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Evaluation of Societal Health Utility of Facial Palsy and Facial Reanimation

Callum Faris, MBBS, FRCS, ORL-HNS; Oren Tessler, MD; Alyssa Heiser, BA; Tessa Hadlock, MD; Nate Jowett, MD

IMPORTANCE The severity of a health state may be quantified using health utility measures. The utility of flaccid unilateral facial paralysis and unilateral moderate to severe postparalytic facial nerve syndrome with synkinesis may be challenging to discern from photographs alone.

OBJECTIVE To determine the societal health utility of flaccid unilateral facial paralysis, unilateral moderate to severe postparalytic facial nerve syndrome, and post–facial reanimation using standard video.

DESIGN, SETTING, AND PARTICIPANTS This survey study was conducted at the Massachusetts Eye and Ear and the Harvard Decision Science Laboratory from June 14, 2017, to August 3, 2017. Healthy adult naïve observers were recruited through advertising in the Cambridge, Massachusetts, area. Participants (n = 298) completed the web-based, interactive survey in person. The survey comprised clinical vignettes consisting of symptom summaries, videos, and pictures depicting 5 health states.

MAIN OUTCOMES AND MEASURES Adult naïve observers ranked the utility of 5 randomized health states (flaccid unilateral facial paralysis, unilateral moderate to severe postparalytic facial nerve syndrome, post–facial reanimation, monocular blindness, and binocular blindness) according to the visual analog scale (VAS), standard gamble (SG), and time trade-off (TTO) measures. Standard videos of patients' facial function were used.

RESULTS In total, 377 naïve observers were recruited and completed the survey in its entirety. Of the 377 participants, 298 (79.0%) were included for analysis. Among the 298 participants, 151 (50.7%) were female, 146 (49.0%) were male, with a mean (SD) age of 33.0 (15.1) years. No differences in health utility scores (SD) were observed between flaccid unilateral facial paralysis and unilateral moderate to severe postparalytic facial nerve syndrome (VAS: 0.598 [0.213] vs 0.629 [0.207]; SG: 0.714 [0.245] vs 0.748 [0.237]; TTO: 0.716 [0.248] vs 0.741 [0.247]). Both health states rated substantially worse than monocular blindness (VAS: 0.691 [0.212]; SG: 0.817 [0.204]; TTO: 0.826 [0.196]) and post–facial reanimation (VAS: 0.742 [0.189]; SG: 0.833 [0.206]; TTO: 0.838 [0.19]).

CONCLUSIONS AND RELEVANCE Health utility scores for flaccid unilateral facial paralysis and unilateral moderate to severe postparalytic facial nerve syndrome appeared to be equivalent and worse than that for monocular blindness, whereas scores for post–facial reanimation were substantially higher than the scores for the 2 facial movement disorders. These findings may provide insights into the societal advantages of facial reanimation surgery.

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Video

Author Affiliations: Division of Facial Plastic and Reconstructive Surgery, Department of Otolaryngology, Massachusetts Eye and Ear, Boston (Faris, Heiser, Hadlock, Jowett); Harvard Medical School, Boston, Massachusetts (Faris, Heiser, Hadlock, Jowett); Department of Otolaryngology and Head and Neck Surgery, Tübingen University, Tübingen, Germany (Faris); Section of Plastic and Reconstructive Surgery, Department of Surgery, Louisiana State University Health Sciences Center, New Orleans (Tessler).

Corresponding Author: Callum Faris, MBBS, FRCS, ORL-HNS, Department of HNO, Tübingen University, Tübingen, Germany (callumfaris@gmail.com).

Facial palsy comprises static and dynamic impairment of facial function and aesthetics. In addition to facial asymmetry, impairments in corneal protection, nasal breathing, articulation, and oral competence are experienced by patients with flaccid unilateral facial paralysis, whereas those with unilateral moderate to severe postparalytic facial nerve syndrome[1-6] report involuntary eye closure, gustatory epiphora, and facial discomfort. Expressional cues critical to nonverbal communication are distorted or lost.[7] A high aesthetic penalty is paid,[8] with profound psychosocial consequences for affected individuals.[9,10] Pre-intervention and post-intervention quality-of-life measures have been documented among patients with facial palsy,[11-18] but the economic burden of these health states has not been well characterized to date.

The severity of a health state may be quantified using a health utility measure, which represents a preference or desirability for a given health state.[19] Health utility is typically expressed as a continuous number between zero (death) and 1 (perfect health) derived from instruments, including the visual analog scale (VAS), as well as standard gamble (SG) and time trade-off (TTO) techniques.[20-22] Health utility is an attractive tool for economic evaluation as it permits the comparison of vastly different health states, which may aid in allocating scarce health care resources. An estimate of the economic implication of a disease or an intervention may be obtained by transforming health utility into quality-adjusted life-year.[22-24] The utility of a given health state may be assessed among patients, health care professionals, or naïve observers. The latter is typically favored, given the argument that allocation of public funds should be guided by societal preference.[25]

In this study, the utility of 2 unilateral facial movement disorders, flaccid unilateral facial paralysis and unilateral moderate to severe postparalytic facial nerve syndrome with synkinesis,[1-6] and a post-facial reanimation health state was assessed by naïve observers. This assessment was performed through an interactive web-based survey including standard videos of representative facial function for each condition or health state.

Key Points

Question What is the societal health utility of flaccid unilateral facial paralysis, unilateral moderate to severe postparalytic facial nerve syndrome with synkinesis, and post–facial reanimation as assessed through a web-based survey including standard video?

Findings In this web-based survey involving 298 adult naïve observers, societal perception of flaccid unilateral facial paralysis and unilateral moderate to severe postparalytic facial nerve syndrome health states is equivalent, with both states rated worse than monocular blindness. The post-facial reanimation health state had substantially higher health utility scores than the flaccid unilateral facial paralysis and unilateral moderate to severe postparalytic facial nerve syndrome states.

Meaning Substantially higher health utility scores for the post–facial reanimation state may provide insights into the societal advantages of facial reanimation surgery.

Methods

Participants and Survey Tools

From June 14, 2017, to August 3, 2017, the Harvard Decision Science Laboratory (Harvard University, Cambridge, Massachusetts) enrolled healthy adult volunteers (n = 377) through local advertising. Individual demographic data were captured and written informed consent was obtained from all naïve observers. The study was approved by the Massachusetts Eye and Ear Institutional Review Board.

Participants completed the survey in the Research Electronic Data Capture (REDCap) graphical user-interface environment (**Figure**), hosted by secure firewalls at Massachusetts Eye and Ear.[26] REDCap is a secure, web-based application that is designed to support data capture for research studies. It provides (1) an intuitive interface for validated data entry, (2) audit trails for tracking data manipulation and export procedures, (3) automated export procedures for seamless data downloads to common statistical packages, and (4) procedures for importing data from external sources.[26]

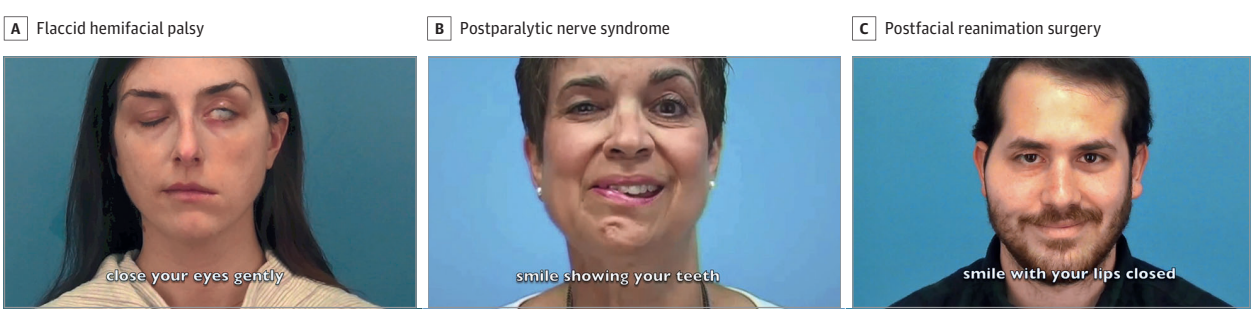
Survey Optimization and Delivery

After the initial transcription into REDCap, the survey was tested by a spectrum of volunteers with and without medical training (n = 30). The survey was assessed for language, duration, and comprehension. Language and syntax were amended to improve comprehension before survey administration. Participants were recruited from throughout the Cambridge, Massachusetts, area and then brought to the Harvard Decision Science Laboratory for survey completion. A trained proctor was on hand during the survey for additional clarification. Survey completion times ranged between 8 and 15 minutes.

Clinical Vignettes

The survey comprised clinical vignettes consisting of a 1-paragraph summary of symptoms and a video of the facial function of a patient with flaccid unilateral facial paralysis, a patient with unilateral moderate to severe postparalytic facial nerve syndrome, and a patient who had surgical post-facial reanimation. To reduce bias from the compounding effects of facial aging or lack thereof at the extremes of age, representative videos were chosen from among young adult and middle-aged patients. The flaccid hemifacial palsy vignette depicted a 34-year-old woman with dense, flaccid unilateral facial paralysis 2 months after the onset of pregnancy-associated Bell palsy (**Video 1**). The postparalytic facial nerve syndrome vignette depicted a 56-year-old woman with unilateral moderate to severe postparalytic facial nerve syndrome with synkinesis 3 years after extirpation of a vestibular schwannoma (**Video 2**). The surgical post-facial reanimation vignette depicted a 24-year-old man with dense, flaccid unilateral facial paralysis. He underwent free gracilis transfer innervated by the nerve-to-masseter muscle for smile reanimation and eyelid weighting for static blink reanimation 3 years after extirpation of a vestibular schwannoma (**Video 3**).

Figure. Graphical User Interface of Health Utility Survey



A, Frame grabs from video vignettes demonstrating flaccid unilateral facial paralysis (A), unilateral moderate to severe postparalytic facial nerve syndrome with marked synkinesis (B), and post-facial reanimation surgery by eyelid weighting for eye closure and free functional gracilis transfer for smile reanimation (C). A standard set of facial expressions may be viewed in their entirety in **Video 1**, **Video 2**, and **Video 3**. D, The visual analog scale, a standard gamble iteration, and a time trade-off iteration are demonstrated. Visual cues (yellow happy faces, red sad faces, and large X's) were employed to facilitate survey participants' understanding of risk percentages.

Facial function in each clinical vignette was assessed and scored by 2 expert clinicians (C.F. and N.J.) using validated clinician-reported facial palsy scales: House-Brackmann (grade scale: I-VI, with the highest grade indicating total paralysis), Sunnybrook (score range: 0-100; 100 represents normal, 0 indicates complete facial paralysis with severe facial disfigurement), and eFACE (score range: 0-100; 100 represents normal, 0 indicates complete facial paralysis with severe facial disfigurement); see **Table 1**.[27-30] Each video shows a representative patient performing a standard set of facial expressions—(1) face at rest, (2) elevation of eyebrows, (3) light-effort eye closure, (4) full-effort eye closure, (5) closed-mouth smile, (6) full-effort smile, (7) lip pucker, and (8) lower lip depression—and reciting 3 phrases (1, 2, 3; Happy Birthday; Hello, it's a pleasure to meet you). Two vignettes for monocular and binocular blindness were also included and consisted of a summary of symptoms and pictorial representation of these health states; the latter were included as internal controls.

Table 1. Facial Grading Scores for Clinical Vignettes

Vignette	House-Brackmann Scale[a]	Sunnybrook Scale[b]				eFACE Scale[c]			
		Resting Symmetry	Symmetry Voluntary Movement	Synkinesis	Composite	Static	Dynamic	Synkinesis	Composite
Flaccid unilateral facial paralysis	VI	20	24	0	4	61	27	100	56
Unilateral moderate to severe postparalytic facial nerve syndrome	IV	15	60	11	34	85	56	19	54
Post-facial reanimation	III	15	56	0	41	86	67	100	81

[a] House-Brackmann grade scale: I-VI, with the highest grade indicating total paralysis.

[b] Sunnybrook score range: 0-100, 100 represents normal, 0 indicates complete facial paralysis with severe facial disfigurement.

[c] eFACE score range: 0-100, 100 represents normal, 0 indicates complete facial paralysis with severe facial disfigurement.

The text and media of the 5 clinical vignettes were uploaded into the survey. The order of presentation of the vignettes was randomly assigned to reduce learning bias and potential survey fatigue.

Health Utility Assessment

Health utility for each condition was assessed by the naïve observers using the VAS, SG, and TTO measures. Participants were asked to imagine themselves in the given health state described in each clinical vignette. For VAS, participants were asked to rate their perceived health in that state using a continuous sliding scale (range: 0-100, with 0 representing death and 100 representing perfect health). For SG, participants were asked to choose between living in the particular health state for the remainder of their life and electing to undergo treatment that carried a risk of immediate death but would completely correct the diseased state. For TTO, participants were asked to imagine they had 36 years of life remaining and then to choose between living in the particular health state for the remainder of their life and undergoing treatment that would completely correct the diseased state but would shorten their life span.

The chosen years of life remaining (ie, 36 years) correlated with the mean life expectancy (approximately 80 years) from the median age of Bell palsy onset (45 years)[31] and from the mean age at first presentation to our center (44 years) of patients with various etiologies of facial palsy.[32] For SG and TTO, a 6-level iterative ping-pong approach with pictorial cues was used. In this approach, the possibility of death from a surgical intervention (in the SG scenario) or life-years to trade off (in the TTO scenario) alternated between high and low values over consecutive iterations to determine the inflection point.

Statistical Analysis

Responses that met the following quality control conditions were included for analysis: survey completed in its entirety, binocular blindness rated as having equal or worse utility than monocular blindness on all 3 measures, and absence of identical responses across scenarios. Scores were normalized to a continuous 0 to 1 scale; the VAS utility by the formula VAS/100; the SG utility by the formula (100 - the percent risk of death at the point of indifference) / 100; and the TTO utility by the formula (36 years - number of years traded off at the point of indifference) / 36 years. Normality was assessed using the Shapiro-Wilk test. Means and SDs were calculated, and differences between the utility of the different health states were sought using 1-way analysis of variance with post hoc pairwise comparison using the Tukey method. A 2-tailed $\alpha = .05$ was used for all tests, which were performed using SPSS Statistics, version 22 (IBM Corp).

Results

Demographics

During a 7-week study period (June 14, 2017 to August 3, 2017), 377 naïve observers were recruited and completed the survey in its entirety. Of these participants, 298 surveys (79.0%) met quality-control conditions and were included for analysis. Seventy-nine surveys (20.9%) were excluded from analysis as respondents rated binocular blindness as having higher utility than monocular blindness on one or more of the VAS, SG, or TTO measures, indicating a data-entry error or respondent failure to comprehend the survey techniques. Among the 298 naïve observers, 151 (50.7%) were female, 146 (49.0%) were male, with a mean (SD) age of 33.0 (15.1) years (**Table 2**). Compared with the US population, survey participants' age distribution was skewed toward younger individuals with higher educational levels (Table 2),[33] and self-identified race/ethnicity was skewed toward lower percentage of whites and higher percentage of Asians and people from the Indian subcontinent.[34,35] Income distribution of participants was comparable to that of the US population as a whole.[36]

Health Utility Scores

Utility ratings for the 5 clinical vignettes are reported (score [SD]) in **Table 3**. Significant differences were detected among the 5 health states for all measures. Post hoc comparisons demonstrated no differences between naïve observer-assessed health states for flaccid unilateral facial paralysis and unilateral moderate to severe postparalytic facial nerve syndrome (VAS: 0.598 [0.213] vs 0.629 [0.207]; SG: 0.714 [0.245] vs 0.748 [0.237]; TTO: 0.716 [0.248] vs 0.741 [0.247]). Both states ranked substantially worse than monocular blindness (VAS: 0.691 [0.212]; SG: 0.817 [0.204]; TTO: 0.826 [0.196]); Table 3) and favorably over binocular blindness. The post-facial reanima-

Table 2. Participant Demographics

Variable	Participants, No. (%) (n = 298)
Age, mean (SD), y	33.0 (15.1)
Sex	
Female	151 (50.7)
Male	146 (49.0)
Unknown	1 (0.3)
Race/ethnicity	
White	141 (47.3)
Asian/Pacific Islander	54 (18.1)
African American	29 (9.7)
Mixed race	6 (2.0)
Latino or Hispanic	31 (10.4)
Arab	1 (0.3)
Bangladeshi, Pakistani, Indian	32 (10.7)
Native American/Aleut/Aboriginal	1 (0.3)
Other	1 (0.3)
Declined to answer	2 (0.7)
Educational level	
Some high school	0 (0)
High school diploma or GED	7 (2.3)
Some college	69 (23.1)
Associate's degree	11 (3.7)
Bachelor's degree	92 (30.9)
Some graduate school	30 (10.1)
Graduate or professional degree	83 (27.9)
Professional certification	5 (1.7)
Declined to answer	1 (0.3)
Annual household income, USD	
≤15 000	44 (14.8)
15 001-25 000	24 (8.1)
25 001-35 000	39 (13.1)
35 001-50 000	36 (12.1)
50 001-75 000	35 (11.7)
75 001-100 000	37 (12.4)
≥100 001	46 (15.4)
Declined to answer	37 (12.4)
Marital status	
Single	203 (68.1)
Committed relationship	36 (12.1)
Married	41 (13.8)
Separated	3 (1.0)
Divorced	12 (4.0)
Declined to answer	3 (1.0)

Abbreviations: GED, General Equivalency Diploma; USD, US dollar.

tion state was substantially favored over flaccid unilateral facial paralysis and unilateral moderate to severe postparalytic facial nerve syndrome states (VAS: 0.742 [0.189]; SG: 0.833 [0.206]; TTO: 0.838 [0.19]) and was scored similarly to monocular blindness on SG and TTO measures and higher on the VAS measures (Table 3 and **Table 4**). In addition, substantially higher utility scores were reported for the post-facial reanimation state (VAS: +0.143 [95% CI, 0.096-0.192]; SG: +0.118 [95% CI, 0.066-0.171]; TTO: +0.122 [95% CI, 0.069-0.175]; $P < .001$), compared with the flaccid unilateral facial paralysis state (Table 4).

Discussion

Several studies have investigated the health utility of facial palsy among naïve observers using static facial photographs.[20,21,37] Grading facial function using standard video of facial expressions has demonstrated high agreement with in-person assessment,[38,39] but to our knowledge only one previous study has used video to assess the health utility of facial palsy,[37] and the health utility of the post-facial reanimation state has not been characterized. In our study, we used standard videos of facial expressions to assess societal preference for 3 manifestations of facial palsy. We assumed that presentation of photographs alone would result in suboptimal understanding of the various dynamic impairments of these health states among naïve observers. Health utility scores for flaccid unilateral facial paralysis reported here tended to be lower for SG and TTO measures than those previously reported by groups that used only static photographs (Sinno et al[20]: VAS, 0.56 [SD, 0.18]; SG, 0.79 [0.21]; TTO, 0.78 [0.21]. Su et al[21]: VAS, 0.43 [0.21]; SG, 0.74 [0.30]; TTO, 0.77 [0.25]).

Of the 3 common techniques for assessing preference-based health utility (VAS, SG, and TTO), not one is ideal, and resulting health utility scores often differ.[40] The SG and TTO methods require respondents to make a treatment choice based on risks or concessions and are therefore considered to be "preference based."[41] The VAS method is not considered a real preference-measurement instrument because it does not require participants to consider the potential downsides of therapy. As a result, the VAS method is not typically recommended for economic evaluation studies[41] or for public health policy applications.[42] It has been suggested that using all 3 techniques to measure health utility scores minimizes the inherent weaknesses of any one test.[21,43] The only study that demonstrated flaccid unilateral facial paralysis with video used only VAS estimations of health utility (Dey et al[44]: VAS, 0.53 [0.22]).

Table 3. Calculated Utility Scores of Health States

Method	Score (SD)				
	Monocular Blindness	Binocular Blindness	Flaccid Unilateral Facial Paralysis	Unilateral Moderate to Severe Postparalytic Facial Nerve Syndrome	Post-Facial Reanimation
VAS	0.691 (0.212)	0.507 (0.249)	0.598 (0.213)	0.629 (0.207)	0.742 (0.189)
SG	0.817 (0.204)	0.640 (0.278)	0.714 (0.245)	0.748 (0.237)	0.833 (0.206)
TTO	0.826 (0.196)	0.616 (0.282)	0.716 (0.248)	0.741 (0.247)	0.838 (0.193)

Abbreviations: SG, standard gamble; TTO, time trade-off; VAS, visual analog scale.

Table 4. Relevant Post Hoc Comparisons Between Health States

Health Utility Compared With Flaccid Unilateral Facial Paralysis			
Comparison Health State	Method	Mean Difference (95% CI)	*P* Value[a]
Monocular blindness	VAS	+0.093 (0.045 to 0.141)	<.001
	SG	+0.103 (0.050 to 0.156)	<.001
	TTO	+0.110 (0.057 to 0.163)	<.001
Binocular blindness	VAS	-0.912 (-0.139 to -0.043)	<.001
	SG	-0.074 (-0.127 to -0.022)	.001
	TTO	-0.100 (-0.153 to -0.048)	<.001
Unilateral moderate to severe postparalytic facial nerve syndrome	VAS	+0.030 (-0.018 to 0.078)	.42
	SG	+0.034 (-0.019 to 0.087)	.40
	TTO	+0.025 (-0.077 to 0.028)	.70
Post-facial reanimation	VAS	+0.143 (0.096 to 0.192)	<.001
	SG	+0.118 (0.066 to 0.171)	<.001
	TTO	+0.122 (0.069 to 0.175)	<.001

Abbreviations: SG, standard gamble; TTO, time trade-off; VAS, visual analog scale.

[a] *P* < .001 indicates statistical significance.

To our knowledge, until our study, no previous investigations included SG or TTO to measure health utility of facial palsy using video.

Postparalytic facial nerve syndrome (sometimes referred to as partial or incomplete facial palsy) represents the bulk of facial movement disorders in the community.[45,46] It is the result of aberrant nerve regeneration following high-grade facial nerve insult (eg, severe Bell palsy, Ramsay Hunt syndrome, Lyme disease-associated facial palsy, surgical insults)[18] and is characterized by permutations of residual facial muscle weakness, hypertonicity, synkinesis, myokymia, and mass hemifacial contractions.[6,47] Although 2 studies have analyzed the health utility of incomplete facial palsy states, they both used static photographs instead of video of facial function.[21,37] Dynamic visualization of facial function is required for adequate assessment of postparalytic facial palsy[38] and is optimally achieved through in-person or video assessment of a series of facial expressions. Many clinicians may assume that flaccid unilateral facial paralysis is worse than unilateral moderate to severe postparalytic facial nerve syndrome, but this study found no differences in their naïve observer-rated utility. Past studies have employed the House-Brackmann grading scale to characterize the severity of facial palsy vignettes presented to respondents,[21,37] but interpretation and translation of the results can be challenging as House-Brackmann grades III and IV may include facial weakness without synkinesis or facial hypertonicity with marked synkinesis.

The naïve observers in this study claimed they were willing to undergo a procedure carrying a risk of death of 29% to reverse flaccid unilateral facial paralysis and to sacrifice 28% of their remaining life or a risk of death of 25% to reverse unilateral moderate to severe postparalytic facial nerve syndrome and sacrifice 26% of their remaining life to reverse these conditions. These figures correspond with lower utility scores reported for chronic health states, including end-stage renal disease after renal transplant at 0.78,[48] stage II HIV infection at 0.75,[48] and cleft lip and palate at 0.84.[49] Substantially higher utility scores were reported for the post-facial reanimation health state compared with the flaccid unilateral facial paralysis health state (Table 4). Assuming that reanimation surgery increased the utility of the hemifacial palsy health state by 0.12 over the remaining mean life expectancy of 36 years, a net gain of 4.32 quality-adjusted life-years would result. With a cost-effectiveness threshold of $100 000 per quality-adjusted life-year,[22] facial reanimation surgery resulting in such a health utility improvement would be cost-effective at $432 000, assuming the procedure was 100% successful and without complications.

Limitations

This study has several limitations. First, survey participants were not entirely representative of the US population because they skewed younger and had higher educational levels. Second, as in other studies that examine the health utility of facial deformities,[20,49,50] the clinical vignettes in this study included a single adult patient example for each health state. Although we made efforts to minimize confounders between video vignettes by selecting individuals from a common race/ethnicity and avoiding extremes of age, we did not explicitly account for race/ethnicity, sex, age, and attractiveness, so care must be taken in extrapolating the results of this study. Multiple examples of varying degrees of facial palsy and reanimation outcomes across a wide spectrum of patients could be used with mixed-effects regression model analysis to account for such potential confounders, as previously reported.[8,51] Third, although this study provides insights into observer preferences for various manifestations of hemifacial palsy, it offers no definitive conclusions about the absolute utility values of these health states among those afflicted in the general population. The videos are available to readers, and they may be helpful in assessing the suitability of applying our findings to the readers' own patient population.

Conclusions

No differences in societal perception of flaccid unilateral facial paralysis and unilateral moderate to severe postparalytic facial nerve syndrome were demonstrable. The post-facial re-

animation state of a patient demonstrated substantially higher societal health utility than the flaccid unilateral facial paralysis and unilateral moderate to severe postparalytic facial nerve syndrome states. Further studies are required to characterize the utility of facial palsy health states across age, sex, and race/ethnicity spectrums as well as to clarify their economic burden and determine the cost-effectiveness of current management strategies.

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Study concept and design: All authors.
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