

Request for Information: Request for Information: Soliciting Input on Areas of Health Disparities and Inequities in Neurological Disease and/or Care in the United States Across the Lifespan

RFI #: NOT-NS-20-026

Requesting Organization: National Institute of Neurological Disorders and Stroke (NINDS)

Response: Hypersomnia Foundation

Date: June 8, 2020

Brief Description of Gap in Research: NINDS/Office of Global Health and Health Disparities (OGHHD) in the Division of Clinical Research (DCR) is embarking on a strategic planning process to guide NINDS's research efforts in this space for the next 5-10 years. One input into this strategic planning process is to create a prioritized, comprehensive list of known health disparities and inequities in neurological diseases, treatment, and care. Second, NINDS is identifying knowledge gaps where new research investments will be critical to advancing the understanding and elimination of health disparities and inequities in neurological disorders.

1. Identify area(s) of health disparity and/or inequity (including race/ethnic, sex/sexual orientation/gender identity, regional/geographic, age, educational attainment, or socio-economic differences) in neurological disease, treatment, care, and/or service in the U.S.

The primary concern of the Hypersomnia Foundation in responding to this RFI is to determine whether racial/ethnic minority individuals in the U.S. are experiencing greater barriers in obtaining a central hypersomnia diagnosis and accessing treatment. We have noted that the patient community interacting with the Hypersomnia Foundation is overwhelmingly White and female. This is particularly problematic when recruiting for clinical trials – there are very few diverse individuals in our patient community to recruit.

The Hypersomnia Foundation maintains a global patient database (via CoRDS, Coordination of Rare Diseases at Sanford), which is used for research and recruiting for clinical trials. As of June 2020, there were 976 U.S. residents in the database who have idiopathic hypersomnia (IH) or Kleine-Levin syndrome (KLS) and have shared their racial/ethnic information. While the 2000 U.S. census data shows that 60.4% of U.S. residents are White, 92.8% of the IH/KLS patients in our database are White. Blacks/African Americans comprise 13.4% of the U.S. population, but only 1.7% of our IH/KLS patient database is Black. Hispanics comprise 18.3% of the U.S. population but only 3.4% of our IH/KLS patient database is Hispanic.

It is not yet known whether hypersomnias might affect races differently (*UpToDate*, 2020). Assuming they do not, we would expect that if people of all backgrounds had equal access to high quality healthcare, probability of obtaining a diagnosis, and socio-economic status (such that they had the

means to enter their data in our registry), then the racial/ethnic profile of our patient community would be a reflection of the U.S. population. We believe that lack of equal access to healthcare and bias in the exam room and elsewhere is preventing people of racial/ethnic minorities from ever obtaining an idiopathic hypersomnia or KLS diagnosis.

Central hypersomnias are believed to be more common in females than males. The U.S. population is 50.8% female, but the Hypersomnia Foundation's IH/KLS patient dataset is 84.8% female. We believe this difference is due to the known higher prevalence in females, although it is possible that males may experience more barriers in obtaining a diagnosis.

To further test for disparities in central hypersomnia diagnosis, we were interested to determine whether the racial and gender profiles of hypersomnia patients would be different in a population that had more equal access to high quality healthcare. The Veterans Health Administration (VHA) has a unique population in which people of all racial and ethnic backgrounds have more equal access to healthcare. The Hypersomnia Foundation approached Dr. Todd Bishop of the Veterans Administration Center of Excellence for Suicide Prevention to analyze the incidence of central hypersomnia diagnoses in the Veteran population.

Diagnostic and treatment utilization data was extracted for all Veterans receiving services between FY16 and FY19, including 72,592 Veterans who carried an ICD-10 diagnosis of a hypersomnia disorder in their medical record (i.e., idiopathic hypersomnia; primary hypersomnia; hypersomnia unspecified; hypersomnia not due to a substance or known physiological condition; and Kleine-Levin syndrome). Cases where the hypersomnia was attributed to a mental health or substance use disorder were excluded.

In contrast to the Hypersomnia Foundation database analysis, Dr. Bishop's analysis of the VHA population determined that hypersomnia disorders are disproportionately **increased** among individuals identifying as a racial or ethnic minority. In agreement with the Hypersomnia Foundation database analysis, the incidence of central hypersomnias in the VHA database is higher among females.

While females make up approximately 9.6% of all Veterans that utilize VHA services (National Center for Veterans Analysis and Statistics, 2020), they comprised 13.1% of hypersomnia diagnoses during FY16-FY19. Likewise, while Veterans identifying as Black/African American make up around 11.8% of the Veteran population, they represented 21.4% of hypersomnia diagnoses. The disparity is even greater when breaking hypersomnia disorders out by specific diagnoses. For example, female Veterans comprised 31.2% of cases of idiopathic hypersomnia, more than three-fold what would be expected if the rates of the disorder mirrored the demographics of the Veteran population.

Veterans frequently struggle with sleep disorders, which are among the most common presenting problems within Veterans Health Administration (VHA) primary care (Jenkins et al., 2015; Troxel et al., 2015). A significant, 50% increase in the number of Veterans diagnosed with a hypersomnia was observed between fiscal year (FY) 2000 and FY2010 (Alexander et al., 2016). Should current utilization and diagnostic trends continue, the VHA could expect to see disproportionate increases in sleep disorders, including hypersomnia disorders, among the Veteran population in the years to come. For

example, the number of female Veterans who engaged VHA services increased by 51.8% from FY08 to FY17, while the number of male Veterans only increased by 9.8% in the same time period (National Center for Veterans Analysis and Statistics, 2020). Similarly, the number of Veterans identifying as racial minorities, particularly those identifying as Black/African American or Hispanic, is projected to substantially increase between 2014 and 2043 (Department of Veterans Affairs, 2017).

The adverse impact on women and other minority Veteran populations takes on heightened significance when we consider the relationship of sleep disorders to the development and course of co-occurring disease processes. As sleep disorders are associated with the onset or exacerbation of conditions ranging from cardiovascular disease to depression and suicide, it stands to reason that those groups disproportionately impacted by sleep disorders will therefore bear the additional burden of these diseases as well, inasmuch that they are related to the underlying sleep disorder. Further, the early identification and treatment of sleep disorders is even more important now that research is uncovering a strong link between sleep problems and neurological disorders such as Alzheimer's disease, autism, schizophrenia and many more.

2. Identify determinants that help explain this/these health disparities and /or inequities

Little research has been done on the determinants of disparities in hypersomnia diagnosis and treatment specifically. We can only speculate based on anecdotal evidence from our patient community and from the research on disparities that has been conducted on other diseases with similarities to hypersomnias. We believe the following determinants contribute to the disparities in hypersomnia diagnosis and treatment:

1. Lack of equal access to high quality healthcare and adequate insurance coverage. As hypersomnias can lead directly to unemployment and loss of insurance coverage, the problem of equal access to healthcare and medication is particularly acute in our patient population.
2. Lack of access to primary care physicians and sleep specialists who identify as a racial or ethnic minority. It has been noted, for example, that a lack of diversity among physicians leads to health disparities (<https://hbr.org/2018/08/research-having-a-black-doctor-led-black-men-to-receive-more-effective-care>). In addition, field experiments show that when the doctor and patient have the same race, trust and communication increases, leading to better care outcomes.
3. Racial bias in medicine has been demonstrated in numerous studies. With an "invisible disease" like idiopathic hypersomnia, for example, providers may be prone to put off an extremely sleepy patient as being lazy or depressed instead of experiencing a significant neurological problem. While people of every racial background tell anecdotal stories of their sleep complaints not being taken seriously by doctors, it is possible that people who identify as a racial or ethnic minority experience even more difficulty getting doctors to believe that their sleep issues are real and require appropriate testing and diagnosis.

Data from the Hypersomnia Foundation's patient registry indicates that in the general U.S. population, hypersomnia disorders are disproportionately diagnosed in individuals who are White. In contrast, the

Department of Veterans Affairs medical record data suggests that among Veterans, hypersomnia disorders tend to be disproportionately diagnosed in individuals who identify as a racial or ethnic minority. Of the three potential determinants listed above, we know that Veterans have much better healthcare access and insurance coverage than the general U.S. population. We do not know whether Veterans have better access to doctors of their own race or if bias in VHA exam rooms is lower.

While the three determinants outlined above contribute to disparities in treatment of neurological sleep disorders, it is also worth mentioning here that sleep disorders, both common and rare, are likely dramatically underdiagnosed in ALL populations. Doctors receive, on average, only 2-3 hours of formal education on sleep during their many years of training, and most primary care physicians do not regularly screen their patients for sleep disorders. Most are also understandably unfamiliar with many rare neurological disorders and therefore may fail to refer potential patients to the sleep specialists that can diagnose and care for them. Until sleep disorder awareness in the PCP population is addressed, patients of all backgrounds will continue to suffer without diagnosis and treatment.

3. Identify evidence-based research strategies, health services, policies, and other interventions that address these disparities/inequities in neurological disease, treatment, service, or care

Some solutions to the problems of healthcare inequities are in great measure a matter of public policy and require redesign of the extremely complex healthcare and insurance industries. This is not our unique area of expertise and we must defer to policy experts to suggest changes that will solve the problems. For example, this article by Jamila Taylor of The Century Foundation, [Racism, Inequality, and Health Care for African Americans](#), suggests specific policy and program changes that are needed to reduce disparities for African Americans.

On the topic of primary care physician ability to recognize and respond to sleep disorders in their patient population, we believe there is a significant gap in research on this topic. How can PCP knowledge of sleep disorders be increased? Should there be universal sleep disorder screening of all primary care patients? What types of follow up tests and referrals are needed for those flagged during screening? Can PCPs adequately care for someone with a sleep disorder, or is the expertise of a sleep specialist required? Dr. Bishop's team within the VHA has led research efforts in this area in some regard, adapting full length CBTi (cognitive behavioral therapy for insomnia) into a brief, 4-session version that can be easily implemented within the primary care setting. Further, VHA has made great strides in regards to primary care mental health integration (PCMHI), one facet of which is to imbed behavioral health providers within the primary care setting. The PCMHI program represents a unique platform from which to deliver brief interventions aimed at addressing sleep disorders. In addition, members of the Hypersomnia Foundation's Medical Advisory Board are currently developing a behavioral approach aimed at improving quality of life among people with hypersomnias (Ong et al., 2020), which the VHA team and others may be able to adapt to use in the primary care setting.

An NCBI search on the early identification and treatment of sleep disorders returns very few published studies, but they all conclude that increased screening and responsiveness to potential sleep disorders is sorely needed:

- Research on the potential for sleep disorder screening in the primary care setting to greatly increase patients identified and referred for diagnosis and treatment: <https://www.ncbi.nlm.nih.gov/pmc/articles/PMC3041630/>
- Research demonstrating how referral to sleep professionals improves treatment as compared to treatment for sleep disorders solely by primary care physician: <https://www.ncbi.nlm.nih.gov/pmc/articles/PMC4678353/>
- Research demonstrating that pharmacies can be an effective place to screen for sleep disorders: <https://www.ncbi.nlm.nih.gov/pmc/articles/PMC4076224/>
- Sleep disorder screening programs in the workplace can also improve health and safety: <https://www.ncbi.nlm.nih.gov/pmc/articles/PMC6084754/>

4. Identify knowledge gaps about disparities/inequities in neurological conditions

We do not know if central hypersomnias are equally likely to occur among individuals of different races or ethnic backgrounds. Based on our preliminary data discussed above, we strongly suspect a significant disparity in diagnosis and treatment of hypersomnias between racial and ethnic populations. Based on available demographic data from the Hypersomnia Foundation's registry, we strongly believe that the problem of underdiagnosis and inadequate treatment is much more severe for racial/ethnic minority patients.

5. Identify potential approaches for addressing these neurological disparities and/or inequities through ongoing or new research collaborations or interventions

The diagnosing and treatment of sleep disorders in general and neurological sleep disorders in particular could be greatly improved by research and improvements in clinical practice. If sleep screening was as common as blood pressure screening, and PCPs were better trained to identify, treat and refer the estimated 10% of Americans who have sleep problems, millions of Americans would live healthier lives.

Additional epidemiological research is certainly warranted in order to obtain additional clarity on the etiology of hypersomnia disorders. A better understanding of the disease can only lead to improved diagnostic capability. Development of biomarkers and improved testing techniques could greatly improve diagnostic certainty, provided that barriers to receiving the tests are addressed so that the undiagnosed, including racial/ethnic minorities, may benefit from the new technology.

One methodological approach that has yielded useful findings for the treatment of other disease processes is an examination of the potential heterogeneity of treatment effects that may be observed for existing hypersomnia treatments. Patients may differentially benefit from available hypersomnia

interventions based on several individual or group characteristics. Were information regarding these effects made available to patients and providers, each would be able to make more informed decisions regarding patient care. Further, such knowledge may help to tailor existing, or develop novel, interventions aimed at reducing hypersomnia symptoms and improving quality of life in the adversely impacted patient population.

6. Additional comments

None.

7. Post-submission addition

On June 30, 2020, the Hypersomnia Foundation contacted the administrator of this RFI and requested permission to add information about a relevant clinical trial. On July 2, 2020, the administrator confirmed that the following citation and comments were added to this submission.

Here is the link to the clinical trial: <https://clinicaltrials.gov/ct2/show/NCT03018912>

Our response to the RFI stated that we believe bias in the exam room to be a contributing factor to disparities in the diagnosis and treatment of neurological sleep disorders. Asking specific questions and applying algorithms consistently to all patients who enter the doctor's office could be an excellent method of reducing bias.

I do not believe the trial was designed to reduce bias in the diagnosis of sleep disorders. However, it would certainly be possible to bring in racial/ethnic data to see if the use of the sleep vital sign and referral algorithm reduced disparities.

References

Alexander M, Ray MA, Hébert JR, Youngstedt SD, Zhang H, Steck SE, Bogan RK, Burch JB. The National Veteran Sleep Disorder Study: descriptive epidemiology and secular trends, 2000–2010. *SLEEP* 2016;39(7):1399–1410.

Jenkins MM, Colvonen PJ, Norman SB, Afari N, Allard CB, Drummond SP. Prevalence and Mental Health Correlates of Insomnia in First-Encounter Veterans with and without Military Sexual Trauma. *Sleep*. 2015;38(10):1547-1554.

Minority Veterans Report: Military Service History and VA Benefit Utilization Statistics. Data Governance and Analytics, Department of Veterans Affairs, Washington, DC. March 2017

National Center for Veterans Analysis and Statistics. *VA Utilization Profile FY 2017*. May 2020. Data accessed on June 8th, 2020 at:
https://www.va.gov/vetdata/docs/Quickfacts/VA_Utilization_Profile_2017.pdf

Ong, J C, et al. "0754 Cognitive Behavioral Therapy for Hypersomnia (CBT-H): A Feasibility Study for Improving Health-Related Quality of Life." *Sleep*, vol. 43, no. Supplement_1, 2020, doi:10.1093/sleep/zsaa056.750.

Troxel WM, Shih RA, Pedersen ER, et al. Sleep in the Military: Promoting Healthy Sleep Among U.S. Servicemembers. *Rand Health Q.* 2015;5(2):19.

UpToDate, www.uptodate.com/contents/idiopathic-hypersomnia.