Allotransplantation; QOL, quality of life; SOT, solid organ transplantation; VCA, vascularized composite allotransplantation.

specialists, psychologists, dermatologists, and institutional or administrative leaders. There were no exclusion criteria other than individuals unable or unwilling to provide commentary or participate in the study. All potential participants and staff received an introductory email inviting participation in an online interview. Written informed consent was provided by the final participants.

A total of four bilateral transplanted patients as well as seven healthcare professionals of the Innsbruck VCA team were interviewed. Overall, four close relatives/partners were potentially eligible to take part in an interview; however, only one family member was enrolled in the study. The wife of the first transplanted patient did not provide informed consent (due to missing skills to realize the online interview), the second patient was living alone (without a partner), and the fifth patient lived with his mother who was almost 90 years old. Only the sister of the third patient provided informed consent to participate in this qualitative research study.

All study activities were approved by the Ethics Committee of the Medical University Hospital, Innsbruck (vote 1044/2020). Recruitment and interviews followed a similar process to that at Brigham and Women's Hospital (Harvard Medical School) published by Kinsley et al. (2022), representing the largest UE VCA samples investigated in this way.

Interviews

We conducted a total of 12 interviews: 7 with UE VCA healthcare professionals, 4 with UE VCA transplanted patients, and one with a patient's relative. A trained interviewer conducted the interviews using a semi-structured guide. The interview guide consisted of open-ended questions that asked participants about their perceptions of factors that either favored or hindered a successful transplant outcome. Topics included functional status before surgery, experience with the preparation for transplant, decision process and information transfer, rehabilitation and functional outcome after surgery, and family and social support. Interviews were conducted online and recorded with the consent of interviewees. Subsequently, a qualitative analysis of the interview transcripts was performed.

Thematic analysis

Thematic analysis followed routinely-accepted methodology in qualitative research (Kinsley et al., 2020). Six researchers collaboratively created a coding scheme consisting of 51 codes and 10 subcodes. The codes consisted of single words or short phrases to produce sections of text that meaningfully related to the study's guiding question, "What factors influence the success of upper extremity VCA?" These codes were applied to all transcript data by six members of the Innsbruck and Harvard research group.

Members of the Innsbruck team grouped the previously created codes into themes, which were formulated as directed hypotheses. Care was taken to ensure that they had sufficient internal homogeneity and external heterogeneity to be sufficiently grouped or distinct from the other themes. We used qualitative analysis software (Atlas.ti) to extract citations supporting the themes. The Innsbruck team listened to the taped recordings and identified themes, using the qualitative analysis software to index their digital location in the recordings, creating codes that have been connected and grouped to themes. All investigators agreed upon a thematic map showing relationships between the individual themes and the guiding questions of the study (for details please see Figure 1).

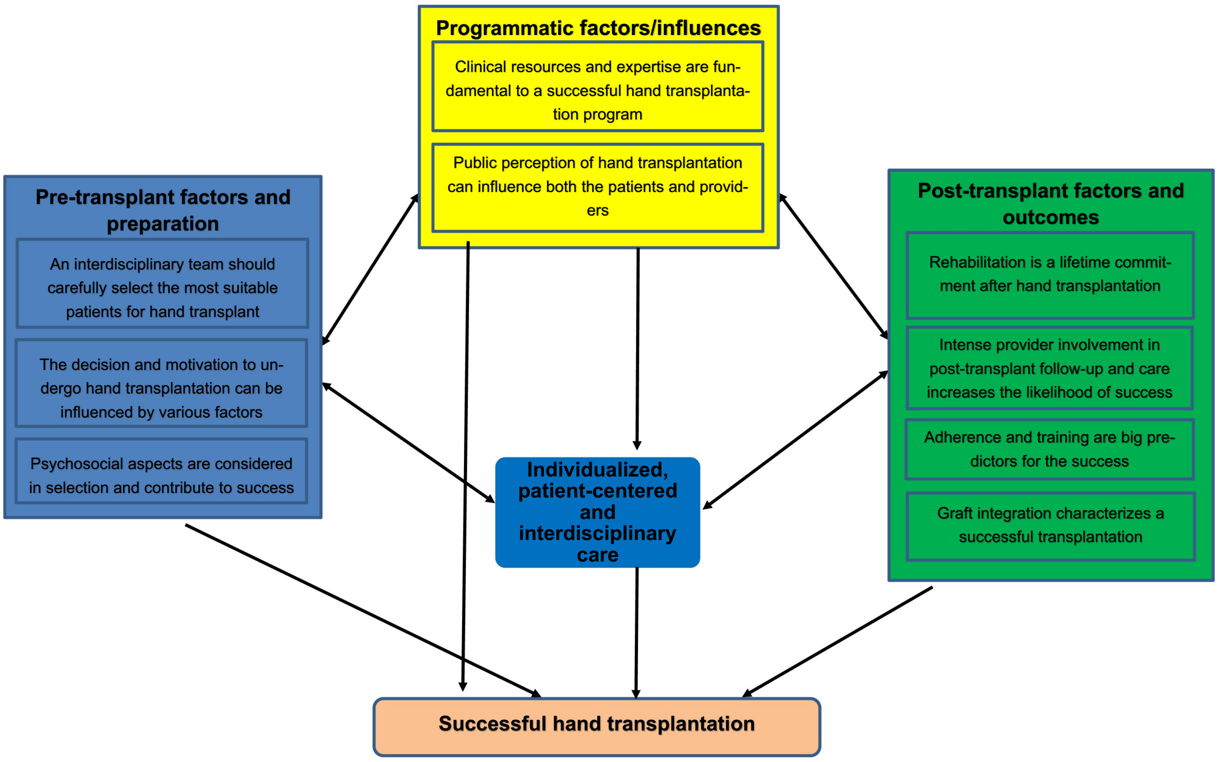

FIGURE 1
Thematic map.

Results

Participants

The study sample consisted of 4 bilateral upper limb VCA patients, the sister of an upper limb VCA patient, and 7 upper limb VCA healthcare professionals. This latter group consisted of 5 transplant surgeons and 1 rehabilitation physician. Four participants were female and 8 were male.

Thematic analysis of participant commentaries led to the identification of three main psychosocial domains: pre-transplant factors and preparation, programmatic factors and influences, and post-transplant factors and outcomes. For each of these three main domains, individual themes were established; these are summarized in detail below and illustrated in Figure 1. Representative participant quotations for each of the domains and themes are illustrated in Table 1.

Pre-transplant factors and preparation

Patient selection

One main outcome of this qualitative research was the identification of the role of an interdisciplinary team that carefully optimizes and selects the most suitable patients for UE VCA. For pre-transplant patient selection, it can be noted that a higher level of adherence to protocols and willingness to adapt to an intense training and rehabilitation process are important psychosocial predictors of a successful outcome (2C, 3C). The independent involvement of each discipline in the UE VCA with subsequent interdisciplinary discussion, treatment planning, and task distribution enables a holistic evaluation of the patient, prevents the forgetting of information, and leads to better pre-and post-transplant treatment and overall psychosocial outcomes (4C, 5C).

Motives

The decision and motivation to undergo UE VCA transplantation can be influenced by various factors. Motives may include a desire to feel whole again or a desire for (gain of) functionality and sensibility. Presumably, since a certain level of function is achievable with prosthetics, sensibility and the sense of wholeness were more often referenced over functionality (1R, 6C, 2R, 7C, 3R, 8C, 4R, and 9C). The desire to no longer to be perceived as “handicapped” or “disabled” as well as one’s own demands and goals for the future influence the motives for undergoing a UE VCA (6C, 9C).

Psychological factors

Psychological factors are not only considered in selection but also contribute to success of UE VCA. Factors that had a positive influence on UE VCA were the pursuit of a regular daily routine, family support, and a high degree of self-discipline (10C, 13C, 14C, 15C). On the other hand, addictive behavior and a lack of cognitive abilities were associated with a worse psychosocial and functional outcome and could be considered as areas to be addressed pre-transplant and/or as relative contraindications (11C, 12C).

Programmatic factors and influences

Clinical resources

Clinical resources and expertise are fundamental to a successful UE VCA transplantation program. An advanced understanding of the field, including literature and exchange with colleagues (16C), and a functional and very experienced transplant team is essential to provide the necessary infrastructure to develop a VCA program (17C).

Public perceptions

Public perception of UE VCA transplantation can influence both patients and providers. Patients can be influenced by the public, as they primarily perceive the loss of a hand as something negative (19C). However, a successful UE VCA is often viewed as sensational by the public (19C, 20C). As UE VCA becomes more common and routine, this may reduce the sensationalism and provide more reassurance to the patients (19C, 20C).

Post-transplant factors and outcomes

Rehabilitation

Rehabilitation is particularly important and a lifetime commitment after UE VCA transplantation. Rehabilitation can be positively influenced by a high level of pre-transplant activities of daily living; however, the rehabilitation process can be experienced as a ‘difficult road’ paved with a variety of complications and challenges (21C, 5R, 24C). Physiotherapy and a high degree of self-motivation lead to steady improvements, in terms of sensibility and motor skills, even after several years (22C, 23C, 25C).

Follow-up and care

Intense provider involvement in post-transplant follow-up and care increases the likelihood of success. The relationship between the primary healthcare professionals and UE VCA patients differs from ‘regular’ doctor-patient relationships in intensity (26C). Maintaining a close relationship between the primary healthcare professionals and the UE VCA patient is very time consuming and demanding, but due to the often time-sensitive and critical nature of patient issues, it may be necessary to prevent complications (26C, 27C, 28C).

Adherence and training

Both adherence and training are big predictors of success. It is essential that patients trust primary healthcare professionals regarding immunosuppressive treatment and other medical management (6R, 7R). Moreover, it is important that patients continue rehabilitation training achieve their highest potential functional improvement (7R, 29C).

Graft integration

Graft integration characterizes a successful transplantation. In order to be motivated through intensive rehabilitation, patients do better when they accept grafts as their own hands (30C, 8R).

In summary, the identified important psychosocial factors that lead to a better overall outcome are typically met when a VCA program provides individualized patient-centered and interdisciplinary care.

Discussion

In this qualitative research on psychosocial predictors and outcomes of patients that underwent UE VCA, three main psychosocial factors have been identified: pre-transplant, programmatic, and post-transplant factors. These psychosocial factors are discussed and contrasted to findings in recent literature.

TABLE 1 Categories and themes from the thematic map, associated hypotheses and supporting quotations from the interview transcripts.

Categories	Themes	Sample of supporting text from transcripts
Pre-transplant factors and preparation		
Patient selection	An interdisciplinary team should carefully select the most suitable patients for hand transplant	*"Mobility is one thing, but I think the main indication for hand transplantation is the sensation that the patients then have in their hands, so that they can feel things and people. And that is what the hand transplant provides, mobility varies greatly depending on compliance and motivation, but a prosthesis can do that too." (1C)* *"I think transplanting a hand for someone who's been waiting for it and who says life only gets good if they have their hands transplanted is something to think twice about. (…) After that, he is not disciplined enough to train or perform in such a way that it actually has a benefit for him in terms of movement. (…) But if it's a patient who says he wants the hands because he wants to feel and he's not restricted in his life in any way because he has designed his daily routine such that he could theoretically get by without hands, but he really wants to feel, I would say ok. He's a reflective person who knows exactly why he wants that and who has organized his life in such a way that it also works without hands." (2C)* *"A congenital malformation is something that should not be transplanted. (..) Then there are patients who have psychological problems, who do not fit into the rehabilitation scheme, who are not compliant, do not perceive control and take medication irregularly, and do not go to occupational therapy. These are things that should definitely be filtered out preoperatively." (3C)* *It is very important that each discipline sees the patient and then discusses collectively because patients tell different doctors different things. Because, for example, they have a better personal basis for conversation, for example with another doctor. Or you can talk to one doctor very easily about the medication, but you do not dare say that you have a functional problem. Some patients want to impress certain colleagues, while others do not like them. And that's why interdisciplinarity is so important, because we are all human. (4C)* *"I have the image in my head that we have managed to set up a football team in which everyone is aware that in order to win this match we are a team and we have to stick together." (5C)*
Motives	The decision and motivation to undergo hand transplantation can be influenced by various factors	*"The most important thing is that you have a hand at all and do not walk around with stumps. That was a disaster, that was a real eye-catcher." (1R)* *"The patient wanted bodily integrity - he was not considered complete in his village community since he has a deficit." (6C)* *"Even if you hug someone with your hands, it's flesh and skin as it should be and not plastic." (2R)* *"Sitting at the inn table and being able to put both hands on the table was certainly more important to him, because he had a functioning hand with sensibility and motor skills, and because of that the optics and aesthetics were most important for the patient." (7C)* *"It was also an important concern for me, when I stroke my wife's hair with my hands, whether I will feel it too." (3R)* *"Sensibility is the main argument for hand transplantation from a professional point of view. Functionally, prostheses can do a lot, only biofeedback is missing, sensibility feedback is missing" (8C)* *"It goes without saying that I had the unspeakable desire to ride a motorcycle again." (4R)* *"And the second reason was his wife's Christmas tree plantation, on which he wanted to continue working. And for that he needed a strong, functioning hand, since he was never able to work with his prosthesis." (9C)*

(Continued)

TABLE 1 (Continued)

Categories	Themes	Sample of supporting text from transcripts
Psychological factors	Psychosocial aspects are considered in selection and contribute to success	*"I would not like to transplant someone who does not have a regular daily routine, someone who has no idea what they would like or could do in the future. (..) So I would like to transplant someone who says I have a job that I go to every day. I have a group of friends that I meet up with regularly. So someone who has very regular routines, who does not live just for the moment, who still lives his life even though he has no hands, is suitable." (10C)* *"Any kind of addictive behavior in terms of substance use and alcohol should be an absolute contraindication. Smoking should also be an absolute contraindication. (..). Of course, this is a patient who is vulnerable, who perhaps has less self-discipline, who does not have such an orderly life, and I think that should be a contraindication." (11C)* *When we talk about hand transplants, we must not only assume absolute contraindications. Of course there are. Take someone who has severe dementia and has had a serious accident as a result and lost both hands. (..). Where you simply have to say that this cannot work due to dementia and the lack of cognitive abilities. This is an absolute contraindication." (12C)* *"I think it's the most important attitude is the self-discipline. (..) So self-discipline is what brings the maximum benefit to the patient." (13C)* *"I think family and supporters, who of course were already there before the operation, are crucial. So a catchment area for physical and mental problems in the immediate family and circle of friends is extremely important." (14C)* *"I think the family should be behind it because the transplant and everything around it does not stop with the transplant, it continues throughout life. (..) I think that it makes the whole situation and the whole project easier when the partner the family, or the parents are behind it and participate, because that promotes compliance." (15C)*
Programmatic factors/influences		
Clinical resources	Clinical resources and expertise are fundamental to a successful hand transplantation program	*"The first important thing is specialist knowledge, i.e., I have to know the literature, what are the others doing, what hardware do I need. Between the lines, I need to go where there is a lot of transplanting and talk to other people." (16C)* *"The technical know-how alone is not enough. I also need resources, I need a structured program, I need team players so that it can work." (17C)*
Public perception	Public perception of hand transplantation can influence both the patients and providers	*"But they always think [hand transplant is] great. The public thinks that's great. (..) It's also simply fascinating." (18C)* *"Before the transplant, your brother received attention in a negative sense, so you have lost something, you are handicapped, you are limited, you look different. And through the hand transplant, you get attention from the outside, but weighted more positively, in the sense that something special has been done." (19C)* *"We doctors also make a lot of mistakes, (..) [but instead of reflecting them] we try to surpass each other with the most amazing and spectacular operations. (..) This also provokes a certain reaction and fear in society. If this becomes a routine procedure, (..) it will no longer be so sensational and you can no longer satisfy the media with it, but it is more of a reassurance for the patients. That's why I'm a fan of standardization." (20C)*
Post-transplant factors and outcomes		

(Continued)

TABLE 1 (Continued)

Categories	Themes	Sample of supporting text from transcripts
Rehabilitation	Rehabilitation is a lifetime commitment after hand transplantation	*"You have to mention again and again that this is a long road that can be paved with complications. (21C)* *"The most important thing is good physiotherapy and rehabilitation. (22C)* *He still reports improvement. That has never stagnated. (..) I know that the patient was always motivated to work with his hands." (23C)* *"Not a year or two years. You have to work a lot. You have to know that it will be a very difficult road, hard work." (5R)* *"This was a patient, who had a high level of activities of daily living (ADLs), and that is also important. And this is also important for the rehabilitation phase." (24C)* *"In the beginning, I think it's normal to make huge moving progress. If you follow the measurements of physical therapy or occupational therapy, there are slight but measurable noticeable improvements every year. In the beginning, the successes were great, big steps, but also changes are apparent year after year, even today. Improvements can be seen in terms of strength, grip, feeling, warmth and perception of cold." (25C)*
Follow-up and care	Intense provider involvement in post-transplant follow-up and care increases the likelihood of success	*"You have to be able to work with the patients, in the sense that they have to enter into a partnership with the doctor who treats them. (..) You need even more trust than usual in doctor-patient relationships. The transplant patients have to report quickly if something does not fit. The doctor must be available. Such a patient is a task that requires a team. That demands a lot from the medical staff. If you are not willing to do this, you will not get good results." (…) (26C)* *It takes someone willing to deal with these patients 24 h a day, 7 days a week, 365 days a year. (..) If these patients have a problem, it can very quickly end in a downward spiral." (27C)* *"I find the support to be very, very time-consuming. The patients require an extremely large amount of time and effort (..)You go on vacation and then you get the messages and the phone call while you are on vacation. (..) that is very time-consuming. Because no finding should be overlooked or forgotten." (28C)*
Adherence and training	Adherence and training are big predictors for the success	*"You have to trust the doctors 100% and do everything the doctors say. No fantasies of your own, the doctors said 5 mg, that means 5 mg." (6R)* *"I train, I do physiotherapy, they work with me and even if it hurts, nothing happens for a long time, there comes a crucial point and a lot of things get better." (7R)* *The functionality is different. It's very related to what you do with your hands and how much you train them." (29C)*
Graft integration	Graft integration characterizes a successful transplantation	*"A successful transplant is when the patient accepts their transplant. (..) That is the first step and the second is when you are ready to deal with the transplant." (30C)* *"These are my new hands and with these new hands I will continue my new life." (8R)*

Pre-transplant factors and preparation

Numerous other studies have supported the importance of patient selection in optimizing patient outcomes (Brau and Clarke, 2006; Kinsley et al., 2021, 2022). Often overlooked are two key points raised by our patients and teams. Firstly, patient selection is a dynamic not a static process. Longitudinal evaluation and ongoing optimization of a potential candidate's psychosocial circumstances is fundamental. For example, patients with a history of substance abuse can be appropriately counseled and supported peri-operatively to help them recover without relapse. Secondly, providers have an important role in optimizing patients' outcomes, ensuring that preparation and support is adequate

In two separate studies conducted in the US, Kinsley et al. interviewed UE VCA patients, primary caregivers, and healthcare providers to evaluate perceived predictors of transplant access. These included realistic expectations of life after transplantation, strong social support, and positive framing of one's situation. A deep desire for limbs or an unrealistic expectation of transplant function can both pose a major barrier to accepting a limb transplant that may be imperfect despite intensive rehabilitation and side effects from lifelong immunosuppression. Patients relied heavily on their caregivers and health providers for both physical and emotional support, while expressing the desire to communicate with other transplant recipients to better set expectations.

Programmatic factors and influences

Programmatic factors have proven challenging for almost all teams globally (Gordon et al., 2009; Gordon and Siemionow, 2009; Kinsley et al., 2021). There are a small number of patients with bilateral upper-extremity amputations who are ready medically and psychosocially for this major intervention. Matching patients who are optimized with teams able to provide the complex care necessary is an ongoing challenge (Gordon et al., 2009; Gordon and Siemionow, 2009; Siemionow and Gordon, 2010; Kumnig et al., 2012, 2013, 2014; Kumnig and Jowsey-Gregoire, 2016; Kinsley et al., 2021).

These large teams also have their own interdisciplinary challenges. We have found that while teamwork is one of the most rewarding aspects of VCA, it can also be one of the most difficult parts. Groups such as the International Society for Vascularized Composite Allotransplantation, the American Society for Reconstructive Transplantation, and the Chauvet Workgroup, all provide collaboration internationally to help educate ourselves within this small field (Jowsey-Gregoire et al., 2016; Kumnig et al., 2022). Furthermore, the International Registry on Hand and Composite Tissue Transplantation provides a superb repository of data that can further encourage collaboration (Kumnig et al., 2022).

In the US, transplant recipients have emphasized how critical it is to have timely access to a dedicated medical care team for long-term wellbeing (Brau and Clarke, 2006). Geographic barriers pose significant logistical challenges for ongoing care, particularly when compounded on financial and compliance issues. These factors are important for healthcare providers during preoperative discussions about continuity of care. Despite the expertise of multidisciplinary programs, providers struggle with setting realistic expectations of rehabilitation and recovery and predicting recipient compliance.

Post-transplant factors and outcomes

Post-transplantation care is often focused on medication adherence and physical rehabilitation. In our study, we notice the importance of psychosocial support in the follow-up. Rehabilitation is a lifelong commitment after UE VCA, with ongoing steady improvements in sensorimotor function continuing years after transplantation in self-motivated patients who continue their physiotherapy regimens. Maintaining a close relationship between transplant recipients and healthcare providers, although time consuming and demanding, may be necessary to prevent complications, promote adherence to immunosuppression despite adverse effects, and maximize overall functional success.

Similar findings were noted by Kinsley et al. (Brau and Clarke, 2006) in the US. The “intimate” and “special” relationship with the caregiver team was described by some patients as crucial for their practical and emotional needs, particularly during times of feeling alienated. Patients recognized their dependence and the sacrifices they may place on caregivers and care providers, and this recognition may serve as a motivating factor to maximize their independence. The desire for more involved psychological evaluation and therapy was also expressed, congruent with testaments of resilience, positive attitude, purposeful rehabilitation, and strong social supports being favorable psychosocial factors for a good functional outcome.

Limitations and proposed directions for future research

Future research efforts that are directed at sharing similar evaluation strategies across centers are needed to establish universal guidelines, pathways, and assessments for candidate evaluation and recipient evaluation (Dew et al., 2007; Kumnig et al., 2014). Another important component of interdisciplinary screening should be the identification of at-risk candidates. Intervention strategies to assist these candidates might then lead them to be eligible for this treatment and might especially be beneficial in supporting their ability to succeed with medication adherence and overall QOL post-transplantation (Kumnig et al., 2012, 2014; Kumnig and Jowsey-Gregoire, 2016).

The citations in Table 1 are more originating from VCA healthcare professionals compared to VCA patient families. However, the number of quotations from each group is proportional to the number of study participants from each group: of 12 participants in our study, 4 UE VCA patients, 7 were healthcare professionals, and only 1 was a patient family member. Our clinical observations show that data from patient families are difficult to collect and thus of particular value to the field, but we want Table 1 to be reflective of our actual data pool. We will certainly endeavor to continue collecting qualitative data from patient families in future studies.

Conclusion

Psychosocial factors are important elements in the assessment and follow-up care for UE VCA. To best capture psychosocial elements of care, protocols must be individualized, patient-centered, and interdisciplinary. Recent research has shown that proposed directions for future research should particularly focus on adherence, training, and close relationship with healthcare providers in the pre-and post-transplant course. The importance of psychosocial factors cannot be overlooked when assessing prospective UE VCA patients preoperatively and optimizing recovery and functional rehabilitation postoperatively. As with all QoL interventions, patients’ subjective experiences are relevant to assessing whether an intervention achieves its aim. Investigating psychosocial predictors and collecting outcomes is, thus, critical to justifying UE VCA as a medical intervention and to providing accurate and salient information to candidates considering the procedure.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Ethics statement

The studies involving human participants were reviewed and approved by the Ethics Committee of the Medical University of

Innsbruck (vote nr.1044/2020). The patients/participants provided their written informed consent to participate in this study.

Author contributions

NH: writing the manuscript, performance of the research, and contributed new insights. KZ : writing and editing the manuscript. ST, SK, and MK: participated in research design, wrote the manuscript, performance of the research, and contributed new insights. ZZ: participated in research design and contributed important insights to perform the research and to wrote this manuscript. JK: contributed important insights to perform the research and wrote this manuscript. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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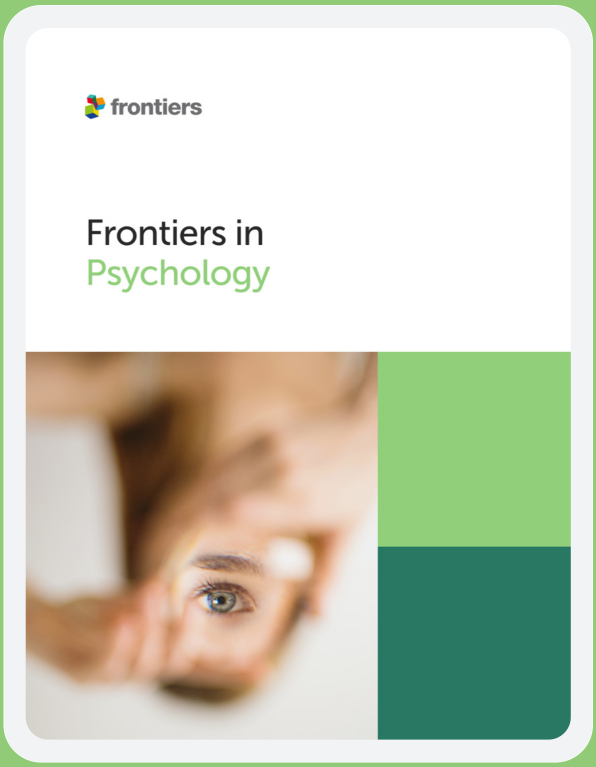
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