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Conceptualizing Quality of Life in Reconstructive Transplantation Ethics

From the Editor

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LETTER FROM THE EDITOR

Assessing Quality of Life in Reconstructive Transplantation

Emily Herrington, PhD, MA and Jessica Benham, MA

When we met as graduate students at the University of Pittsburgh, both of us pursuing an MA in bioethics concurrently with a PhD in communication, we often found ourselves together in courses and colloquia. As mutual concerns and interests arose through classroom encounters, we recognized that many of our most dissatisfying—and often most heated—discussions with peers and professors circled around questions of what constitutes a particular quality of life (QoL) or health, from whose perspective QoL or health is assessed, and what values or measurements QoL or health is based on. We recall several early seminar-style conversations about disability and quality of life in which the second author (J.B.) would ask of our readings and of the group, "Where are the patient perspectives? Where are the voices of people with this condition?" In communication studies and in bioethics—but especially in bioethics—the insights of persons affected by a condition or a medical intervention were in many cases elided, replaced by less descriptive, "sanitized" medical terminology or ruled out by academic research journals' preference for objective, quantitative data.

For this theme issue of the *AMA Journal of Ethics*, we wanted to open a multidisciplinary conversation on the related issues of QoL, representation of outcomes, and knowledge generation in vascularized composite allotransplantation (VCA) using a wide range of empirical and conceptual tools. We have been gratified in our efforts to bring together voices from bioethics, surgery, psychiatry and behavioral health, disability studies, communication and rhetoric, religious studies, and the VCA patient population (in the form of oral history data collected by the first author, E.H.).[1] We believe the convergence of diverse viewpoints on the multifaceted nature of VCA bioethics is useful both practically (for generating productive discourse) and symbolically (for underscoring the multiple points of view that can be relevant if not critical to a holistic bioethics of VCA).

VCA, sometimes called reconstructive transplantation—which includes transplants of the hand, face, penis, larynx, and uterus—is primarily conducted to improve a patient's quality of life. Yet ethical inquiry into QoL aspects of VCA is lacking, even though it is relevant to topics such as patients' lived outcomes, the impact of social acceptance or stigma on recipients' experiences of physical disfigurement or functioning, and the effects of disparate access to personal and financial support on a person's candidacy for VCA. For one thing, as authors in this theme issue point out, developing "objective" scientific assessments of QoL in the field of VCA faces challenges, including low numbers of geographically isolated patients (fewer than

200 VCA surgeries have been performed worldwide as of 2015[2]) and the complicated, long-term nature of these interventions. For another, the patient's QoL is not the priority of every stakeholder. As discussed in one of the essays that follows, although it may seem "intuitive and self-evident that VCA candidates' and recipients' QoL and autonomy should be priorities in decisions about engaging in or disengaging from experimental surgery," competing interests can arise when the goals of medical researchers, institutions, and patients differ and are not recognized openly (eg, patients harboring undisclosed restoration fantasies or professionals or programs targeting scientific endpoints).

For all of these reasons, in cases in which patients have experienced burdensome side effects or adverse results in the long postoperative phase of the VCA process, which lasts the rest of the recipient's life or as long as the allograft is retained, VCA surgeons must navigate difficult decisions about whether and how to share unfolding information about participants' QoL with their peers and the public. For instance, should researchers publish data that are measurable by existing tools for assessing QoL, or would narrative methods be more useful for understanding how possible complications played out in the course of treatment? Because VCA has intensive postoperative requirements (eg, immunosuppression, rehabilitation) and a lifelong risk burden for participants, several contributors to this issue argue that narrative data from patients receiving these treatments can be germane to understanding how well or poorly treatment protocols promoted the health and QoL of VCA patients.

The second author's background as a member of a vulnerable group and as a researcher who works almost exclusively with narratives from marginalized populations proved valuable in informing the development of this issue. The small number of VCA patients is significant not only because this paucity makes quantifying outcomes problematic (highlighting the importance of individual narratives) but also because the medical research context privileges perspectives of surgeons. And the smaller is the number of people receiving treatment, the less likely they are to be able to find each other and share their stories in spaces unmarked by the language, power, or expectations of medical researchers. As feminist bioethicist and disability scholar Margrit Shildrick and collaborators have written, "transplant professionals need to question the limits of what is seen as unproblematically therapeutic, and to look beyond conventional data."[3]

Taken together, the essays in this theme issue comprise a unique and timely collection of perspectives on VCA bioethics; considered individually, they address perennial challenges to authentic representation of vulnerable others that are germane to other areas of research and health care ethics. It is our sincere hope that, in addition to contributing to the literature on VCA ethics and medical ethics, this theme issue of the *AMA Journal of Ethics* will draw attention to why quality of life is an area of inquiry that can help us both challenge our assumptions about what data count in a particular setting and reorient studies of bioethics and medical practice to the importance of subjective, contextualized narrative data.

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CASE AND COMMENTARY

In Experimental Hand Transplantation, Whose Views About Outcomes Should Matter Most?

Andrea DiMartini, MD and Mary Amanda Dew, PhD

Abstract

Consent to any experimental procedure, even when offered as therapeutic, involves extensive discussion between patient-subjects and clinician-researchers. Decision making should be shared with a focus on potential risks and benefits of enrolling in a protocol. Just as patients who underwent nonexperimental interventions might experience regret or reconsider autonomously made choices, patient-subjects who are undergoing or who have undergone experimental therapies should be afforded latitude to reconsider their decisions. Although clinician-researchers tend to be deeply invested in gathering data about patient-subjects' experiences, they are obligated to express respect for patient-subjects' fundamental right to stop being enrolled in research.

Case

After losing his hand in an industrial accident, R sees a television special about a hand transplant recipient who regained the ability to type. He inquires about the experimental surgery, undergoes multiple evaluations, and learns about potential risks and benefits. Although the surgeon stresses limitations of the transplanted hand, which might never regain full strength and sensibility, R hopes his outcome will allow him to return to work.

Two years later, after extensive rehabilitation, R is disappointed with the graft's functioning. His employer does not feel he can safely return to work and advises him to take permanent disability. R is frustrated with other aspects of his posttransplant quality of life: he has dietary restrictions, medications that cause nausea, a directive to avoid crowds, and he does not enjoy many outdoor activities due to the extra care he must take with his graft. He is tired of regular appointments and tests and of his surgeon-researcher's surveillance of his progress.

When he asks about surgical removal of his hand, his surgeon is surprised and says, "Your function is well above what we anticipated and you have had few complications. I don't recommend amputation. You would have to endure another surgery and recovery before being outfitted with a prosthesis. This has been a huge investment for you. What you've got now is probably the best you can get and it would be unlikely you would get a second chance. However, the side effects and risks of your medications—including kidney damage, infections, and cancers—should be considered."

R says, "I definitely can do things I couldn't do with my prosthetic. But I don't think a hand transplant is right for me in the long run. What I have to do to take care of the hand interferes too much with how I want to live my life. All things considered, I think it was better for me before." The surgeon thanks R for explaining his concerns and suggests they take some time to think things over.

Commentary

Vascular composite allograft (VCA) transplantation is an emerging, still-experimental field in transplantation. VCA transplants can restore function and appearance to patients with severe injuries, disfigurement, and malformations. Most VCAs are upper limb or hand transplants, although craniofacial, uterus, penile, and lower limb transplants have been performed in the United States and worldwide.[1] Since 2014, 39 VCA surgeries have been conducted in the United States,[2,3] with 11 VCA transplants in 2018 alone.[3]

What makes VCA transplantation clinically and ethically different from most types of solid organ transplantation is that VCA transplants are intended to be life enhancing, not lifesaving. In consequence, VCA decision making requires more intense focus on quality of life (QoL) than on extending life. Specifically, as an experimental elective surgery, VCA transplantation requires greater consideration of risks and potential impacts on recipients' QoL because VCA recipients require intensive posttransplant rehabilitation, integration of many self-care tasks into their daily living, and meticulous adherence to lifelong immunosuppression medication regimens that might undermine their QoL. Some transplant recipients, like R in this case, might find requirements like these to be too burdensome or not what they expected. With the exception of craniofacial transplantation, for which graft removal is less feasible, VCA transplants provide unique opportunities in experimental surgery to consider when and which exit strategies should be developed for patients.

Clinical and surgical researchers obviously have a stake in the success of VCA transplantation. Opportunities to improve the functional status and QoL of persons with disabilities drives their desire to explore innovative, cutting-edge advances. However, VCA researchers' early declarations that "functional outcomes exceeded expectations" and that "VCA recipients enjoy a quality of life" unattainable with conventional reconstructive surgery were based on follow-up of fewer than 100 cases[4]; substantial quantitative data on either short or long-term outcomes is lacking.[5] Thus, the spectrum of possible outcomes will not be fully realized until the experimental procedure is well developed.

The uncertainty of outcomes creates important ethical considerations for VCA transplantation. How should clinical researchers maintain equipoise[6] when offering an experimental procedure with an uncertain outcome? How should candidates considering experimental VCA procedures think about their preferences, evaluate unknown risks, and weigh their hopes for improved function and QOL against these risks? Importantly, having participated in an experimental procedure, how should candidates, recipients, and clinical researchers consider exit strategies, including explantation?

Informed Consent to Experimental Surgery

Appropriate expectations for outcomes, such as physical functioning and QoL, begin with thorough explanation and discussion of a proposed procedure, its potential risks and benefits, recovery, and patient-subject responsibilities and self-care. Informed consent requires that a patient-subject have good comprehension of potential risks and benefits and be capable of voluntary decision making. To be informed, a patient-subject also needs time to understand and process complex information and to reflect on risks and benefits relative to his or her personal preferences. To ensure shared decision making, a researcher must not only disclose all information known about the experimental procedure but also consider a patient-subject's unique values, preferences, and expectations when making a recommendation.[7]

The experimental nature of VCA transplantation can complicate informed consent. The elective nature of plastic surgery necessitates disclosure of realistic odds of obtaining desired results[7] and appropriate management of expectations—especially in experimental VCA transplantation, given its substantially heightened high risks and uncertain results. As surgical experience with VCA outcomes grows, however, the risk-benefit ratio will likely change—hopefully in favor of benefits to patient-subjects—which would affect informed consent discussions. Moreover, to fully inform a potential VCA candidate, a clinical researcher should provide information about all currently known outcomes—not just optimal ones[7]—and their likelihood.[7] Because the number of VCAs performed so far is small and each case is unique, the procedure's experimental nature and possible unknown (and potentially undesirable) outcomes should be emphasized.

Informed consent to experimental VCA can also be complicated by how VCA transplantation is covered by the media. Perhaps unsurprisingly, amazing and courageous stories of VCA recipients and their surgical teams tend to attract media coverage,[8] which can influence the public's and potential VCA patient-subjects' perceptions of the procedure. As with many new interventions, positive media coverage has potential to benefit researchers' careers and their institutions and should be recognized as a possible conflict of interest. Additionally, positive coverage focusing on VCA recipients with the best outcomes may lead to VCA candidates' misunderstanding of surgical risks and outcomes, thereby compromising informed consent and respect for autonomy.[7]

In experimental VCA transplantation, patient-subjects' motives can powerfully influence their decision to proceed; this power should not be underestimated. Some patient-subjects harbor undisclosed fantasies about complete restoration of functioning or cosmesis or expect that, among possible outcomes, theirs will be optimal. Despite some patient-subjects' apparent willingness to accept a less-than-optimal outcome, they really might only be prepared to accept an optimal outcome. Furthermore, it can be difficult for some to fully comprehend and evaluate future demands of postoperative care, rehabilitation, medication regimens, laboratory and procedure monitoring, restrictions and limitations, and daily self-care tasks. When faced with daily realities of these activities and demands, patient-subjects—such as the one in the case—might find them unacceptable over time and feel that their prior QoL, while not optimal, was preferable to their QoL with a VCA transplant. In the case, for example, R continued to hope unrealistically for complete restoration of function and found the demands of postsurgical care too burdensome. Ultimately, his expectations of benefit were not realized and his QoL diminished.

Respect for Autonomy

VCA researchers and subjects must accept that when actual outcomes are not satisfactory to VCA recipients and accommodations cannot be made to improve them, then VCA recipients should be able to terminate postsurgical interventions and request graft explantation. Discussion of such exit strategies and their possible risks and benefits should be part of informed consent prior to surgery or informed refusal after surgery. Prior to experimental VCA transplantation, patient-subjects should be made aware that graft removal could be recommended by the research team. Circumstances that would possibly or definitely require graft explantation should therefore be discussed. Likewise, circumstances for which explantation is not possible should be considered by the research team and discussed with a VCA candidate.

In the case, a clinical researcher should inform R that explantation risks could include those related to the surgery itself, extended recovery, difficulty in fitting a new prosthetic, compromised functionality relative to presurgery functionality, and inability to be considered for retransplantation. Importantly, the researcher in the case reviewed possible benefits of explantation, including termination of chronic immunosuppression medications with their significant risks.[9] Providing time for R or any VCA recipient to process and reflect on this information should be allowed to ensure that the patient-subject's ultimate decision is not impulsive. The patient-subject's perceived QoL and perceived deviations from expected QoL after surgery are individual, subjective, and worthy of respect.

It might seem intuitive and self-evident that VCA candidates' and recipients' QoL and autonomy should be priorities in decisions about engaging in or disengaging from experimental surgery. Patient-subjects might decide that their outcomes did not meet their expectations, or they might change their minds about how acceptable specific outcomes are after they experience them. However, a researcher might believe that explantation would introduce new risks and harms and be inclined to strongly recommend against it. The researcher must weigh this potential recommendation against potential conflicts of interest when he or she is highly invested in developing a novel surgical technology or has concerns about the impact of a poor outcome on a study. Would such an outcome negatively affect the continuation of the research, cause early termination of a research protocol, or lead to greater oversight? Given the substantial individual and institutional investment of time and resources in experimental VCA, it can be difficult—though it is essential—for clinical researchers to be mindful of their own hopes when discussing risks and benefits with VCA candidates or recipients.

Future Considerations

There are several ways to better prepare VCA candidates and to reduce the likelihood of their being dissatisfied with their outcome. Importantly, the risks and benefits of explantation should be emphasized during the informed consent process. Additionally, psychological counseling—conducted independently of the VCA team to allow candidates to reflect on their decision and prepare for and adapt to the demands of VCA transplantation—might improve satisfaction and acceptance of outcomes. Opportunities for VCA candidates to speak with VCA recipients who have had a range of positive and negative outcomes could also help inform their decision. Additionally, similar to policy for living donor transplantation programs,[10] independent advocates could help evaluate VCA candidates' understanding of the procedure's risks and benefits and help temper clinician-researchers' influence on candidates' decisions. As VCA experimental surgery evolves, inclusive approaches will be needed to safeguard candidates' and recipients' autonomy and optimize their QoL outcomes.

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The case to which this commentary is a response was developed by the editorial staff**.**

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CASE AND COMMENTARY

Should a Caregiver's QoL Be Considered in Decisions About Whether a Patient Has an Experimental Double-Hand Transplant?

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Abstract
A goal of hand and upper extremity transplantation is to return motor and sensory function to an amputee. Given the integral roles of one's hands in activities of daily living and social interaction, however, restoring psychosocial well-being should also be a priority. Based on the authors' experience, double-hand transplantation success depends significantly on strong social support, physical rehabilitation, medication adherence, and social integration. Because caregiving is demanding, tasks should be distributed among members of a patient's family and social network. This article analyzes how to respond to an overwhelmed caregiver by drawing on solid organ transplant literature about caregiver fatigue.

Case

C is a quadrilateral amputee who lives with her partner, N. After 2 years of working with prosthetics and outfitting their apartment with assistive devices, N and C have a life together they enjoy.

When C is offered an opportunity to be evaluated for an experimental double-hand transplant by Dr PT, her long-time physical therapist (who is working with a newly formed research team at their hospital), N struggles with the idea of a radical change in her own and C's living situation. Despite N's devotion to C, she is worried about how she would manage additional caretaking responsibilities if an experimental surgery does not turn out well for C. N agrees with C, however, that having C be evaluated would give them an opportunity to consider possible benefits and risks.

During C's evaluation to consider whether she meets the criteria for enrolling in the double-hand transplant research protocol, N tries to gain information about possible effects that a double-hand transplant would have on her own quality of life without seeming self-absorbed or unsupportive of C's enthusiasm about the protocol. N asks, "What will C's recovery process be like?"

Dr B, the study's principal investigator, responds, "I wish I could tell you it's going to be quick and easy. But as you can probably imagine, it will likely be neither. In above-elbow amputees, like C, we expect nerve regeneration to take 1½ to 2 years.[1] Nerves grow at a rate of about a millimeter per day. So, in the first year after the transplant surgery, C will need help with almost all aspects of self-care and rehabilitative therapy."

N struggles to keep her expression neutral and optimistic, but she is distressed by this information. The transplant team's physical therapist, Dr PT, who is helping to facilitate the conversation, notices N's distress as the team concludes the evaluation.

Dr PT remarks to Dr B later, "N was clearly upset and seemed overwhelmed, and who wouldn't be. I wish we had brought N into the conversation more about whether hand transplant is the right choice for C and for them as a couple. Highly involved partners and family members provide critical support to our study participants' abilities to adhere to postsurgical care recommendations. They'll both need a lot of support in the follow-up years if C participates in our study. It seems we should somehow be incorporating the caregiver more in our evaluations. How should we do that?"

Commentary

Hand and upper extremity transplantation restores form and function to bilateral amputees,[1] with recipients experiencing good motor and sensory outcomes 6 or more years posttransplant.[2,3] It persists as the most common type of vascularized composite allotransplant. More than 100 such transplants had been reported worldwide by 2018.[4] Although patient survivability surpasses 95% at 10 years,[5] unlike solid organ transplantation, this procedure's primary aim is not to extend life in relatively ill patients but to improve quality of life in relatively healthy patients.[6,7,8,9,10] For both kinds of transplants, life-long immunosuppression, which is necessary to prevent graft rejection or loss, increases the risk of developing oncologic malignancy and metabolic disorders like diabetes mellitus and heightens susceptibility to infections.[11] Nevertheless, acute allograft rejection is nearly assured within the first year in 4 of 5 hand transplant recipients, although it is reversible when treated promptly.[12] Although some candidates might be informed of these statistics during preoperative consultation and informed consent processes,[13] evidence is limited of patients' and caregivers' understanding of, expectations for, and experiences of surgery and its postoperative demands.

Efforts are being made to study hand and upper extremity transplant recipients and caregivers outside of laboratory settings.[14] Herrington et al explored the narratives of caregivers who often felt burdened by demands of providing care,[15] despite their being crucial for long-term patient well-being and allograft survival.[16] Caregiving imposes physical and psychosocial demands on caregivers that affect their health and well-being sufficiently to impact the care they provide.[17,18] This caregiver burden is amplified in nonprofessional caregivers who are often untrained and unprepared to perform skilled medical tasks.[19] Given these potential burdens, should finding a balance between a patient's need and desire for bilateral hand transplantation and a nonprofessional caregiver's well-being become the responsibility of the treatment team?

With little written about caregivers in hand transplantation, our aim as members of a reconstructive transplant team is to reflect on that question and share insights from our 10 years of experience. In this case commentary, we elucidate the unique role of nonprofessional caregivers, such as N, in hand transplantation. We then highlight positive behavioral characteristics of potential caregivers and concerning red flags. Finally, we provide suggestions to improve the assessment of caregiver burden and its prevention.

Understanding the Caregiver Role

In this case, N has limited experience in providing medical care as she is not a professional caregiver, such as a physician or a visiting nurse. Yet after 2 years of working with prosthetics and assistive devices, N has managed to balance her caregiving and personal responsibilities to develop an enjoyable life with C. Now that C is considering bilateral hand transplantation to meet her individual treatment goals, which for many candidates involve complex considerations and decisions,[20] N realizes her role as a nonprofessional caregiver could change.

Nonprofessional caregiving for prosthetic users and hand transplant recipients differs in several respects. If needed at all, nonprofessional caregiving for prosthetic users is of much shorter duration than the 1½ to 2 years required for bilateral hand transplant,[1] and it presents caregivers with different physical and financial responsibilities than would a bilateral hand transplant.[21] In addition, hand transplant patients' morbidity, mortality, and degree of social reintegration are dependent on the nonprofessional caregivers' long-term commitment and social support.[16]

After learning the risks of the surgery, that roughly 17% of unilateral and bilateral hand transplant recipients suffer graft loss,[22] and that additional caretaking responsibilities would be required if everything were to go well, N realizes the impact that the transplant could have on her well-being. Nerve regeneration might prolong the need for aiding C with activities of daily living.[23,24] This possibility, combined with the strenuous rehabilitative regimen,[25] would hinder N's ability to participate in social activities—potentially straining her relationship with C.[26] Given these burdens, consultation must consider the needs of nonprofessional caregivers like N to be an extension of the patient's needs via their social relationship.

Positive Caregiver Characteristics and Red Flags

No quantifiable or standardized assessment of caregivers exists for hand transplantation. Instead, our treatment team discusses whether caregivers can fulfill the obligations of their role, including assisting in medical management that frequently changes after tailoring the immunosuppression regimen, performing wound care and occupational therapy at home, providing psychosocial support and a second set of ears during hospital visits, monitoring the health of the allograft, and assisting with activities of daily living such as transfers, bathing, cooking, cleaning, and hygiene. These tasks might overwhelm a nonprofessional caregiver who has other life and work responsibilities. However, the burden of these tasks can be mitigated by sharing caregiving responsibilities among a group of nonprofessional caregivers working as a caregiving team.[27]

Based on our experience, positive characteristics of nonprofessional caregivers include

- Willingness to be present, to learn, and to help the hand transplant recipient.
- Dutifully executing the tasks listed above.
- Demonstrating the stability and resilience needed to focus on the patient's needs.
- Clearly expressing that one's obligation is to the patient, not the patient's team of professional caregivers (ie, the transplant team).
- Honesty or warranted trust in the treatment team.

A caregiver of one of our patients demonstrated these characteristics by taking a leave of absence from work to provide care for her spouse. This unfaltering support continued throughout the first 2 years posttransplant, a time when poor caregiver support has been shown to increase medical nonadherence in cardiothoracic transplantation.[28] Once the rehabilitative routine eased up, the caregiver returned to work and integrated her new caregiving responsibilities into her daily routine, remaining an excellent caregiver to her spouse. The strong social relationship between patient and caregiver contributed to the overall success of our case as well as to the couple's alacrity in forming a trusting relationship with our treatment team; a trusting patient-clinician relationship has been linked with improved pain control, functionality, and mental health for knee replacement recipients.[29] In N's case, Dr PT recognizes that early consideration of the patient-caregiver unit would build the rapport needed to ascertain the appropriateness of hand transplantation for both N and C.

In contrast, proceeding with transplantation when the behavior of the patient's nonprofessional caregiver puts up red flags could jeopardize the postoperative health of the recipient and the allograft. From our experience, concerning red flags include

- Impeding therapy or consultation.
- Financial impoverishment to a degree that would hinder postoperative care.

- Living geographically distant from the recipient without plans to relocate after transplant.
- A general lack of preparedness.
- Dishonesty or an unwarranted distrust of the treatment team.

These red flags factored into a decision at our institution to deny candidacy, despite the patient's meeting medical necessity guidelines for transplantation.[30] The couple showed general lack of preparedness for temporarily relocating and maintaining financial stability for themselves and their young children. This behavior conflicted with our duty to treat. Related to the lack of preparedness, the couple's expectations of financial security from medical celebrity after hand transplantation were unfounded. Additionally, the caregiver would seldom accompany her partner to consults, but, when present, the caregiver perseverated on the inability to find a donor—a not uncommon happening[31]—and often became argumentative. Differences between the nonprofessional caregiver and the treatment teams impeded the development of trusting relationships, which led to our institution's declining this patient's candidacy for transplant. If nonprofessional caregivers like N seem unwilling to speak up, treatment teams should consider scheduling appointments specifically with those caregivers to better assess their ability to be a supportive team player. Transplant teams that do not secure reliable support for patients like C risk unsafe hand transplantation.

Our method of assessment is not perfect. One particular case at our institution blindsided the treatment team the day of transplantation. The patient and caregiver came to appointments together over months of pretransplant consultations, despite living far away, as many reconstructive transplant patients do.[32] The nonprofessional caregiver demonstrated willingness to provide supportive care and showed no indication of an inability to cope with the burdens of transplantation. On the day of the surgery, however, the caregiver arrived intoxicated and incoherent. After 3 days of similar behavior, it became evident that the caregiver would be unable to fulfill the obligations demanded of the role, which speaks to the difficulty of ascertaining coping styles in caregivers.[33] Another member of the patient's nonprofessional caregiving team was contacted and came immediately to meet with the patient and treatment team, eager to learn what would be helpful in assuming the role of primary caregiver. Fortunately for this patient, the caregiving transition did not negatively impact the postoperative course.

Significance of Caregiver Burden and Suggestions for Prevention

Given nonprofessional caregivers' significant impact on graft survival and overall patient well-being and their crucial rehabilitative role outside of the hospital,[16] a moral obligation exists to include caregivers and their well-being in considerations of a patient's candidacy for hand and upper extremity transplantation. It would disrespect the transplant recipient's autonomous desire to undergo a successful rehabilitation if the treatment team neglected to provide forward-thinking guidance to the nonprofessional caregiving team. Furthermore, it could undermine the recipient's attempts to regain personal independence and self-identity if caregiver burden is not addressed. Besides being a sound choice, caring for the burdened caregiver is upheld by our professional medical ethos.[34] As such, understanding the ability of caregivers to appraise their experiences, to access resources or support, and to care for themselves aids treatment teams in recognizing and affirming their vital role in supporting transplant recipients' physical, psychological, social, and spiritual well-being.[35]

Specific assessment for, and early detection of, caregiver burden hinges on serial evaluations with members of the treatment team and on implementation of evidence-based interventions.[36] Implementing the following recommendations would help determine an individual's ability to be a reliable nonprofessional caregiver to a hand transplant recipient.

1. *Completion of a social contract*. In lieu of a medicolegal informed consent form, a social contract could be completed by nonprofessional caregivers. Such a document is not legally binding but articulates nonprofessional caregivers' social bond with and obligations to their friend or loved one. A failure to complete the form would be a red flag to the transplant team signaling the individual's unwillingness to take on caregiver responsibilities.

2. *Expanding in-depth psychosocial assessments*. Psychosocial assessments could be expanded to include nonprofessional caregivers in preoperative consultation, with a particular focus on coping abilities. Implementing such an in-depth psychosocial evaluation might improve our approach and could assist in determining the coping abilities of nonprofessional caregivers like N.

3. *Development and adoption of a hand transplant-specific tool.* Similar to the Family Caregiver Activation in Transitions tool,[37] a hand-transplant-specific tool could guide interventions intended to enhance caregiver preparation and confidence during care transitions. For caregivers like N, this tool could highlight areas for which another nonprofessional caregiver or the transplant team could provide additional support.

Successful Nonprofessional Caregiving

Anecdotal evidence from our experience is consistent with the literature and suggests that the success of hand and upper extremity transplantation depends significantly on strong social support from a nonprofessional caregiving team that assists with physical rehabilitation, medication adherence, and social integration. In consulting with individuals like N and C, program-specific treatment teams like ours should remain cognizant of the burden to individuals like N of becoming a nonprofessional caregiver to a hand transplant recipient. Understanding caregivers' circumstances can only support our aim of providing excellent care and necessary expertise in reconstructive transplantation from preoperative consultation through individualized postoperative rehabilitation.

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CASE AND COMMENTARY

How Should Surgeons Balance Transplantation Innovation With Acceptance of a Trauma Survivor's Appearance?

Carly Parnitzke Smith, PhD

Abstract

Clinical and ethical issues involved in counseling a patient about reconstructive surgery for a traumatic and disfiguring injury require special consideration. This article proposes prioritizing 2 considerations: (1) the influence of traumatic experiences on a survivor's cognitive processes and (2) insights into a survivor's acceptance of his or her posttrauma appearance or consent to high-risk or experimental surgery, which can be gained from dialectical behavior therapy. This article argues that these priorities should be explicitly discussed by plastic surgeons counseling patients whose appearances are altered by trauma.

Case

About 40% of Dan's face was burned in an accident many years ago. Several sites on Dan's body have also been scarred by skin-harvesting from numerous reconstructive surgeries. Although many years have passed since the accident, Dan still suffers long-term grief and feels profoundly depressed about not having a romantic partner. Dan also feels hopelessness and sadness about his surgeons having said they've exhausted traditional reconstructive options on his face. Specifically, Dan remembers his plastic surgeon once stating, "There's nothing else we can do" to improve appearance, ability to speak, or ability to eat easily.

Dan's feelings of grief, longing, hopelessness, and sadness are made even more complex by his feeling guilty about not being able to just accept his face as it is. Dan feels solidarity with other burn survivors, whom he meets occasionally at conferences and support groups, and he feels it is important to resist cultural and social pressure both to medicalize his survivorship more than necessary and to try to meet unrealistic standards of "normal" physical appearance. He feels torn between wanting to accept his appearance as it is and wanting it to be good enough for a prospective romantic partner to find attractive.

Dan has read about face transplantation in various online news sources, and he now researches the procedure with more interest. He knows that whatever aesthetic, manual, and social enhancement he could gain through a face graft would come with high costs, substantial risks, and demand for lifelong adherence to prescription medications. Dan also knows his health insurance is not likely to cover surgery costs or aftercare, but he feels it could be worth the cost if it helps him find a romantic partner. He takes advantage of a free consultation with a plastic surgeon, Dr P, who has some experience with face transplantation, to explore his options. Dr P listens to Dan's story and wonders how best to respond to his conflicting feelings.

Commentary

Trauma exposes people to the unacceptable: safety from physical and psychological harm is not a given or is perhaps illusory. Injuries sustained in traumatic accidents leave outwardly visible physical scars, but psychological scars are often hidden. Although only physical scars fall under the purview of a plastic surgeon, both are reminders of the painful lesson that terrible things can and do happen. For Dan, a desire to have his physical appearance restored or "normalized" might not be easily disassociated from his psychological injuries, as he is distressed that his physical appearance can no longer be "improved" by reconstructive surgery. A good trauma-informed practitioner would probably begin to wonder how many of Dan's scars are psychological ones that remain unaddressed.

This commentary provides guidance—from the perspective of a clinical psychologist who specializes in traumatic disorders—on the cognitive changes associated with traumatic experiences and how they are relevant to counseling patients considering high-risk or experimental plastic surgeries.

Changes in Cognition Following Trauma

In response to a traumatic experience, relatively predictable shifts in cognition occur as people attempt to integrate that experience into their understanding of the world and their place in it.[1] In the (even distant) aftermath of a traumatic event, such as a catastrophic burn, 2 common changes in thinking style attempt to re-establish a sense of safety and predictability: all-or-none thinking (eg, "I have to either accept how I look or continue trying to change my face") and a sense of a foreshortened or bleak future (eg, "I will never find a romantic partner if I look like this"). It is also common for people who have experienced trauma to make nonspecific autobiographical future projections. That is, rather than imaging doing or even trying to do specific things, such as applying for a job or graduating from school, people who have experienced trauma report general outcomes[1] (eg, "I would not be able to handle going back to the site of the accident" or "It doesn't really matter what I try, things will go wrong").

These changes in thinking style have implications for therapy with patients recovering from traumatic events, particularly for exposure-based therapy, in which patients work to approach a previously avoided experience (known as an *exposure*) in order to regain control over their emotional reactions. It is common for them to overestimate the terror they will feel in a future situation and later to minimize how nervous they were going into that same situation, thereby defeating their sense of having faced their fear. Because I am aware of cognitive changes that follow trauma, I ask patients to carefully monitor their predictions about their responses to an exposure as well as their before-exposure ratings of distress as they work to approach reminders of a trauma or triggers of a traumatic memory and their after-exposure ratings of distress. Over time, patients can abandon faulty catastrophic predictions their mind naturally offers up as they adjust their predictions about how tolerable different situations are likely to be based on new evidence they accumulate during therapy.

Understanding how shifts in cognitive style occur is important for plastic surgeons, particularly as they discuss treatment options for patients who have experienced trauma and weigh potential risks and benefits of surgery. Such patients are likely to overestimate risks associated with reminders of trauma and to underestimate their coping abilities or others' acceptance of their appearance.[2]

Recognizing Dan's Hidden Scars

In Dan's case, his predictions about his future dating prospects are particularly vulnerable to cognitive distortions. He might also experience another common trauma-related cognitive change: discounting the predictive value of positive autobiographical memories (eg, prospective dating partners in his past who had expressed interest in him even with his burn scars). Positive memories are more likely to be discounted compared to negative memories and related predictions.[1,2] These cognitive changes account for Dan's vague, unrealistic goal for improved physical appearance as the sole means of improving his dating prospects. They also account for the hopelessness Dan experienced when he was told that no further improvement could come from traditional surgical options.

As Dr P learns Dan's history and hears his conflicting feelings, she would likely consider 2 options: advising him to consider surgery (perhaps even face transplantation) or working with him on acceptance of his current appearance without further surgeries. If Dr P agrees with Dan about the impact of his appearance on his prospects for a romantic partner, she might be guided by the principle of beneficence and favor surgery. Indeed, plastic surgeons are vulnerable to the same biases about people with disfigurements as others[3] and might be more susceptible to assuming they understand a patient's goals about having an "ideal" appearance, given the frequent conversations they have with patients about treatment goals. However, if Dr P is unsure whether Dan is viewing potential benefits of a face transplant realistically, given his belief that his appearance causes his singleness, the principle of nonmaleficence could guide her to counsel Dan against surgery.[4]

In either case, by taking a trauma-informed perspective, Dr P might recognize the distorted nature of Dan's all-or-nothing thinking, as well as her own bias against external scars and her desire to provide treatment that could free Dan from external reminders of his traumatic injury. Even this last approach alone, however, fails to provide a means of subverting a decision-making process framed as binary—that is, one that fails to account for the possibility that Dan could be desperate to change his physical appearance *and* be able to seek and find a romantic partner without changing it or the possibility that he could accept his current appearance *and* still make changes to it. To embrace these polarities, Dr P and Dan need to adopt a dialectical perspective.

Dialectics and Decisions

Instead of framing decision making as a choice between extremes, a dialectical approach advocates a middle path in which truths of both extremes are acknowledged and synthesized. Dialectical behavior therapy (DBT) was developed in the 1990s by Marsha Linehan, a psychologist who recognized that patients and clinicians are both vulnerable to thinking in terms of extremes when faced with the urgency and life-or-death stakes of suicidal behavior.[5] A dialectical therapist might validate a patient's urge to escape from unbearable pain while also trying to help a patient solve problems that are making his life unbearable. In DBT, patients synthesize polar opposites in their thinking in order to change their lives; with a therapist's help, they do so by acknowledging—and, by extension, accepting—the very things, including trauma, that have made their lives intolerable. Levins and Lewontin call this synthesis of apparent opposites *dialectics*: "These are the properties of things that we call dialectical: that one thing cannot exist without the other, that one acquires its properties from its relation to the other, that the properties of both evolve as a consequence of their interpretation."[6]

Dan's thinking suggests an unresolved dialectic. He feels torn and conflicted about his competing desires: to accept himself as he is, on one hand, and to meet an aesthetic standard that presumably will make him acceptable to a romantic partner, on the other. Desire for romantic connection is as human as the tendency to value physical attractiveness in a partner. That Dan views this desire as being in conflict with self-acceptance is evident in what he does and says. Acceptance, as Dan has been practicing it, seems conditional; that is, in the absence of a surgical option, he "has to" accept his face as is. Although the solidarity he feels with other burn survivors suggests he accepts his facial appearance as part of his history and identity, it is also a part of his identity he would readily shed for the chance to have a romantic relationship. What Dr P might explore with Dan is the degree to which Dan has set acceptance and change of his appearance at odds with one another.

What might synthesis look like for Dan? An ideal partner for Dan might be one who accepts his appearance and would also support his choice for surgery. Given Dan's focus on dating as a successful face transplant outcome and the potential influence of trauma-related cognitive biases on his decision of whether to have a face transplant, Dr P might ask Dan to describe some of his predictions and experiences up until this point: Is Dan making a prediction about being rejected based on his appearance or has rejection actually occurred on this basis in the past? What was Dan's dating history like before the accident? How has Dan's personal life been affected more generally by his injuries, multiple surgeries, and recovery? Dan's responses to these questions would help both Dr P and Dan identify polarities in his thinking about dating and his appearance.

Dialectics and Ethics

Two additional considerations are of note when taking a trauma-informed, dialectical approach to decision making with Dan. First, if Dan's thinking is sufficiently compromised by cognitive distortions to undermine his capacity to give informed consent or refusal, this limitation should be recognized by clinicians helping him assess the appropriateness of surgery. Helping Dan confront his cognitive distortions is perhaps best done with a DBT therapeutic intervention, which Lineman calls "entering the paradox."[5] To enter the paradox is to acknowledge without irony that 2 opposites may simultaneously be true—that is, to reject the rightness or wrongness of any single perspective—and instead to focus on maintaining a middle path between them. Dr P must identify the type of all-or-none thinking associated with trauma-related changes in Dan's cognition so that she can help Dan make an informed decision about surgery not unduly influenced by his cognitive distortions. Dr P can then help Dan find a middle path between changing and accepting his face in a way that overrides effects of his cognitive biases. She could advise Dan, for example, that she cannot support his consent to surgery unless he creates a loving, steady support system. This kind of response invites Dan to find a middle way in which his quality of his life is not conditional on his appearance.

Second, it might seem as though a trauma-informed, dialectical approach to Dan's thinking and decision making should be facilitated by a psychologist or other mental health professional rather than a plastic surgeon. Dan's negative reaction to a previous plastic surgeon's statement ("'There's nothing else we can do' to improve appearance, ability to speak, or ability to eat easily") demonstrates the clinician's failure to take a dialectical approach with Dan by exploring whether these were Dan's or his own goals for further surgery (and particularly whether the two shared an idea of what an "improved appearance" would entail). Discussing his conflicting desires with a plastic surgeon could validate Dan's experience of the intense societal pressure to look "normal" (which plastic surgeons are uniquely suited to acknowledge, given their livelihood) while also enabling him to see how changeable that definition is[7] (which plastic surgeons again are uniquely suited to discuss based on shifting norms in the field). At the very least, some consideration of the impact of passing off or "turfing" patients such as Dan is warranted if the main motivation is avoiding an uncomfortable discussion, as turfing has a negative impact on patients' perceptions of their care and recovery.[8] This outcome is particularly relevant to patients with trauma histories, who are especially vulnerable to feeling abandoned and betrayed by health care institutions or individual clinicians.[9] Taking the time to counsel Dan about how past trauma could influence his decision making about and expectations for surgery would be critical and well within a plastic surgeon's scope of practice.

Conclusion

Patients who have experienced traumatic injuries like Dan's need clinicians who will allow time and space to navigate paradoxes during decision-making processes. Clinicians who can help patients like Dan seek a middle path between acceptance and change can (1) avert harm by avoiding procedures that are not clinically indicated or could expose patients to unnecessary risk and (2) help patients identify and resolve conflicts generated by posttraumatic cognitive biases.

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IN THE LITERATURE

How to Help Patients Considering VCA

James L. Benedict, PhD and Rolf N. Barth, MD

Abstract

Patients who might benefit from some form of vascularized composite allotransplantation (VCA) can be expected to have prior long-standing relationships with one or more primary care professionals or specialists who are well-positioned to help patients make well-informed decisions about whether and when to pursue VCA. Helping patients decide requires becoming familiar with VCA, its various forms, eligibility criteria, prior and possible outcomes, and potential risks and benefits. This article shares key points for helping patients.

History of Vascularized Composite Allotransplantation

Vascularized composite allotransplantation (VCA) is the name now used for transplantation of complex anatomical structures composed of multiple tissue types. The term *reconstructive transplantation* has also been used in the literature since around 2009.[1] Reconstructive transplantation reflects the involvement of plastic surgeons who perform other reconstructive procedures. Until recently, the term *composite tissue allotransplantation* was more common.[2,3] Several different forms of VCA have been attempted over the past 23 years, with varying degrees of success in terms of both graft viability and patient quality of life. In 1996, a German team began a series of 6 knee transplants, all of which failed due to vasculopathy.[4] In September 1998, a French team performed a unilateral hand transplant on a man from New Zealand who experienced loss of his original hand in an accident with a circular saw. At first considered a success, the graft had to be removed 28½ months later because the recipient had discontinued antirejection medications.[5] The first genuinely successful VCA was a unilateral hand transplant performed in 1999 by Warren C. Breidenbach and his team at Louisville in the United States.[6] That graft has now been maintained with good function for more than 20 years.[7] In addition to knee and upper extremity VCAs, the field includes lower extremity,[8] esophagus,[9] larynx,[10] abdominal wall,[11] penis,[12] uterus[13,14,15] and craniofacial transplants.[16]

All forms of VCA remain relatively rare. Upper extremity VCA is the most common, with approximately 100 patients worldwide having undergone the procedure by 2018.[17] Craniofacial transplantation, which began in 2005, had been performed 44 times by the end of 2018.[18] And uterus transplantation is currently being performed in an increasing number of patients.[13] Upper extremity, craniofacial, and uterine VCA are intended to address the needs of individuals whose upper extremities have been lost or are absent; major craniofacial disfigurement, including loss of function; and primary uterine infertility. Thus, these 3 types of VCA and their impacts on recipient quality of life will be the focus of attention in the remainder of this article.

Eligibility and Patient Selection

Like solid organ transplant (SOT) candidates, potential VCA recipients must meet various criteria for general health and be matched with an appropriate donor.[19] Payment for VCA transplantation is not available through private insurance, Medicare, or Medicaid, so patients in the United States are selected for grant-based programs (eg, Department of Defense grants for upper extremity and face or institutional grants for uterus) after rigorous evaluation of their physical condition, psychosocial well-being, and social support.[16,17,20]

Because living with any transplanted organ involves substantial burdens and significant risks, it is especially important that prospective patients demonstrate emotional stability, adaptability, and strong coping skills.[21] Ironically, this requirement means that many individuals who might be excellent candidates are not interested in VCA, as they have adapted well to their current condition and are uninterested in taking on VCA's risks and burdens. Meanwhile, many who are eager or even desperate to undergo VCA are not good candidates. Failures in patient selection have been blamed for several poor outcomes in VCA, including poor function and graft loss resulting from recipients choosing not to participate fully in physical rehabilitation and failing to adhere to the immunosuppression protocol.[22] It should be noted that recipients' noncompliance can arise from the tremendous demands recipients face and the burdensome side effects of immunosuppression, which will be explored in more detail below. Nonetheless, proper patient selection has been a challenge from the outset and remains so.[12,23,24]

Outcomes

Outcomes in upper extremity transplants in the United States and Western Europe have ranged from excellent to dismal. In the United States, as of 2011, 2 patients had maintained their grafts for 10 years or longer with good function—that is, function superior to prostheses though not equal to that of the natural extremity.[25] On the other end of the spectrum, 4 patients have lost their grafts as a result of rejection.[26] Other patients have experienced outcomes that fall between these 2 extremes.

Craniofacial transplant outcomes have been generally good both aesthetically and functionally, but patients have experienced a variety of complications including chronic rejection and renal insufficiency or failure, and 5 recipients have died since their transplants.[16,27,28] Volumetric changes in the facial tissue of craniofacial transplant recipients appear to mimic accelerated aging, resulting in a noticeable difference in appearance.[29] Of special concern for craniofacial transplant recipients is the fact that graft loss might not be survivable unless a new donor is found and a second transplant is done.[30]

Uterus transplantation is unique among VCAs in that it has a clear definition of success: the live birth of a healthy child. Outcomes have been encouraging, with several births resulting from live donors in both the United States and Sweden,[31] and recently the first live birth following transplant from a deceased donor was reported from Brazil.[32] However, significant complications have been reported, including bleeding, thrombosis, and infection requiring urgent graft removals.[31,33]

Because VCA is not necessary to preserve or extend life, its justification is that it might improve the quality of life. Unfortunately, in the literature much more attention has been given to measurements of graft viability and function than to quality of life assessment. Herrington and Parker addressed this lacuna recently in a report of 5 narrative case studies, but more research is needed.[34]

Burdens, Risks, and Potential Benefits

Upper extremity, craniofacial, and uterine VCA each present significant burdens and risks while holding the potential for substantial benefits. These benefits include functional, aesthetic, and psychological improvements. Upper extremity recipients can gain greater independence in activities of daily living and, along with craniofacial recipients, might gain confidence in their appearance, which allows them to be less self-conscious while socially active. Craniofacial recipients might experience restoration of the ability to eat by mouth and have improvements in speech. The obvious benefit to uterus transplant recipients is realizing their goal of giving birth to a healthy child.

However, the potential for such benefits is accompanied by significant burdens and risks. Upper extremity VCA—and, to a lesser extent, craniofacial VCA—requires rigorous and time-consuming physiotherapy to restore function.[16,35] All forms of VCA share the burdens and risks of major surgery and long-term immunosuppression. In uterine VCA, the burdens and risks associated with immunosuppression are more limited because the uterus is removed and immunosuppression discontinued after the recipient has given birth to the desired number of children. (Current research protocols permit a maximum of 2 children per recipient due to risks of long-term immunosuppressant use.[31,33,36]) For upper extremity and facial VCA, immunosuppression is required as long as the graft remains in place. As is known from solid organ transplantation (SOT), the likelihood is high that long-term immunosuppression will lead to serious complications (ie, viral, fungal, and bacterial infections; hypertension; new-onset diabetes after transplantation; dyslipidemia; chronic kidney disease; and malignancy).[37,38,39,40] The high risk of such complications could be acceptable when the goal of transplantation is to extend as well as to enhance life. However, as mentioned, VCA aims only at improving quality of life, and the complications of immunosuppression can negatively affect a recipient's posttransplant quality of life and even lead to an earlier death. Common side effects of immunosuppression, such as oral ulcerations, gastrointestinal problems, weight gain, hirsutism, hair loss, depression, or heightened anxiety, might also have a profound negative effect on the recipient's quality of life.[41,42,43]

What is also known from SOT is that nearly all transplanted tissues are eventually rejected, despite immunosuppression. The half-life of major transplanted organs (ie, kidney, heart, liver, lung) ranges from 6 to 15 years.[44] It is reasonable to anticipate a similar half-life for VCA. Potential recipients should therefore expect the eventual loss of the graft. As noted above, uterus transplants are intended to be removed before rejection can reach a critical stage. For craniofacial recipients, graft loss due to chronic rejection can be fatal.[16] For upper extremity recipients, rejection typically leads to a significant decline in function before the grafts are removed.[4]

The psychological burdens and risks of upper extremity, craniofacial, and uterus transplantation are also substantial. As the literature on adherence in SOT shows, living with a transplant is psychologically as well as physically demanding. Complex immunosuppression and physical therapy regimens can become burdensome.[42] Stress and a desire to escape some of the side effects of immunosuppression can cause some patients to take "medication holidays" despite the increased risk of rejection.[45] The difficulties of posttransplant life are often underestimated by candidates,[46] who, once they become recipients, might become discouraged and experience decreased desire to participate in rehabilitation or even to retain the graft.

Conclusion

For those who meet the rigorous eligibility requirements, an informed decision to undergo any form of VCA requires both extensive knowledge and careful weighing of burdens, risks, and potential benefits. Most potential patients are unlikely to be able to gather and analyze this information, in part because it is difficult to access and in part because it is difficult for those without some professional training to understand. In particular, potential patients might have difficulty understanding how long-term physiotherapy, a strict regimen of immunosuppression, and the side effects of immunosuppression can impact their quality of life. Medical professionals, including primary care practitioners and specialists who have a long-standing relationship with potential patients, can play a critical role in facilitating robust informed consent processes by exploring these issues with them.

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STATE OF THE ART AND SCIENCE

What's Missing in Our Thinking About Quality of Life in VCA?

Emily Herrington, PhD, MA

Abstract

Drawing on the principles of respect for autonomy and beneficence, many scholars have argued that despite significant drawbacks of immunosuppression and surgery, vascularized composite allotransplantation (VCA), such as hand and face transplantation, has the potential to enhance the lives of patients who meet appropriate criteria and are well supported. This article provides a brief overview of the literature on VCA with a focus on hand transplantation (HTx) and offers a critique of the lack of empirical data on HTx patients' perspectives.

Quality of Life in Hand Transplants

Within a few years after the publication of the first journal articles arguing for the ethical and scientific viability of hand transplantation in the modern era of immunosuppression,[1,2,3] hand transplantation (HTx) and other forms of vascularized composite allotransplantation (VCA) had become technically possible,[4,5,6] and they now are viewed as beneficial for properly selected and supported recipients.[7,8,9,10] Because reconstructive transplantation, such as hand and face transplantation, does not afford the lifesaving or life-extending benefits that have so far characterized transplant medicine, the permissibility of VCA resides in the potential of these transplants to enhance recipients' subjective quality of life (QoL) for as long as they have their allograft. Realizing potential gains in QoL is complicated, however, by the burdens of lifelong immunosuppression to prevent graft rejection and loss, which can also potentially limit longevity.

This essay presents a brief overview of QoL conceptions in the literature on VCA, with a focus on HTx, especially in terms of how HTx patients' quality of life has been imagined and represented by stakeholders writing on the ethicality of these interventions. I argue that the sparse characterization of HTx patients' pre- and posttransplant QoL in papers reporting outcomes data for HTx is an obstacle to knowledge generation and ethical analysis in the field; systematic efforts to describe and understand patients' lived experiences are needed to ground professional discourse on the ethicality of HTx and VCA more generally.

Making QoL a Goal of Transplantation

The emergence of QoL as a goal of organ transplantation has been called "a quiet revolution in organ transplant ethics" by Arthur Caplan and Duncan Purves.[11] Although some medical and ethical professionals remain skeptical as to the permissibility of HTx and other types of VCA[12,13,14] and many ethicists and health care professionals voiced strong misgivings throughout the early years of human experiments in the field,[1516,17,18,19] reports on the outcomes of the first cases of VCA—2 unilateral hand transplants in Lyon, France, and Louisville, Kentucky—celebrated these interventions' surgical success and the idea that hand transplants with a normal level of immunosuppression could be effective for some patients.[20,21,22] In these initial case reports—published 6 to 24 months after surgery—success is presented in terms of technical *proof of concept* (ie, the idea that HTx that yields functional returns and extended graft survival is scientifically possible) but not in terms of HTx patients' own understanding of their postoperative wellness and satisfaction with results.

Despite the lack of subjective QoL data in early HTx and other VCA outcomes reports, many of these papers were cited as showing proof of concept for performing further hand transplants and for the diversification of composite tissue allotransplantation (an early name for VCA) to other conditions.[4,23,24,25] In 2004, bioethicist Françoise Baylis criticized the thin knowledge base among those arguing that success with hand transplants[24] supported new types of VCA interventions: "Wiggins and colleagues do not show unequivocally that hand transplantation is morally acceptable—the fact that something is done does not in itself constitute evidence of its moral acceptability." Baylis concluded, "in their haste to persuade others … they focus on the technical aspects of facial transplantation and issues relevant to the research ethics review process."[26]

Because HTx was the original VCA and successful HTx continues to be referenced to justify both performing other types of VCA and continued use of HTx as a treatment for amputees,[24,25,27] the question of the extent to which HTx restores patients' QoL in the long-term is important to consider.

Gaps in the Literature on HTx and VCA

Although attempts have been made to describe transformations in the health and quality of life of HTx patients using self-reported, survey-based methods or thematic analysis of psychiatric semistructured interviews,[28,29,30] these approaches do not necessarily perform the essential work of representing the viewpoints of recipients and their families before and after hand transplant surgery. In a 2012 review, "Quality of Life Considerations in Upper Limb Transplantation," Sally E. Jensen and colleagues consulted approximately 250 academic papers on HTx, 27 of which had quality of life as their main topic and were included in their analysis.[29] Only 3 of the 27 papers on hand transplant patients' QoL included interviews with hand-graft recipients. None of the studies were conducted by nonprogram-affiliated researchers using open-ended questioning techniques, and none of them quoted patients directly regarding their experiences. In some cases, no attempt was made to approach experimental participants for their point of view even when the stated goals of the research were compatible with doing so.[30]

The poor quality of QoL outcomes reporting in the first 2 decades of HTx and VCA experimentation has been remarked on by health care policy and behavioral health researchers and HTx practitioners.[17,19,31] Martin Kumnig and colleagues wrote in a 2014 review paper:

> Despite a thorough analysis of the literature, the lack of relevant published information in the psychosocial domain of transplanted patients is a significant limitation…. The majority of articles do not address the psychosocial assessment in any greater than passing detail, so the conclusion that can be made from these highly descriptive, mostly empiric studies in the current literature is limited.[31]

According to United Kingdom hand transplant surgeons Simon Kay and Daniel Wilks, "Of the large number of [hand and face] transplants completed now, outcome data of value is to be found in few."[32]

Why Does What's Missing Matter to the Ethics of HTx and VCA?

HTx professionals and ethicists agree that objective evaluation of outcomes in HTx is fraught with difficulties,[33,34] in part because fewer than 80 hand transplant surgeries had been performed around the world as of 2013[35] and meaningful statistical analysis cannot be performed on such limited data. Perhaps surprisingly, there has been little interest in preserving and parsing those data that *can be gathered* on outcomes for HTx, including insights from patients describing their experiences. The difficulty of collecting and analyzing subjective observations of hand transplant recipients and family members might account for the absence of patients' voices in the literature on HTx outcomes. Nevertheless, since so few hand graft recipients' first-person accounts have been published, HTx surgeons and ethicists must work from insufficient data to imagine what the particular physiological and subjective psychosocial costs and benefits might have been for HTx patients who faced the trade-offs of HTx in their lives.

Justifying the Need for Patient Perspectives in HTx and VCA

To begin to address lack of subjective outcomes data on HTx and VCA, I conducted oral histories with hand transplant recipients and caregivers (17 interviews total).[36] Hand transplant recipients almost unanimously experienced functional gains with a hand transplant that were impossible with the prosthetics they had tried. However, these gains were made *within the parameters of the posttransplant lifestyle* (including restrictions on diet, lifestyle, budget, and freedom to travel in the short- or long-term). How narrators perceived their QoL pre- and posttransplant depended on several factors, including whether their personal conception of the value of the HTx process evolved with their experiences. Far from the straightforward, ostensibly objective accounting of outcomes of HTx reported in medical journals, the oral history interviews I collected with HTx patients and their caregivers paint a much more complex and deeply human picture of "success."

I argue in closing this essay that short- and long-term QoL trade-offs for HTx and VCA more generally are not yet well understood, especially given the impact of managed complications on QoL for patients receiving these types of grafts. Because VCA is focused primarily on improving recipients' QoL though such gains are attended by significant risks and long-term challenges, broadening the question of success to include VCA patients' self-reported experiences of (dis)ability and the ongoing construction of their identity—along with insights generated by tests of kidney function and hand strength-motility, for example—would seemingly be appropriate to discussion of the ethicality and value of reconstructive transplantation. Insights from patients' perspectives are needed if the field of VCA is to develop person-centered knowledge of the effectiveness of different research protocols and the large-scale impact of these surgeries on participants' lives.

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POLICY FORUM

Why Quality-of-Life Data Collection and Use Should Be Standardized When Evaluating Candidates for Hand Transplantation

Martin Kumnig, PhD, MSc, Emma K. Massey, PhD, and Lisa S. Parker, PhD

Abstract

This article argues for 3 mutually reinforcing interventions in the field of hand transplantation (HTx): (1) collection of qualitative data about hand transplant recipients' subjective quality of life (QoL) outcomes, (2) multicenter standardization of data collection, and (3) use of data to develop evidence-based, standardized protocols for HTx candidate evaluation and information disclosure. These interventions are needed to improve candidate evaluation and informed consent processes in HTx, wherein the highly personal nature of desired outcomes justifies holding a candidate's consent to a standard approaching authenticity rather than the usual minimal standard of being informed and voluntary.

Quality-of-Life Data

Because the primary goal of hand transplantation (HTx) is maximizing transplant recipients' functional, emotional, and social quality of life (QoL),[1,2] it is ethically, clinically, and scientifically critical to assess the potential for HTx to improve a recipient's QoL. As with all QoL interventions, patients' subjective experiences are relevant to assessing whether an intervention achieves its aim. If HTx generally or routinely fails to improve hand transplant recipients' QoL, it might not (yet) be ethical to offer it, especially outside of experimental protocols. Collecting QoL outcomes data is thus critical for justifying HTx as a medical intervention and for providing accurate and salient information to candidates considering the procedure. Without QoL data, candidates are unable to evaluate the risk-benefit ratio and thus to give informed consent. In addition to collecting QoL data, 2 other interventions are needed to improve candidate evaluation and informed consent processes: multicenter standardization of QoL outcomes data collection and use of QoL data to develop evidence-based, standardized protocols for HTx candidate evaluation and information disclosure. This article discusses these interventions and argues for holding a candidate's consent to a standard approaching authenticity rather than the usual minimal standard of being informed and voluntary.

Data Collection Standardization

Collection of QoL outcomes data is needed to identify factors that predict successful HTx outcomes—including not only graft survival, functionality, and absence of comorbidities, but also improved QoL—and to use these factors to develop tools for use in candidate evaluation. There are no standardized guidelines for HTx candidate evaluation, and existing health status survey instruments (eg, the SF-36 by Ware and Sherbourne[3]) fail to capture the existential, identity-related, and interpersonal aspects of recipients' pre- and post-HTx life experiences that are critical to their QoL. Professionals who conduct psychosocial evaluations (PSEs) of HTx candidates and evaluate their social support and financial preparedness need to know which factors are useful for predicting positive HTx outcomes, including improved QoL. Beyond improving recipients' capacities to accomplish activities of daily living, goals such as the ability to feel a child's skin, to look "normal," to feel whole, or to return to vocational or avocational activities might be of critical importance to particular candidates.[4,5] Assessing rehabilitation demands prior to HTx is also important. For example, some candidates might welcome the sense of control their rehabilitation regimen can offer, but others might find it onerous or a necessary evil at best. The subjective, individualized, even idiosyncratic nature of QoL benefits to hand transplant recipients suggests that an outcomes registry that includes QoL outcomes data is needed as a first step to develop an evidence base. An evidence base is critical not only for developing standardized instruments for evaluating candidates but also for improving information disclosure and decision making during informed consent processes.

A number of specific psychosocial domains are emerging as important and predictive of posttransplant outcomes.[6,7,8,9] Yet key psychosocial challenges faced by HTx candidates and recipients are not well characterized despite some reports of QoL improvements[10] and negative psychosocial sequelae, including reactivation of psychiatric disorders, family discord, substance dependency issues, nonadherence, and dissatisfaction.[6] Currently, there are no psychosocial instruments designed specifically for use in this unique population.[6,10,11,12,13] In consequence, a variety of PSE protocols are used by individual transplant centers (see Supplementary Appendix). Standardized collection of subjective QoL outcomes data would likely increase confidence in research findings on factors predictive of improved QoL. Yet no standardized guidelines for collection of QoL outcomes have been developed for HTx.

Qualitative research is often used to generate hypotheses, theme-based criteria, or questions to be used when standardizing assessment or survey instruments.[14,15] Qualitative research on patient-reported subjective dimensions of QoL should be used to develop new standardized—perhaps even quantitative—assessment tools for evaluating candidates and collecting post-HTx data about QoL. Findings from such standardized assessments should in turn be used to improve informed consent and decision-making processes for HTx candidates.

Fairness and Candidate Evaluation Standardization

Given the subjective, individual, and even idiosyncratic nature of QoL benefits candidates seek from HTx, each candidate must be carefully evaluated. Indeed, concern for patients' well-being supports developing evidence-based, standardized instruments and protocols for PSE that would facilitate transfield comparison of surgical, functional, and QoL outcomes. Standardization of evaluation instruments and processes can also promote fairness in several ways.

The ethical importance of treating similar patients similarly supports incorporating standardized candidate evaluation (including PSE) instruments into all vascular composite allograft programs.[13] Standardization would minimize the impact of personal biases (eg, about whether a candidate is likeable, sympathetic, or "difficult") on evaluation. Moreover, developing and employing standardized assessment tools based in part on factors of subjective importance to past candidates and recipients would mitigate the impact of scientific biases in candidate evaluation.

Standardization of candidate evaluation processes through use of standardized assessment tools would enable—indeed, force—HTx programs to clarify whether a candidate's ineligibility for HTx is based on factors that are team focused, candidate focused, or a combination of the two. Different programs might justifiably accept or reject candidates based in part on a team's particular expertise, but a lack of "fit" between a candidate and a transplant team should result in referral to another HTx program rather than a declaration that the candidate is ineligible for HTx.

Fairness and concern for patients' well-being also requires that decisions about candidates' access to HTx be based on their medical needs and desired medical and QoL outcomes. History or presence of psychopathology, for example, should not categorically exclude HTx candidates; instead, this factor should be taken to indicate that additional support might be necessary during and following HTx. Indeed, a candidate's psychopathology is particularly relevant when the need for HTx derives from significant trauma. Similarly, while strong social support is associated with positive HTx outcomes, fairness demands that this fact not lead to the categorical exclusion of candidates lacking traditional familial support structures. Instead, teams should recognize the possibility that less traditional support structures may be adequate or should work creatively to identify social services to fill this need.

Authenticity of Informed Consent

Improving informed consent should be a goal of developing and using standardized tools to both evaluate HTx candidates and assess recipients' QoL outcomes. Informed consent requires disclosure of potential risks and benefits of an intervention and its alternatives, including refusal of treatment. Clinicians are obligated to help HTx candidates accurately assess this information and consider the relevance of both risks and benefits to their specific situation. Some candidates might overestimate HTx's potential to improve their QoL or underestimate demands of long-term rehabilitation and life-long immunosuppression regimens, for example. Other candidates might not fully comprehend the nature and scope of surgical risks or the potential for re-amputation or re-transplantation in case of graft loss.[4,6] A standard outcomes-assessment tool (based on previous HTx candidates' and recipients' expectations for and concerns about HTx as well as their QoL reports) could help clinicians better inform and support candidates' decision making.

An evidence base of subjective QoL outcomes could put flesh on the skeleton of the HTx risk-benefit ratio, which currently focuses on functionality in terms of activities of daily living and clinical risks. For some HTx candidates, factors such as aesthetics, identity, a sense of wholeness, facility performing specific functions, and relative facility interacting with others with a prosthesis vs HTx may be equally or more important than facility performing activities of daily living. Candidate evaluation and informed consent must elicit HTx candidates' personal goals and expectations, and candidates must be informed about the likelihood of their being met.

Given the subjective, individualized—even idiosyncratic—nature of QoL benefits candidates seek from HTx, there should be a strong correlation between these potential benefits and candidates' values, deeply held preferences, and specific goals. Therefore, the informed consent process should go beyond ensuring that the candidate's decision is informed and voluntary, which are the typical requirements for informed consent.[16] Instead, the candidate's decision should approach the ideal of authenticity—that is, it should be reflective of the candidate's personality, character, deeply held values, and view of a life worth living. By providing data about the subjective QoL outcomes of HTx and seeking a consent decision that is authentic, clinicians can help ensure that candidates' decisions promote their well-being as they themselves define it and that HTx achieves its goal of improving recipients' functional, emotional, and social quality of life.

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MEDICINE AND SOCIETY: PEER-REVIEWED ARTICLE

How to Integrate Lived Experience Into Quality-of-Life Assessment in Patients Considering Facial Transplantation

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Abstract

Facial transplantation has gained increasing acceptance as a treatment option to improve quality of life (QoL) for persons suffering from severe facial disfigurement. Despite its growth, the field has yet to establish a consistent approach to assessing QoL in face transplant candidates and recipients that includes integration of meaningful patient-reported outcomes. The published literature suggests that face transplant programs currently use a wide variety of assessment tools and strategies. Moreover, confusion remains as to how best to weigh patients' lived experiences and incorporate them into QoL assessments. Qualitative research can illuminate the dimensions of QoL that are meaningful to face transplant candidates and recipients. Coupled with collaboration and data sharing across face transplant programs, qualitative research will help to bring conceptual clarity and transparency to the assessment process.

Background

Persons living with severe facial disfigurement, whether congenital or acquired, suffer not only from debilitating functional limitations—including difficulty speaking and swallowing—but also from intense social stigma related to their visible difference. Social stigma in the form of ostracism frequently leads to extreme social isolation and is associated with adverse mental health outcomes, including depression, anxiety, posttraumatic stress, and suicidal thoughts or actions.[1] Facial transplantation (FT) as a means of improving quality of life (QoL) can enable recipients to re-engage socially, restoring their social identities as active, integrated members of their communities. Bramstedt has even argued that severe facial disfigurement could be akin to a form of social death and thus that FT can be considered lifesaving.[2]

Although more than 40 FT procedures have been performed worldwide and ethics discourse about FT has evolved beyond the risk-benefit ratio of a procedure that seeks to improve rather than extend life,[3] the field has yet to reach consensus regarding how to best define and assess QoL in FT recipients—including how to incorporate the meaning and value of patients' lived experiences of facial disfigurement and FT into assessments—as illustrated by the multitude of QoL measures that FT programs have used to evaluate FT candidates and recipients.[4] The lack of conceptual clarity about QoL also applies to other forms of vascularized composite allotransplantation (VCA), including upper extremity, penile, and uterine transplantation.[5] Nevertheless, there is increasing recognition of patient-reported outcome (PRO) measures as valuable, and some standardized PRO instruments such as the FACE-Q have been developed to assess QoL for a variety of aesthetic facial interventions.[6,7]

Addressing how to define and measure QoL for FT—including how to develop standardized PRO measures—raises important ethical considerations about how and by whom such choices are made, how patients' lived experiences should be incorporated into assessment approaches, the feasibility of developing standardized measures for a small population, and the value of transparency and data sharing across programs. We argue that assessing QoL in FT must be grounded in patients' experiences of living with severe facial disfigurement if QoL measures are to be useful in responding to the full scope of patients' needs.

Influence of Facial Disfigurement on QoL

Public perception, social stigma, and QoL are closely intertwined for persons with extensive facial disfigurement. Daily social interactions are characterized by continuously evolving and highly dynamic perceptions of self and others. Erving Goffman hypothesized that everyone strives generally to put his or her best face forward in social settings,[8] and recent scholarship has examined physical appearance as a form of cultural capital influencing social standing.[9] Social stigma can be considered a form of rejection resulting from *spoiled identity*, in which a person is excluded from many meaningful forms of social participation.[10] Exclusion from social participation or reduction in social standing can be particularly pronounced for persons with facial disfigurement, as facial differences can significantly interfere with social interactions and relationships.[11,12] Data on patients affected with cleft lip and palate and on patients undergoing oncologic head and neck procedures and reconstructions suggest that such patients frequently suffer from negative self-perception, impairment in interpersonal relationships, and mental health issues including anxiety, depression, self-harm, and an increased risk of mortality and suicide.[13,14,15,16,17,18]

Overabundance of QoL Measures

The published literature on FT reveals that FT programs use many instruments to assess QoL. Recently, Aycart et al systematically reviewed methods used to measure and report the impact of FT on patients' QoL.[4] QoL outcomes for only 14 FT recipients (37.8%) worldwide were reported in original peer-reviewed publications.[4] Of 17 articles reporting QoL outcomes, only 10 reported both pre- and posttransplant QoL outcomes. Eleven articles relied on subjective or descriptive accounts, and 6 used validated quantitative instruments. A total of

26 generic or reconstructive surgery-specific QoL instruments were identified, including the Short Form-36 Health Survey, the Rosenberg Self-Esteem Scale, the Dyadic Adjustment Scale, the Facial Disability Index, and various depression and anxiety scales.[4] Overall, QoL was reported to improve following FT.[4] However, the paucity of reported outcomes, risk of bias, variability in evaluators, and heterogeneity in instruments and assessment time points significantly limit the comparison and generalizability of results.

Developing a Standardized Instrument for FT

PRO measures have gained traction as components of value-based health care decision making, including in assessment of comparative effectiveness and shared decision-making support in areas such as breast cancer surgery.[19,20] There is even discussion of third-party payers incorporating PRO data into value-based reimbursement schemes.[21]

Standardized tools incorporating patients' perspectives have been developed and validated to assess QoL for patients with many conditions. For example, PRO measures, such as Body-Q scales, have been used to assess QoL in patients who have undergone bariatric surgery and body contouring procedures.[22] The definition of QoL used in these measures evolved through a phased approach, beginning with systematic review of extant literature and qualitative methods (including in-depth patient interviews) and progressing to development of scales, field testing, and psychometric validation and evaluation.[23,24] This approach has helped identify meaningful, replicable outcome measures,[22,23] and published minimum standards help promote appropriate uses of these PRO measures.[25] This approach has also been applied in developing and validating scales for a number of facial conditions, including the FACE-Q for aesthetic facial procedures[6,7,26] and the CLEFT-Q for cleft lip and palate.[24] Some have suggested that the FACE-Q might eventually be adapted for use in FT,[27] but existing PRO measures, such as those used in FACE-Q, have not been validated in the FT population due in part to small numbers of FT candidates and recipients. Eventually, as more procedures are undertaken, PRO measures might be developed for FT. A standardized PRO instrument for FT would help support the procedure's eventual transition from research to standard of care and would help facilitate reimbursement by third-party payers.[28,29]

Integrating Patients' Lived Experiences

That numerous QoL assessment tools are reported to be in use in FT likely reflects not only the lack of a standardized instrument validated for use in this population, but also differing perspectives about which dimensions of QoL are most significant. Although some dimensions of QoL lend themselves readily to assessment by widely accepted standardized means, other dimensions of patients' lived experiences of facial disfigurement and FT that are less amenable to traditional quantitative measurement are nonetheless deeply meaningful to FT recipients.[4] For example, patients' perceptions of the impact of facial disfigurement and FT on their social integration are not easily quantifiable. Moreover, these perceptions might vary over time and even across patients with similar levels of disfigurement and social support.

Historically, Western biomedicine's predominantly positivist paradigm has favored objective or quantitative information over subjective or qualitative information, such as patients' reports of their experiences.[30] This epistemological focus can be seen in how tools are developed and administered to assess dimensions of clinical functioning based on the assumption that universal knowledge of objective facts is both possible and preferable. This assumption can be useful for capturing some kinds of quantitative data, such as lab values and certain functional measures, but patients' experiences and knowledge claims are also valuable and increasingly recognized as such in health care.

As FT programs consider how best to integrate patients' lived experiences in definitions of QoL and in setting parameters for QoL assessment, particularly with regard to standardization of measures, the field should consider how best to account for variation and to avoid the pitfalls of privileging some ways of knowing over others. For instance, because demographic differences among patient populations can influence perceptions of QoL, the FT field will need to determine how such variations should be accommodated or accounted for when validating assessment tools. Traditional approaches to validating standardized tools might not be feasible given the small number of FT candidates and recipients. Moreover, developing validated measures of certain dimensions of patients' lived experiences might not be an appropriate goal. Qualitative research with persons with severe facial disfigurement, FT candidates, and FT recipients can illuminate patients' lived experiences and help identify dimensions of QoL that are most relevant and meaningful to potential FT candidates and recipients.

Collaboration is Key

FT programs' willingness to collaborate is key to meaningful assessment of QoL. Given that each FT procedure is unique and that few procedures are performed, programs should commit to sharing outcomes data and approaches to QoL assessment and monitoring to ensure ethical and sustainable progression of the field.[28] The Organ Procurement and Transplantation Network encourages data reporting for VCA procedures,[31] which include FT; this encouragement is a step toward the collaboration and transparency that is needed to advance the field of FT. The Chauvet Workgroup also has considered standardization of psychosocial assessment for VCA, including measures of QoL,[5] and reported preliminary findings for upper extremity transplantation.[5,32] The field of FT must integrate multiple epistemological stances and include a range of experiences to ensure that QoL assessment captures data that are meaningful and useful for FT programs, payers, candidates, and recipients.

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MEDICINE AND SOCIETY

What Are Good Guidelines for Evaluating Uterus Transplantation?

Margaret Horvat, MA and Ana Iltis, PhD

Abstract

Recent advances in uterus transplantation (UTx) suggest it is on a trajectory toward becoming an accepted clinical practice to treat absolute uterine factor infertility (AUFI). Additional uses have been envisioned but not studied. UTx programs thus far have relied largely on ethical frameworks associated with clinical research, surgical innovation, organ transplantation, and assisted reproductive technologies, as reflected in the Revised Montreal Criteria and the Indianapolis Consensus. This article argues that it is time to develop integrated guidelines that incorporate existing evidence, acknowledge and address tensions among the ethical frameworks that have informed judgments of UTx for AUFI thus far, identify and address ethical questions on which existing frameworks are silent, and anticipate future ethical issues in UTx research.

Introduction

Recent advances in uterus transplantation (UTx) suggest that it is on a trajectory toward becoming an accepted clinical practice to treat absolute uterine factor infertility (AUFI), which Brännström and Díaz-García describe as "infertility that is completely attributable to uterine absence (congenital or surgical) or an abnormality (anatomic or functional) that prevents embryo implantation or completion of pregnancy to term."[1] UTx is a type of vascularized composite allotransplantation (VCA) for the purpose of assisted reproductive technology (ART), and the uterus is classified as an organ subject to the National Organ Transplant Act.[2] The goal and desired outcome of UTx are similar to those of ARTs—specifically, gestational surrogacy—yet UTx largely relies on ethical guidelines that are specific to solid organ transplantation (SOT). Translating UTx to the clinical setting requires developing clinical practice guidelines specific to UTx that incorporate existing evidence; acknowledge and address tensions among the ethical frameworks that have informed judgments of UTx for AUFI thus far; identify and address ethical questions on which existing frameworks are silent; and anticipate future ethical issues in UTx research, including possible applications other than AUFI.

Clinical Practice Guidelines

Clinical practice guidelines are meant to translate reliable evidence into recommendations to improve quality, reduce variation in treatment, constrain costs, empower patients to make decisions, and inform third-party payers' coverage decisions.[3] Although the evidence base for the efficacy of UTx for treating AUFI is limited, it is important to promote quality, consistency, and transparency in UTx clinical programs, recognizing that guidelines will evolve over time.

The revised Montreal Criteria for the Ethical Feasibility of Uterine Transplantation and the Indianapolis Consensus are the most comprehensive recommendations available that are specific to UTx.[4,5,6] Despite differences among these 2 sets of recommendations for ethical UTx practice—and differences among UTx research program descriptions, which indicate that some programs' practices deviate from these recommendations—both draw on ethical frameworks from clinical research, surgical innovation, SOT, and ARTs, among other fields. For instance, the revised Montreal Criteria call for the recipient to be deemed "likely to take antirejection medication and follow up with the treating team in a responsible manner,"[5] which mirrors factors measured by SOT eligibility screening tools.[7] The Indianapolis Consensus recommends that the recipient have AUFI that has "failed all current gold standard and conservative therapy,"[5] a criterion that also stems from SOT frameworks.[8] In addition to these influences, both sets of recommendations suggest, in the words of the revised Montreal Criteria, that a recipient must "not exhibit frank unsuitability for motherhood,"[5] which is rooted in ART frameworks.[9] Moreover, the Indianapolis Consensus states that UTx would need to fulfill the criteria for surgical innovation, should require approval by "a duly constituted ethics committee" as recommended or required of any research study or innovative surgery, and should carefully consider risks to living donors and recipients.[6] UTx's reliance on guidelines from several different fields—and the tensions and ambiguities that could arise from this reliance—call for the development of a set of UTx-specific guidelines.

Developing UTx Guidelines

Because the International Society of Uterus Transplantation (ISUTx) gathers and disseminates information about UTx with a view to developing the field, it could facilitate guideline development. Establishing clinical guidelines for UTx to treat AUFI will involve consideration of stakeholders; criteria for recipient and donor eligibility, including risks and benefits; data collection; and posttransplantation management.

1. *Stakeholders.* An important first ethical step in developing guidelines for UTx to treat AUFI is identifying the stakeholders. Who counts as having AUFI? UTx studies have been limited to genetic females with AUFI seeking to gestate at least one pregnancy. [10,11,12] Some have suggested that transwomen also have AUFI or that all genetic males have AUFI and should be included.[13] Deciding whether to include transwomen or men

as stakeholders in this process and the priority to be given their interests involves ethical judgments. In UTx, potential living and deceased donors and their families also have relevant interests.

2. *Eligibility and organ allocation requirements.* Other ethical considerations involve criteria for recipient and donor eligibility and organ allocation. Will recipients be required to have produced their own genetic embryos, as appears to be the case in existing trials,[10,11,12] or will the use of donor eggs (or the eggs of a female partner) be permissible? Will potential recipients' suitability as parents be assessed and, if so, by whom and how? Must a recipient find adoption and surrogacy unacceptable or is a preference or desire to gestate a child sufficient? If living donors are permissible, will the eligibility requirements differ depending on whether recipients have a willing living donor (LD) or instead rely on a nondirected LD or deceased donor (DD)? How will organs from DD and nondirected LDs be allocated among eligible recipients, and what factors will be considered in prioritizing recipients?

3. *Risks and benefits.* Which risks (eg, hemorrhage, damage to internal organs, general anesthesia)[14] and potential benefits will be considered in establishing eligibility for LDs? How will the significance of these risks be assessed? Will the eligibility requirements for directed and nondirected LDs differ and, if so, how and why? In SOT, paired exchanges—in which an incompatible LD-recipient pair exchanges organs with another LD-recipient pair—are permissible, as are donor chains when incompatible LD-recipient pairs are linked with other pairs to form a donation chain. Would either of these types of exchanges be allowed in UTx? What if the paired exchanges varied in organ type? For example, would a woman be permitted to identify a willing kidney donor who would donate a kidney to someone in exchange for the kidney recipient providing a uterus donor?

4. *Data.* Which data should be gathered and reported as part of the UTx registry maintained by ISUTx,[15] and for how long will LDs, recipients, and future children be followed?

5. *Posttransplantation management.* The expectations of donors and recipients posttransplantation also raise ethical issues. Will recipients be compelled to have the transplanted uterus removed after one or two live births, as currently recommended due to the risks of long-term use of immunosuppressants?[5] How will this requirement be enforced? What if a woman desires more than 2 children? Which risks and potential benefits will be considered in determining when and how many embryos can be transferred post-UTx? What contact, if any, will be facilitated between LDs or deceased donor families and recipients?

These are among the questions that should be addressed in developing clinical practice guidelines for UTx to treat AUFI. The answers to these questions depend not only on medical judgments but also on ethical judgments, which have significant implications for the future of UTx and all potential stakeholders.

Comparison of UTx to SOT and ART

Because 2 UTx clinical trials in progress involve LDs,[11,12] it is likely that as UTx moves to the clinical setting, it will not be restricted to DDs. Here we discuss how the clinical practice of UTx could draw on guidance on the use of directed and nondirected LDs in SOT and ARTs. Each of these fields has different priorities and norms, which could lead to competing understandings of what is ethically permissible or obligatory in practicing UTx in the clinical setting.

Payment. The strict standards governing living nondirected organ donation limit the authority of donors and recipients, whereas the norms governing ARTs offer more latitude in negotiating the terms of the donor-recipient relationship. The National Organ Transplant Act prohibits organ donors from receiving “valuable consideration” for the organ.[16] Under the act, payment for a uterus donation would be illegal, but coverage of certain donor expenses may be permissible.[17] By contrast, payments to gamete (sperm and egg) providers and gestational carriers are routine in some jurisdictions.[18] One might argue that donating a uterus is not substantially different from donating an egg or serving as a gestational carrier and that therefore payment to a uterine donor may be acceptable.[18]

Recipient characteristics. Organs donated by nondirected LDs are allocated according to the criteria of the Organ Procurement and Transplantation Network (OPTN), which specify that donors may not stipulate recipient characteristics.[19] If SOT guidelines apply to UTx, then nondirected LDs would not be allowed to restrict who might receive their donated uterus. Gestational carriers in the United States, on the other hand, are free to choose with whom they are comfortable entering into a donor-recipient relationship.[20] This freedom allows a gestational surrogate to restrict her services based on her preferences; a gestational carrier may decide that she will only carry a child for a gay male couple or that she will not carry a child for single men. If we view UTx for treatment of AUFI through the lens of ARTs, we might conclude that a nondirected LD should be permitted to choose among potential recipients or restrict who receives her uterus. These decisions will be more complex if UTx is offered to nongenetic females or for reasons other than pursuing pregnancy.

Future contact with donor. Nondirected LDs and their recipients do not know each other’s identity and receive little information about each other, and future contact must be established through the organ procurement organization.[19] Neither party can set conditions on future contact before the donation, and they can choose to remain anonymous and restrict contact. In the case of uterus donation, this restriction would mean that the donor could not agree to donate only on the condition that she be informed of the recipient's future pregnancies and their outcomes or receive updates about future children. In gestational surrogacy, however, the parties may not only meet but also become involved in each other's lives, and surrogacy contracts may include provisions for future information about or contact with a child.[21] If we view UTx through the lens of ARTs, particularly surrogacy, we might conclude that potential donors and recipients should be able to negotiate contract terms rather than be governed by blanket prohibitions typical of SOT.

Ethical issues. UTx also raises ethical questions that are not easily addressed by the SOT and ART frameworks. One question is how to allocate uteri from nondirected LDs and DDs. Uterus allocation could be based on a first-come, first-served basis; motherhood status; child-rearing capacity; likelihood of being able to carry a pregnancy to term; or age.[22,23,24] Some of the ethical principles that govern the allocation of solid organs do not map neatly onto UTx. To promote the equitable allocation of organs, the OPTN relies heavily on the principle of utility, whereby an action or practice is deemed morally right "if it promotes as much or more aggregate net good than any alternative action or practice."[25] Applying the principle of utility to organ allocation involves taking into account all possible goods and harms, including patient survival.[25] Unlike many cases of SOT, UTx is not lifesaving or life extending.[25] Identifying other factors to be taken into account involves making decisions about the appropriate goals of UTx and ranking those goals. Thus far, the pursuit of pregnancy has been assumed to be the only acceptable primary goal, but another possibility is achieving a sense of bodily integrity or wholeness. This goal could contribute to quality of life, which the OPTN considers part of the utility assessment.[25]

The allocation framework used for SOT does not map neatly onto UTx for a second reason. The OPTN prohibits consideration of "social aspects of utility" and especially the "social worth or value of individuals."[25] Yet assessing the recipient's capacity for child-rearing—which could be seen as resembling social worth assessments—is part of the UTx evaluation recommended by some scholars and practitioners.[22,24] Someone deemed unworthy of the social role of being a parent would be rejected. Such assessments could be riddled with judgments about what makes a good parent and easily could lead to ranking of potential recipients based on suitability for child-rearing.

Summary. Ad hoc reliance in UTx on ethical frameworks from SOT and ART means that ethical guidance may be applied differently in UTx than it is in SOT or ART and that determinations of legitimate applications of UTx might change over time. For this reason, these 2 frameworks are not sufficient to guide UTx as it moves to the clinical setting. Clinical practice guidelines specific to UTx are needed.

Conclusion

Developing UTx practice guidelines would have a number of benefits. First, guidelines would foster a greater degree of consistency in UTx practice. Variation in UTx practice can arise in criteria for donor and recipient eligibility, time between transplantation and first embryo transfer, the use of living vs deceased donors, the permissibility of using donor gametes, and the number pregnancies or attempted pregnancies permitted. The UTx trials listed on ClinicalTrials.gov reflect this variation. For example, the age requirements for recipients range from 18-45,[10] 20-35,[11] and 18-39[12] at time of transplantation. Second, although variability in practice can be reasonable, it also can lead to mistrust, inequitable treatment, and inequitable outcomes.[23,24] Developing comprehensive guidelines for UTx will thus promote transparency, equity, and trust among those who consider themselves stakeholders in this new procedure. Third, developing practice guidelines also is an important starting point for establishing the future research trajectory of UTx and anticipating the ethical implications of expanded uses of UTx. Just as has been the case with SOT and ART guidelines, UTx guidelines will evolve as more becomes known about the procedure. However, changes to the guidelines should be anticipated and—like the initial guidelines—they should be implemented in an ethically consistent manner.

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What Hand Transplantation Teaches Us About Embodiment

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Abstract

While stakeholders in hand transplantation (HTx) recognize the importance of assessing quality of life (QoL), QoL has historically been inadequately defined and measured in such assessment procedures. Current conversations related to QoL in HTx could be enhanced by a phenomenological account of the lived body—namely, by illuminating the ways in which humans develop a holistic QoL through meaningful orientation in their interactions with the world and others. This meaningful orientation involves many factors; this essay considers how QoL is shaped by temporality (how past and future inform present satisfaction), embodiment (habituated, generally unconscious, meaningful attunement to the world), and intersubjectivity (how our identity as selves is constructed through social relationships).

Hands-on Phenomenology

Stakeholders in vascularized composite allotransplantation—specifically, hand transplantation (HTx)—acknowledge that standard assessment of quality of life (QoL) and providing long-term enhancement of QoL to hand transplant recipients is essential for the future of HTx.[1,2] However, there is ongoing debate regarding how QoL is to be defined and measured in such assessment procedures and, furthermore, how assessments can be standardized across the unique circumstances of individual transplant recipients.[2,3] Concerns over how to assess QoL are magnified by the tendency among stakeholders and the media to overemphasize cases that have been particularly successful and without incident.[2,3] For example, the website for the Louisville program[4] provides links to the success stories of its 10 HTx patients but does not mention that one recipient completed suicide, 2 have had their hand transplants removed, or that another feels that his hand transplant is effectively useless.[2,3] Furthermore, current tools that are used to track the progress of HTx patients (Carroll; Disabilities of the Arm, Shoulder and Hand, or DASH; and Hand Transplantation Score System, or HTSS, which still needs validation[1,5]) are primarily concerned with functionality of the hand and lack adequate metrics for evaluating QoL.

Current QoL assessments of hand transplant recipients fail to appreciate how our habituated, multifaceted, and generally unconscious facilitation of our hands provides a familiarity with and attunement to the world that shapes how we comport ourselves toward objects and create meaningful human experience. This article argues that current conversations related to QoL in HTx could be enhanced by the phenomenological method, which insightfully highlights how humans establish a meaningful orientation toward the world. Phenomenology, developed by Edmund Husserl, argues that a descriptive, first-person account of our lived experience can produce essential features about human existence. Phenomenology investigates how humans encounter the world in the immediate and primary modes of being, in contrast to scientific investigation, which is a derivative and secondary activity compared to our everyday being-in-the-world.

One salient observation phenomenologists have stressed is that humans are meaning makers. Martin Heidegger observes that what seems to make humans unique is our thoroughgoing concern to make meaning of our existence, make meaning of our circumstances, and illuminate meaning in our world.[6] Furthermore, phenomenologists observe that we always already expect the world and our experiences to be meaningful. We perceive the world and others within a meaningful orientation, structure,[7] milieu, or directionality[8] that tends toward significance.

This capacity for meaning making is essential for understanding and defining QoL. For example, when one hand transplant recipient "wakes each day to two transplanted hands that he feels are utterly useless,"[3] such uselessness is not merely about function but about meaningfulness. His hands are useless because their presence makes no sense, provides no orientation in his daily life. The phenomenological method has produced extensive insight into how humans interpret meaning and what QoL entails, especially with regard to 3 categories: how humans make meaning from their physical spatiality in the world (embodiment), from their experience of and orientation toward time (temporality), and from their sense of identity and purpose constructed through relating to others (intersubjectivity).

Temporality

Making meaning is only possible because a present moment retains elements of the past and anticipates the future. Heidegger suggests that humans are primarily oriented toward the future, or Being-ahead-of-itself.[6] Consider, for example, how often you are compiling a to-do list in your head while walking, driving, or sitting. More positively, humans find meaning through the imaginative creation of future possibility. We lose all sense of meaningfulness if we sense ourselves to be incapable of imagining new goals or creating new possibilities.

Temporality is critical for evaluating QoL. It illuminates the fact that, by its very nature, QoL is comparative and ongoing. QoL cannot be measured in a discrete moment; it requires an evaluation of the past and future. For example, the first hand transplant recipient of the Pittsburgh program initially had a “successful” HTx. In 2009, not long after his transplant, his new hand provided him a positive futural orientation. One reporter asked the hand transplant recipient, “What do you see in this different future?” He responded, “The same future I saw before I got hurt. I don’t feel broken anymore.”[9] However, when he requested the removal of his transplanted hand in 2013, he perceived the hand as inhibiting the meaningful future he had imagined—of becoming a mechanic and spending his time differently than driving to hospital visits and undergoing therapy.[10] Similar narratives have been shared by at least 3 separate hand transplant recipients who did not or could not imagine a meaningful future.[2,3] If a recipient’s QoL goals or future desires dramatically change over time, when can one say whether an HTx was successful?

Temporality adds a qualitative element to quantitative metrics of hand functionality. On average, hand transplant recipients in France have achieved 69% functionality in dominant hands and 55% functionality in nondominant hands, based on Carroll scores.[1] But percentages alone fail to capture whether such functionality translates into a sense of meaningfulness. Furthermore, there is no clear relation between degree of functionality and individual satisfaction with hand transplants. For example, one hand transplant recipient reported that he is quite happy with his hand transplant, yet he only has 55% functionality compared to his original hands.[11] Doctors predicted the first hand transplant recipient in the Pittsburgh program would regain about 65% functionality in his hand, and he initially exceeded that; however, despite that quantitative measure of achievement, 4 years later he still chose to have the transplanted hand removed.[10] Another hand transplant recipient, deeply dissatisfied with his hand transplant, reported that years of weekly sessions and grueling physical therapy could yield only 50% to 55% functionality.[2] And yet another patient completed suicide because the functionality of his hand transplant failed to afford him renewed life “purpose.”[2] If patients’ hand functionality plateaus, proves limiting, or fails to meet their expectations, patients have difficulty creating meaning out of their circumstances as their future possibilities become diminished.

Embodiment

As humans, our encounter with the world is thoroughly embodied. René Descartes claimed that the body is merely a machine that does whatever the mind tells it to do and that thinking is a task distinct from the body.[12] For Maurice Merleau-Ponty, however, I do not merely have a body; I am my body.[7] My perception of the world is situated by the physicality I inhabit. A growing body of research suggests that cognition itself is embodied and extended. Within this framework, much of what qualifies as *thinking* is not merely brain function; our bodies are part and parcel of the process.[13] Thinking is full-bodied and multidirectional, a continual body-brain feedback loop.[14] Memory and trauma are stored in the physical body.[15] The habituated and unconscious activities we perform throughout the day without thinking reflect a form of body intelligence or embodied know-how, suggesting that the body is as "smart" as the brain.[16]

Embodiment is essential in QoL discussions. We don't just have hands like we have a tool; we identify with them, and seeing a transplanted hand as part of oneself is critical.[3] With our hands, we extend our thinking into space and illuminate both ourselves and the world. Common references to our hands in figures of speech, as well as the myriad ways we communicate nonverbally and symbolically through hand gestures, suggest how deeply essential our embodiment is for creating meaning and communicating with others. One hand transplant recipient alludes to the profound symbolism of handedness, stating: "There's so many figures of speech and whatnot that relate to our hands and our feet…. If you don't have them [hands and feet], you have to find a way that when people use them, they don't feel uncomfortable. They just put their foot in their mouth, so to speak. See what I mean?"[2] Or as Rosemarie Garland-Thomson states, "Hands appear to help us conceptualize our world. Available items are on hand, handy, or in hand."[17] To heap praise on another, we offer a "thumbs up." A handshake symbolizes peace, agreement. My friend is "handy" because he can facilitate tools to perform mechanical tasks. We express ourselves with countless hand gestures: pointing, shushing, or a stiff palm meaning, "Stop!" People who use American Sign Language wield not only an entire grammar, syntax, and vocabulary with their hands but also conceptualize an entire world. Similarly, the extent to which hand transplant recipients see their new hand(s) as their own, express themselves by using their hands, and communicate gesturally with others dramatically affects their ability to make meaning of their world. Given the pivotal role hands play in human expression, it is astonishing that none of the assessment tools used for hand transplant recipients ask about gestures.

Embodiment is relevant to QoL for another reason: because our lived experience is direct and immediate to us, which means it is not captured in assessment tools, many of the ways we create meaning often go unnoticed. We are so adept with our hands that it often takes injuring them for us to fully appreciate them. As Boris Ladwig notes of a Louisville program hand transplant recipient, "Rickelman said people who have two hands don't realize how often they use both hands, from putting on clothes in the morning to eating with knife and fork to operating a mobile phone. 'It's a two-handed world,' he said. 'You say you can (do anything), but you can't.'"[18]

When we are attuned to the world around us, we do not really have to think about our hands and their affordances to achieve daily tasks. Hence, while current metrics for assessing HTx patients evaluate limb functionality and difficulty in performing tasks, they do not quantify the additional mental burdens hand transplant recipients carry that are not manifested in conventional handedness. With conventional handedness, I don't have to think about whether these hands are mine, worry about constant infection, worry about whether I can do something, worry about whether people will notice the scars, or spend extra time recalling how to shape my hand to perform particular tasks. Hence, the mental exhaustion the task exacts, the extra amount of time that's required to achieve it, and the difficulty in developing the intuitive recall to perform the task are obfuscated or ignored in assessment questions related to difficulty in performing tasks.

Intersubjectivity

Discussion regarding QoL also requires acknowledging intersubjectivity. Phenomenologists highlight that our being-in-the-world and individual identity are thoroughly shaped by our intersocial relationships. We are not autonomous subjects; rather, our sense of self develops within an interwoven array of familial, social, and cultural networks. Emmanuel Levinas stresses that one's subjecthood is not an established fact or foregone conclusion; it is an event realized through one's ethical attunement toward others.[19]

Intersubjectivity has numerous implications for QoL, but given space constraints, I will mention one. Meaning making happens in community, alongside others, with others. Our sense of identity and belonging is developed through relationships. And yet, on my reading of articles on the "success" of HTx, researchers appear to be thoroughly concerned about whether individual hand transplant recipients have achieved independence[5,20,21,22] in tasks like eating or driving—ignoring that these are thoroughly social activities that we regularly perform with or alongside others and that we learn to perform by watching others. While a certain level of autonomy is critical for personal satisfaction, an holistic assessment of QoL requires not only close proximity to hand transplant recipients and an evaluation of what tasks they can perform, but also evaluation of their ability to derive meaning from intersubjective and social encounters as well as ongoing conversations with their support community.

More Complete QoL Deliberation

While discussion regarding defining and assessing QoL in HTx is ongoing, this paper has highlighted factors that are essential to illuminating one's psychosocial health or QoL and that are not currently included in QoL assessment tools. These factors include 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.

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MEDICINE AND SOCIETY

What Do Unusual Faces Teach Us About the Ethics of Recognition?

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Abstract
With close attention to the film *Wonder*, this article examines how a narrative of community acceptance offers sustaining relationships for people with unusual facial appearance. This article argues that premodern responses of wonder can help reframe modern understandings of looking different.

Responses to Unusual Faces

In his practical advice book, *Changing Faces: The Challenge of Facial Disfigurement,* James Partridge gently but insistently counsels people with newly acquired facial injuries about how to manage other people, a task that Partridge suggests is more demanding than managing one's own facial injuries.[1] Of concerned friends and family, Partridge advises, "They have usually come to help you, but you may well find that you have to help them."[1] Of the many visual interlocutors one must face out in the world, Partridge concludes, "One secret of changing faces is to realize that *you* have to help them to break out of their scared-ness and meet you face-to-face."[1] Partridge's instruction manual lays out the social agency that patients must cultivate in order to achieve a quality of life that can be equal to that expected for people without disabilities. His insight into the work of what medicine considers rehabilitation—and what psychology considers adjustment for people with disabilities—comes from his own experience of living with an unusual facial appearance.

The 2017 film *Wonder* complements Partridge's practical wisdom for people with unusual faces by portraying how a 10-year-old boy with Treacher Collins Syndrome develops the social and life management skills that I call *dignity maintenance*.[2] *Wonder* is at once an extended case study in managing unusual facial appearance and a *bildungsroman* detailing how the fledgling hero ripens from a callow child into a mature young man equipped with the self-knowledge, direction, and worldly wisdom to lead a good and moral life. *Wonder* offers transplant recipients a lesson both in how to manage social relations while maintaining their dignity and in how to conceptualize the acceptance and recognition they need to rebuild a sense of self-value. The story thus offers a model for how people with unusual appearances can cultivate interdependent relationships and high quality of life.

Managing Others' Perceptions

In *Wonder*, Auggie Pullman, whose rare genetic condition and 27 surgeries have given him a very unusual face, enters school for the first time in fifth grade after being homeschooled by his devoted mother. After the shock of his birth, Auggie's family has come to love him—as many parents of children born with disabilities affirm in their memoirs[3,4,5]—because he is theirs and his distinctiveness makes him the son they love. We, like Auggie, are loved not for our normality—which is a statistical composite that no one actually fully embodies—but rather for our particularity. Sheltered and supported by his loving and economically secure family, Auggie has accrued the essential inner resources to become a sturdy and secure person. Inside his family circle, he is an ordinary kid playing games, teasing his sister, and sharing his day with his parents. Upon entering the community of peers at school, however, he is subjected to rejections that he has never before needed to navigate. He must endure rude stares, curiosity, bullying, and every kind of unwanted attention a kid as different as Auggie must endure. Up until he enters fifth grade, Auggie, who loves science and space travel, always has gone out of his home wearing an astronaut helmet that conceals what geneticists term a "coarse" appearance in which the proportions, shape, and placement of his features—riven now with scars from multiple surgeries—diverge far from ordinariness. Auggie gives up that astronaut helmet as he leaves the sheltered familiarity of his family and enters a community of anonymous peers.

Like Partridge's newly facially injured advisees, Auggie must learn to help his peers accept his extraordinary appearance. He must learn to take control of his own narrative, to help others understand his uniqueness as valued rather than damaged. As his older sister, Via, wisely advises him after the first day of school, "You can't blend in when you were born to stand out." Through his experience of entering into the school community of peers and teachers, Auggie ultimately learns the social skills that sociologists call *stigma management*[6] by developing his own distinctiveness, by welcoming the attention that his singular appearance provokes, and by understanding his face as the emblem of his unique Auggieness. In other words, Auggie learns to cultivate in others outside his family circle the emotional response of *wonder*, the awe people feel when they witness something they've never seen before. He does this by persisting in presenting his unusual appearance as part of his distinct self rather than hiding behind the astronaut helmet or in the safe space of his family who recognize and love him as he is.

Reframing Unusual Appearance

Wonder, Auggie's *bildungsroman* suggests, can be a productive, even redemptive, frame through which people with significant appearance impairments can be understood, accepted, and appreciated in today's modern social world. Wonder is an affect that was elicited in response to phenomena in the natural world that were rare—even singular—and often taken to be omens or divine signs during the premodern period. Large-scale dramatic phenomena such as earthquakes, comets, or similar inexplicable and unexpected visitations provoked wonder, understood as a psycho-emotional state of awe that brought together terror and fascination, a version of what the Romantics called the *sublime*.[7,8] Prodigies, divination, oracles, soothsayers, shamans, and all manner of worship rituals gained social traction from their power to inspire wonder. Ancient wonders such as the star of Bethlehem and the Cyclops Polyphemus, as well as hybrids such as Pegasus, Minotaurs, or angels are still prevalent in our received cultural archive. Early modern cultures of collection such as the cabinets of curiosity or the precursors to our contemporary museums harnessed the power of wonder by shifting communal rituals focused on singular objects or occurrences to private ownership of powerful objects that drew intense attention and speculative interpretation.[9,10,11,12] Human wonders such as so-called monstrous births, which we now understand as congenitally disabled newborns, as well as singular embodiments or the kinds of bodily shapes that could be interpreted as hybrids of animals and humans, were prized as occasions for prophecy or worship. A newborn with a face like Auggie's arriving in the premodern world—like other rare physiological forms such as conjoined twins, cycloptic stillborns, the hirsute, babies with fewer or more than the ordinary 10 fingers, or all manner of what now counts as abnormality—would have elicited wonder as a form of intense attentiveness from which meaning—revelation of a curse, blessing, transgression, election, or some other form of singularity—could be drawn and then debated.

The development of science with its systems of classification and measurement brought wonders down to earth and out of the realm of mystery and superstition. The very qualities of rarity and singularity that made the wide range of natural, animal, or human wonders culturally valuable and venerated in premodern cultures are the qualities that the modern system of medical science deems *abnormal* and aims to eliminate. Modernity disenchants the wonder, transforming the narrative of the marvelous into a narrative of the deviant.[13,14] As modernity develops in Western culture, the prodigious monster becomes the pathological specimen; what was once a revelation becomes a birth defect; what aroused awe now inspires horror; what was once to be valued is now to be cured. But wonders endure in modern societies as what cognitive scientists call *novel stimuli*, unfamiliar or rare phenomena that draw attention and interest. Even while societies value predictability, expect regularity, and extol the normal as a desired achievement, the power of human singularity endures, and wonder can be invoked as a stigma management strategy for people with a facial appearance like Auggie's.[15]

Stigma Management

When Auggie went out into the world hiding his face behind his astronaut helmet, he was exercising what the conversation analyst Harvey Sacks calls "doing being ordinary" and what the sociologist Erving Goffman describes as seeking civil disattention.[16,17] In modern anonymous communities whose size extends beyond a knowable kinship circle, being ordinary is the social benefit of avoiding undue attention from or being interrupted by the unknown others who constantly surround us. Auggie must leave that helmet with his father when he enters the schoolyard, where his peers stare at and torment him. Auggie's story of development begins as he literally shows his peers who he is: his distinctive self that is made up of his knowledge of science, his astronaut costume on Halloween, his kindness, his humor, his limitations—and his unusual face. After the first day of school, he cries to his mother, "Why do I have to be so ugly?" But day after day of presenting his face to the outside world of other kids transforms him from ugly to wondrous in their eyes, distinctively Auggie. At first, his radically unusual face was unreadable to his peers as anything else but ugly. But as Auggie's particular persona emerged day after day, his face came to be understood by the other kids as not so much ordinary as familiar—as the face of their friend and classmate, unusual and extraordinary as it was. He became one of them, and his face became the emblem of his distinctiveness. He amazed them and was—quite literally—*awesome*, not because his face was so strange but because he was the person they had come to care about.

The disability rights lawyer, activist, and storyteller Harriet McBryde Johnson, a person with an unusual appearance, offers a story about social management strategies for helping people move from seeing her as ugly to seeing her as wondrous. "It's not that I'm ugly," she writes in the *New York Times Magazine* in 2003, "It's more that most people don't know how to look at me."[18] "Two or three times in my life," Johnson continues, her distinctive appearance has evoked the response of wonder, an appreciation for what she calls "a rare kind of beauty … some people call me Good Luck Lady: they consider it propitious to cross my path when a hurricane is coming and to kiss my head just before voting day."[18] This sense of his own "rare kind of beauty" is what Auggie learns to recognize and accept, not only within the safe haven of his family circle but also in the community of peers at school.

The film's final scene narrates interpersonal validation as recognizing and appreciating the distinctive "rare kind of beauty" of people with unusual and highly stigmatized appearances. Sometime after Auggie has made a place for himself in the work and social environment of school, he asks his father about the long-missing astronaut helmet. Having hidden Auggie's helmet at his office, his father explains to his beloved son that when the boy started wearing the helmet, "I never got to see you anymore." In what I found the most moving moment in the film, Auggie's father says, "I missed your face." Acknowledging his son's own struggle, Auggie's father continues, "I know you don't always like it [your face], but I love it. It's my son's face. I want to see it." This affirmation of the distinctive, individual human face echoes the philosopher Emmanuel Levinas's premise that to regard the face of the other is the fundamental moral task of being human.[19] Auggie's face is beloved because it is different from every other face; it is the means through which his father knows his cherished son. Such a response that recognizes particularity is an example of the ancient affect of wonder, a cleaving to the strangeness of the other—here, of the other's face. To his father, Auggie's face can never be ugly but is instead the face of Johnson's "rare kind of beauty," a beauty like no other.

At the end of his life, dying from cancer, the neurologist Oliver Sacks published a series of essays reflecting on the experience of dying and the wisdom it brought him. A lifetime observer of the human condition, Sacks concluded that the essence of being human is our distinctiveness. "When people die, they cannot be replaced." Sacks, our time's most important observer of wonder, wrote, "They leave holes that cannot be filled, for it is the fate—the genetic and neural fate—of every human being to be a unique individual, to find his own path, to live his own life, to die his own death."[20] For Sacks, our human distinctiveness is a source of wonder. That all human uniqueness, perhaps especially that which is unusual or rare, can evoke wonder is what gives meaning to our lives and the work of being human. This is what Auggie has learned when he tells his father he doesn't really need to find that astronaut helmet after all.

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ART OF MEDICINE

Representing Communication, Compassion, and Competence in the Era of AI

Antonio Yaghy, MD, Jerry A. Shields, MD and Carol L. Shields, MD

Abstract
Artificial intelligence (AI) is now integrated into a variety of fields, including medicine. AI applications raise numerous ethical questions, particularly about quality of care and patient-clinician relationships. This article accompanies 2 digital photo-paintings that address these ideas narratively and visually, with special emphasis on communication, compassion, and competence.

Three *C*'s

Excellence is widely regarded as an attribute of good health care quality, often defined in terms of communication, compassion, and competence.[1] Restricted time is an obstacle to acquiring and mastering these 3 *C*'s if health care delivery systems focus too narrowly on volume and ignore patients' and clinicians' needs for compassion.[2]

AI Power

Artificial Intelligence (AI) is currently being adopted in many sectors, including health care. The power of AI technologies is evident in innovations in processing huge amounts of data, deep-learning algorithms, and expansion of machine learning capacity to learn in less time than it takes a human being.[3] AI applications in health care include faster and often more accurate diagnosis through image recognition, pathology detection, and diagnostic assessment.[3]

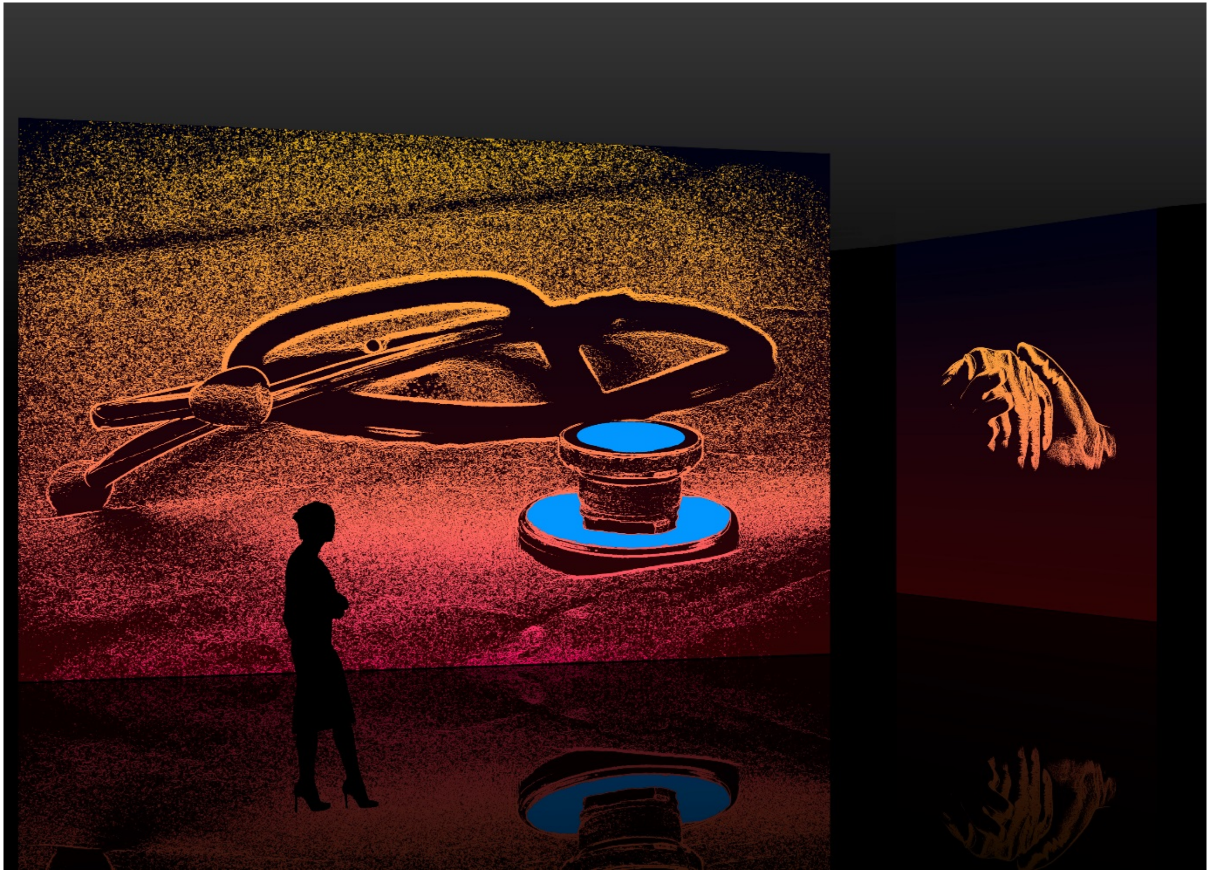
Quality Care and Patient-Clinician Relationships

Ethical questions have been raised about AI's capacity to undermine or extinguish compassion and human connection in health care.[4] If properly implemented in caregiving, however, AI could nurture excellence and motivate organizational cultures in which the 3 *C*'s could be practiced and cultivated with rigor, diligence, and care. For example, while an AI application is executing tasks, clinicians might be able to spend time communicating with patients, expressing compassion, and delivering care with competence and satisfaction.[2,3] Incorporating AI-based decision aids could also boost clinicians' confidence in care management decisions.[3]

Representing the 3 *C*'s Visually

The first image invites a viewer to consider communication and compassion as features of everyday patient-clinician encounters. These skills are displayed on screens suggestive of AI in the form of a digitized symbol of traditional professional caregiving—a stethoscope—and in the form of a digitized pair of holding hands, a traditional symbol of solidarity and care.

Figure 1. *Communication and Compassion*



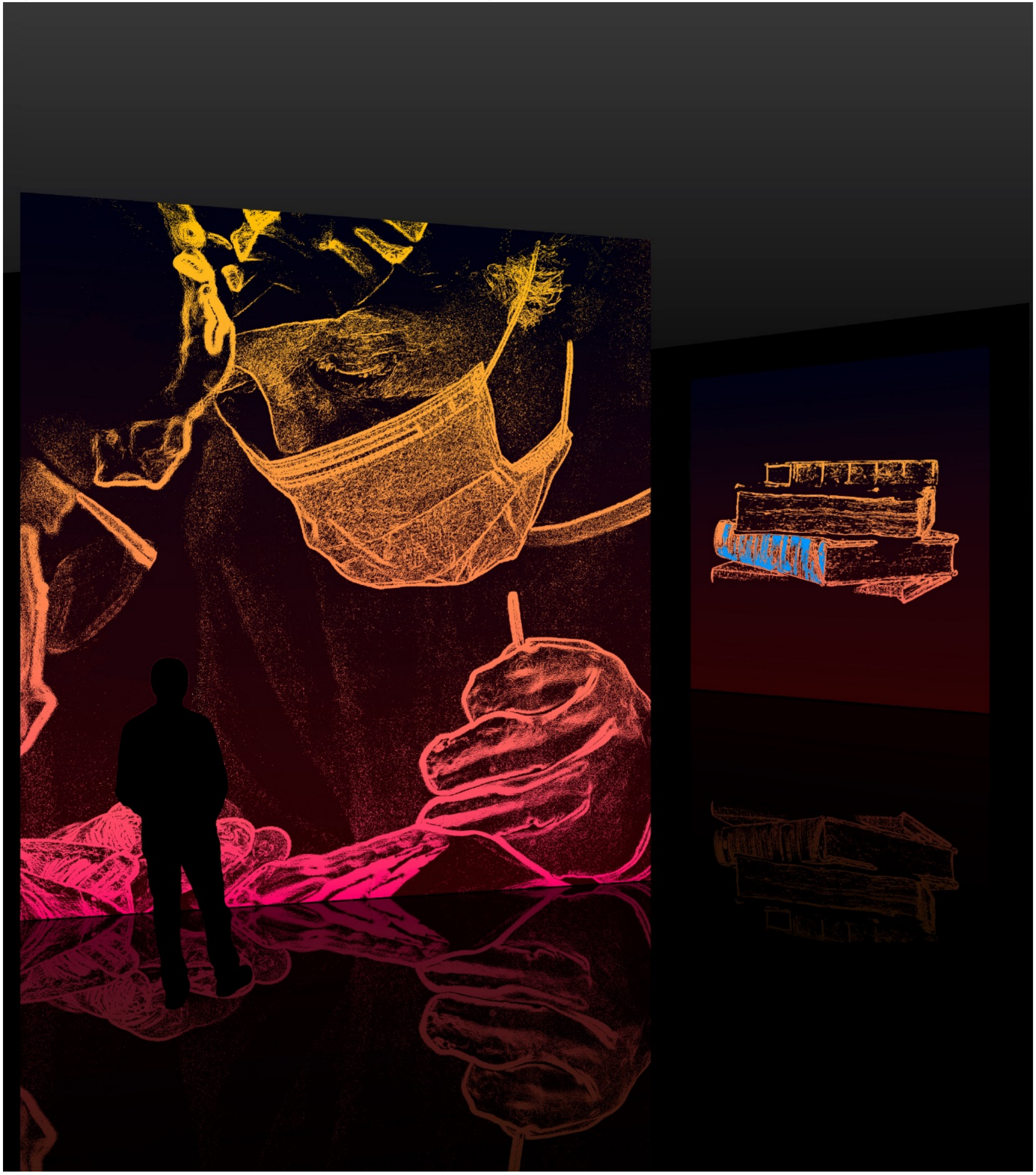
Media

Digital photo-painting.

In the foreground of the image, the woman—perhaps a patient or a clinician—might regard this larger-than-life stethoscope as a means of emphasizing listening as a key to good communication in any relationship. The blue upper surface of the stethoscope's bell and diaphragm suggests how listening can be healing in the same way that water quenches thirst.

A second image portrays competence in 2 forms: experience, represented as gained in surgical practice; and knowledge, represented in books.

Figure 2. *Competence: Experience and Knowledge*



Media
Digital photo-painting.

In the foreground of this image, a man regards a larger-than-life surgery in progress. This surgeon's upper eyelids and nose bridge suggest years of accumulated experience and knowledge put into the service of patients. At the right of the image, a book's blue spine suggests this time that, as water quenches a clinician's thirst for knowledge, it flows later to patients during clinical encounters.

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