

Defining Utility Values for Chorea Health States in Patients With Huntington's Disease

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SUPPLEMENTARY MATERIAL

Table S1. Vignettes for Health States of Chorea

	<u>Severe chorea</u>	<u>Moderate/severe chorea</u>	<u>Moderate/mild chorea</u>	<u>Mild chorea</u>
General symptoms				
General	• You are anxious, irritable, and depressed • You are not interested in seeing friends or doing hobbies • You have trouble concentrating, multitasking, and learning new skills, and take more time to process what people are saying	• You are anxious, irritable, and depressed • You are not interested in seeing friends or doing hobbies • You have trouble concentrating, multitasking, and learning new skills, and take more time to process what people are saying	• You are anxious, irritable, and depressed • You are not interested in seeing friends or doing hobbies • You have trouble concentrating, multitasking, and learning new skills, and take more time to process what people are saying	• You are anxious, irritable, and depressed • You are not interested in seeing friends or doing hobbies • You have trouble concentrating, multitasking, and learning new skills, and take more time to process what people are saying

Movement symptoms				
Arm & hands	• Arms move all the time • Wild, uncontrolled arm-waving and irregular movements of hands, which are worse when you are stressed	• Arms move very often • Uncontrolled and irregular movements of arms and hands, but don't swing wildly in the air	• Arms move often • Hands are very fidgety and cannot sit still	• Arms move occasionally • Looks like normal fidgeting of your hands, and causes you to occasionally drop things
Legs	• Legs move all the time • Walking is very unstable, which makes you appear drunk sometimes (you were arrested once because a police officer thought you were drunk in public) • Fall frequently	• Legs move very often • Walking unstable • Fall often, and need a walker to help you walk	• Legs move often • Walking unstable • Fall occasionally • Need physical therapy for gait, and your participation in some of the exercises is limited	• Legs move occasionally • Sometimes make walking unstable • Lose balance occasionally but don't fall • Able to participate in rehabilitation exercises

Torso	• Torso jerks or sways from side to side often	• Torso moves throughout the day, some people think you are fidgeting	• Torso movement is infrequent, but worse with stress	• Torso is not affected by the disease
Face	• Uncontrollable, irregular, jerky movements of the tongue, face, or neck • Speech sounds slurred	• Eyebrows raise once in a while and mouth often stays open while speaking	• Eyebrows raise and nose scrunches occasionally	• Facial muscles tighten occasionally
Impact on daily activities				
Sitting	• Cannot sit still in a chair and often fall out or slide off	• Can sit in a chair without falling out of it	• Can sit in a chair without falling out of it	• Can sit in a chair without falling out of it
Sleep	• Movements interfere with you falling asleep and make you very restless at night • Sometimes fall out of bed; your mattress was put on	• Movements interfere with you falling asleep and make you restless at night. You wake up early when the	• Movements sometimes interfere with you falling asleep • Do not fall out of bed	• Movements occasionally interfere with you falling asleep • Do not fall out of bed

	the floor so you don't get hurt	movements bother your sleep • Do not fall out of bed		
Eating	• Difficulty swallowing makes it hard for you to eat; need to be very careful not to choke • Cannot feed yourself	• Some difficulty swallowing makes it hard for you to eat; need to be careful not to choke • Cannot feed yourself due to uncontrollable arm movements	• Swallowing is a little difficult; need to be careful not to choke while eating • Can feed yourself with minimal assistance	• Can swallow and eat with some adaptations to your diet • Can feed yourself without assistance, but sometimes drop your fork or cup
Daily activities	• Need help with all of your daily activities • It is difficult for your caregiver to help you get dressed, move to the	• You might need help with most or all of your daily activities • Your caregiver may need to help you get dressed, move to the shower, get out of	• Need help with some of your daily activities • You can get dressed, move to the shower, get out of bed, and go to the bathroom with only some assistance	• May need help with a few activities • You can get dressed, move to the shower, get out of bed, and go to the bathroom with minimal assistance

	shower, get out of bed, and go to the bathroom	bed, and go to the bathroom without difficulty		
Driving & working	• Cannot drive • Cannot work independently	• Cannot drive • Cannot work independently	• You can drive, but with some restrictions • Work accommodates for your disability	• Driving is not restricted • You can work independently

Table S2 Sensitivity Analysis Results^a

Utility values, mean (SD)	Illogical responses excluded ^b		All participants and responses included	
Severity of chorea	TTO ^c	VAS ^d	TTO ^e	VAS ^e
Severe	0.09 (0.53)	0.20 (0.18)	0.10 (0.54)	0.21 (0.18)
Moderate/severe	0.28 (0.49)	0.32 (0.19)	0.25 (0.52)	0.33 (0.20)
Moderate/mild	0.47 (0.49)	0.46 (0.20)	0.46 (0.51)	0.47 (0.20)
Mild	0.61 (0.46)	0.59 (0.21)	0.59 (0.47)	0.58 (0.21)

SD, standard deviation; TTO, time tradeoff; VAS, visual analogue scale.

^aVAS scores have been converted from a scale of 0 to 100 to a scale of 0 to 1 for easier comparison with TTO utility values.

^bAnswers that demonstrated a misunderstanding of the health states by rating a more severe health state better than a less severe health state have been removed from the sample; participants' logical answers have been retained.

^cIncludes responses from 189, 176, 182, and 194 participants for severe, moderate/severe, moderate/mild, and mild chorea, respectively.

^dIncludes responses from 197, 189, 189, and 199 participants for severe, moderate/severe, moderate/mild, and mild chorea, respectively.

^eIncludes responses from all 205 participants.

Table S3 TTO and VAS Utility Values by the Order of Health States Presented^a

Utility values, mean (SD)	Participants who saw health states from most to least severe ^b		Participants who saw health states from least to most severe ^b		<i>P</i> value ^c [A] vs [B]	
	[A] (n = 70)		[B] (n = 85)			
Severity of chorea	TTO	VAS	TTO	VAS	TTO	VAS
Severe	0.07 (0.56)	0.22 (0.18)	0.07 (0.49)	0.16 (0.16)	0.97	0.01
Moderate/severe	0.25 (0.56)	0.36 (0.20)	0.26 (0.44)	0.29 (0.17)	0.85	0.02
Moderate/mild	0.50 (0.52)	0.51 (0.22)	0.46 (0.43)	0.43 (0.18)	0.60	0.01
Mild	0.68 (0.42)	0.65 (0.21)	0.61 (0.39)	0.54 (0.19)	0.30	<0.001

SD, standard deviation; TTO, time tradeoff; VAS, visual analogue scale.

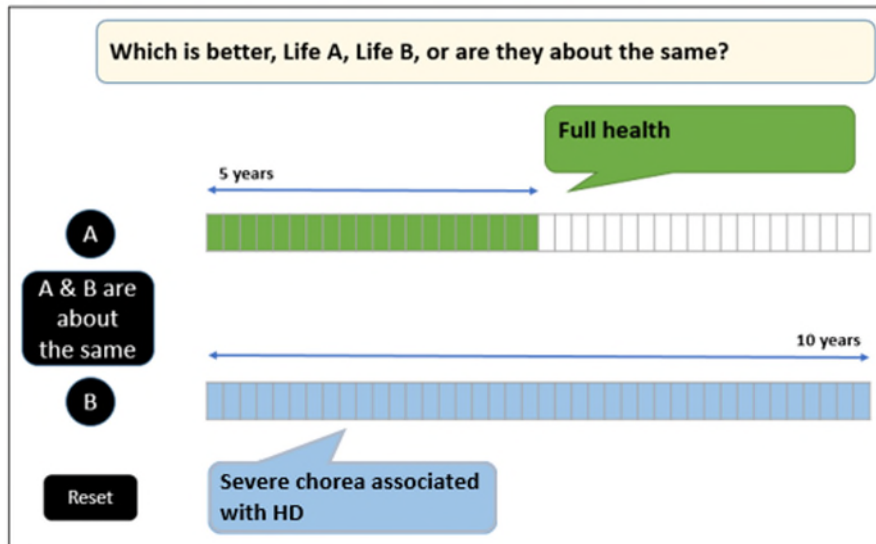
^aVAS scores have been converted from a scale of 0 to 100 to a scale of 0 to 1 for easier comparison with TTO utility values.

^bParticipants that demonstrated a misunderstanding of the health states by rating a more severe health state better than a less severe health state in the TTO or VAS have been removed from the sample.

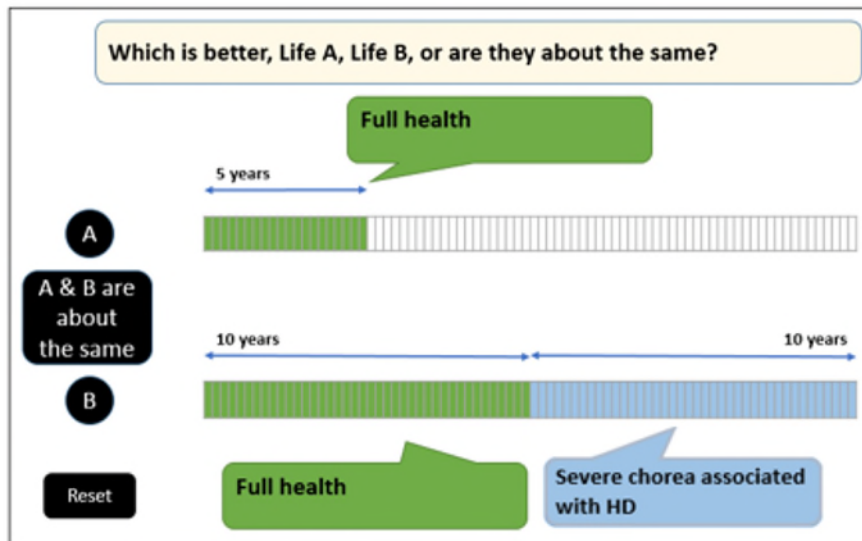
^c*P* values were obtained from two-sample *t* tests comparing the order by which health states were presented.

Figure S1 Example Visual Aids for Time Tradeoff (TTO) Questions in the Survey

a) Valuation of States Considered to be Better Than Death

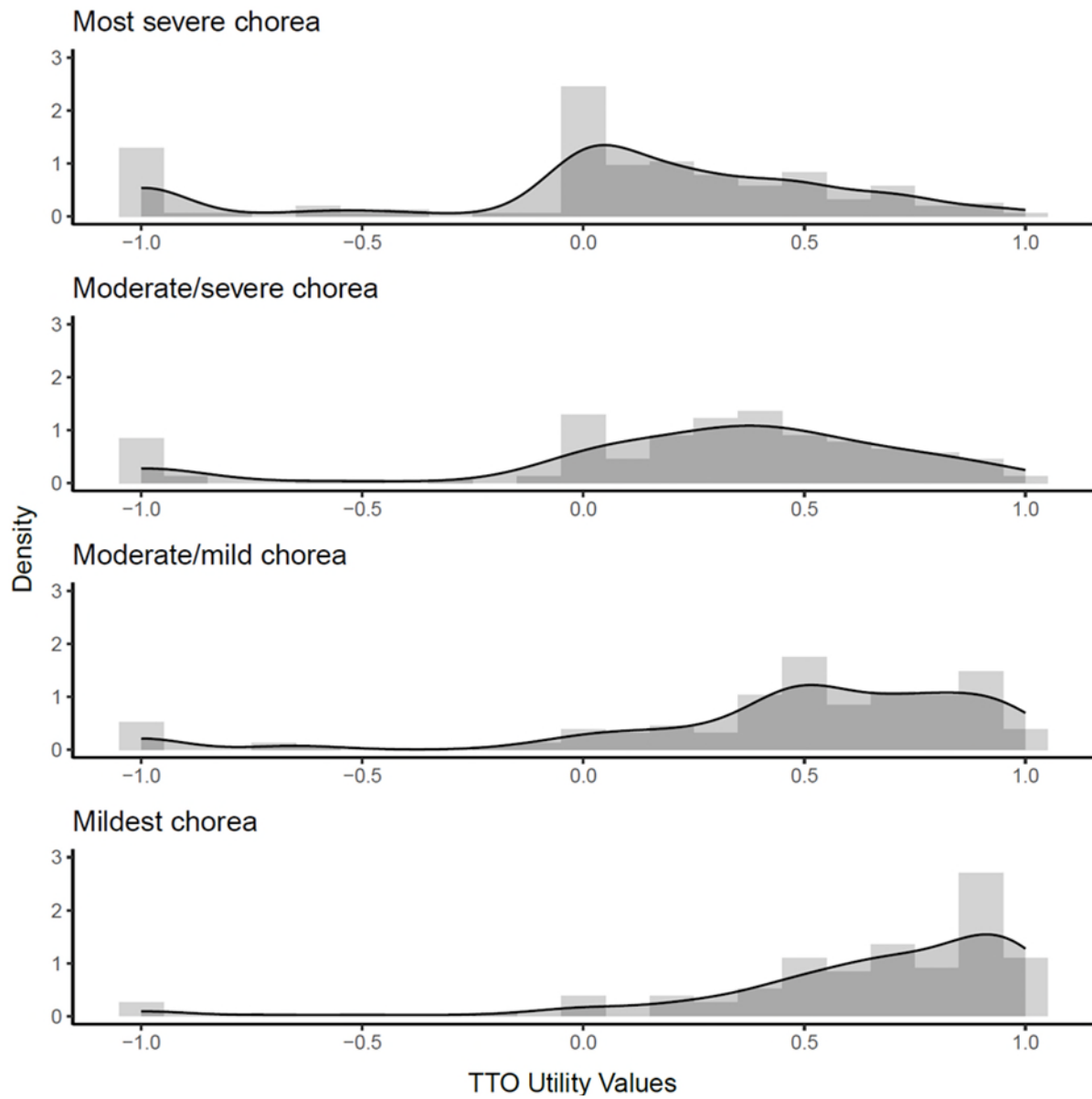


b) Valuation of States Considered to be Worse Than Death



HD, Huntington's disease.

Figure S2 Distribution of Time Tradeoff (TTO) Responses for the Primary Analysis (N = 155) by Health State



Supplementary Appendix

TTO Data Collection Procedure

Based on the EuroQol Valuation Technology (EQ-VT) protocol (Oppe M, et al. EuroQol protocols for time trade-off valuation of health outcomes. *Pharmacoeconomics* 2016; 34(10): 993-1004), the time tradeoff (TTO) data collection procedure was as follows:

- 1) Participants were first asked to choose between:
 - a. Life A: 10 years of life in full health followed by death, and
 - b. Life B: 10 years of life in the target health state followed by death.
- 2) Participants were asked to choose between:
 - a. Life A: 0 years of life in full health (i.e., immediate death), and
 - b. Life B: 10 years of life in the target state followed by death.
- 3) If participants chose life A in Task 2, then they were asked to choose between:
 - a. Life A: 5 years of life in full health followed by death, and
 - b. Life B: 10 years of life in full health followed by 10 years of life in the target health state, followed by death.

If participants chose life B in Task 2, then they were asked to choose between:

 - a. Life A: 5 years of life in full health followed by death, and
 - b. Life B: 10 years of life in the target health state followed by death.
- 4) Following Task 3, the procedure continued with 1-year incremental changes to life A, followed by a 6-month correction at preference reversal.

The iteration terminated when participants state preferential indifference.