

## MEDICINE AND SOCIETY: PEER-REVIEWED ARTICLE

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

Laura L. Kimberly, MSW, MBE, Allyson R. Alfonso, Rami S. Kantar, MD, Elie P. Ramly, MD, Arthur L. Caplan, PhD, and Eduardo D. Rodriguez, MD, DDS

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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.<sup>1</sup> 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.<sup>2</sup>

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,<sup>3</sup> 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.<sup>4</sup> The lack of conceptual clarity about QoL also applies to other forms of vascularized composite allotransplantation (VCA), including upper extremity, penile, and uterine transplantation.<sup>5</sup> 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.<sup>6,7</sup>

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,<sup>8</sup> and recent scholarship has examined physical appearance as a form of cultural capital influencing social standing.<sup>9</sup> 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.<sup>10</sup> 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.<sup>11,12</sup> 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.<sup>13,14,15,16,17,18</sup>

### **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.<sup>4</sup> QoL outcomes for only 14 FT recipients (37.8%) worldwide were reported in original peer-reviewed publications.<sup>4</sup> 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.<sup>4</sup> Overall, QoL was reported to improve following FT.<sup>4</sup> 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.<sup>19,20</sup> There is even discussion of third-party payers incorporating PRO data into value-based reimbursement schemes.<sup>21</sup>

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.<sup>22</sup> 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.<sup>23,24</sup> This approach has helped identify meaningful, replicable outcome measures,<sup>22,23</sup> and published minimum standards help promote appropriate uses of these PRO measures.<sup>25</sup> 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<sup>6,7,26</sup> and the CLEFT-Q for cleft lip and palate.<sup>24</sup> Some have suggested that the FACE-Q might eventually be adapted for use in FT,<sup>27</sup> 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.<sup>28,29</sup>

### **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.<sup>4</sup> 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.<sup>30</sup> 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.<sup>28</sup> The Organ Procurement and Transplantation Network encourages data reporting for VCA procedures,<sup>31</sup> 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,<sup>5</sup> and reported preliminary findings for upper extremity transplantation.<sup>5,32</sup> 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.

## References

1. Rifkin WJ, Kantar RS, Ali-Khan S, et al. Facial disfigurement and identity: a review of the literature and implications for facial transplantation. *AMA J Ethics*. 2018;20(4):309-323.
2. Bramstedt KA. A lifesaving view of vascularized composite allotransplantation: patient experience of social death before and after face, hand, and larynx transplant. *J Patient Exp*. 2018;5(2):92-100.
3. Rifkin WJ, David JA, Plana NM, et al. Achievements and challenges in facial transplantation. *Ann Surg*. 2018;268(2):260-270.
4. Aycart MA, Kiwanuka H, Krezdorn N, et al. Quality of life after face transplantation: outcomes, assessment tools, and future directions. *Plast Reconstr Surg*. 2017;139(1):194-203.
5. Jowsey-Gregoire S, Kumnig M. Standardizing psychosocial assessment for vascularized composite allotransplantation. *Curr Opin Organ Transplant*. 2016;21(5):530-535.
6. Klassen AF, Cano SJ, Scott A, Snell L, Pusic AL. Measuring patient-reported outcomes in facial aesthetic patients: development of the FACE-Q. *Facial Plast Surg*. 2010;26(4):303-309.
7. Pusic AL, Klassen AF, Scott AM, Cano SJ. Development and psychometric evaluation of the FACE-Q Satisfaction With Appearance scale: a new patient-reported outcome instrument for facial aesthetics patients. *Clin Plast Surg*. 2013;40(2):249-260.
8. Goffman E. *The Presentation of Self in Everyday Life*. Garden City, NY: Doubleday; 1959.
9. Jarrin A. *The Biopolitics of Beauty: Cosmetic Citizenship and Affective Capital in Brazil*. Oakland, CA: University of California Press; 2017.
10. Goffman E. *Stigma: Notes on the Management of Spoiled Identity*. New York, NY: Simon & Schuster; 1963.
11. Rumsey N, Harcourt D. Body image and disfigurement: issues and interventions. *Body Image*. 2004;1(1):83-97.
12. Robinson E. Psychological research on visible differences in adults. In: Lansdown R, Rumsey N, Bradbury E, Carr A, Partridge J, eds. *Visibly Different: Coping With Disfigurement*. Oxford, England: Butterworth-Heinemann; 1997:102-111.
13. Crerand CE, Sarwer DB, Kazak AE, Clarke A, Rumsey N. Body image and quality of life in adolescents with craniofacial conditions. *Cleft Palate Craniofac J*. 2017;54(1):2-12.
14. Millard T, Richman LC. Different cleft conditions, facial appearance, and speech: relationship to psychological variables. *Cleft Palate Craniofac J*. 2001;38(1):68-75.

15. Turner SR, Thomas PW, Dowell T, Rumsey N, Sandy JR. Psychological outcomes amongst cleft patients and their families. *Br J Plast Surg*. 1997;50(1):1-9.
16. Christensen K, Juel K, Herskind AM, Murray JC. Long term follow up study of survival associated with cleft lip and palate at birth. *BMJ*. 2004;328(7453):1405.
17. Teo I, Fronczyk KM, Guindani M, et al. Salient body image concerns of patients with cancer undergoing head and neck reconstruction. *Head Neck*. 2016;38(7):1035-1042.
18. Fingeret MC, Yuan Y, Urbauer D, Weston J, Nipomnick S, Weber R. The nature and extent of body image concerns among surgically treated patients with head and neck cancer. *Psychooncology*. 2012;21(8):836-844.
19. Broderick JE, DeWitt EM, Rothrock N, Crane PK, Forrest CB. Advances in patient-reported outcomes: the NIH PROMIS<sup>®</sup> measures. *EGEMS (Wash DC)*. 2013;1(1):1015.
20. Legendijk M, van Egdom LSE, van Veen FEE, et al. Patient-reported outcome measures may add value in breast cancer surgery. *Ann Surg Oncol*. 2018;25(12):3563-3571.
21. Squitieri L, Bozic KJ, Pusic AL. The role of patient-reported outcome measures in value-based payment reform. *Value Health*. 2017;20(6):834-836.
22. Gilmartin J, Bath-Hextall F, Maclean J, Stanton W, Soldin M. Quality of life among adults following bariatric and body contouring surgery: a systematic review. *JBI Database Syst Rev Implement Reports*. 2016;14(11):240-270.
23. Poulsen L, McEvenue G, Klassen A, Hoogbergen M, Sorensen JA, Pusic A. Patient-reported outcome measures: BODY-Q. *Clin Plast Surg*. 2019;46(1):15-24.
24. Wong Riff KWY, Tsangaris E, Goodacre T, et al. International multiphase mixed methods study protocol to develop a cross-cultural patient-reported outcome instrument for children and young adults with cleft lip and/or palate (CLEFT-Q). *BMJ Open*. 2017;7(1):e015467.
25. Reeve BB, Wyrwich KW, Wu AW, et al. ISOQOL recommends minimum standards for patient-reported outcome measures used in patient-centered outcomes and comparative effectiveness research. *Qual Life Res*. 2013;22(8):1889-1905.
26. Cogliandro A, Barone M, Salzillo R, Persichetti P. Quality of life after face transplantation: outcomes, assessment tools, and future directions. *Plast Reconstr Surg*. 2017;140(5):757e.
27. Klassen AF, Cano SJ, Schwitzer JA, Scott AM, Pusic AL. FACE-Q scales for health-related quality of life, early life impact, satisfaction with outcomes, and decision to have treatment: development and validation. *Plast Reconstr Surg*. 2015;135(2):375-386.

28. Caplan AL, Parent B, Kahn J, et al. Emerging ethical challenges raised by the evolution of vascularized composite allotransplantation. *Transplantation*. 2019;103(6):1240-1246.
29. Kantar RS, Ceradini DJ, Gelb BE, et al. Facial transplantation for an irreparable central and lower face injury: a modernized approach to a classic challenge. *Plast Reconstr Surg*. 2019;144(2):264e-283e.
30. Goldenberg MJ. On evidence and evidence-based medicine: lessons from the philosophy of science. *Soc Sci Med*. 2006;62(11):2621-2632.
31. Organ Procurement and Transplantation Network, US Department of Health and Human Services. VCA data collection. <https://optn.transplant.hrsa.gov/news/vca-data-collection/>. Published August 31, 2015. Accessed May 15, 2019.
32. Jowsey-Gregoire SG, Kumnig M, Morelon E, Moreno E, Petruzzo P, Seulin C. The Chauvet 2014 meeting report: psychiatric and psychosocial evaluation and outcomes of upper extremity grafted patients. *Transplantation*. 2016;100(7):1453-1459.

**Laura L. Kimberly, MSW, MBE** is an assistant research scientist in the Hansjörg Wyss Department of Plastic Surgery at NYU Langone Health in New York City, where she is also an associate in the Department of Population Health Division of Medical Ethics. She studies the ethical and psychosocial dimensions of vascularized composite allotransplantation, including equitable access to care, informed consent, embodied identity, and quality of life.

**Allyson R. Alfonso** is a medical student at NYU School of Medicine and a predoctoral research fellow investigating facial transplantation in the Hansjörg Wyss Department of Plastic Surgery at NYU Langone Health in New York City. Her research focuses on surgical education and quality improvement in plastic and reconstructive surgery.

**Elie P. Ramly, MD** is a surgery resident and postdoctoral research fellow investigating facial transplantation in the Hansjörg Wyss Department of Plastic Surgery at NYU Langone Health in New York City. Within the field of plastic and reconstructive surgery, his research focuses on improving patient safety and quality of care and optimizing surgical outcomes in the outreach setting.

**Rami S. Kantar, MD** is a surgery resident and postdoctoral research fellow investigating facial transplantation in the Hansjörg Wyss Department of Plastic Surgery at NYU Langone Health in New York City. He is interested in academic and outreach craniofacial reconstructive plastic surgery.

**Arthur L. Caplan, PhD** is the Drs William F. and Virginia Connolly Mitty Professor of Bioethics and the founding director of the Division of Medical Ethics at NYU School of Medicine in New York City. He is the author or editor of 35 books and more than 725 papers in peer-reviewed journals and has published widely on

the ethics of organ transplantation, including vascularized composite allotransplantation.

**Eduardo D. Rodriguez, MD, DDS** is the Helen L. Kimmel Professor of Reconstructive Plastic Surgery and chair of the Hansjörg Wyss Department of Plastic Surgery at NYU Langone Health in New York City. He has performed 3 face transplants, and his research interests include the technical refinements of facial transplantation as well as ethical aspects of the procedure.

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