HEALTH RELATED QUALITY OF LIFE (HRQOL) IN NARCOLEPSY & IH

A research update from the: Boston University Narcolepsy & Idiopathic Hypersomnia Patient Perspectives Study (BUNIHPPS)

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Definitions (for this presentation)

Hypersomnia: the condition of too much sleep

Hypersomnia condition: one of the 3 clinical diagnostic groups in BUNIHPPS (NT1, NT2, and IH)

Excessive Daytime Sleepiness (EDS): a symptom required for diagnosis of NT1, NT2, and IH

Sleepiness/Hypersomnolence: the experience of being more likely than most people to take naps during the day, sleep for long periods at night, or fall asleep at times that it is problematic to do so (for example, while driving, at work, or during a conversation).

Fatigue/Tiredness: states of having low energy or having trouble remaining on the same task but not necessarily falling asleep or sleeping too much



BUNIHPPS Methodology

Participants

- 833 adult patients with Narcolepsy Type 1 (NT1), Narcolepsy Type 2 (NT2), or Idiopathic Hypersomnia (IH)
- All participants in the Boston University Narcolepsy and Idiopathic Hypersomnia Patient Perspectives Study (BUNIHPPS), a cross-sectional online survey
- Data collected between October 10, 2015 and January 26, 2016

Survey Measures

- Epworth Sleepiness Scale (ESS)
- VR-36 HRQoL (Veteran's Rand 36; adapted from the first version of SF-36)
- PROMIS Satisfaction with Social Roles and Activities Questionnaire

Have you heard of BUNIHPPS before?

Recruitment Methods – Anonymous survey

- Facebook announcing recruitment, updates, and presentations/publications
- NN Conference 2015 Table Display with how to participate and paper surveys
- Narcolepsy Network, Wake Up Narcolepsy, and Hypersomnia Foundation email blasts and website announcements (NN and HF)
- 10 clinical sleep centers (including Stanford University)

Purpose

- Create a data set for dissertation
- Collect patient-reported data on numerous validated questionnaires, symptoms, sleep/wake behavior, time to diagnosis, and outcomes (HRQoL, satisfaction with social roles, & productivity)
- All variables collected together for greatest options in analysis; anonymity to enhance participation







	Ger	neral Dem	ogra	ohics for I	BUN	IHPPS	
	NT1 (n=338)		N	Г2 (n=210)		H (n=285)	
Variable	N	NT1	Ν	NT2	Ν	IH	p-value
Age	334	42.24 ± 14.78	205	39.21 ± 13.00	282	37.71 ± 12.58	0.0002
BMI	317	28.49 ± 8.22	198	26.40 ± 6.56	274	27.90 ± 7.15	0.0084
ESS	338	15.2 ± 5.0	209	14.2 ± 4.9	282	14.6 ± 4.6	0.0607
Gender	337		210		285		
Female		279 (82.8)		178 (84.8)		249 (87.4)	0 2834
Male		58 (17.2)		32 (15.2)		36 (12.6)	0.2004
Marital Status	338		210		285		
Married/LT		192 (58.4)		131 (63.3)		146 (52.1)	0.0446
Single		137 (41.6)		76 (36.7)		134 (47.9)	
Children	323		199		276		
Yes		178 (55.1)		104 (52.3)		123 (44.6)	0.0324
No		145 (44.9)		95 (47.7)		153 (55.4)	
Working Status	337		208		283		
Employed		189 (56.1)		159 (76.4)		172 (60.8)	<0.0001
Not Employed		148 (43.9)		49 (23.6)		111 (39.2)	
** p < 0.01; * p <	0.05. LT	= living togeth	er.				



Chi square analysis for individual conditions/disease groups; p < 0.05 for all reported "more" conditions.

	Та	ble 1.3: Pł	narmaco	ological T	reatmer	nt					
	Group To	otal (n=833)	NT1	(n=338)	NT2	(n=210)	IH (n=	285)			
Madia tian Variable											
Medication variable	N	Total (%)	Ν	NT1 (%)	Ν	NT2 (%)	Ν	IH (%)	p-value		
Currently Use Medication	803	86.9	285	89.6	184	88.0	229	83.0	0.0002		
Modafinil/armodafinil	325	39.0	131	38.8	90	42.9	104	36.5	0.3543		
Methylphenidate	142	17.1	65	19.2	29	13.8	48	16.8	0.2586		
Mixed amphetamine salts	201	24.1	68	20.1	72	34.3	61	21.4	0.0003		
Dextroamphetamine	58	7.0	31	9.2	9	4.3	18	6.3	0.0799		
Methamphetamine/ selegiline	14	1.7	7	2.1	2	1.0	5	1.8	0.6080		
Sodium oxybate	184	22.1	126	37.3	49	23.3	9	3.2	<0.0001		
Tricyclic antidepressants	14	1.7	11	3.3	1	0.5	2	0.7	0.0138		
SSRIs	130	15.6	63	18.6	33	15.7	34	11.9	0.0711		
Pregabalin	16	1.92	8	2.4	1	0.5	7	2.5	0.2105		
Other Medication	182	21.9	79	23.4	40	19.1	63	22.1	0.4878		
p < 0.05 bolded. Abbreviations: SSRI = selective serotonin reuptake inhibitor.											





HRQoL: Health Related Quality of Life

What is HRQoL?

How do those with hypersomnia disorders measure on HRQoL? How do NT1, NT2, and IH groups compare?

Una	djusted	Healt	h-related Q	uality	of Life (VR	-36)		
VR-36 Domain	Total (n=833)	N	Г1 (n=338)	NT	Γ2 (n=210)	II	l (n=285)	
	Ν	Ν	Mean ± SD	Ν	Mean ± SD	Ν	Mean ± SD	p-value
Physical Function	762	300	68.3 ± 28.4	197	75.0 ± 26.1	265	65.7 ± 26.1	0.0011
Role Physical	760	298	49.0 ± 30.5	198	50.6 ± 29.8	264	39.8 ± 30.2	0.0001
Body Pain	759	297	58.5 ± 24.8	199	60.3 ± 25.2	263	56.3 ± 25.6	0.2270
General Health	750	294	49.3 ± 24.5	194	50.4 ± 23.0	262	43.3 ± 23.8	0.0021
Vitality	753	296	26.4 ± 19.9	195	23.0 ± 18.0	262	16.5 ± 16.6	<0.0001
Social Functioning	759	297	47.2 ± 28.3	199	48.0 ± 26.5	263	40.7 ± 27.7	0.0056
Role Emotional	758	297	65.2 ± 30.7	198	62.9 ± 28.8	263	63.1 ± 32.6	0.6209
Mental Health	753	296	62.6 ± 20.1	195	56.7 ± 19.0	262	58.5 ± 20.8	0.0033
Physical Composite (PCS) ¹	746	293	39.8 ± 11.2	192	42.5 ± 10.9	261	37.5 ± 10.5	<0.0001
Mental Composite (MCS) ¹	746	293	39.2 ± 11.9	192	35.7 ± 11.2	261	36.6 ± 12.1	0.0029
p < 0.05 bolded. ¹ Scale scores made according to	o norm bas	sed sco	oring (standarc	lized t :	score transfor	mation	with a mean of	50 ± 10).

HRQOL Unadjusted Results

HRQOL Comparisons to the General Population

- NT1, NT2, and IH groups reported lower mental and physical health than the average population
- Composite scores under 50 indicate all diagnostic group's mean scores for physical and mental health were comparatively worse than the average scores for the general population
- MCS and PCS scores were between 35 and 42 between groups, with standard deviations of approximately 11, indicating that the cohort is functioning approximately one standard deviation lower than the average population on mental and physical health
- These results do not account for known differences in HRQOL that may be influencing the results, such as gender, age, BMI, and comorbidities



BUNIHPPS Participant Demographics by Diagnosis										
Variable		1 (n=338)	NT	⁻ 2 (n=210)	IH	(n=285)	p-value			
	Ν	NT1	Ν	NT2	Ν	IH				
Current Age	334	42.2 ± 14.8	205	39.2 ± 13.0	282	37.7 ± 12.6	0.0002			
BMI	317	28.0 ± 7.4	198	26.3 ± 6.3	274	27.8 ± 7.0	0.015			
ESS	338	15.2 ± 5.0	209	14.2 ± 4.9	282	14.6 ± 4.6	0.0607			
Gender	337		210		285					
Female		279 (82.8)		178 (84.8)		249 (87.4)	0.2834			
Male		58 (17.2)		32 (15.2)		36 (12.6)				
Currently Use Medication	338		210		285					
Yes		285 (89.6)		184 (88.0)		229 (83.0)	0.0002			
No		53 (10.4)		22 (12.0)		56 (17.0)				
No. of Comorbidities	338	2.9 ± 2.1	210	3.0 ± 1.8	285	3.0 ± 1.9	0.6134			
Physical Activity Level	290		191		258					
Regular		100 (34.5)		57 (30.0)		87 (34.0)	0 7204			
Occasional		102 (35.0)		66 (34.5)		85 (33.0)	0.7304			
Never		88 (30.5)		68 (35.5)		86 (33.0)				

Comparing Diagno	stic Groups	by Adjusted H	RQOL Domain	n Scores
VR-36 Domain	Overall p-value	NT1 vs NT2 p-value	NT2 vs IH p-value	NT1 vs IH p-value
Physical Function	0.0053	0.2662	0.0037	0.1489
Role Physical	0.0004	0.7937	0.0011	0.0038
Body Pain	NA	NA	NA	NA
General Health	0.0194	0.8147	0.0263	0.0736
Vitality	<0.0001	0.1542	0.0045	<0.0001
Social Functioning	0.0224	0.8640	0.0324	0.0721
Role Emotional	NA	NA	NA	NA
Mental Health	0.0240	0.0177	0.3298	0.3447
Physical Composite (PCS) ¹	<0.0001	0.0962	<0.0001	0.0245
Mental Composite (MCS) ¹	0.0306	0.0236	0.4004	0.3245
p < 0.05 bolded. ¹ Scale scores made according to mean of 50 ± 10).	o norm based s	coring (standardiz	ed t score transfo	ormation with a



Adjusted HRQOL Summary

NT1 vs. NT2

• Mental Health significantly differed, and NT1 had higher scores than NT2

NT2 vs IH

- Physical aspects of health significantly differed, with NT2 scoring higher on PCS, PF, and RP compared to IH
- Social Functioning and Vitality were significantly different, with NT2 scoring higher on both scales compared to IH

NT1 vs IH

- Physical aspects of health significantly differed, with NT1 scoring higher on PCS and RP compared to IH (PF no longer was significantly different)
- Vitality was significantly different, with NT1 scoring higher compared to IH







Conclusions

Lower HRQoL scores across physical and mental domains were reported by NT1, NT2, and IH patients compared to healthy US adults.

NT2 reported significantly lower mental health functioning (MH and MCS) despite having better physical functioning (PF, RP, and PCS) compared to NT1.

Those with the most reported physical activity had the highest HRQoL scores on physical (PF, PCS, and RP), GH, and SF domain scores with a minimum of 5 points separating each group's scores. A modest improvement in physical activity level may confer a clinically meaningful improvement in HRQoL among those with NT1, NT2, and IH.

The benefit of physical activity on HRQoL mental health (MH and MCS), VT, and RE domains was greatest in those who reported the most physical activity.

A physical activity intervention should be considered in addition to medication as a behavioral intervention in NT1, NT2, and IH.

Limitations

Methodology

- · Was not able to substantiate diagnosis since this was an anonymous survey
- Cross-sectional nature of the survey represents one time point in participants' lives, and changing attitudes and health status over time could not be collected
- Self-selection to participate without compensation may have resulted in bias based on the type of respondents who participated (i.e. female predominance)
- Data collection primarily from social media and patient advocacy organizations may also have biased data collected
- While purpose-driven, the study was created using more hypothesis-generating methods than necessarily hypothesis-driven methods, thus significant differences observed between groups should be replicated using other methodologies

Survey Measures

- All standardized questionnaires needed to be freely available or permissions in place for free use (VR-36)
- Additional questions related to sleep/wake behavior were not standardized

Thank You

1301 BUNIHPPS Participants

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