CHILDREN SLEEP HABITS PRIOR, DURING, AND AFTER ACTIVE OPSOCLONUS MYOCLONUS ATAXIA SYNDROME (OMAS): AN INTERIM ANALYSIS.

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Background and Objective:

Sleep disturbances are common in individuals with OMAS, persisting variably even during remission. This study aims to characterize sleep behaviors before, during, and after active OMAS to inform care strategies and identify potential long-term consequences of disease.

Methods:

Between Aug 2024 to Jan 2025, 121 respondents completed an abbreviated Children's Sleep Habit Questionnaire (CSHQ) for pre-OMAS (retrospective), active OMAS (retrospective or concurrently), and/or post-OMAS (concurrently) through the OMSLife Patient-Reported Registry hosted by the National Organization of Rare Disorders. [FIGURES 1-4] Final analysis set (FAS, n=83) was limited to caregivers of children <12 years old at symptom onset either with active (n=18) or in remission OMAS (n=65). Response scale for each of the 22 CSHQ questions was based on frequency of event but converted for consistency in analysis to 0 (desired behavior) to 4 (undesired behavior). Normality of response distribution assessed by Shapiro-Wilk. Repeated measures were evaluated via Wilcoxon signed-rank (2 timepoints) or Friedman with post-hoc Conover's test (3 timepoints). Fisher Exact or Chi-Square tests were used for assessing categorical variables between subsets.

FIGURE 1. Survey Instrument Selection

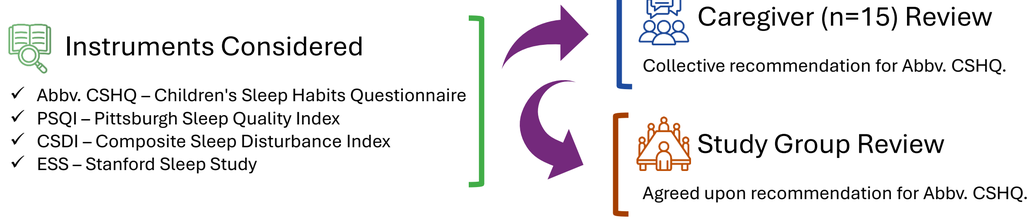


FIGURE 2. Abbreviated CSHQ (22 questions)

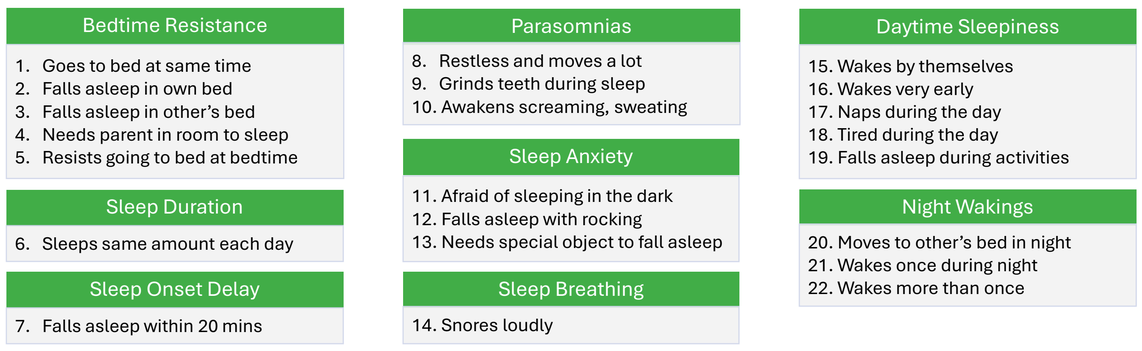


FIGURE 3. Study Schematic

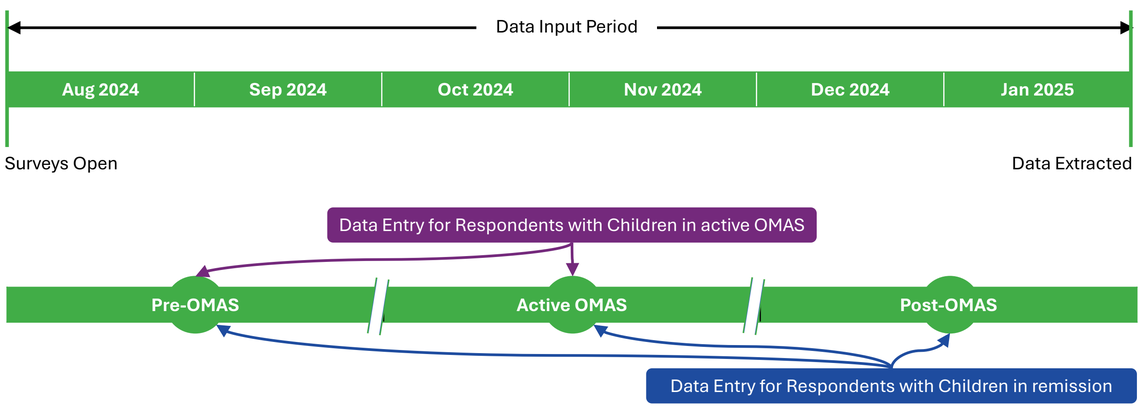
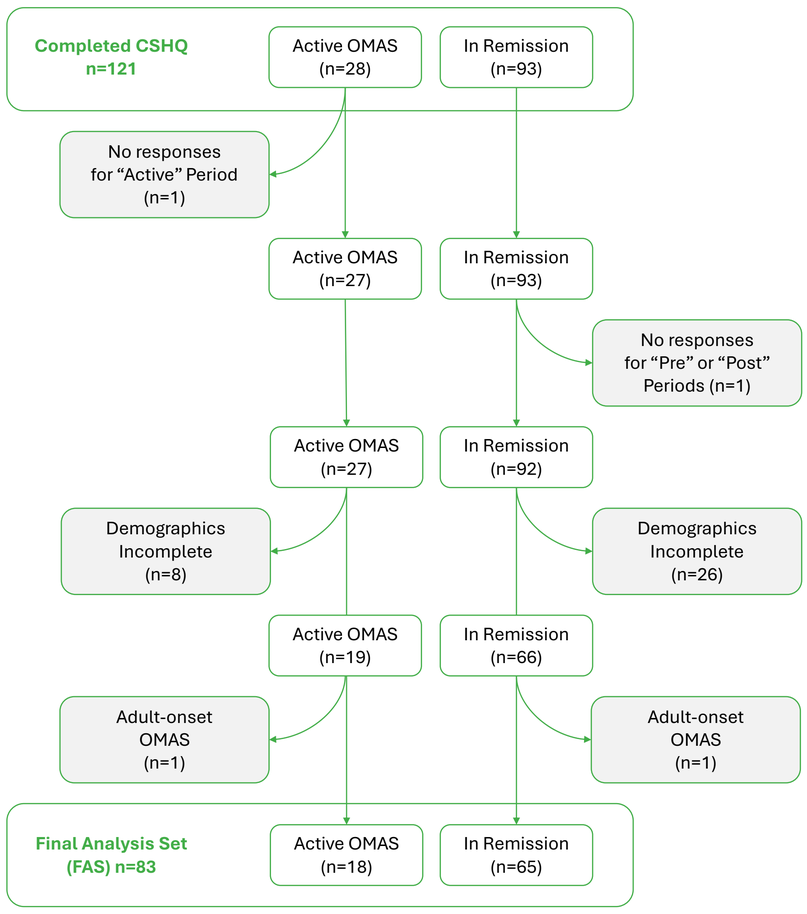


FIGURE 4. Respondent/Patient Disposition



Results:

Patient (n=83) characteristics: predominantly female (64%), white (86%), US resident (84%), and 0-3 years old at OMAS symptom onset (94%).

TABLE 1. Patient Characteristics at Time of Study

Characteristics	Active OMAS (n=18)		OMAS in Remission (n=65)		FAS (n=83)	
	n	%	n	%	n	%
Female	13	72%	39	60%	52	63%
Race						
Unspecified	0	0%	5	8%	5	6%
American Indian or Alaska Native	0	0%	1	2%	1	1%
Asian	1	6%	4	6%	5	6%
Black or African American	0	0%	1	2%	1	1%
White	17	94%	54	83%	71	86%
Ethnicity						
Unspecified	2	11%	17	26%	19	23%
Not Hispanic or Latino	15	83%	43	66%	58	70%
Hispanic or Latino	1	6%	5	8%	6	7%
Country Residence = US	15	83%	55	85%	70	84%
Insurance (US only)						
Medicaid (+/- Private)	4	27%	10	18%	14	20%
Medicare (+/- Private)	1	7%	3	5%	4	6%
Military (+/- Private)	0	0%	4	7%	4	6%
Private	10	67%	35	64%	45	64%
Other or Unspecified	0	0%	3	5%	3	4%
Total	15	100%	55	100%	70	100%
Age at Symptom Onset						
0-3 y	17	94%	61	94%	78	94%
4-6y	0	0%	3	5%	3	4%
7-11y	1	6%	1	2%	2	2%

Subsets were not significantly different for these characteristics at p≤0.05 by Fisher Exact or Chi-Square Tests.

Scores for were statistically higher (worsened, shaded red) for 18/22 questions and lower (improved, shaded green) for 1/22 question for active OMAS compared to pre-OMAS (n=70 to 83 respondents).

TABLE 2. Aggregate Response Scores between Pre- and Active OMAS (n=70 to 83)

Response Scores (0 desired to 4 undesired, based on habit frequency)	Pre-OMAS		Active OMAS		n	p*
	Mean (SD)	Median (q1-q3)	Mean (SD)	Median (q1-q3)		
1. Goes to bed at same time	0.77 (0.66)	1 (0-1)	1.78 (1.05)	2 (1-3)	81	< .001
2. Falls asleep in own bed	1.27 (1.37)	1 (0-2)	2.56 (1.43)	3 (2-4)	82	< .001
3. Falls asleep in other's bed	1.19 (1.44)	1 (0-2)	2.01 (1.74)	2 (0-4)	80	< .001
4. Needs parent in room to sleep	2.08 (1.51)	2 (1-4)	2.96 (1.36)	4 (2-4)	80	< .001
5. Resists going to bed at bedtime	1.13 (1.07)	1 (0-2)	2.4 (1.32)	2 (1-4)	80	< .001
6. Sleeps same amount each day	0.84 (0.66)	1 (0-1)	2.1 (1.08)	2 (1-3)	81	< .001
7. Falls asleep within 20 mins	1.01 (0.96)	1 (0-1)	2.42 (1.05)	2 (2-3)	81	< .001
8. Restless and moves a lot	1.58 (1.15)	2 (1-2)	3.36 (0.99)	4 (3-4)	77	< .001
9. Grinds teeth during sleep	0.33 (0.81)	0 (0-0)	1.23 (1.5)	0 (0-2)	70	< .001
10. Awakens screaming, sweating	0.35 (0.68)	0 (0-1)	1.96 (1.35)	2 (1-3)	80	< .001
11. Afraid of sleeping in the dark	1 (1.31)	0 (0-2)	2.04 (1.75)	2 (0-4)	76	< .001
12. Falls asleep with rocking	1.66 (1.47)	2 (0-3)	1.6 (1.59)	1 (0-3)	82	0.709
13. Needs special object to fall asleep	2.15 (1.71)	2 (0-4)	2.4 (1.74)	3 (0-4)	80	0.162
14. Snores loudly	0.43 (0.83)	0 (0-1)	1.01 (1.31)	0 (0-2)	79	< .001
15. Wakes by themselves	0.81 (1.06)	0 (0-1)	0.78 (1.15)	0 (0-1)	83	0.787
16. Wakes very early	1.49 (1.17)	1.5 (0.75-2)	2.53 (1.33)	3 (2-4)	80	< .001
17. Naps during the day	3.03 (1.36)	4 (2-4)	2.26 (1.38)	2 (1-3)	81	< .001
18. Tired during the day	1 (0.92)	1 (0-2)	2.58 (1.07)	2 (2-3)	79	< .001
19. Falls asleep during activities	0.33 (0.68)	0 (0-0.75)	0.71 (1.07)	0 (0-1)	78	0.002
20. Moves to other's bed in night	0.91 (1.15)	0 (0-1.5)	2.12 (1.68)	2 (0-4)	75	< .001
21. Wakes once during night	1.64 (1.22)	1 (1-2)	2.84 (1.17)	3 (2-4)	81	< .001
22. Wakes more than once	1.31 (1.22)	1 (0-2)	2.53 (1.32)	3 (2-4)	81	< .001

*Wilcoxon signed-rank

Scores for 20/22 questions for post-OMAS were statistically lower (improved, shaded green) compared to active OMAS (n=61 to 65).

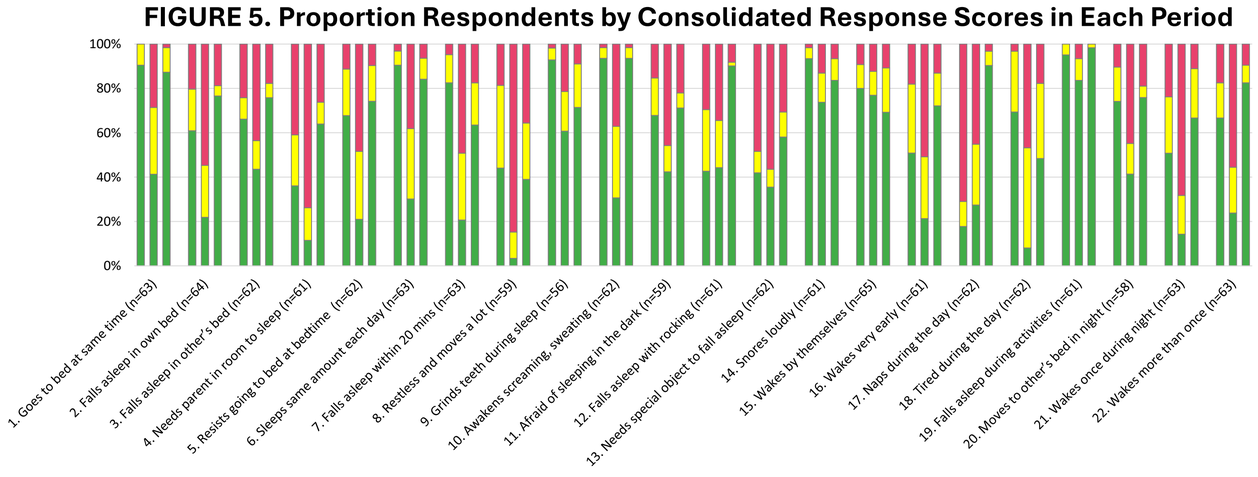
TABLE 2. Aggregate Response Scores between Active and Post-OMAS (n=61 to 65)

Response Scores (0 desired to 4 undesired, based on habit frequency)	Pre-OMAS		Active OMAS		n	p*
	Mean (SD)	Median (q1-q3)	Mean (SD)	Median (q1-q3)		
1. Goes to bed at same time	1.88 (1.11)	2 (1-3)	0.94 (0.61)	1 (1-1)	64	< .001
2. Falls asleep in own bed	2.59 (1.39)	3 (2-4)	0.95 (1.42)	0 (0-1)	65	< .001
3. Falls asleep in other's bed	1.98 (1.71)	2 (0-4)	0.94 (1.49)	0 (0-1.5)	63	< .001
4. Needs parent in room to sleep	3.02 (1.29)	4 (2-4)	1.29 (1.65)	0 (0-2.5)	63	< .001
5. Resists going to bed at bedtime	2.45 (1.3)	2 (2-4)	1.03 (1.11)	1 (0-1.25)	64	< .001
6. Sleeps same amount each day	2.17 (1.08)	2 (1-3)	0.97 (0.83)	1 (0-1)	65	< .001
7. Falls asleep within 20 mins	2.43 (1.13)	3 (2-3)	1.42 (1.18)	1 (1-2)	65	< .001
8. Restless and moves a lot	3.46 (0.89)	4 (3-4)	1.93 (1.33)	2 (1-3)	61	< .001
9. Grinds teeth during sleep	1.32 (1.56)	0 (0-2)	0.87 (1.25)	0 (0-2)	63	0.003
10. Awakens screaming, sweating	2.08 (1.37)	2 (1-3)	0.33 (0.64)	0 (0-0.25)	64	< .001
11. Afraid of sleeping in the dark	1.93 (1.76)	2 (0-4)	1.16 (1.46)	1 (0-2)	61	< .001
12. Falls asleep with rocking	1.8 (1.57)	2 (0-3)	0.48 (0.99)	0 (0-1)	61	< .001
13. Needs special object to fall asleep	2.42 (1.75)	3 (0-4)	1.52 (1.64)	1 (0-3)	62	< .001
14. Snores loudly	0.8 (1.17)	0 (0-1.25)	0.61 (0.95)	0 (0-1)	64	0.065
15. Wakes by themselves	0.83 (1.14)	0 (0-1)	1.05 (1.1)	1 (0-2)	65	0.165
16. Wakes very early	2.44 (1.35)	3 (2-4)	1.16 (1.26)	1 (0-2)	62	< .001
17. Naps during the day	2.27 (1.43)	2 (1-4)	0.52 (0.87)	0 (0-1)	64	< .001
18. Tired during the day	2.62 (1.02)	2 (2-3.5)	1.59 (1.04)	2 (1-2)	63	< .001
19. Falls asleep during activities	0.66 (1.07)	0 (0-1)	0.11 (0.37)	0 (0-0)	62	< .001
20. Moves to other's bed in night	2.2 (1.67)	2 (0-4)	0.92 (1.37)	0 (0-1)	61	< .001
21. Wakes once during night	2.84 (1.19)	3 (2-4)	1.23 (1.18)	1 (0-2)	64	< .001
22. Wakes more than once	2.55 (1.36)	3 (2-4)	0.86 (1.15)	1 (0-1)	64	< .001

*Wilcoxon signed-rank

Comparisons of same respondent entries for all 3 periods (for each question, L→R: pre-OMAS, active OMAS, post-OMAS). Score consolidation to 3 levels (scores 0 to 1 = positive [green], 2 = intermediate [yellow], 3 to 4 = negative [red]). For most assessments, favorable sleep habits (green) were experienced by fewer respondents during active OMAS compared to pre- and post-OMAS.

FIGURE 5. Proportion Respondents by Consolidated Response Scores in Each Period



Conclusion: In this interim assessment, overall sleep habits worsened during active OMAS and largely improved upon disease remission. Further analyses are needed, with consideration of other factors (age, time since active OMAS, etc.), to shed light on care practices that may hasten the return to favorable sleep habits upon remission.