Prevalence Of Periodic Limb Movement Disorder In Intellectually Disable Population

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Abstract: Background: Periodic limb movement Disorder (PLMD) is defined as presence of repetitive limb movements during sleep, seen in 4-11% in general population. PLMD is known to cause sleep fragmentation leading to daytime consequences. Intellectual developmental disorder is a disorder known to have intellectual and adaptive functioning deficits. In case, PLMD is present in Intellectual disability then the problems faced by such population further magnify. Method: Local areas of Thane and Nasik were approached. After consent from parents 27 participants underwent nocturnal polysomnogram. Scoring of the study was done as per AASM criteria. Result**:** Participants positive on PLMD were 51.48%. Polysomnography findings showed TST 6hrs, sleep onset 38.35 mins, wake after sleep onset 42.23 mins, Stage 1-1.6%, Stage 2- 49.35%, REM- 17.82%, Sleep Efficiency- 77.5%. Conclusion**:** The paper highlights results pertaining to determining prevalence of periodic limb movement disorder in intellectually disabled population by polysomnography. The results have qualitatively improved perception of practitioners of medicine as well as parents and guardians of intellectually disabled persons with regard to physical disorders among them. [Irani N, Natl J Integr Res Med, 2019; 10(6):1-5]

Key Words: Intellectually Disabled Population, PLMD, Sleep disorders, polysomnography, stages of sleep.

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Introduction: Sleep is not state of unconsciousness but rather there are controlled set of neurobiological events with REM sleep showing signs of brain activity similar to that of waking state[1]. Both, a good quality & quantity of sleep are essential for physical, emotional, and cognitive development[2]. Sleep disorder/s are known to cause daytime sleepiness[3], abnormal neurobehavioral functioning[3], being anxious or depressed[3], weight gain[3], self-injury especially in case of children[4,5]. One such major sleep disorder is Periodic Limb Movement Disorder (PLMD), which is characterized by repetitive movements, mostly observed in lower limbs along with brief muscle twitches, jerking movements or an upward flexing of the feet. PLMD episodes may last from a few minutes to several hours[6]. PLM index is defined as number of movements for each hour of total sleep time, as determined by polysomnography; an index of 5 or more is regarded as abnormal[7].

Intellectual disability is a disorder with starts during developmental period that will include both cognitive and adaptive functioning deficits in conceptual, public, and practical domains[8]. Prevalence of Intellectually Disabled population is observed in 1% of the general population and its prevalence differs based on the age. In India as per The National Sample Survey Organization, NSSO prevalence of mental retardation in rural areas is 31.0 per 1000 while in urban are 9.0 per 1000. The DSM-IV-TR (APA, 2000) estimates the gender ratio in the population of Individuals with MR to be 1.5:1 with males having a higher incidence than females.

Mentally challenged population has been observed to have sleep disturbances which are constantly prevalent which have also shown to be affecting family members and care takers[9, 10]. Questionnaire and interview based studies among the mentally retarded show that approximately 15–50% of adults[7,11] and 30–67% of children[12,13] have sleep problems. Sleep problems in such population are referred to more as “behavior problems” rather than a diagnosis in such population. Literature survey on PLMD and intellectual disabled population reported children with PLMD had significantly lower percentage of rapid eye movement (REM) than control[14].

Association of PLMD and ADHD was made evident by Chervin *et. al*. by questionnaire based study and found a direct correlation between PLM scores and inattention/ hyperactivity scores[12]. There are no clinical symptoms for predicting PLMD and thus polysomnographic studies remain gold standard for diagnosis of PLMD in patients with insomnia or hypersomnia.

At times underlying cause of insomnia or bedtime resistance due to PLMS may go unnoticed due to patients being unaware of their child’s limb movement and may attribute to their bedtime behavior or low IQ. There have not been many studies which discuss about periodic limb movement in intellectually disabled. To our knowledge this is first polysomnogram based

study in intellectual disabled population in India specifically looking at PLMD.

Materials And Methods: The study protocol was approved by the Ethics Committee of International Institute of Sleep Sciences located in Thane district of Maharashtra state, India. Three centers of India in local areas of Thane and Nasik were approached which are working with intellectually disabled students. In all, 275 students were addressed for the study. The purpose and procedure of the study was explained to authority in-charge and parents; which were later called for the meeting. After getting consent from the authority and parents/caretakers, students were enrolled for the study. As we wanted to observe prevalence of PLMD in Intellectual Disable we took representative of general population and thus inclusion criteria were not much stringent. Parents those giving consent and subjects being physically fit to undergo polysomnogram were considered based on evaluation by a Sleep Specialist. Exclusion criteria were subject's with 60yrs and above as elderly populations. Participants were allowed to take his/her time to sleep instead of sedating them.

Nocturnal Polysomnography included of Electroencephalogram (EEG) (according to the International 10–20 System) C3-M2; C4-M1; Cz-M2; O2-M1; O1-M2; F3-M2;F4-M1; Fz-M2; T3-M2; T4-M1. Electro-occulogram (EOG left and right eyes), Electrocardiogram (ECG left central and right central), sub-mental electro-myogram (EMG), inter-costal EMG, leg EMG (cross referenced over the anterior tibialis muscle of both legs specific to diagnose PLMD), respiratory airflow (thermistor signals), respiratory effort (thoracic movement and abdominal movement), body position, arterial oxygen and snore sensor (microphone taped in place on the neck near the larynx). Recordings were taken from bedtime to awakening in the morning. Sleep stages were manually scored according to AASM guidelines. Criteria for scoring PLMD was PLM last duration of 0.5 to 5 seconds which occur within 5 to 90secs of each other and at least 4 of this movement occur in a series. Required amplitude of these limb movements is at least 8 uV higher than the resting EMG amplitude[5].

Results: Table 1 a) & b) shows demographic details and polysomnographic findings of total ID participants respectively. Individuals with PLMI ≥ 5 were 14 (51.85%). Results of demographics and polysomnographic findings for children positive for PLMD are noted in table 2 (a) and 2 (b) respectively. Micro arousal index when compared with those negative for PLMI showed statistical significance with p= 0.04.

Table 1 (a): Demographics

No.	Variable	Demographic details
1.	Age (in yrs)	17.88 ± 9.61(4-43yrs)
2.	Sample size (N)	27
3.	Gender	Males (17)- 62.96% Females(10)- 37.03%
4.	Body mass index (BMI), kg/m2	7.66

Table 1 (b): Polysomnographic Findings:

No.	Variables	Mean ± S.D.
1.	Total Bedtime (TBT), mins	430.59 ± 45.13
2.	Total Sleep Time (TST), mins	337.85 ± 74.80
3.	Sleep onset (SOL), mins	51.22 ± 63.23
4.	Wake after sleep onset, mins	35.23 ± 33.92
5.	Stage 1, %	1.51 ± 1.47
	Stage 2, %	48.86 ± 15.48
	Stage 3, %	38.07 ± 12.56
	REM, %	15.29 ± 17.78
	Micro arousal index, /hr	12.4 ± 17.68
	Sleep Efficiency, %	± 15.98

Table 2 (a): Demographics:

Variable No.	Variable	PLM>5 /Hr Mean± SD
1.	Age (yrs)	15.85 ± 8.4 (4-30yrs)
2.	Gender	Male- 6(42.85%) Female-8 (57.14%)
3	Body mass index (BMI), kg/m^2	21.04 ± 4.45

Table 2(b): Findings of evaluation of sleep architecture in PLMD positive subjects

Variable No.	Variable	PLM>5 /HR Mean± SD
1.	Total Bedtime (TBT), mins	417.71 ± 40.02
2.	Total Sleep Time (TST), mins	329.5 ± 65.34
3.	Sleep onset (SOL) ,mins	38.35 ± 35.9

4.	Wake after sleep onset (WASO), mins	42.23 ± 34.57
5.	Stage 1, %	1.6 ± 1.3
6.	Stage 2, %	49.35 ± 15.45
7.	Stage 3, %	38.03 ± 12.74
8.	REM, %	17.82 ± 23.03
9.	Micro arousal index	17.77 ± 23.39
10.	Sleep Efficiency, %	77.5 ± 11.22

Discussion: Children with ID are usually neglected in the medical field. This is mainly because of their poor ability to communicate and social disadvantages[15]. They are frequently excluded from clinical, etiologic, and treatment studies[15]. Most polysomnogram studies in this population have focused on Obstructive Sleep Apnea and not on PLMD. To our knowledge this is first study which will create awareness about prevalence of Periodic Limb Movement Disorder (PLMD) in Intellectual Disabled.

Our results show that overall prevalence of PLMD is significantly higher 51.85 % v/s 4- 8 % in general population[16] refer Table 1 (b). As observed in table 2.b the prevalence was found to be higher in subjects age group of 4 to 30 yrs of age with female in age group of 20 to 30yrs observe to be more positive. Possible explanation for this could be Iron deficiency secondary to menstrual cycle[17]. Also PLM can be due to an under activity of dopaminergic function in certain central nervous system pathways which needs to be studied in this population.

However micro arousal index was significantly elevated in PLMD population denoting fragmented sleep. Non-restorative sleep due to sleep fragmentation from micro-arousal is known to cause daytime sleepiness[18]. Sleep quality in PLMD is impaired to such a degree that it is known to cause serious consequences such as daytime sleepiness, hypertension, autonomic alteration anxiety disorder, depression, affects fine motor activity, mental performance, numerical memory, attention variability and hyperactivity[19,20]. Intellectual Disability which already known to have deficit in adaptability of which one includes fine motor skill, intellectual functioning is learning memory and onset of both these deficits at an early age. As PLMD have also been reported to risk factor for hypertension[21] one of probable reasons for early mortality rates in ID could be due to PLMD.

Bedtime resistance and nighttime awakenings are usually attributed to bedtime behavior problems and not paid much attention upon[22]. Sleep initiation and fragmentation can be result of many different factors[11]. Infact, increased PLM in children, even in the absence of restless legs syndrome, may be associated with insomnia and sleep disturbances[23]. At times underlying cause of insomnia or bedtime resistance due to PLMS may go unnoticed due to parents being unaware of their child's limb movement as their ward may not understand or able to express their symptom.

Study carried out by Sforza *et.al.* showed that PLMD occurrence is highly reliable across nights suggesting that a single night is sufficient to confirm diagnosis and associated sleep disturbances in these patients[24]. Hence one night study was enough to study PLMD episode in such population. Also Polysomnography studies which is gold standard measurement for diagnosis of PLMD to prove the disorder objectively[25].

This is first record of polysomnography studies in India on intellectually disabled Indian population. The studies highlights efforts taken to create awareness among parents and practitioners regarding sleep disorder prevalent in such population and also to stress the fact that it is not often that the obstructive sleep apnea is the main cause of sleep fragmentation. Also, studies suggest that parents and teachers should pay attention that daytime sleepiness or inactiveness during the day could be due to sleep disorder.

It is imperative that the study had limited number of participants due to economically challenging situations that the parents/ guardians of intellectually disabled persons undergo. Correlation between low ferritin level and PLMD has been well documented. We did not study ferritin level in our population.

Conclusion: Our studies show much higher prevalence of PLMD in intellectually disabled cohort under study than in normal population. It is associated with increased micro arousals, fragmented sleep and daytime behavioral consequences.

Limitation: our evaluation studies were largely of observational nature. More studies are needed to complement our findings. Intervention studies treating PLMD should be undertaken to see if the daytime behavior improves significantly.

Polysomnographic studies before and after treatment to objectively measure any difference is measured in various sleep parameters. The results have qualitatively improved perception of practitioners of medicine as well as parents and guardians of intellectually disabled persons with regard to physical disorders among them.

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