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EXTRANODAL NON-HODGKIN LYMPHOMA MASQUERADING AS OBSTRUCTIVE SLEEP APNEA

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Introduction: Presentation of patients with Non Hodgkin’s Lymphoma (NHL) could either be acute or subacute. In children, NHL is primarily an extranodal disease. Most frequently involved systems are the gastro intestinal tract, Waldeyer’s ring, lung, liver, spleen, bone and skin. Diffuse large B-cell lymphoma is a mature B-cell neoplasm that represents 10% to 20% of pediatric NHL with better prognosis than adults. Here we report an uncommon presentation of tonsillar lymphoma as obstructive sleep apnea.

Materials and methods: An 11-year old boy presented to Ear, Nose and Throat-Head & Neck Clinic with a 3 week history of worsening loud snoring and apnea in supine position. He needs to sleep in propped up position. There were also symptoms of sorethroat, odynophagia, dysphagia and halithosis. On examination he was a thin boy with mouth breathing and had a muffled voice. Oral examination revealed a right tonsillar hypertrophy with bluish discolouration which occluded the airway. Neck examination revealed shotty cervical neck nodes.

Results: Flexible nasopharyngeal scope examination showed mild hypertrophic adenoids with an enlarged right tonsil occupying the anterolateral part of the oropharynx. A simple oxygen monitoring was performed in supine position revealed clusters of desaturations more than 85% > 3. Computed tomography scan of the neck showed highly vascularized right tonsillar hypertrophy and adenoid enlargement. Magnetic resonance imaging of the neck has rule out tonsillar hemangioma and showed bilateral hypertrophic tonsils in which the right was more prominent than the left. He underwent an uneventful tonsillectomy. Histopathological examination reported as diffuse large B-cell lymphoma, germinal centre phenotype. He was then referred to haematologist for further treatment.

Conclusions: This case highlights the importance perioperative management of obstructive sleep apnea with suspected tonsillar tumour.

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TWO STAGE SURGERY IN UNRESOLVED PAEDIATRIC SLEEP APNEA (A CASE REPORT)

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Introduction: OSA in children is most commonly associated with adenotonsillar hypertrophy. The recommended initial treatment consists of surgical removal of the adenoids and tonsils. However, not all children who undergo adenotonsillectomy (T&A) for OSA are cured. Persistent obstructive sleep apnea (OSA) is demonstrated in 40% of children after adenotonsillectomy. One of the reason for persistent obstructive symptom is lingual tonsillar hypertrophy. Here we share our experience managing lingual tonsillar hypertrophy.

Materials and methods: A 9-year-old girl presented to our clinic for snoring and witness apnea more than 6 months duration. She does not have any history of runny nose, blocked nose or any ear symptom. On examination she is a thin girl with neither syndromic or micrognathic features. Tonsil and adenoids were enlarged. Sleep study showed features of severe OSA in polysomnogram. An early adenotonsillectomy was performed and postoperative recovery was uneventful. Repeat sleep study 6 months later still showed persistent severe OSA with AHI >9. Drug Induced sleep apnea assessment was performed and revealed lingual tonsil hypertrophy. Subsequently she undergone coblator assisted lingual tonsillectomy and postoperative recovery was uneventful.

Results: At day 14 post operation she had torrential bleeding from the oral cavity. Emergency exploration was performed to secure the bleeding. Blood was trasfused as she had 2 gram of blood loss. The bleeding was arises from raw areas of the lingual tonsil region. There was no bleeding points noted. Direct laryngoscope was performed with difficulty as there was pooling of blood. The base of tongue region was packed with adrenaline to reduced bleeding. Cauterization was done over the raw areas and bleeding was stop. She was discharged well and currently free from sleep apnea.

Conclusions: Adenotonsillectomy is still the main treatment for OSA in children. Lingual tonsil hypertrophy should be considered in all cases of OSA if symptom persist after surgery. Delayed complication can be fatal.

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SETTLING AND WAKING BEHAVIOURS IN CHILDREN WITH SMITH-MAGENIS AND ANGELMAN SYNDROMES

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Introduction: Smith-Magenis (SMS) and Angelman syndromes (AS), two rare genetic syndromes associated with intellectual disability have a prevalence of 1 in 15,000 and between 1 in 10,000 and 1 in 40,000 respectively. Difficulties with sleep onset and night waking are more common in individuals with these syndromes, compared to both typically developing and heterogenous intellectual disability peers. The heightened prevalence in these syndromes suggests additional internal and environmental causative and/or maintaining factors of sleep difficulties which warrant further investigation. Little is known about the behaviours children with AS and SMS demonstrate when settling to sleep and during night waking, though insight into these would improve understanding of the potential causative and/or maintaining factors of the sleep difficulties.

Materials and methods: Videosomnography and actigraphy data were collected for 12 participants with AS (M chronological age = 8.01) and 11 with SMS (M chronological age = 8.80). Children with SMS showed greater adaptive functioning (M=65.90, SD=11.44) than children with AS (M=47.16, SD=7.94). The groups were well matched on number of nights of footage (t(21) = 1.575, p=.130), and mean sleep duration across these nights (t(21) = 2.073, p=.051). Settling (30 minutes prior to sleep onset) and night waking were coded for twenty-one behaviours using a live coding scheme, relating to pain, challenging behaviour and interactions with caregivers. In total, 173 clips were coded (5,190 minutes).

Results: Inter-rater reliability for the behavioural codes was good (mean κ = 0.74; range 0.54-1.00). Significant differences in behaviours shown during settling and waking between groups and between individuals were obtained. Lag sequential and Yule’s Q analyses demonstrated that children and parents used a hierarchy of behaviours when attempting to re-settle to sleep after waking.

Conclusions: The results highlight indicators of pain and environmental factors which may differentially maintain sleep problems in children with AS and SMS. These differences have implications for the underlying aetiology of sleep problems in AS and SMS and potential intervention approaches.

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STUDY WITH POLYSOMNOGRAPHY, MULTIPLE SLEEP LATENCY TEST AND ELECTROENCEPHALOGRAPHY ACTIVITY IN KLEINE-LEVIN SYNDROME: A CASE REPORT

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Introduction: Kleine-Levin syndrome is included in the group of Central Disorders of Hypersomnolence in the International Classification of Sleep Disorders (ICSD-3). It has been defined as a severe recurrent hypersomnia that also associates hyperphagia and hypersexuality. However, symptoms such as apathy or derealization in these patients are increasingly described instead of hyperphagia and hypersexuality. Actually, there are no criteria that define typical polysomnographic or EEG characteristics in this group of patients.

Materials and methods: We present a clinical case of a 15-year-old boy with Kleine-Levin syndrome. The patient presents severe periodic hypersomnia episodes since he was 12. The first episode started with severe hypersomnia (17 hours sleeping) accompanied by confusion, apathy and derealization, lasting 5-6 days and repeating at 40 to 75 days interval. Some episodes are accompanied by hyperphagia and he has gained 15 Kg from the first episode. Hypersexuality has never occurred and the amnesia of the episodes is repeated in all of them. Between episodes he is asymptomatic and the day before he has hypersomnia he is able to perceive derealization and confusion. In some episodes he has also presented headache. Blood test, LCR and MR was normal. SPECT findings were a right fronto-temporal hypoperfusion during symptomatic episodes. Polisomnography and Multiple Sleep Latency Test (MSLT) has been performed in different days of different episodes: first, second and third-fourth day from the onset of the symptoms. Sleep latency was reduced in all three studies. MSLT mean sleep latency was normal in the study of third-fourth day, whereas in the studies of the first and the second symptoms day it was short. REM was present in two subtest just in the study of the second day. The main difference between studies in different days was the EEG activity. Background activity was 7.5-8Hz in three studies. During sleep-wake transition and light sleep occurred high-amplitude low-frequency waves mainly in the bilateral frontal and frontotemporal areas. This activity was more expressive and persistent in the study of the second day and very isolated on the first day. The actual treatment in this patient is lithium. In the last episode, Amantadine was tried on the first symptom day in order to abort the episode without success.

Conclusions: EEG, Polysomnography and Multiple Sleep Latency Test findings could be a useful tool in the diagnosis of Kleine-Levin if we correlate it with the clinical course and the day in which it is performed from the onset of symptoms. In our case, the second day from the onset of symptoms was the best suited to find the findings described in Kleine-levin patients.
Introduction: Sleep is important for learning, memory and underlying neural plasticity. Studies support that children utilize a dual memory system when acquiring and integrating new vocabulary, and sleep (especially sleep spindles, slow waves sleep and REM sleep) is important for this process.

Materials and methods: Descriptive analysis was performed in children with specific language impairment (from January 2011 to December 2016). The inclusion criteria were diagnosed of specific language impairment with development quotient > 70 in Brunet-Lezine Scale, normal cranial MRI, without epileptiform discharges in EEG, no hearing impairment, no other disorders and with parents in medium-high sociocultural level. Children participated in a polysomnographic (PSG) sleep recording. Sleep disorders like Obstructive Sleep Apnea Pediatric (OSAP), Periodic Limb Movement during Sleep (PLMS) and parasomnias, and sleep characteristics like sleep stages percentage, spindles characteristics, presence of significant alpha rhythm during sleep (alpha-delta sleep), microarousal index, sleep efficiency and awakenings episodes were analyzed.

Results: 80 children (60 boys and 20 girls) with 3-5 years old. We found OSAP only in 3 patient, and parasomnias in 2 patients. However, it was relevant data about PLMS, because 83.3% present a significant number of them with an average index of 8.01 (6.81±9.28). We found an average of 4.07%±3.41 of stage N1, 40.76±8.89 of stage N2, 25.381±6.91 of stage N3, 25.587±6.91 of stage REM, 89.24±41.93 of body movements, 3.39±1.81 awakening episodes ≥5 min, 82.47±15.62 of sleep efficiency (66.66% presented sleep efficiency <90%), 39.31±11.53 of microarousal index, 66.7% present significant percentage of alpha rhythm during sleep and 66.7% presented unsuitable spindles.

Conclusions: We observed that although the sleep architecture in terms of the macrostructure, like time spent in the different stage of sleep, appears to be not consistently altered, it is very important the analysis of the microstructure because specific features would be altered in these patients and could be in relation with the problem of language skills. Specific treatment for specific sleep disorder like PLMD and treatment to stabilize sleep structure, could improve the symptomatology in these patients.
RESPIRATORY POLYGRAPHY IN CHILDREN: FEASIBILITY IN CLINICAL PRACTICE IN AN ENT DEPARTMENT AND VALUE OF THE AUTOMATIC DETECTION OF RESPIRATORY EVENTS

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Introduction: The use of respiratory polygraphy (RP) in children for the diagnosis of obstructive sleep apnea hypopnea syndrome (OSAHS) can be challenging: reluctance of children, frequent mismatch of sensors, lack of surveillance. In adults, RP interpretation is usually based on an automatic signal analysis, followed by manual correction. In children, the automatic analysis is proposed by the manufacturers but has never been evaluated. The primary objective of this study is to determine the feasibility of pediatric RP in an ENT department routine practice. The second objective is to evaluate the reliability of the automatic signal analysis in children.

Materials and methods: Retrospective single center study of 50 children (32 boys and 18 girls, mean age 5.5 ± 2.3 years) who benefited from an overnight RP in an ENT hospitalization department between January and August 2016 for OSAHS suspicion. The recordings were all performed in an ENT hospitalization department. The following informations were collected: clinical data, indication for RP, acceptance of the device, mismatch of sensors during recording, signal quality. A quality signal >50% was considered technically acceptable. All recordings were manually interpreted by the same ENT specialist trained to pediatric RP analysis. The results of this analysis were compared to the automatic detection of respiratory events.

Results: Indications for RP were a non-contributory medical interrogation in 26% of the cases, a discrepancy between interrogation and physical examination in 22% of the cases, a systematic evaluation for obese or syndromic patients in 34% of the cases, a systematic evaluation before surgery in high risk patients in 4%, and a postoperative control in 14% of the cases. The device was well accepted in 98% of the cases. There was a mismatch of the nasal sensor during a part of the night in 52.2%, the digital sensor in 21.7%, and both in 26.7% of the cases. The examination was technically acceptable in 76% of cases, with an average signal quality of 70.8%. Patients under 3 years old had a lower rate of technically acceptable RP: 25% (n=2/8) versus 86% (n=36/42) for children >3 (p<0.005). There was statistically no correlation between manual and automatic analysis, except for central apneas (Spearman coefficient 0.43 p=0.0015). 100% of patients presented OSAHS according to automatic detection, compared to 32% according to manual detection (p<0.005).

Conclusions: Pediatric RP is feasible in routine practice in an ENT department, with good acceptance and satisfactory signal quality in children older than 3. The automatic analysis of respiratory events in children is unreliable, except for central apneas. Manual detection must be performed for obstructive events, and manual correction for central events.
CONTRIBUTION OF DRUG INDUCED SLEEP ENDOSCOPY (DISE) IN THE MANAGEMENT OF PEDIATRIC OBSTRUCTIVE SLEEP APNEA HYPOPNEA SYNDROME (POSAHS)

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\textbf{Introduction:} DISE is a nasal fiberoptic flexible endoscopy of the upper airway using anesthetic agents that approximate physiological sleep. Its aim is to look for one or more pharyngeal laryngeal obstructive sites. DISE use in the assessment of POSAHS is still being evaluated. The main objective of this study was to determine the influence of DISE on the therapeutic decision when facing a POSAHS. A secondary objective was to compare vigil physical examination and DISE performances in the detection of obstructive sites. Another secondary objective was to search for a correlation between DISE findings and sleep recording results.

\textbf{Materials and methods:} This is a prospective single-center study of 22 children (14 boys and 8 girls, mean age 7.4 ± 4.1 years) who benefited from a DISE between September 2015 and September 2017 for SDB assessment. Beforehand all patients were clinically evaluated by a pediatric otolaryngologist, and recorded by a polysomnography (PSG) or respiratory polygraphy (RP). Therapeutic strategies were confronted respectively before and after DISE. The obstructive sites identified at vigil physical examination and during DISE were compared. A correlation was searched between the obstructive apnea-hypopnea index (OAHI) and the number of obstructive sites found during DISE.

\textbf{Results:} DISE led to a change in therapeutic management in 40.9% of cases (n = 9/22). A perfect concordance between vigil physical examination and DISE was found in 27.3% (n = 6/22). A multisite lesion was found during DISE in 63.6% of cases (n = 14/22). No correlation was found between the number of obstructive sites at DISE and the OAHI (r= 0.13, p=0.6). The OAHI was 4.83 on average in multi-site POSAHS, and 1.8 in uni-site POSAHS (p = 0.26).

\textbf{Conclusions:} DISE contributes to an improvement in the management of POSAHS. It allows a better detection of obstructive sites, thus guiding the therapeutic decision. However, its implementation requires standardization of the obstructive sites evaluation method and the anesthetic protocol.
A SYSTEMATIC REVIEW AND META-ANALYSIS OF BEHAVIOUR CHANGE INTERVENTIONS FOR SLEEP DIFFICULTIES IN CHILDREN WITH NEURO-DEVELOPMENTAL DISORDERS

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Introduction: The aim of this systematic review and meta-analysis is to examine the efficacy of behaviour change interventions in managing sleep difficulties in children who have been diagnosed with neuro-developmental disorders.

Materials and methods: Data sources: Four databases were searched; the Cochrane Library, Embase, Ovid MEDLINE, and PsycINFO in February 2015. Eligibility criteria for selecting studies: We selected randomized controlled trial (RCT) designs that were published between 2004 until February 2015 in English, including children aged between two and fourteen years with neuro-developmental disorders, such as ADHD, Autism and intellectual disabilities. The selected studies tested behavior change interventions, and a measure of sleep difficulties was included. Data extraction: Two independent reviewers extracted data on sleep difficulties, interventions, outcomes measures and the results. The included studies were assessed in accordance with the Cochrane Collaboration’s tool for assessing risk of bias and in accordance with the Grading of Recommendations Assessment, Development, and Evaluation (GRADE) to evaluate the quality of evidence.

Results: Only four studies met the inclusion criteria for this review (n = 377), revealing limited research in area. However, the studies were assessed using the GRADE approach, and the quality of the evidence for managing sleep difficulties in children with neuro-developmental disorders was judged to be high, and indicated a large and positive effect of behaviour change interventions. In contrast, low quality evidence found no evidence for a corresponding reduction in ADHD symptoms in two of the four included trials that reported this outcome. As such, this review reveals clear and positive evidence for the role of behaviour change interventions to improve sleep in children with neurodevelopmental disorders, but does not enable us to conclude that such interventions have a corresponding effect in the reduction of ADHD symptoms in this context.

Conclusions: Further high quality trials are urgently required to facilitate the development of effective evidence based interventions to improve sleep, and test for effects on concomitant outcomes, in this population.

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OVERNIGHT POLYSOMNOGRAPHY IN PATIENTS WITH MÔBIUS SEQUENCE

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Introduction: Möbius sequence (MbS) is a rare congenital disorder with primary diagnostic criteria of congenital facial and abducens nerve palsy. Mental retardation and autism, involvement of other cranial nerves and limb malformations are also described in MbS. This sequence is frequently described as a sporadic condition, but dominant, recessive autossomal and even X-linked recessive inheritance have been described. In our series most cases are related to the misuse of misoprostol. The results of many studies are consistent with a lesion of the central pathways in the brainstem, as a part of the syndrome. It has been associated with both central and obstructive sleep apnoea, also with brainstem abnormalities. Despite of that there is an evidence of lesion of the central pathways in the brainstem, as part of the syndrome, it is not clear the impact of this syndrome in the sleep processes. The aim of this study was to describe the sleep aspects of MbS using overnight polysomnography compared to control group.

Materials and methods: Our study compared 15 (9 girls) patients with MbS with 18 (8 girls) healthy controls, and age ranged were respectively 5.3±4.8 and 12±3.8. All subjects underwent overnight polysomnography with standard parameters. Sleep stages and respiratory events were scored according to standard criteria. Central tendency measures were expressed as mean and standard deviation. The level of significance for the variance analyses was set at p 0.05.

Results: We found differences between patients and control group. PSG parameters were respectively: Sleep efficiency (%): 85.4±8.2 vs 90.7±3.7, p=0.02; Sleep total time (min): 354.3±80.2 vs 446±40.7, p=0.07; Sleep latency 12.9±24.2 vs 23.5±21.9, p=0.03; REM latency 196±82.8 vs 5128.5±57, p=0.03; N1% 7.3±5.6 vs 5.0±2.8, p=0.02; N2% 49.3±12.6 vs 43.9±6, p=0.09; N3% 34±11.8 vs 23.1±11.3, p=0.02; REM % 9.4±4.1 vs 17.2±5.3, p=0.02; Arousal index 8.9±4.8 vs 7.3±5, p=0.02; Apnea-hypopnea index: 5.3±4.8 vs 1.2±0.8, p=0.03.

Conclusions: We found changes in sleep architecture by overnight polysomnography, particularly in the REM sleep expression. Our study suggests that patients with MbS have a reduced sleep efficiency, an increased REM latency and reduced REM amount. Interestingly, the pattern of sleep breathing was not severely disturbed. As REM sleep is generated by a complex neuronal interaction in the brainstem, which is the principal site of lesion in these patients, it is quite challenging the understanding of sleep mechanisms in these patients.
AGE DEPENDENT RISK FACTORS OF SLEEP-DISORDERED BREATHING IN SCHOOL AGE TO ADOLESCENT DOWN SYNDROME PEOPLE

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Introduction: Sleep-disordered breathing (SDB) is frequently comorbid with the people with Down syndrome (DS) because of their craniofacial hypoplasia and muscular hypotension. On the other hand, obesity is well known risk for SDB in general population. Since the change in the influence of factors of SDB in DS depending on their age was unclear, this study assessed these impacts of age for risk of SDB in DS.

Materials and methods: The nation-wade questionnaire based study conducted in Japan. We sent out 2,000 questionnaires to DS people and their caregiver(s), belonged to Japanese Down syndrome society. We received 1,222 questionnaires form the caregiver(s). We excluded 594 and 54 questionnaires since the DS people were <6 years old and > 19 years old, and the questionnaires had any missing data, respectively. Finally, we analyzed 574 questionnaires (Male: 56.6%, Age: 11.9±3.7 years). The questionnaire includes question about demographic data and SDB symptoms (snore, apnea, nap, and nocturia). We stratified the people into 4 groups (6-9 years/10-12 years/13-15 years/16-18 years).

Results: In the all age groups, high prevalence of snoring (72-80%), apnea (31-37%), nap (23-30%) and nocturia (13-25%) were observed. Body structure indexed by Rohrer index and BMI increased as the age increased. There was no age related difference in snoring and apnea. Though there were no significant relationship between SDB symptoms and age, sex or Rohrer index in younger groups (6-9 years and 10-12 years), there were significant relationship between Rohrer index and snoring (OR: [95%CI] 2.13 [1.13-4.03], P=0.02) and apnea (OR: [95%CI] 1.78 [1.04-3.04], P=0.04) in the group of 13-15 year and between BMI and snoring in the group of 16-18 years (OR: [95%CI] 2.07 [1.02-4.18], P=0.04).

Conclusions: This result indicates that other factors than obesity is the main determinant of SDB under 12 year but obesity has significant impact on SDB in DS over 13 years. Thus, caution should be paid according to the age group to sensitively diagnose SDB in DS.
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SLEEP-RELATED MINOR MOTOR EVENTS IN N3 SLEEP: DIAGNOSTIC CHALLENGE AND POLYSOMNOGRAPHY CONTRIBUTION -CASE REPORT

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Introduction: Routine EEG presents limitations to diagnose epileptic seizures and epileptiform paroxysms during N3 sleep, a situation in which polysomnography may be very useful for diagnostic clarification.

Materials and methods: Medical history and videopolysomnography review.
Results: HGS, a 5-year-old male patient was evaluated at the Children’s Sleep Disorders Outpatient Clinic due to muscular spasms occurring every night, exclusively during sleep since his first year of life. Previous medical history was marked by delayed neuropsychomotor development without defined etiology. Extensive research was inconclusive, including two routine electroencephalograms, magnetic resonance imaging of the brain and spinal cord, electroneuromyography of the four limbs, research on inborn errors of metabolism, and evaluation at the outpatient clinic of medical genetics. Polysomnography with an extended montage of electroencephalogram and electromyogram of the 4 limbs was performed and it revealed dozens of axial and appendicular, stereotyped, symmetrical and recurrent, muscle spasms, isolated and in cluster, sometimes evolving to probable global, tonic, brief, both symmetric and asymmetric seizures, occurring almost exclusively in N3 sleep. The EEG showed a mild disorganization of the background activity, due to slowing of the posterior rhythm (7 Hz), and frontocentral spike-wave paroxysms bilaterally. Slow-wave bursts were recorded during the muscular spasms, sometimes associated with fast rhythms and, on some occasions, with the described paroxysms. During the tonic events, maintenance of the described pattern and, sometimes, accentuation of fast rhythms in frontal regions were observed. On one occasion, following the tonic event, the patient sat up in bed and cried, with the maintenance of slow waves on the EEG, clinically compatible with confusional arousal. The sleep efficiency was normal (94.6%), there was an increase in the number of arousals (42.9/ h), an increase in N3 sleep (38.3%), snoring, an increase in the obstructive apnea - hypopnea index (1.9 / h), an increase in the respiratory events index (4.8 / h), with minimal peripheral oxygen saturation of 93%. Treatment with carbamazepine was initiated after the polysomnography analysis. After 14 months the patient presented 3 daytime seizures characterized by behavioral arrest and, on one occasion, with global tonic posture of prolonged duration.

Conclusions: Recurrent sleep-related minor motor events, occurring almost exclusively in N3 sleep, sometimes associated with parasomnia, constitute a diagnostic challenge. The above-mentioned case involving the use of polysomnography with extended montage, by examining a prolonged period of N3, was able to identify events activated by this stage of sleep, therefore contributing decisively to the diagnosis. The stereotyped pattern of motor events, the presence of interictal epileptiform paroxysms and the occurrence of daytime seizures support the hypothesis of the epileptic nature of the nocturnal phenomenon observed.
CLINICAL CHARACTERISTICS OF CHILDREN WITH MILD SLEEP RELATED BREATHING DISORDERS

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Introduction: Pediatric sleep apnea may be associated with clinical, behavioral and cognitive outcomes and other sleep disorders. We analyzed the clinical characteristics of children with mild respiratory sleep disorder in relation to a comparison group.

Materials and methods: Cross-sectional study carried out at Ribeirão Preto Medical School - University of São Paulo. Participants included 48 children aged 4 to 12 years old (mean of 8.9), divided into a group with complaints of snoring or apnea (n = 28), and a comparison group, without respiratory complaints and with nasal breathing (n = 20). Clinical interviews were performed by a neurologist experienced in Sleep Medicine, including Epworth Scale and Abbreviated Conners Scale. Sleep disorders were identified according to the International Classification of Sleep Disorders – 3aed. Pediatric Obstructive Sleep Apnea was confirmed or excluded by polysomnography (scored according to AASM 2.4 manual).

Results: 51% were female. Sleep Habits: there was a similar frequency of sleep-disruptive habits in both groups (25.8% symptomatic group/SG and 22.5% in comparison group/CG). Sleep latency was prolonged in 25% of the SG and 10% of the CG. Sleep was non restorative in 64.3% of the SG and 33.3% of the CG. Obstructive Sleep Apnea: in the symptomatic group (mean AHI index 2.3/h), frequent snoring occurred in 71.4% and witnessed apnea in 53.7%. Agitated sleep occurred in 60.7% in symptomatic children and 5% in the CG. Headache of any frequency occurred in 28.6% in symptomatic children and 5% in the CG. Behavioral Insomnia of Childhood: occurred in 35% in SG and 30% in CG. Movement Disorders: restless legs syndrome was diagnosed in 7.1% of symptomatic children and 5% in CG. Sleep related bruxism occurred in 28.5% of symptomatic patients and 5% in CG. Parasomnias: confusional arousals were diagnosed in 25% of symptomatic children and 30% in CG. Sleep terror was diagnosed in 7.1% of symptomatic patients and was not found in the CG. Only 0.15% of all presented sleepwalking. Nightmares 5-7 times per week occurred in 3.6% in symptomatic patients and didn’t happened in CG. There was no diagnosis of REM sleep behavior disorder. Sleep talking occurred in 57.1% in symptomatic patients and 45% in CG. Hipersomnias of Central Origin: were not diagnosed. Epworth Scale: scores were increased in 17.9% of symptomatic children and 5% in CG. The most discrepant situations regarding the chance of falling asleep were postprandial moments, while reading and as a transit passenger. Abbreviated Comners Scale: abnormal scores suggestive of Attention Deficit Hyperactivity Disorder occurred in 25% of SG and in none of CG.

Conclusions: There was a similar frequency of sleep-disruptive habits in both groups, but the complaint of non restorative sleep was more important in the symptomatic group. In respiratory group, witnessed apnea occurred only in 53.7% and it may be important to ask about restless sleep. Children with mild respiratory disorder had higher frequencies of other associated disorders, notably headache, bruxism, excessive sleepiness, and attention deficit and hyperactivity symptoms.
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AMBULATORY HOME SETTING OF NASAL CONTINUOUS POSITIVE AIRWAY PRESSURE THERAPY (NCPAP) IN NON-SYNDROMIC CHILDREN WITH OSAS: ADHERENCE UP TO 24 MONTHS

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Introduction: The prevalence of obstructive sleep apnea syndrome (OSAS) in otherwise healthy children and teenagers may be as high as 5%. The diagnosis should be made in the presence of snoring associated with restless sleep, academic or behavioral difficulties, stunting or destabilization of asthma or epilepsy … In case of persistence of OSAS after adeno-tonsillectomy or in the absence of surgical indication, nCPAP treatment may be transitorily proposed. No data is available concerning adherence to nCPAP when initiated in an outpatient procedure (ambulatory)

Materials and methods: Our main objective was to describe nCPAP adherence in children with OSAS when initiated in an outpatient procedure during the first 24 months. Our secondary objectives were to determine the predictive factors of adherence and evolution of adherence to nCPAP. Children with OSAS were diagnosed by polysomnography in multidisciplinary regional networks. nCPAP was proposed in patients who were not candidates for adeno-tonsillectomy or in patients with persistent OSAS after surgical treatment. We report a retrospective longitudinal follow-up of 154 children under 18 years old who started nCPAP at home or after day-time short therapeutic education session (child + family), with either automated-mode or fixed pressure. They were reassessed at M1, M3, M6 and M12 and M24 during follow-up visits.

Results: Mean time of use of nCPAP at 1 month was more than 6.5 hours and did not decrease at 3, 6, 12 (N=83) and 24 months (N=46). The cumulative rate of discontinuation of nCPAP was 5.3% at M3, 9 % at M6, 9.7% at M12 and 13% at M24. Age was inversely correlated with nCPAP adherence during the first 12 months follow-up. Higher initial polysomnographic AH index (OSAS severity) was significantly associated with better adherence during the first 12 months of treatment (p<0.05). The compliance of primary school children tended to be better over time compared to middle and high school students. Compliance was also better in obese as compared with non-obese subjects (p <0.05). Finally, adherence to nCPAP was similar regardless of the mode of introduction of nCPAP (home vs. therapeutic education in day-time hospital) and the automated vs. fixed-pressure mode.

Conclusions: The use of nasal CPAP in OSAS children, when initiated in an outpatient procedure is safe, acceptable, optimal and prolonged, reflecting the tolerance and perceived benefit of children and their families. It can be proposed until weight loss, physiotherapy and / or orthodontic therapy or surgery are effective. It is crucial to increase access to the diagnosis and treatment of children and teenagers with OSAS.
SLEEP DISTURBANCES IN ROMANIAN CHILDREN DIAGNOSED WITH ASPERGER SYNDROME

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Introduction: Sleep disorders are common in children with different forms of autism spectrum disorders (ASD). Children with Asperger Syndrome (AS) present difficulties in initiating and maintaining sleep, shorter sleep duration, earlier morning awakenings, and parasomnias more frequently other autistic children. Specifically, AS was linked mainly with disruptions of sleep continuity. Very few studies evaluated sleep microstructure in children with AS and showed the presence of subtle alterations of NREM sleep. Alterations in sleep architecture have been linked with cognitive and behavioral deficits described in AS as well as with anxiety who is very often comorbid. Also hyperserotonemia and low melatonin levels in urine, plasma and pineal gland have been described in ASD individuals. The literature review revealed that current treatments for insomnia in children who have ASD show promise for behavioral/educational interventions and melatonin trials. Pediatric sleep specialist in Romania started to be a part of a multidisciplinary team only a few years ago. Our effort was toward to increase the level of awareness about the therapeutic options available worldwide for the management of sleep disorders in children with ASD and build up multidisciplinary teams for the management of these issues.

Material and methods: We will present three cases of children diagnosed with AS and sleep disturbances. The first case is a 3-year-old girl with difficulties in initiating and maintaining sleep. The second case is a 6-year-old girl with high level of sleep anxiety and insomnia, and the third case is a boy who has difficulty concentrating attention and restless sleep. In the first two cases we applied melatonin, behavioral interventions, and parent education/education program interventions. In the third case we performed sleep study.

Results: In the first two cases sleep problems have improved significantly after parent education about setting up an appropriate sleeping routine, melatonin administration and pharmacological decrease of anxiety level. Moreover, there has been an improvement in the level of daily social functioning. In the third case changes in sleep architecture were observed and abnormal spindle characteristics in NREM sleep and in REM sleep, long REM sleep latency.

Conclusions: Quality sleep is imperative for the maintenance of good health, and sleep loss can lead to or exacerbate existing behavioral problems associated with AS. A question is whether the sleep pathways are developmentally changed in AS or whether disruptions in sleep are a consequence of changes in daily anxiety levels. In addition, the changes observed in the sleep architecture can be explained by abnormal brain connectivity. After we started to perform pediatric sleep study in our country we have encountered many different situations. Our efforts are to continue this ongoing educational process in Romania, both for patients and doctors, regarding the management of sleep problems in children who have AS. This should begin with parent education in the use of behavioral approaches as a first-line approach; pharmacologic therapy may be indicated in certain situations.
HYPNAGOGIC FOOT TREMOR AND ALTERNATING LEG MUSCLE ACTIVATION IN CHILDREN: CASE SERIES

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Introduction: Hypnagogic foot tremor (HFT) and alternating leg muscle activation (ALMA) are characterized by rhythmic muscle activity related to sleep, with short-term muscle activations, grouped into high-frequency series. Currently available studies have not evaluated the prevalence of these motor phenomena in the pediatric population.

Materials and methods: Retrospective study based on the review of 120 polysomnograms (PSGs) for research on HFT or ALMA according to the diagnostic criteria of the most recent AASM manual (version 2.4), of children under the age of 14 referred to the sleep laboratory of Ribeirão Preto Medical School - University of São Paulo between January and December 2017.

Results: Sequences of HFT and/or ALMA were identified in PSGs of 16 individuals (13%), whose ages ranged from 2 to 12 years old, with a mean of 8 (years old). Among these, the indication for the examination was obstructive sleep apnea (OSA) in 88%, abnormal movements in 6%, and evaluation of excessive daytime sleepiness in 6%; 94% of PSGs were diagnostic and 6% were positive pressure titration exams. The main comorbidities of the HFT / ALMA group were allergic rhinitis (56%), asthma (19%), obesity (38%), overweight (13%), gastroesophageal reflux disease (25%), epilepsy (19%), migraine (13%).

ALMA was identified in 14 cases (11.7%), with a minimum age of 2 years old, maximum of 12 years old, and a mean of 8 years old, 10 patients were male. The number of ALMA sequences recorded per patient ranged from 1 to 86, and lasted between 1.1 and 101 seconds. The duration of individual muscle activation ranged from 0.075 to 3.2 seconds, with the majority between 0.1 and 0.5 seconds. The mean frequency in each sequence ranged from 0.5 to 3.0 Hz. ALMA sequences occurred both in presleep wakefulness and in all sleep stages, mainly related to arousals. In one children there was video recording of flexion and extension movement of the foot and great toe during the ALMA sequence.

HFT series were recorded in 10 cases (8.3%), with a minimum age of 3 years old, maximum of 12 years old, and an average of 8.3 years old, 8 patients were male. In 50% of the children, both ALMA and HFT sequences were recorded. The number of recorded HFT sequences ranged from 1 to 42 per patient, and they occurred in pre-sleep wakefulness, N1 and N2 stages, ranged in duration from 2.1 to 34.3 seconds. Only in the case with the highest number of HFT series there were events in both legs, but with clear predominance on the right (95% of the events). The duration of muscle activation varied from 0.075 to 1.4 seconds, and the frequency 0.4 to 4 Hz.

The final polysomnographic diagnosis was OSA in 15 of the 16 patients.

Conclusions: The present study demonstrates the occurrence of HFT and ALMA in children. The pathophysiology and clinical significance of these motor events are still not fully understood. Further studies in healthy children are needed to clarify the general prevalence and clinical significance of these motor phenomena.
CARDIORESPIRATORY AASM PARAMETERS IN TERM NEWBORNS: PRELIMINARY RESULTS

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Introduction: Cardiorespiratory parameters in children are age-dependent and there are few studies in the neonatal period. Neonates have polyphasic sleep, with cycles of active sleep (analogous to adult REM sleep) and quiet sleep (analogous to NREM sleep) of approximately one hour duration. For this reason they can be evaluated through daytime examination with neonatal polyographies of short duration. In this study, we analyzed cardiorespiratory parameters in diurnal polygraphs of full term neonates, according to AASM criteria.

Materials and methods: A prospective study conducted at the Women's Health Reference Center - Ribeirão Preto Medical School - University of São Paulo. Daytime neonatal polygraphs of 1 to 2 hours duration were performed in healthy term infants. The polygraphic parameters were acquired and analyzed according to AASM manual, version 2.4. Additionally, central events with durations of 3 seconds, 10 seconds, 15 seconds and 20 seconds were quantified.

Results: 40 polygraphs of neonates between 37 to 44 weeks of postconceptional age and 0 to 10 days of legal age were analyzed, being 57% female. The mean total apnea (AI) index was 2.6/h. The highest index of central apnea (CAI) with duration ≥ 3 seconds was 7.3/h (mean 2.2/h). The highest CAI with duration ≥ 10 seconds was 1.2/h (mean 0.1/h). There were no respiratory pauses lasting 15 seconds or longer. The highest obstructive apnea index was 1.2/h and the highest mixed apnea index was 2.7/h. Central hypopneas were quantified, and the mean total AHI was 14.7/h (1.8/h-40.8/h). The mean duration of respiratory events in each polygraph ranged from 5 to 10.5 seconds, and the maximum duration ranged from 7.4 to 22.2 seconds.

The average desaturation index ≥ 3 seconds was 17.9/h. The average oxygen saturation (SATO2) in each polygraph varied between 94 and 99.9%. The mean saturation nadir was 90.6% and the minimum oxygen saturation was 85%. The mean of total sleep time in periodic breathing was 0.13%, ranging from 0 to 2.1%.

The mean heart rate in sleep was 118.6 bpm and varied in each polygraph between 102.5 and 149.5 bpm. The heart rate nadir ranged from 82 to 123 bpm, and the maximum heart rate at each exam ranged from 116 to 200 bpm. Events of bradycardia <100 bpm were rare (0/h at 4.4/hr). There were no episodes of bradycardia <80 bpm.

Conclusions: In our sample analyzed so far, there were discrepancies in the total apnea + hypopnea index, mainly due to the variability of the occurrence of central hypopnea events. The predominant apnea events were of the central type, being rare those with duration greater than 10 seconds or associated with nadir of saturation <90%. The total sleep time in periodic breathing was below the currently proposed cutoff point for infant apnea (5%). Bradycardia events were less expressive in frequency and intensity.
SLEEP ARHITECTURE DISTURBANCE IN CHILDREN WITH CROUZON SYNDROME AND ACHONDROPLASIA

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Introduction: Patients diagnosed with Crouzon syndrome and achondroplasia, consecutive to craniosynostosis, develop changes of the upper airways and intracranial hypertension, followed by respiratory events (central / obstructive) with or without drop of SpO2. Disturbance of sleep architecture is partially explained by these mechanisms.

Materials and methods: We will present three children diagnosed with syndromic craniosynostosis, which were polysomnographic investigated for sleep disturbance (two cases with Crouzon syndrome) or for achondroplasia cardio-respiratory monitoring, prior diagnosed with mild obstructive sleep apnea. Respiratory events and sleep staging where noted according to the guidelines of the American Academy of Sleep Medicine.

Results: In all three cases we observed a low percentage of REM sleep ( <12% for Crouzon syndrome, 19.2% for achondroplasia) and Stage III NREM sleep (19.4- 23.4%). In both cases with Crouzon syndrome AHI were >15/h, and achondroplasia had AHI 3.1/h.

Conclusions: Polysomnographic monitoring of Crouzon and achondroplasia syndromes is useful for quantification of AHI and sleep stages / sleep architecture. Altogether these changes might be important in therapeutic approach for children with craniofacial malformation. In children cases, both quality of sleep and respiration are important for overall health and long term development.
SLEEP AND PSYCHIATRIC SYMPTOMS IN ADOLESCENTS WITH ANOREXIA NERVOSA HOSPITALIZED IN A CHILD AND ADOLESCENT PSYCHIATRY UNIT

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Introduction: Sleep disturbances is a very prevalent complaint, evaluated at 50.3% of anorexic patients. The studies performed in patients with anorexia nervosa (AN) confirm that this disorder is associated with a pattern of sleep alterations and particularly in an increase in Wake After Sleep Onset (WASO). To date no study has investigated the links between sleep abnormalities and clinical symptomatology of adolescents with AN. Notably, we don’t know if consequences of short sleep duration described in the adolescents without psychiatric symptoms are the same in adolescents with AN. In this study our aim is to compare sleep disturbances and psychiatric symptoms in the two groups of anorexic adolescents with and without short sleep duration (SSD) to determine how the SSD interact with symptoms in anorexic adolescents.

Materials and methods: We recruited 50 anorexic inpatients aged 11 and 18 years old into two groups: SSD (N=12) with a Total Sleep Time (TST)<7 h and No-SSD. Sleep parameters were assessed with the Pittsburgh Sleep Quality Index (PSQI), the Pediatric Daytime Sleepiness Scale (PDSS) and the Horne-Ostberg questionnaire (HO). The psychiatric symptomatology was assessed with the Eating Disorder Inventory 2 (EDI2) and some anxiety, depression, self-esteem, assertiveness and temperamental scales. All questionnaires were filled four weeks after admission. We compared the different parameters between the two groups.

Results: In our sample, the TST was 446.2 ±114.6 min, sleep latency was 31.8 ±34.5 min and WASO was 53.8 ±76.1 min. This sample had a moderately morning chronotype (HO mean score=59.6 ±8.5) with sleep disturbances (PSQI mean score =7.1 ±4.6) and without daytime sleepiness (PDSS mean score= 10.3 ± 5.2). The SSD group was significantly more sleepy (p<0.001), had a significantly longer sleep latency (p<0.001) and WASO (p=0.008) and more sleep disturbances at PSQI (p<0.001) than No-SSD group. In the psychiatric questionnaires, the SSD group was significantly more anxious and depressed (p<0.001) and with a lower self-esteem and assertiveness (p<0.05). The SSD group was also more active in the day and night with a bigger rigidity (p<0.05) than No-SSD group. All the features of AN were significantly increased in SSD group (0.001<p<0.05) except the maturity fear and bulimia. Finally we found no significantly difference between the two groups for the body mass index (BMI).

Conclusions: Our results confirm sleep disturbances in adolescents with AN and particularly a short TST mainly explained by a long WASO. As it has been described in general population of adolescents, the TST less than seven hours seems to impact on adolescents with AN: increase in sleepiness, anxiety, depression, physical activity and rigidity and decrease in self-esteem. Contrary to adolescents general population, adolescents with AN didn’t present a BMI increase. Moreover, it is interesting to note that reduced sleep duration seems to aggravate many features of AN.
THE DIAGNOSTIC EXAMINATIONS OF THE OBSTRUCTIVE SLEEP APNEA HYPOPNEA SYNDROME (OSAHS) OF THE CHILD

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Introduction: The diagnosis of OSA in children depends both on daytime and nighttime symptoms which vary between individuals and also on the results of nocturnal monitoring. Nocturnal respiratory monitoring aims to increase diagnostic precision and enable the physician to judge the severity of OSA.

Materials and methods: Daytime and nighttime symptoms, Nocturnal monitoring, Polysomnography

Results: Nocturnal monitoring measures the apnea and hypopnea index (AHI): if the score is >1.5/hour in a child, the diagnosis of OSA is confirmed and the score is key in determining management. Nocturnal respiratory monitoring alone (without sleep recording via EEG) can confirm OSA, but cannot identify upper airway resistance syndrome (UARS) where apneas and hypopneas are often absent but increased airway resistance leads to a micro-arousal.

Conclusions: The clinician plays a vital role in interpreting the results of nocturnal monitoring as the different techniques have different sensitivity and specificity, and can thus adopt a patient centered approach in the management of sleep disordered breathing.
ROLE OF THE ORTHODONTIST IN SCREENING FOR OBSTRUCTIVE SLEEP APNEA (OSA) SYNDROME IN CHILDREN

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Introduction: In children, the syndrome of obstructive sleep apnea is common and poorly tracked. The orthodontist, within the multidisciplinary team, is strategically positioned to detect a respiratory disorder and suspected OSA syndrome. The objective of this poster is to consider for young patients a medical treatment. In fact, parents do not always report their child’s snoring, it is for the orthodontist to open dialogue.

Materials and methods: The radiological anatomy, The physiology of the masticatory system, The clinical examination of young patients.

Results: The knowledge of the orthodontist in radiological anatomy, physiology of the masticatory system and used to observe the different functions in the clinical examination of young patients will alert parents to orient to the ENT Doctor or sleep and possibly consider taking early treatment.

Conclusions: The orthodontist is in the front line of screening since he frequently follows children. For this from the clinical examination of young patients, he must observe the cranio-facial morphology and functional elements predisposing and consider possible early multidisciplinary therapeutic management involving the ENT doctor, the pulmonologist and the pediatrician working in collaboration with the orthodontist.
CONSEQUENCES OF PAEDIATRIC OBSTRUCTIVE SLEEP APNEA SYNDROME ON DENTO-MAXILLO-FACIAL MORPHOGENESIS

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Introduction: The obstructive sleep apnea syndrome (OSAS) in children is defined as a disorder of breathing during sleep characterized by prolonged partial upper airway obstruction and/or intermittent complete obstruction (obstructive apnea) that disrupts ventilation.

Materials and methods: The facial patterns and craniofacial morphologies were assessed by a cephalometric analysis of the lateral head X-ray using teleradiography. The craniofacial morphology and airway and dental occlusions, is accomplished by including craniofacial analysis. The cephalometry for the detailed study of skeletal craniofacial morphology.

Results: Craniofacial caracteristiques in the pediatric obstructive sleep apnea syndrome are siviral such as dolichocephaly, mandibular plane, maxillary constriction, an elongated soft palate, a narrow mandible, and an enlarged tongue base.

Conclusions: Craniofacial morphology plays an important role in obstructive sleep apnea syndrome (OSAS). These findings may be relevant in preventing sleep apnea in adult.
THE ONSET OF MOTOR MILESTONES CHANGES INFANTS’ SLEEP PATTERNS

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Introduction: During infancy, a rapid and intense period of learning and skill acquisition, sleep is the primary brain activity. This is likely no coincidence as sleep functions to consolidate newly learned information and skills into memory. The developmental trajectory of the regulation of sleep-wake states in infancy is protracted and unstable across the early years and is shaped by changes in infants’ physical and motor development (Scher, 2005; Scher & Cohen, 2005; Lampl & Johnson, 2011). For example, cross-sectional comparisons by motor ability (i.e., pre-crawling vs. crawling infants without respect to experience) found increases in nightwaking upon the acquisition of independent locomotion (Scher 2005; Scher & Cohen 2005; Scher & Ratson, 1998). Our primary aim was to better understand the relationship between sleep regulation and motor development and to obtain precise measurements of motor experience relative to sleep measures. To do so, we took a microgenetic approach to studying the relationship between the onset of new motor skills and sleep patterns.

Materials and methods: We used dense, longitudinal sampling of motor milestone onsets and sleep experiences of 3 infants over their first year. Parents kept a daily checklist diary from before infants could crawl until after they could walk independently. For the purposes of this study, we documented 21 motor skills, number of nightly wakings, and duration of night sleep. Completing the checklist took less than 20 minutes/day. Families participated for 313, 197, and 296 days, respectively.

Results: We identified two sleep profiles that described the shape of infants’ sleep trajectories. Two infants showed stable phase shifts, where changes in number of night wakings shifted from many to moderate to low and with stability in each phase. The other infant had a consistent profile with little variation or fluctuation.

We applied Autoregressive Integrated Moving Average (ARIMA)-Intervention methods (Box & Tiao 1975; Box & Jenkins 1976) to evaluate the dynamics of infant sleep changes. Sleep duration and night-wakings are considered fluctuating dependent time series spanning infants’ participation in the study. Milestone onsets were treated as individual interventions along this series in two ways: first day of occurrence or mastery day (milestone first performed over a continuous sequence). First day coincided with mastery for 72% of the skills measured. For the infants with stable phase shifts, six major milestones (sitting with hands, transitioning from sitting to prone, crawling less than 10 feet, crawling greater than 10 feet, pulling-to-stand, and walking greater than 10 feet) indicated a significant intervention effect on night-wakings (all p’s < .05). Night waking of the consistent infant was not significantly interrupted by milestone onset. No statistically significant relationships between milestone onsets and sleep duration were found.

Conclusions: The onset of key motor milestones changed the ongoing pattern for night wakings, suggesting that changes in sleep patterns and trajectories reflect changing motor ability. Additionally, this study reveals the potential for this statistical method to address developmental research questions.
Introduction: Individuals with neuropsychiatric / neurodevelopmental conditions often display disruptive behaviours e.g. hyperkinesia, hypermotor-restlessness, hyper/hypo-arousability and hypermotor events (H-behaviours). This terminology, introduced by the IPSA Video-Working-Group, allows an in-depth phenotyping of structured behavioural observations. We investigated how to develop a shared, neutral, annotation language for describing rather than interpreting H-behaviours, using qualitative freehand and pictogram-based annotations (FH-a; P-a) during the Suggested Clinical Immobilization Test (SCIT).

Materials and methods: Using REDCap data management, seven research assistants without prior formal training analyzed: (A) three ‘Fidgety Philip’ (FP) cartoons, using qualitative FH-a and reviewed possible applications for P-a, and (B) 12 SCIT snapshots of five participants using FH-annotations. This was repeated 2 days later in a randomized order; (C) and the same procedure repeated with P-a. Inter-/Intra-observer variability of (A) & (B) results were investigated and data were separated into descriptive vs. interpretive categories. (D) Finally, 18 videos lasting approximately 20 minutes with two embedded SCITs were analyzed by two trained RAs and compared with Restless Legs Syndrome Severity Score (RLSSC) rankings of observed individuals and a non-trained senior RA.

Results: (A) FP-cartoons: FH-a were divided into descriptions (n=168, mean=8.0±3.8) and interpretations (n=106, mean=5.0±3.1); with each cartoon, the number of descriptions increased. (B) SCIT snapshots: descriptive vs. interpretive results: FH-a (median=6/7 vs. median=1/1) and P-a (median=1/2 vs. median=2/1); pictogram categorization was reassessed and developed further. (C) Intra-observer reliability for descriptive/interpretative FH-a was low (61.9%/36.6%). Inter-observer reliability of P-a for overarching categories was higher (intraclass correlation coefficients: head 0.895; upper limbs: 0.823; lower limbs: 0.878) but low for body tension in dependency of posture (0.588). Factors leading to rating tension differently were investigated. (D) All video-recorded volunteers were on the RLSSC spectrum and indicated an urge-to-move (SCIT-1: 72%; SCIT-2: 83%); tension and/or movements were observed by trained RAs (SCIT-1: 84%; SCIT-2: 85%) versus the non-trained senior RA (60% vs. 50%). After excluding the three cases with disagreement between the two trained RAs, correlation coefficients increased to 0.975 and 0.998.

Conclusions: (A) and (B) The ability to describe and reduce interpretations increased with experience, P-a enhanced this process. (C) Descriptions via FH-a yielded low, P-a higher inter-observer consistency. P-a characterizing movements achieved higher, those for body posture and interpretative movements lower inter-observer consistencies. (D) The training helped to optimize body posture ratings - still there is a need for two independent raters in order to analyze inconclusive SCIT results. This exercise is the foundation for the Video-Annotation-Training-Module that our Working Group has developed.

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PRECISION MEDICINE IN ASSESSMENT OF SLEEP/WAKE-BEHaviours: PICTOGRAM-BASED CONSENT FORMS FOR ENHANCING PARTICIPATORY RESEARCH

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Presentation preference: Poster

Introduction: Observational sleep and wake-behaviour medicine can greatly benefit from qualitative and quantitative analysis of video recordings during structured and/or naturalistic behaviour observations. The Formal Suggested Clinical Immobilization Test (SCIT) was developed based on the lab-based Suggested Immobilization Test (SIT) and supplements standardized observations. The SCIT investigates with "emplotted narratives" the countless descriptions of the various "urgeto-move" patterns in patients who can express the "movement-soothes" sensation in their own words. For those who lack the ability to verbalize their sensations, an informal SCIT test via third party behavioural observations can be administered.

Materials and methods: Before implementing SCIT or SIT video recordings in clinical practice, a best practice for informing participants has to be established. Any recording has to be consented by the 'observed' or 'investigated' individual; obtaining oral consent is the minimum while obtaining written consent is the recommended practice. The main problem with written consent forms is their wordiness and explanation of obvious content, often for legal reasons.

Results: We are suggesting a digital pictogram-based consent form, where hover boxes as a graphical control element are activated when the user moves or "hovers" the pointer over its trigger area. This way the user decides, which text he/she will read. See SCIT procedure figure 1a without any hover boxes and 1b explaining the first step of the activity with the instructions presented in the hover box.

Conclusions: The SCIT and SIT have the potential to be used as a standard in all sleep/wake-behaviour assessments, because they investigate measureable changes in behaviours in a structured way. Further, both use a common basis, namely the video recordings, for negotiating the observed symptoms between the affected individual and professionals. However, the process of consensual information sharing, which should initiate collaborative, participatory exploration of symptoms is counteracted by dense, difficult to read consent forms, which contradict the original idea of informing participants in the most efficient way.

In summary, we propose a new concept, which utilizes graphical elements and enhances focused reading. In our Vancouver Summer School 2018 Project, we will investigate the feasibility of these new forms.

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THE ROLE OF POLYSOMNOGRAPHY IN RESPIRATORY MANAGEMENT OF OBSTRUCTIVE SLEEP APNEA IN INFANTS WITH PIERRE ROBIN SEQUENCE: ESSENTIAL OR NOT?

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Introduction: Currently, polysomnography (PSG) is the gold standard for the diagnosis of obstructive sleep apnea (OSA), but its access is still difficult. A polygraphy (PG) combined with pulse oximetry and transcutaneous PCO2 (TcPCO2) allow the diagnosis of OSA. Until this day, the respiratory management of infants with PRS and OSA is not standardized. The aim of this study is to compare the OSA therapeutic decision in infants with Pierre Robin sequence (PRS) depending on PSG versus PG combined to oximetry and TcPCO2.

Materials and methods: This study was conducted in infants with PRS in the sleep unit of the pediatric university hospital of Lyon, France, from 2015 to 2017. A nocturnal polysomnography with measurement of gas exchange was performed for each PRS infants during their routine follow-up. Each anonymized PSG were analyzed by a single reviewer. After suppression of sleep data, 1 month after, anonymized raw data were analyzed by the same single reviewer in order to obtain only polygraphy data. The first OSA therapeutic option selected with PSG by 3 pediatricians was notified (sleep position, non-invasive ventilation support, nasopharyngeal tube NPT). The new OSA therapeutic option selected with PG by the same 3 pediatricians was notified. Respiratory index [obstructive apnea index (OAI), obstructive hypopnea index (OHI), obstructive apnea hypopnea index (OAHI)] were compared between PSG and PG. Index expressed the mean of events per hour.

Results: PSG was performed in 20 infants at 66 days (mean, 14-189 days, minimum and maximum). 15 patients had isolated PRS, 5 patients had associated or syndromic PRS. All patients had confirmed OSA. Therapeutic option selected after PG combined to oximetry and TcPCO2 and compared to decision after PSG recording was identical for 17 infants, similar for 1 infant and different for 2 infants. No significant difference was observed between OAHI measured by PSG versus PG (41/h versus 32/h, p=0.458). OAI was under-estimated in PG versus PSG (3.2/h versus 15.4/h, p<0.0001).

Conclusions: An overnight PG recording in a sleep unit, combined to oximetry and TcPCO2, lead to select the adequate therapeutic option to treat OSA in PRS infants in the 8 first months of life. In PRS infants, PSG could be performed in a second time, in case of negative PG, unconformity or respiratory management difficulties.
P#53-Saturday
CHANGES OF BEHAVIORAL AND EMOTIONAL PROBLEMS IN SCHOOL-AGE CHILDREN WITH CHRONIC INSOMNIA AFTER GROUP CBT-I: LONG-TERM EFFECTS OF A RANDOMIZED CONTROLLED STUDY

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Introduction: The present work is the first randomized controlled study providing evidence for the long-lasting efficacy of treatment for chronic insomnia on behavioral and emotional problems in school-age children. At first assessment, the total sample of 112 children with chronic insomnia indicated increased values for internalizing and overall behavioral and emotional problems. Problems of initiating and maintaining sleep as well as arousal predicted internalizing and externalizing problems of children.

Materials and methods: All children were randomly assigned to a treatment (CBT-I) or waitlist (WL) condition. Treatment consisted of Cognitive Behavior Therapy for Insomnia (CBT-I) for children. Besides sleep measurements, also mental health was evaluated before and after treatment as well as at follow-up measurements.

Results: Compared to children in the WL condition, children in the CBT-I condition indicated greater improvements in behavioral and emotional problems. Improvements after CBT-I persisted over the 3-, 6-, and 12-month follow-up assessments. Besides enhancing sleep behavior, CBT-I was also efficient to improve mental health in children.

Conclusions: Results point out the importance of adequate sleep behavior for mental health in school-age children.
Introduction: Sleep problems in children with developmental disorders are common, with up to 80% of children experiencing some kind of sleep disturbance during their development. Sleep problems are particularly prominent in children with Autism. These include delayed sleep onset, sleep fragmentation with prolonged night-time wakings, and daytime sleepiness. However, we still know very little what impact can poor sleep of a child pose on maternal wellbeing. Hence the main aim of this study was to characterise the relationship between child’s sleep and daytime functioning in relation to maternal sleep and wellbeing of school-aged children with and without ASD diagnosis. The second question was to examine if there are ethnic differences in sleep by comparison two groups: UK and Saudi Arabia.

Materials and methods: Participants included 95 mothers of children with ASD (50% from UK) and 96 mothers in control group (no ASD, 50% from UK). Inclusion criteria were that all children had to be of school age and have current diagnosis of ASD. Maternal sleep was measured using sleep diary for 7 days and further assessed using a large battery of Sleep Questionnaires such as The Pittsburgh Sleep Quality Index. Maternal mental status was measured using Becks Inventory and Parenting Stress Index. Each mother filled in Childhood Sleep and Habits Questionnaire about their child’s sleep patterns as well as Childhood Autism Rating Scale.

Results: As expected children with ASD had significantly higher sleep onset delay, more frequent and longer night-time wakings in comparison to the control groups. Mothers in both ASD groups suffered from sleep onset delay, frequent night wakings and shorter sleep. Mothers of children with ASD also reported higher parenting stress, lower physical quality of life and emotional quality of life in comparison to the mothers of typically developing children. Comparison of the UK and Saudi Arabia groups showed that children in all groups had poorer sleep in the Saudi sample yet negative maternal outcomes were less prominent than the UK ASD group.

Conclusions: The current results showed sleep issues of a child should not be taken in isolation and use of holistic approach is necessary when examining public health issues. The relationship between sleep and wellbeing in the life of mothers are very closely linked with sleep profiles of their children with autism. Implications of this research shed light in areas that healthcare practitioners should consider the effects of sleep in all family members when looking for interventions targeting quality of life. Our further research aims to employ non-invasive objective sleep measures such as actigraphy along with interviews to gain better insights into their sleep patterns in relation to cognitive, mental health and behavioural functioning of families.

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PSYCHOMOTOR PERFORMANCE IN CHILDREN WITH SLEEP DISORDERED BREATHING

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Introduction: Our objective was to evaluate the psychomotor performance in children with Sleep Disordered Breathing.

Materials and methods: It is a cross-sectional study, carried out with children aged 7 to 11 divided into 3 groups: 1) OSAS (Obstructive Sleep Apnea Syndrome), 2) Primary Snoring (PS), and 3) Control Group (CG- No Sleep Disorders). They were evaluated with Polysomnography Study (PSG) and Motor Development Scale (EDM).

Results: 1) OSAS group presented a worse score in global motricity when compared to the PS group (p = 0.045). 2) PS group had worse performance in balance when compared to CG (p = 0.037). 3) OSAS group presented a worse performance in relation to other groups (p = 0.002) in body scheme/speed. 4) PS group presented worse performance in EDM’s classification when compared to CG (p = 0.053).

Conclusions: 1) Body scheme / speed was impaired in children with OSAS. 2) Body scheme/speed was impaired in OSAS group when compared to CG. 3) Global motricity and body scheme/speed had worse performance in OSAS children when compared to PS. 4) Balance was impaired in children with PS when compared to CG. 5) PS group had worse results in EDM’s classification when compared to CG.
THE PAEDIATRIC NARCOLEPSY PROJECT: CHARACTERISING AND COMPARING SLEEP, PHYSICAL ACTIVITY, COGNITIVE FUNCTION AND PSYCHOSOCIAL WELL-BEING IN CHILDREN WITH NARCOLEPSY AND HEALTHY CONTROLS

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Introduction: Narcolepsy is a lifelong neurological sleep disorder characterised by excessive daytime sleepiness and attacks of muscle weakness triggered by emotions (cataplexy). In 2010, alarms were raised about an increase in the incidence rate of narcolepsy diagnosis in children. Subsequent research has confirmed a causal link between the use of the ASO3 adjuvanted pandemic A/H1N1 2009 influenza vaccine (Pandemrix) and cases of narcolepsy in children. Despite the rise in cases, there is very little research investigating the impact of narcolepsy in childhood. The Paediatric Narcolepsy Project is a case-control study designed to characterise and compare the sleep, physical activity, cognitive function and psychosocial well-being of children with narcolepsy and healthy matched controls.

Materials and methods: 23 children with narcolepsy (age: 8-15 years) and 23 healthy gender, age, I.Q and socioeconomic status (SES)-matched controls were recruited from the United Kingdom and The Republic of Ireland. 21 out of the 23 children with narcolepsy were treated with medication at the time of the study. The children were visited in their homes by the researcher (JB) and underwent standardized neuropsychological assessment and home polysomnography (PSG) using a portable PSG system. The children also wore an actigraphy monitor for 8 days to measure their daytime sleep and physical activity.

Results: The polysomnography data showed that the children with narcolepsy were awake for three times as long after sleep onset than healthy controls and as a result had significantly reduced sleep efficiencies. Children with narcolepsy spent significantly more of their sleep in stage N1 sleep and stage N3 sleep than healthy controls. However, they spent significantly less of their sleep in stage N2 sleep compared to healthy controls.

The actigraphy data showed that the children with narcolepsy spent a significantly greater percentage of their time sedentary when awake than the healthy controls. Healthy children spent a significantly greater percentage of their time in moderate physical activity when awake than children with narcolepsy. The neuropsychological assessment revealed that the children with narcolepsy did not have impaired cognitive function compared to healthy controls. However, the results showed that the children with narcolepsy experienced more symptoms of anxiety, depression and anger than the healthy controls. The children with narcolepsy also reported poorer psychosocial well-being and health-related quality of life than the healthy controls.

Conclusions: The majority of children with narcolepsy included within this study were treated with medication at the time of participation. The results show that the children with narcolepsy had more sleep disturbance, were less active when awake and had poorer psychosocial well-being than matched healthy controls. However, there were no significant differences found in cognitive performance between the two groups, suggesting that well-managed children with narcolepsy do not have impaired cognitive function compared to healthy controls. Given these observations, clinician’s may wish to carefully monitor the sleep and psychological health of children with narcolepsy.

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MATERNAL-INFANT SYNCHRONY IN STRESS RESPONSE DURING BEDTIME SEPARATION

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Introduction: Infant sleep interventions that encourage infants to self soothe, are dependent on a mother separating from their infant at bedtime. For many years, parents have reported stress when instructed to separate from their infant at this time. Much debate has developed over how much stress these sleep interventions actually cause infants and mothers but surprisingly, very few studies have specifically measured this stress at the moment of bedtime separation. This study aimed to determine whether maternal assessment of infant distress is confirmed by levels of infant cortisol when separated at bedtime.

Materials and methods: Sixteen mothers of 16 infants (mean age 9.55 +/- 2.0 mths), recorded their infants’ stress subjectively (measured by questionnaire items “Is your baby stressed?”, “Is your baby crying?” and a crying intensity item), and objectively (mothers collected salivary cortisol at bedtime, before and 40 minutes after separation). Cortisol samples were analysed (Salimetrics), to evaluate changes in infant stress levels before and after bedtime separation. In addition, maternal perceptions of infant distress were measured to determine whether these maternal perceptions would mirror physiological measures of infant distress (i.e. cortisol levels).

Results: Four infants were excluded due to poor quality data. Of the remaining 12, maternal self-reported stress was related to infant stress, and maternal assessments of how stressed their infant was, were reflected in higher cortisol levels, although these levels did not reach significance. The study findings also suggest that, for the most part, the bedtime stressor was insufficient to disrupt the maternal-infant relationship.

Conclusions: Maternal-infant separation at bedtime does result in maternal stress, and this maternal stress tends to be a reflection of perceived infant stress as well as infants stress levels as measured by cortisol. Finding variability may reflect the current small sample size, therefore data collection will continue until a larger sample size is achieved to adequately address the research question.

Acknowledgements: This data is from Jo Osborne’s Honours Psychology Thesis.
P#16-Sunday
SLEEP CLASSES IMPACT ON SCHOOL PERFORMANCE: DATA FROM AUSTRALIAN INDIGENOUS CHILDREN

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Introduction: There is a significant gap between Australian indigenous and non indigenous children in health and education. Sleep duration and sleep schedule variability have been related to negative health and wellbeing outcomes in children but little is known about Australian Indigenous children. It is worthy of exploration to see if poor sleep contributes to these performance outcomes in these children. Findings could inform strategies to improve school performance through improving sleep thus decreasing this gap.

Materials and methods: Data for children aged 7-9 years came from the Australian Longitudinal Study of Indigenous Children and the National Assessment Program Literacy and Numeracy (NAPLAN). Latent class analysis determined sleep classes taking into account sleep duration, bedtimes, waketimes, and variability in bedtimes from weekdays to weekends. Regression models tested whether the sleep classes were cross-sectionally associated with grade 3 NAPLAN scores. Latent change score modeling then examined whether the sleep classes predicted changes in NAPLAN performance from grades 3 to 5.

Results: Five sleep schedule classes were identified: Typical Sleep, Early Risers, Long Sleep, Variable Sleep and Short Sleep. Long Sleepers performed best while those with reduced sleep (either Short Sleepers or Early Risers) performing the worse on grammar, numeracy, and writing performance. Variable Sleepers, Short Sleepers (and interestingly Typical Sleepers) also showed consistently poor performance across the school years 3-5.

Conclusions: In this sample, short sleep was associated with poorer school performance in several domains both in the short and longer term. In addition, for the first time, variable sleep has also been shown to also impact performance in a similar way. These data assist in understanding avenues for improving sleep and thus downstream school performance. It is also likely that short and variable sleep will be predictors of poor school performance in non-indigenous children suggesting a common pathway to improved education outcomes for all children via improved sleep schedules.
P#45-Sunday

EFFECTS OF CHRONIC EXPOSITION TO NOISE ON HOMEOSTASIS IN JUVENILE RATS

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Introduction: One third of the developed countries population has a poor sleep quality or reduced sleep duration. One of the cause of these complains is noise. The noise is generally defined as an unwanted sound which can have auditory but also extra-auditory effects on health. Furthermore, in OECD, 100 million peoples go through noise exposure exceeding 65dB(A) whereas levels should not exceed the threshold of 55dB(A) because could provoke awakening. The aim of this study is to see the effects a chronic high level noise exposition on juvenile rats on sleep and homeostasis parameters.

Materials and methods: 24 3-weeks old male Wistar rats (12 control vs 12 noise exposed) were exposed or not to noise 5 weeks at 87.5 dB 50-20.000Hz, mixing environmental and artificial noises during the light-rest period of the rat (12h from 6:00 am to 6:00 pm). The noise level of control was under 65dB. After 3 weeks of exposition, rats were surged to dispose polysomnographic telemetric implants (EEGs and EMG). At 5th week of exposition, 24-hrs polysomnography was performed and total duration (min, % of Total Sleep Time, TST), frequency (ep/h), mean duration of episode (s) for Wake, NREM, REM and Quiet Wakefulness were measured. The ratios NREM/TST and REM/TST were calculated. Food and water intake and weight of the animals were recorded. At the beginning of the 6th week, plethysmography was set for 1 hour (10:00 to 11:00 in non noise-exposed area) and apneas were analyzed in correlation with sleep states. The data were analyzed with Statview software and expressed in Mean ± Standard Deviation (SD). Depending on normality (Kolmogorov-Smirnov), two-way ANOVA with PLSD Fisher post-test or Mann-Whitney analyses were performed. Differences were considered significant at p<0.05. Significant results (p<0.10) are given when consistent.

Results: The weight of noise group animals tended to increase compared to controls (279.5±26.7 vs 290.6±33.3 g, p=0.055). Noise exposed animals ate more (total quantity: +2.72 g/24h, p<0.001) as a result of increased number of meals (+1.2 meals/24h, p=0.001) and increased average duration of a meal (+153 s, p=0.002). Moreover, these animals drunk more (+7.3 mL/24h, p<0.001). The number of apneas significantly increased in noise exposed group (1.1±1.9 vs 3.2±5.4 apnea/h, p=0.04), whatever the sleep states. Chronic noise exposure significantly affected sleep with a diminution of Quiet Wakefulness and NREM episodes (respectively -2.3 ep/h, p=0.05 and -2.4 ep/h, p=0.003) whereas mean duration of NREM episodes increased (17.6 s; p=0.058). The other noise effects on sleep depend on nycthemeral period: during the light-rest period (noise exposure), there was no significant modification of sleep. In contrast, during the dark-activity period (no noise), total duration of NREM sleep decreased (-3.1% of TST, p=0.017) at the expense of REM sleep.

Conclusions: The chronic noise exposure at high-level exposure of 87.5 dB during the light-rest period induced significant sleep disturbances with modification of sleep microstructure without great modification of sleep macrostructure. These effects generally occur during the dark-rest period. Indicators of general homeostasis were also perturbed as shown by increased of body weight, hyperphagia and sleep apnea.
OUTCOMES OF PERSONALISED SLEEP PROGRAMMES DELIVERED BY SPECIALIST SLEEP PRACTITIONERS IN A TERTIARY SERVICE: A 12 MONTH REVIEW

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Introductions: The Southampton Children’s Hospital Sleep Disorder Service provides multi-disciplinary specialist care for chronic sleep disorders. 350 new patients were assessed in 2017. There are three main work streams dependent on the underlying sleep disorder(s): (1) hypersomnias, sleep related movement disorders and parasomnias are managed by medical sleep specialists and a clinical nurse specialist; (2) primary anxiety related insomnia by psychologists; (3) chronic behavioural insomnia (with associated chronic health or developmental disorders) and circadian rhythm disorders by specialist sleep practitioners with expertise in behavioural sleep medicine who deliver personalised sleep programmes following an initial medical assessment. A local comprehensive sleep score (Southampton Composite Sleep -SCS- Score) measuring severity of sleep difficulties and impact on child and parent/carer is routinely completed pre and post intervention.

Materials and methods: We conducted a retrospective review of children managed by the specialist sleep practitioner team (work stream 3) discharged between March 2017 and February 2018. We evaluated outcomes based on the SCS score pre and post intervention using paired samples t-test. Characteristics of families who engaged positively with sleep were compared with those who disengaged from therapeutic work using Mann-Whitney U /Chi-Square tests.

Results: Ninety-eight children (64 male), median age of 5 years (range 1-17) were included in this review. 71% had at least one underlying physical or mental health diagnosis, 31% had a diagnosis of autism spectrum disorder. Primary sleep disorder diagnosis included behavioural insomnia (78%), circadian rhythm disorder (delayed 11%, advanced 1%), and anxiety related insomnia (6%). 36% had more than one sleep disorder.

In line with UK national statistics data, 68% lived in two-parent and 26% in single-parent families. 73% had either no or one sibling; 26% shared a bedroom with sibling(s); 38% co-slept with their parent(s) at initial assessment.

Children received support from specialist sleep practitioners for a median of 29 weeks (range 8 – 177). This included a median of 2 clinic appointments (range 1-27), 1.5 (0-14) telephone and 1 (0-7) email contacts. Eleven patients underwent either full (n=1) or respiratory polysomnography (n=10), 31 patients had actigraphy. Personalised behavioural sleep programmes (66%), phase advancement (32%) and chronotherapy (2%) were the main management approaches; in 30% two approaches were used. 25% of patients received melatonin as either hypnotic or chronobiotic.

The mean total SCS-Score improved from 11.24 (SD 3.6) at initial assessment to 4.86 (SD 5.3) at discharge (p<0.001). Families of older children who had more diagnoses were more likely to complete sleep programmes. An early disengagement rate of 16% was associated with lower number of underlying diagnoses and younger age (p=0.030 and p=0.048 respectively).

Conclusions: Contrary to expectation families of children with greater clinical complexity were more likely to engage positively with a personalised sleep programme. The improvements in composite sleep score were both statistically and clinically significant. Specialist sleep practitioners, skilled in behavioural sleep medicine, can work very effectively with children with complex physical health problems and developmental disorders to deliver successful personalised sleep programmes.
POLYSOMNOGRAPHIC STUDY IN PRIMARY-SCHOOL CHILDREN WITH ATTENTION DEFICIT HYPERACTIVITY DISORDER

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Introduction: Children with Attention Deficit Hyperactivity Disorder (ADHD) display more sleep problems than their peers and 10% of them present persistent sleep problems. Sleep has a recognized role in cerebral plasticity and maturation. Poor sleep may worsen the predisposed attentional dysfunction in ADHD. Anyway, sleep diseases may also mimic ADHD-like symptoms. Correct differential diagnosis (sleep disease versus ADHD) or comorbidity diagnosis (ADHD + sleep disorder) is often complex.

Materials and methods: This retrospective study investigates subjective and objective sleep quality in a group of 22 ADHD children. Aged from 6 to 10 years (mean age 7.5), 18 boys and 4 girls, they were referred to our Sleep Unit for complaints of poor sleep and poor attention. They had not yet a conclusive diagnosis of ADHD and they were all stimulant drug-naïve at the time of PSG. They underwent a clinical interview, conducted by a sleep expert, and one-night polysomnography (PSG).

Results: Children’s family reported: snoring (77%), tiredness (68%), insomnia (41%) and parasomnia (41%). No one complained about clear Restless Legs Syndrome, but 14% reported "restless sleep". 86% complained about at least two sleep symptoms. 50% of families considered sleep problems more important than attention deficit and consulted first for sleep problems.

PSG showed a mean sleep latency of 21 m + 22; a mean sleep efficiency of 89% + 8 and total sleep time of 553 m + 61.
23% of children had a bad night at Sleep Unit with sleep latency > 30 min or sleep efficiency < 80%, with total sleep time < 420 m.
Mean slow-wave sleep percentage was 20% + 7 and Rem sleep percentage was 19% + 4.
PSG showed periodic limb movement disorder during wakefulness in 23% of children and an Apnea-Hypopnea Index between 3-4.9/h in 36%.
No one had a AHI > 5/h.
We had not recorded parasomnia episodes.
Clinical features of all children met the DSM-V diagnostic criteria for ADHD (14 combined and 8 inattentive presentation). Attention deficit was confirmed by a neurocognitive evaluation (Kitap battery).

Conclusions: The present results show that 50% of school-aged drug-naïve ADHD children arriving at our sleep consultation had a priority complaint of poor sleep rather than attention. PSG showed comorbid sleep disorders: parasomnia in 41%, mild OSAS in 36%, restless legs syndrome in 23%, insomnia in 23%.
PSG was a key step in the pathway of care of this population. It was followed by a neurocognitive and ADHD expert evaluation, confirming ADHD.
These results reinforce the idea that children complaining about attention and sleep problems should undergo a cognitive and PSG analysis in order to objectivize the priority symptom.
Sleep expert may be the first physician consulted by ADHD children. He has to propose a multidisciplinary approach in these cases.
P#20-Saturday
PITOLISANT IN CHILDREN WITH NARCOLEPSY: AN ON-GOING DOUBLE-BLIND TRIAL

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Introduction: Narcolepsy is frequent in childhood but there are little satisfactory therapeutic opportunities for Excessive Daytime Sleepiness (EDS) and cataplexy. Pitolisant is a first in class, orally active histamine H3 receptor antagonist/inverse agonist, enhancing the histaminergic transmissions in brain. Given once-a-day to adults, it reduces EDS and cataplexies and is well tolerated (Dauvilliers Lancet Neurol 2013, Szakacs Lancet Neurol 2017). This led us to start a phase 3 trial in children (Clin Trials gov. NCT02611687).

Objectives are to assess the efficacy of pitolisant, up to 36mg/d, or 18mg/d whether weight <40kg (as shown in a former pharmacokinetic study EUDRACT Number: 2013-001505-93), compared to placebo in a double-blind randomized trial, to reduce EDS, measured by Pediatric Daytime Sleepiness Scale (PDSS), Maintenance Wakefulness Test and the number of cataplectic episodes in narcoleptic children treated for 8 weeks; and to determine the safety profile, assessed by follow-up of vital signs, ECG and laboratory tests. Additionally, an open-label extension will assess the long term efficacy and tolerance.

Materials and methods: This international study includes 96 narcoleptic children with or without cataplexy, balanced between two age groups 6-11 and 12-18 years, with EDS measured by PDSS ≥ 15. All other conditions considered as the primary causes of EDS are excluded. Diagnosis is confirmed with polysomnography and Multiple Sleep Latency Test. Children should be free of psychostimulant during the whole study. Nevertheless, patients treated by Xyrem® and anticitatplectics are allowed to continue their treatment if given at a stable dose since at least 4 weeks before inclusion.

The starting dose is of 4.5 mg, increasing to a maximum oral dose of 18 or 36 mg once daily, according to efficacy, tolerability and patient weight. Patients with a weight less than 40kg will be treated with a maximum daily dose of to 18mg. Treatment taken the morning before breakfast. The double blind period is 32 weeks and the open label.

The primary endpoint will be change in EDS measured by the Paediatric Daytime Sleepiness Scale (PDSS) between baseline: \[V_1\) score (D-14) + \(V_2\) score (D0)]/2 and the end of treatment: \[V_6\) score (D49) + \(V_7\) score (D56)]/2. Results between pitolisant and placebo groups will be compared.

Results: The results of this study are expected in 2019, and will give data on the efficacy, the optimal dosage and the safety of pitolisant for narcolepsy children from 6 to 18 years old.

Conclusions: If efficacy of EDS and cataplexy and safety results of pitolisant in this study are confirmed, pitolisant could be considered as a new therapeutic option for narcolepsy children from 6 to 18 years old with or without cataplexy. This study should also confirm that children under 40 kg should receive half dosage of pitolisant compared to adults as it has been previously shown in the pharmacokinetic pediatric study.

Acknowledgements: Bioprojet thanks all the investigators of this study.
P#17-Sunday
TOLERANCE AND PHARMACOKINETICS OF PITOLISANT (WAKIX®), A HISTAMINE H3-RECEPTOR ANTAGONIST/INVERSE AGONIST, IN 24 NARCOLEPSY CHILDREN

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Introduction: Pitolisant has been recently approved in Europe (March 2016) for “treatment of narcolepsy with or without cataplexy”, in adults. Due to its good safety profile and its efficacy on both Excessive Daytime Sleepiness and Cataplexy in adult, it was relevant to study pitolisant in pediatric narcoleptic patients. This trial assessed its tolerance and pharmacokinetics in pediatric patients suffering from narcolepsy with and without cataplexy.

Materials and methods: Four subgroups of 6 pediatric patients equally balanced with gender and age (6-11 years (sub-group I) and 12-17 years (sub-group II)) received an 18mg pitolisant single tablet. Blood samples were collected up to 10 hours post-dose and general clinical, cardiovascular, biochemical and hematological tolerance were assessed. Pitolisant and its main metabolites plasma levels were measured by UPLC/MS. First, the pharmacokinetic parameters were analysed and compared by subgroup and gender. Secondly, these pharmacokinetic parameters were compared to data previously obtained in young healthy male and female adult volunteers who received a single 18 mg pitolisant oral dose.

Results: No significant safety signal was recorded. Cmax values in males and females were respectively 60.5 and 48.5 ng/mL in children, 30.86 and 42.07ng/mL in adolescents. Corresponding AUC0-10h values were 332.02 and 293.72 ng.h/mL in children, 156.42 and 207.95 ng.h/mL in adolescents. Mean Cmax and AUC0-10h were 20% and 12% lower, respectively, in female than in male children patients. Corresponding values were 36% and 33% higher in female than in male adolescent patients. There was no statistically significant gender effect in each subgroup.
Compared to data previously obtained in young healthy male and female adult volunteers, mean Cmax were 68% and 51% lower while AUC0-10h were 70% and 48% lower than in children and adolescents. Tmax (~2h) were similar. These differences were not explained by any direct age, gender nor BMI effects. The sole direct correlation was with bodyweight.

Conclusions: The 18mg pitolisant single dose tolerability was excellent in narcolepsy pediatric patients. No gender nor age effect on drug exposure was observed. Since body weight impacts drug exposure, and despite the excellent pitolisant safety profile, the daily recommended dose should be 18mg for patients weighting less than 40 kg.

Acknowledgements: Bioprojet thanks all investigators attending this pediatrician clinical study.
CHARACTERISTICS OF SLEEP HYGIENE, SLEEPINESS AND SELF-ASSESSED SLEEP QUALITY IN LATVIAN ADOLESCENTS: DIFFERENCES BETWEEN BOYS AND GIRLS

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Introduction: Although data about sleep hygiene and sleep behavior (analyzing Sleep Hygiene Index (SHI), Epworth Sleepiness Scale (ESS)) are available, sleep practices differ among populations/countries and could also change in time, therefore it might be difficult to generalize data to all populations. The objective of the study was to analyze sleep hygiene and sleepiness in Latvian adolescents using SHI and ESS and to analyze possible differences between boys and girls.

Materials and methods: Prospective study was carried out in different schools in Latvia including adolescents attending seventh to twelfth grade. Data were collected using three questionnaires: SHI, ESS and the general demographic data (age, gender) with a part of self-assessed sleep quality. Statistical analysis was performed using Pearson correlation coefficient, Mann-Whitney test, ANOVA and Chi-squared test.

Results: Final sample consisted of 973 respondents (402 (41.3%) male) with the mean age of 15.4 (SD ±1.7) years (median age - 15 years, range 12 to 19 years).

In the total patient sample the mean SHI score was 32.9 (SD ±6.0; range: 14-53), median ESS score - 9.0 (range: 0-24); 197/973 (20.6%) adolescents assessed their sleep quality as bad.

Both SHI and ESS scores increased with age: the mean SHI from 30.0 (SD ±6.7) in 13-year-olds to 39.0 (SD ±6.6) in 19-year-olds (p<0.001) and the median ESS from 7.0 (QR=0.5) in 13-year-olds to 13.0 (QR=0.16) in 19-year-olds (p=0.0025). Scores of SHI showed weak but significant correlation with ESS scores (correlation coefficient = 0.305, p<0.001).

Mean SHI scores and mean ESS scores were significantly higher among girls compared to boys (33.7 (SD ±6.2) vs. 31.7 (SD ±5.6) and 9.4 (SD ±4.4) vs. 7.9 (SD ±4.4); p<0.001 respectively). Significantly more girls assessed their sleep quality as bad compared to boys (128 (65%) vs. 69 (35%) (p=0.04). In particular, girls scored significantly more in the following questions: retiring at different times; exercising to the point of sweating before retiring; energizing activities before retiring; being stressed before retiring; using bed for other things other than sleeping and sex; important work before retiring and thinking, worrying, planning while in bed. Male “bad sleepers” indicated not having enough sleep and feeling tired in the morning as causes of bad sleep.

Conclusions: The findings highlight that the sleep hygiene of Latvian adolescents is relatively good, but both sleep hygiene and sleepiness worsen with age. Females have worse sleep behavior possibly due to emotional factors (like feeling stressed, nervous or anxious before retiring) that possibly affect sleep quality, while boys sleep worse mostly because of not having enough time for sleep.

However, differences between both sexes should be studied more detailed investigating possible influence of other (for example, endocrine) factors.
SCREENS ARE HARMFUL FOR SCHOOL-AGED CHILDREN’S SLEEP AND BEHAVIOR: YES, BUT NOT DURING MORNING HOURS

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Introduction: The light produced by screens in the evening delays melatonin production and therefore affects the quality of sleep of school-aged children evening users of TV, tablets and video-games (Brockman et al., 2010), aggravating a Delayed Sleep-Wake Phase as well. On the contrary, exposure to light in the morning contributes to a better functioning of biological rhythms (Auger et al., 2015). This study aims to (a) confirm the harmfulness of screens on children’s sleep and behavior, (b) determine whether morning, afternoon or evening exposure has a different impact on sleep quality and (c) evaluate the extent to which exposure to screens has an impact on the relation between sleep and behavioral disorders.

Materials and methods: Parents of 375 children aged between 6 and 16 years old completed a questionnaire concerning screen use of their children, the French-version of the Sleep Disturbance Scale for Children (Putois et al., 2017) and the ADHD Rating Scale (DuPaul et al., 1998). Apart from correlation analysis, data were also processed using structural equation modeling, which offers information on the dependence relationship between sleep, screens and behavior.

Results: The presence of a screen in a child’s bedroom is correlated with sleep disorders, and in particular insomnia, parasomnias, daytime somnolence and sleep deprivation as well as inattention and impulsivity in terms of behavior. Using screens during evening hours is correlated with the severity of insomnia and hyperactivity. However, using screens during morning hours had no impact neither on sleep quality nor on behavior. Mediation analysis revealed sleep disturbance as a mediator of the influence of screens on hyperactivity-impulsivity-inattention behavior.

Conclusions: This study replicates the results of Brockmann et al. (2015) using a larger sample and older children. Exposure to screens is harmful for the sleep quality and behavior of children, except during the morning hours. Causal relationships between screens, sleep and behavior are therefore discussed. The results of our study should allow pediatricians, aiming to prevent sleep disorders in school-aged children, moderate their recommendations in terms of children’s screen exposure.

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P#02-Saturday
SLEEP DISORDERS IN CHILDREN WITH ASTHMA

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Introduction: Both OSA and asthma involve airway obstruction, but being at different levels. Airway inflammation, as we know, a characteristic of asthma could be, recently regarded associated with OSA. So we should pay more attention in asthmatic population. The purpose of this study is to assess SDB in children with asthma to assess sleep fragmentation in children with asthma to evaluate the association between sleep in asthma children and severity of lung disease.

Materials and methods: Lung Function: FVC, FEV1, PEF as a percent predict
PSG performed on all subjects before and after asthma controlled

Results: The data suggest that the prevalence of pediatric sleep-disordered breathing and sleep fragmentation could be very high among children with asthma no matter well-controlled or non-well controlled. Non-well controlled asthmatic children had a significant decrease in sleep efficiency [SE; 76.3% (N) vs 79.1% (W); P<0.05] prolonged rapid eye movement (REM) latency [150.5min (N) vs 88 min (W); P<0.05]. Reduction in percentage of REM sleep [12.7 (N) vs 18.3 (W); P<0.05]. Non-well controlled asthmatic children compared with well-controlled asthmatics. Higher apnea-hypopnea index (P < 0.05). Apnea-hypopnea-related arousal index (P < 0.05). SE (indicated the degree of sleep disruption) was correlated with FEV1. There was no significant correlation between SE and end-tidal pCO2.

Conclusions: Asthmatic children have significant sleep fragmentation. OSA may coexist with asthma. Sleep disruption is associated with severity of lung disease. But sleep disruption is not directly correlated with the degree of nocturnal hypoxemia or hypoventilation.

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LONGITUDINAL POLYSOMNOGRAPHIC FINDINGS IN INFANTILE POMPE DISEASE WITH ENZYME REPLACEMENT THERAPY


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Introduction: We initiate the first Pompe disease newborn screening program since 2005, and patients with infantile onset Pompe disease with early introduction of enzyme replacement therapy can now survive much longer. However, the deficiency of acid alpha glucosidase still confounds the neuromuscular function and leads sleep-disordered breathing in these children.

Materials and methods: In this prospective study, we followed all patients (n=12) diagnosed with infantile Pompe disease who have been identified through newborn screening program. The methods of screening, confirmatory process, criteria for initiating ERT, treatment dosage and regimen, and follow-up have been described previously. Patients were scheduled to have overnight polysomnogram (PSG) at Sleep center of National Taiwan University Hospital starting at the age of 1 year and then every year. Repeated PSG was performed if clinically indicated, such as after adenotonsillectomy.

Results: The most severe patient (NBS3) at the baseline PSG study had improvement in obstructive event after the surgery (from 2.1/hr to 0.4/hr), but his hypopnea aggravated (from 9.1/hr to 14.6/hr) with more frequent desaturation event (from 9.1/hr to 16.8/hr) despite less arousal. Therefore he was instructed to have CPAP at night. His condition remained stable until the most recent follow-up at the age of 10.7 years when he lost the ability of independent walking. The most severe patient (NBS6) at the most recent follow-up refused CPAP and his hypopnea developed gradually with more frequent desaturation event, and finally hypoventilation with CO2 retention. Adenotonsillectomy had no effect on this patient, since he presented no obstructive before the surgery, and the hypopnea increased from 2.5/hr to 5.5/hr after the surgery. Two other patients presented moderate-severe OSA during the follow-up, but improved with increasing rhGAA dosage and frequency (NBS5), and adenotonsillectomy (NBS8).

Conclusions: A high incidence of both hypoventilation and obstructive sleep apnea (OSA) present in this group, with relatively stable in both OSA and central sleep apnea. Adenotonsillectomy and CPAP may be equally effective in children diagnosed with mild to moderate OSAS by PSG in children with ERT, despite a long-term effect due to progressive myopathy was not present.
CRANIOFACIAL AND AIRWAY MORPHOLOGY CHANGE AFTER WEARING ORAL APPLIANCE DURING SLEEP FOR ONE YEAR ON A COHORT OF OBSTRUCTIVE SLEEP APNEA CHILDREN

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Introduction: Myofunctional therapy (MFT) for obstructive sleep apnea (OSA) could improve breathing during sleep, and decrease Apnea-Hypopnea Index (AHI) by about 50% in adults and 62% in children. MFT can improve the tongue muscle, particularly in hypotonic premature infants. But the level of compliance for MFT has often been low in children. The oral device with a tongue bead could be a passive MFT appliance and improve clinical respiratory symptom in the OSA children during sleep. The device is designed on the basis that intrusion of a foreign object close to the tip of the tongue stimulates tongue activity at least during light stages of sleep.

Materials and methods: The aim of the present investigation was to evaluate the treatment effect of the passive myofunctional therapy through an oral appliance with a tongue bead and to compare the differences in craniofacial and airway morphology before and after wearing the oral appliance for preterm with full term obstructive sleep apnea children. Twenty-nine children with OSA problem were included and all participants were wearing the oral device during sleep for one year. The lateral cephalometric radiograph was taken to compare lateral craniofacial and airway morphology before (T0), during (T6m), and after (T1y) the oral device treatment.

Results: The skeletal morphology had significantly grown in anterior facial height, posterior face height and the ratio of PFH/AFH after wearing the oral appliance after one year. The airway morphology had been significantly bigger space in nasopharynx.

Conclusions: The oral device could significantly improve nasopharyngeal airway and posterior facial growth.

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P#50-Saturday

IMPACT OF PRONE POSITIONING ON OBSTRUCTIVE SLEEP APNEA IN INFANTS WITH PIERRE ROBIN SEQUENCE

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Introduction: The management of Pierre Robin sequence (PRS) infants is not standardized. Among therapeutic options of obstructive sleep apnea (OSA), the prone position during sleep is widely used but has not been validated. The aim of the study was to highlight by polysomnography the impact of the prone position on both sleep and respiratory events.

Materials and methods: This retrospective study was performed in the sleep unit of the pediatric university hospital of Lyon. A nocturnal polysomnography with pulse oximetry and transcutaneous PCO₂ were performed in prone and supine position for each PRS infants during their routine follow-up. Sleep and breathing were compared between prone and supine position. Sleep criterion and respiratory criterion were analyzed, expressed in mean with standard deviation.

Results: The polysomnography of 21 PRS infants, from 2015 to 2017, were analyzed. The statistical analysis was performed on 18 PRS infants, aged of 61 days ± 50. In the prone position (mean ± SD: 80 ± 15 %) infants had a significantly higher efficiency than in the supine position (65 ± 21 %, p=0.04), and there was trend towards higher efficacy (82 ± 14 % versus 70 ± 20 %, p=0.11). In the prone position there was a trend towards lower respiratory micro-arousals index than in supine position (18 ± 16 % versus 21± 15 %, p=0.06). In the prone position in REM, there was a trend towards lower obstructive apnea hypopnea index than in supine position (62 ± 47 % versus 80 ± 66 %, p=0.05). In prone position in total sleep time, there was a trend towards lower obstructive apnea index (19 ± 26 % versus 34 ± 51 %, p=0.06). In the prone position, desaturation index 4% was lower than in supine position (21 ± 18 % versus 32 ± 29 %, p=0.03). Among 21 included infants, 14 infants presented a better quality of sleep in prone position than in supine. Among 21 included infants, prone position was sufficient for 4 infants. Respiratory support were required for 15: 12 infants were turned on NIV, 2 needed settings of their NIV and 1 needed a nasopharyngeal tube.

Conclusions: In PRS infants, regardless of breathing clinical severity, prone positioning improves both breathing and sleep quality. However, prone position could not be sufficient to correct OSA, even in the absence of OSA symptoms. A sleep recording seems to be indicated for all PRS infants. Because of the concordance between breathing and sleep quality on sleep recording, a simple polygraphy, in first intention, could lead to the appropriate respiratory therapeutic option.
A STUDY TO ASSESS THE NORMAL 3% DESATURATION INDEX (DI) IN HEALTHY CHILDREN UNDER 12 YEARS USING MASIMO TECHNOLOGY

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Introduction: The gold standard for quantifying the amount of sleep disordered breathing (SDB) is polysomnography. These studies report the apnoea hypopnoea index (AHI). Previously scoring hypopnoeas was dependent on a 4% desaturation but in 2012 the criteria were amended such that hypopnoeas are now scored on the basis of a 3% desaturation (American Academy of Sleep Medicine). Pulse oximetry is a simple non-invasive method of monitoring blood oxygen saturations and is often used to determine the presence or absence of SDB. Short pauses in breathing result in transient falls in oxygen saturation. These can be detected by modern pulse oximeters which have short averaging times. Criterion most frequently used to detect abnormality are the mean oxygen saturations (SAT50), the 4% desaturation index (DI4) and the delta 12 index (DI12s). In particular a DI4 >2 has a high predictive value for an AHI >1. With changes to the AASM guidelines, 3% desaturation indices (DI3) are increasingly reported rather than DI4. Normative data for DI3 using modern oximeters are currently not available.

Materials and methods: Healthy children under 12 years of health care professionals underwent nocturnal home pulse oximetry using Masimo Rad-8 pulse oximeters. Sleep onset time, morning wake time, periods of wake throughout the night and the quality of sleep during the study was recorded. Parents completed a questionnaire about their child’s sleep, with a particular focus on symptoms relating to SDB. Data was analysed using Visidownload software, with wake periods and artefact extracted. A minimally acceptable study required artefact free recording time (AFRT) of at least 4 hours. Data was collected on DI3, DI4, mean saturations (SAT50), minimum SpO2 (SATmin) and percentage of time spent with SpO2<88% (SAT88).

Results: Sleep studies were performed in 80 children; 72 studies provided adequate AFRT. No child had clinical evidence of SDB. The median (IQR) age of participants was 7.0 years (5.0 to 9.8 years). The mean (95% CI) value for DI3 and DI4 was 2.81 (2.42 to 3.20) and 1.10 (0.90 to 1.30) respectively. The mean (SD) values for SAT50 and SATmin were 97.59% (0.81) and 90.68% (6.00) respectively.

Conclusions: These results in healthy children support existing data on DI4. This is the largest cohort to report DI3 data using a modern oximeter with short averaging times. This normative data will be helpful in the interpretation of pulse oximetry traces. Further work is required to define the relationship between DI3 on standalone oximeters and AHI as determined by polysomnography.

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EPILEPTIC MYOCLONUS AND SLEEP MYOCLONUS: DIAGNOSTIC CHALLENGE AND USE OF POLYSOMNOGRAPHY WITH EXTENDED MONTAGE - CASE REPORT

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Introduction: The recognition of the reciprocal relationship between sleep and epilepsy is not recent. In certain epilepsy syndromes, this association is even clearer. In practice, the distinction between epileptic and non-epileptic events during sleep can be challenging, and even after a detailed semiological description, it may be not possible to accurately differentiate such events from one another. So it is essential to use a complementary method with video, including videopolissonography (vPSG).

Materials and methods: Medical history review and vPSG analysis.

Results: BFO, male, aged 15, with Generalized Epilepsy probably symptomatic, followed up at the Epilepsy Outpatient Clinic with different types of seizures since he was 1 year and 3 months of age (febrile, tonic-clonic, tonic and myoclonic ones). He had good seizure control, but his mother complained that every night, 30 to 60 minutes after falling asleep, the patient presented highly stereotyped movements, interpreted then as benign sleep myoclonus. Because of the need to distinguish between epileptic and non-epileptic nocturnal events, it was decided to use polysomnography with extended montage. It was observed that, in NREM sleep, fourteen clinical events characterized by segmental axial myoclonus (cervical flexion and / or elevation and proximal abduction of upper limbs) or global broad ones, were electrographically coincident with epileptiform paroxysms of isolated spike and polyspike types followed by slow waves, of diffuse projection and previous accentuation, high voltage (200-350 μV spikes / polyspikes, slow waves of 600-800 μV) and duration between 0.4 and 0.6 seconds. Such paroxysms sometimes occurred without association with clinical phenomena (16 events). In REM sleep, the described axial myoclonic phenomena were observed, as well as global awakening reactions, but without association with epileptiform paroxysms. On 11 occasions, both in NREM sleep and REM sleep, motor episodes were preceded by central or obstructive respiratory events, and on 5 occasions, motor events were followed by them. Sleep efficiency was normal (86.7%); there was an increase in the arousal index (32 / h); mild increase in N1 sleep (12%); increased obstructive apnea + hypopnea index (3.9/h) and respiratory events index (9.8/h); presence of mild desaturations, with a minimum SATO2 of 92%; and presence of REM sleep without atony.

Conclusions: Approximately 20% of patients with epilepsy have nocturnal seizures exclusively. It is known that sleep can facilitate epileptic activity; the thalamic-cortical synchronization during NREM sleep is the probable reason for the generalization and dissemination of epileptic discharges that occur more usually at this stage. Primary generalized epilepsy is one of the epileptic syndromes in which there is suppression of discharges during REM sleep. However, regarding the above-mentioned patient, there was maintenance of sleep-related minor motor events during this stage, manifested similarly to those observed in NREM sleep. Thus, we especulate if the manifestations of the REM stage are only benign sleep myoclonus, or the expression of the same epileptic phenomenon without electrophographical correspondence, which, is supposed be capable of detection only with the introduction of invasive electrodes.
SLEEP RELATED HYPOVENTILATION DUE TO PARTIAL BIOTINIDASE DEFICIENCY - CASE REPORT

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Introduction: Idiopathic sleep related hypoventilation occurs in individuals with hypercapnia during sleep, in normal conditions of the respiratory system, including airways, pulmonary parenchyma, ventilatory muscles and ribcage, in the absence of obesity, other sleep respiratory disorders and use of central nervous system depressors. Cases considered to be idiopathic may be related to unexplained conditions. The differential diagnosis includes any disorder that can cause hypoventilation during sleep. Hypoventilation may be associated to partial biotinidase deficiency, although this relation is very unusual. Biotinidase deficiency is defined as a genetic disorder which affects biotin metabolism and is the result of an autosomal recessive mutation in BTD gene located in 3p25 chromosome. The most typical presentation involves neurological and skin changes. This enzyme partial deficiency represents 10% to 30% of the average activity and it may be manifested later and with atypical symptomatology. This study reports a clinical case of sleep related hypoventilation due to partial biotinidase deficiency.

Materials and methods: Analysis of a three-year old female patient’s records, and relevant findings in literature from PubMed and Scielo databases.

Results: The patient's mother observed respiratory irregularities during sleep since the child was seven months old. She had shallow or periodic breathing, sometimes associated with bradycardia. Few months later, she had some apneic episodes while awake, associated with cyanosis or paleness and syncope. These episodes were triggered by crying or regurgitation. There was a previous history of reflux and ALTE (Apparent Life-Threatening Events) by the age of two months. The general physical and neurological examination were normal and neuropsychomotor development was appropriate. The capnography registered ETCO2 > 50mmHg during 60.4% of total sleep time, compatible with diagnosis of hypoventilation. The general metabolic investigation showed increased blood level of ammonia (107uG%) and serum lactate level was higher on two occasions (2.6 mmol/L e 4.8 mmol/L). Simultaneously, a genetic investigation was performed by exome sequencing which showed two BTD gene heterozygosis mutations. Drug based treatment was initiated early and consisted of 20 mg of biotin intake per day, with resolution of the described symptoms.

Conclusions: The importance of the present clinical condition lies in biotinidase deficiency with an isolated cardiorespiratory condition in a neurologically normal child, a case where the etiology of Breath Holding Spells was the deficiency of this enzyme. Both clinical conditions are possible. However, in practice they are not normally correlated due to this atypical manifestation. This relation was confirmed after central hypoventilation resolution when biotin was introduced. Biotinidase deficiency should be considered in the differential diagnosis of ALTE, breath holding spells, and sleep related breathing disorders in children, including "idiopathic" cases of hypoventilation. Besides, it illustrates the ALTE symptoms which terrify the observer and even professionals, have a simple resolution: the intake of biotin.
CAN FUNCTIONAL JAW ORTHOPEDIC BASED ON NEURO OCCLUSAL REHABILITATION CONTRIBUTE TO A MULTIDISCIPLINARY TEAM IN SLEEP-RELATED BREATHING DISORDERS PREVENTION?-CASE REPORT

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Introduction: Sleep Related Breathing Disorders (SRBD) in children is a frequent disease for which optimal diagnostic methods are still being defined. Habitual snoring, has been common parent complaints at pediatric dental practice. Some patients with this complaint can present narrow maxilla and mouth breathing related to respiratory allergy, as: rhinitis, sinusitis, tonsillitis adenoid hypertrophy. Treatment of SRBD in children should include improvement in upper airway space and craniofacial growth, resolving respiratory symptoms, and preventing the development of the disease in the adult years. The mouth is a peripheral projection of the central nervous system, which provides somatosensory sensitivity and motor activity, very well represented through Penfield & Rasmussen's homunculus. The Functional Jaw Orthopedic (FJO) based on Neuro Occlusal Rehabilitation (NOR) works with four natural forces: growth and development, dental eruption, tongue and jaw movement and posture. The fundamental principles of FJO based on NOR are neuromuscular excitation, posture change and therapeutic posture change. The stimulus used in FJO based on NOR can guide the stomatognathic system growth and development, modeling and remodeling bone with muscular dynamics, both in quantity and direction, following oral functions balance. According to the functional priority octagon theory, the posture of head, neck, jaw and tongue are closely linked to the air space, inframandibular region and spinal column. In addition, vicious habits produce proprioceptive and exteroceptive stimulus in the orofacial area, leading to neuromuscular compensations and development of inadequate functional patterns producing structural alterations and functional anatomic imbalance. In this way, timely intervention in child is necessary for reestablish neuromuscular patterns without prejudice to growth and development, restoring functional patterns when it is altered by vicious habits. The aim of this case report is to demonstrate how FJO based on NOR can contribute to a multidisciplinary team for SRBD prevention.

Materials and methods: Caucasian patient, male, 5 years old. Mother has had complained of respiratory allergy, vicious habits of mouth breathing and habitual snoring during sleep. Thought oral examination, the patient has mixed dentition with dental class II division 1, overjet and open bite, lips hypotonia and lower tongue posture. The basal teleradiograph analysis demonstrated: hyperdivergency of facial grow with severe divergence of maxillo-mandibular planes, increase in vertical facial dimension, prevalence of facial height over depth and presence of skeletal open bite and a narrow upper airway space. This patient was treated at the end of lower secondary prevention level with FJO based on NOR using three kinds of FJO appliances and Plana's direct tracks. For maximized response the patient was accompanied by multidisciplinary team. The patient was sent to the otorhinolaryngologist to treat respiratory allergy and to the physiotherapist to stimulate nasal breathing.

Results: The results of 3 years and a half treatment and 23 years follow-up were registered by cephalometry teleradiographs: increase in posterior air space, improvement of head and neck posture, advancement of mandible, hyoid and tongue. Intraoral photography showed permanent dentition, class I with improvement in dental arcs, correction of open bite and overjet and lip sealing. The initial mother complaints were solved: habitual snoring was remised, and nasal breathing was reestablished.

Conclusions: In this case report FJO based on NOR contribute to a multidisciplinary team in sleep-related breathing disorders prevention. Timely treatment and stability are necessary, intervening in the parts, verifying the causes, defining the effects and changes, aiming to establish functional balance. Additional studies in FJO based on NOR treatment should be performed with objective data related to SRBD to optimal diagnostic methods and criteria.
ERYTHROCYTOSIS WASN'T CLINICALLY SIGNIFICANT IN A GROUP OF PEDIATRIC PATIENTS WITH OBSTRUCTIVE SLEEP APNEA SYNDROME

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Introduction: It is though that intermittent nocturnal hypoxemia in obstructive sleep apnea syndrome (OSAS) might function as a sufficient hypoxic stimulus to increase hematocrit (Htc) levels, but there isn’t consistent clinical evidence of this. Sleep-disordered breathing has been shown to occur in up to 20% of patients with unexplained erythrocytosis and OSAS is usually considered a common cause of secondary erythrocytosis. Recent studies showed a positive correlation between nocturnal hypoxemia severity and Htc levels, but not clinically significant erythrocytosis.

The aim of this study was to determine the prevalence of erythrocytosis in a sample of pediatric patients with OSAS who needed noninvasive ventilation (NIV); correlating the severity of OSAS with Htc levels and evaluating the variation of Hct, hemoglobin (Hb) and red blood cell (RBC) count with NIV treatment.

Materials and methods: Retrospective study, including patients with OSAS diagnosed by clinical and polysomnographic criteria and treated with NIV, followed in a Sleep Laboratory from 1999-2017. Patients with conditions causing erythrocytosis or changes in blood count were exclude. The following variables were studied: age at diagnosis and of NIV beginning; body mass index; comorbidities; blood count before and after NIV; polysomnographic parameters according to AASM scoring manual. OSAS classification was: mild if apnoea/hypopnea index (AHI) 1-5, moderate if AHI 5-10, severe if AHI >10. Statistical analysis was performed using the SPSS® software and statistical significance was defined as p<0,05.

Results: Eighty-eight patients (65% male) were included, mean age at diagnosis being 9,9 ±5,3 years (0,2-9). Twenty-eight (32%) were obese, 5 (6%) were overweight and 5 had arterial hypertension. 85% patients had other comorbidities. About OSAS severity: 50% had mild OSAS, 19% moderate OSAS and 31% severe OSAS. There weren’t differences between OSAS severity and NIV beginning age, obesity or other comorbidities.

At diagnosis, mean Hb, Htc and RBC was 13,8±1,4 g/dL, 40,9±4% and 4,9±0,4x10³, respectively, with normal results in the majority of patients (86%, 78% and 67% respectively). Only 11% of patients had increased Htc for age (6% with mild, 3% with moderate, 2% with severe OSAS). The Htc was increased in 8,7% of patients with severe OSAS. The lowest SaO2 recorded ranged from 12- 98%.

There wasn’t significant correlation between lower SaO2 and Htc, Hb or RBC values (p>0,05). AHI mean was higher in the group with higher Htc, but without statistical significance (p=0,209). We found significant differences in Htc between the different OSAS groups, with higher values in moderate compared to mild and severe OSAS (p<0,05), but this might be explained by our small sample.

We didn’t find differences between Htc levels before and after NIV beginning (p= 0,954).

Conclusions: We didn’t find a significant relationship between erythrocytosis and OSAS. Only 11% of our patients had clinically significant erythrocytosis. We found no correlation between OSAS severity or nocturnal SaO2 and haematological parameters.

OSAS alone doesn’t seem to be sufficient to explain secondary erythrocytosis. The use of polysomnography as a diagnostic tool in finding erythrocytosis cause should be more judicious, especially when resources are limited.
NON-INVASIVE VENTILATION IN CHILDREN WITH UPPER-AIRWAY RESISTANCE SYNDROME

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Introduction: In the last decades, upper airway resistance syndrome (UARS) has been recognized as an important cause of daytime sleepiness, fatigue and cognitive morbidities, with a substantial impact in quality life. Some authors consider it a part of sleep-disordered breathing spectrum, others referring that it's a different respiratory disorder, usually present in non-obese patients and predisposed by genetic factors and smaller upper airways.

Paediatrics' prevalence is not well known, but some authors consider it more common than obstructive sleep apnoea syndrome (OSAS).

It has been associated with respiratory events combining absence of oxygen desaturation despite frequent nocturnal arousals related to abnormal respiratory effort and absence of OSAS.

The authors present 3 cases of UARS in children who needed non-invasive ventilation (NIV) for symptom control.

Materials and methods: Retrospective review of clinical information of 3 children with UARS. Diagnosis was set following clinical (snoring, daytime sleepiness/fatigue) and full-night polysomnographic findings: apnoea/hypopnea index (AIH) ≤1, oxygen saturation > 92%, presence of respiratory effort-related arousals (RERAs) and flow limitation events (non-apnoea/hypopnea events based on nasal cannula curve analysis), Respiratory Disturbance Index (RDI)>1.

Results: Two boys and one girl aged 12, 5 and 7 years-old at time of beginning CPAP were included. UARS diagnosis was established 14 months before (mean). They complained of snoring, restless and disrupted nocturnal sleep, difficulty to wake in the morning, diurnal sleepiness/fatigue, inattention and poor school performance (girl), and frequent sleep walking episodes (one boy). They had positive history for upper airway infections and 2 had allergic rhinitis. Two had been submitted to adenotonsillectomy, snoring recurrence occurring between 3 months and 2 years. All had family history of sleep-related breathing disorder. They had normal weight and blood pressure for age, but all presented some facial anomalies: elongated face, oral breathing, turbinate enlargement, nasal septum deviation, high and narrow hard palate, low soft palate, crossbite, slight retrognathia. All performed attended full-night polysomnography: total sleep time 457.8± 48 minutes, arousal index 9.3±5.1 ; AHI<1, oxygen saturation ≥95%, RERA index 2.9 ± 2.5, multiple flow limitation events and RDI 3.3 ± 2.2. They had no criteria for ENT surgery and the one with crossbite and high hard palate didn’t met criteria for orthodontic treatment. Under nasal corticosteroids, montelukast and antihistamines they remain symptomatic. NIV with continuous positive air pressure was started, with good tolerance and compliance been achieved. Mean follow-up is 3.8 years and they are symptom-free.

Conclusions: Unlike the description in literature about compliance in adults with UARS, our children maintained NIV with good compliance and symptom control. In paediatric patients some orthodontic and ENT treatments are age-limited so NIV must be an alternative, even temporarily. There has been great acceptance of this entity as an important cause of sleep disruption and impaired daily functioning, so recognizing and treating it in childhood/adolescence is imperative.
USE OF THE EMDR TECHNIQUE TO TREAT CHILD’S NIGHTMARE DISORDER ABOUT THE CLINICAL CASE OF LITTLE MYA

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Hôpital Montfavet (Avignon)

Introduction: According to the DSM 5 (American Psychiatric Association, 2013), nightmares are usually long and complex story like dream sequences that seem real and cause anxiety, fear, or other dysphoric emotions. Occasional nightmares are common among children (10 to 50% of children 3 to 5 years, American academy of sleep medicine, 2014). According to ICSD-3 (American academy of sleep medicine, 2014): Nightmare disorder is a parasomnia characterized by:
A. Recurrent episodes of awakenings from sleep with recall of intensely disturbing dream mentations, usually involving fear or anxiety, but also anger, sadness, disgust, and other dysphoric emotions.
B. Full alertness on awakening, with little confusion or disorientation; recall of sleep mentation is immediate and clear.
C. At least one of the following associated features is present:
   i. Delayed return to sleep after the episodes
   ii. Occurrence of episodes in the latter half of the habitual sleep period.

Materials and methods: EMDR is a psychotherapeutic approach that uses sensory stimulation on both sides of the body, either through eye movement or through auditory or cutaneous stimuli, to induce rapid resolution of symptoms related to past events. This method was discovered in 1987 by Francine Shapiro (Shapiro, 1987). It has been shown to be effective in the treatment of post-traumatic stress disorder (National Collaborating Center for Mental Health, 2005). EMDR is also used today in anxiety disorders, mood disorders or somatic disorders and performance for example (Augeraud, 2017).

EMDR therapy with children is a specialty of generic EMDR Therapy based on the model developed by Tinker and Wilson (1999) taking into account the child's level of development.

In the clinical case described, we realized that Mya's nightmares had started on her return from vacation in Brazil. The nightmares were centered on the fear of dying (for her and / or her family members) and on animal attacks, especially snakes. Disorders, who have been evolving for 4 months before the beginning of the care, were associated with an aggravation of the sleep-onset insomnia and a separation anxiety at bedtime. Moreover, a specific phobia related to snake was associated with this nightmare disorder.

We chose to treat Mya with the EMDR technique, taking the most vivid memory of the nightmare as a starting target.

We therefore used the child-friendly EMDR technique with Mya, combined with a behavioral approach.

Results: We used EMDR on two sessions for Mya. We also taught Mya and her parents relaxation techniques, and we have strengthened bedtime rituals. In children, EMDR sessions can use emotional cognitions to reprocess traumatic content.

We used cognition of fear during the sessions and we treated the episodes of recurring nightmares as well as the fear of snakes.

After two sessions of EMDR, little Mya regained quality sleep, and did not have any more nightmares.

Conclusions: The use of EMDR to treat young children nightmare disorder can be an effective technique associated with a behavioral management supported by parents.

Acknowledgements: Alan Smewing for his help.
Introduction: Pulse oximetry is a simple non-invasive method of monitoring blood oxygen saturations. It is often used to determine the presence or absence of sleep-disordered breathing (SDB) in settings where gold standard polysomnography is unavailable. Short pauses in breathing cause transient falls in oxygen saturation which is detected by modern pulse oximeters. Criteria most often used to detect abnormality are mean oxygen saturations (SAT50), 3% and 4% desaturation indices (DI3, DI4) and delta 12 index (DI12s). In particular DI4 >2 is highly predictive of an apnoea hypopnoea index (AHI) >1. It is currently unclear what constitutes a minimum artefact free recording time (AFRT) on which prediction of SDB can be made. This retrospective study aims to evaluate this.

Materials and methods: Children referred to the clinical service underwent nocturnal pulse oximetry using Masimo Rad-8 pulse oximeters. Data were analysed using Visidownload software, with wake periods and artefact extracted. For the purpose of this study only studies with at least 8 hours of AFRT were included. Mean saturations (SAT50), DI3, DI4, and DI12s were reported. Limits of agreement were calculated for the last 4 and 6 hours of sleep compared to the last 8 hours of sleep. Data at all time points was compared to determine whether individual cases crossed the upper 95 % confidence interval for our healthy cohort (abstract submitted).

Results: Thirty-nine children (15 female), median age of 4.6 years (range 0.4 – 17.4) were analysed. Indications for sleep studies were obstructive sleep apnoea (n=21), titration of ventilation (n = 10), weaning of oxygen (n = 6), assessment for hypoventilation (n = 2).

The mean difference and limits of agreement (SD, 95% CI) between 8 and 6 hours and 8 and 4 hours with regards to (i) mean saturations were 0.16 (0.18, -0.2 – 0.62), and 0.28 (0.47, -0.66 – 1.22) respectively; with regards to (ii) DI3 the difference was 0.36 (1.37, - 2.38 – 3.1), and 0.86 (2.79, -4.72 – 6.44) respectively; with regards to (iii) DI4 the difference was 0.25 (0.98, -1.71 – 1.06), and 0.58 (1.96, - 3.34 – 4.50) respectively; with regards to (iv) DI12s the difference was 0.01 (0.04, -0.07 – 0.09), and 0.04 (0.07, -0.1 – 0.18) respectively.

For DI4 the median (IQR) values were 2.39 (1.13 – 5.44), 2.51 (0.92 – 5.07), and 2.24 (0.75 – 3.71) for 8, 6 and 4 hour AFRT analysed respectively. When compared to the upper 95% confidence interval (CI) for our normative data (1.30), five cases switched from being above or below the 95%CI dependent on the length of time analysed. The remainder studies did not change (12 studies remained normal, 22 abnormal).

Conclusions: Mean differences are small between measurements for different lengths of study. Limits of agreement remain clinically important. This may reflect small sample size and data collection is ongoing. Of 39 children five switched from being normal to abnormal or vice versa. This was not statistically significant. On this basis we recommend that 4 hours of data is enough.
SHOULD CHANGES IN PULSE OXIMETRY AFFECT HOW THESE STUDIES ARE CLINICALLY INTERPRETED?

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Introduction: The American Academy of Sleep Medicine defined oxygen desaturation (OD) as a 3% drop from baseline for paediatric polysomnography in 2007 (1). These 3% ODs are used to identify apnoeas and hypopnoeas, which define the diagnostic outcome variable of polygraphy studies – the Apnoea Hypopnoea Index (AHI). Previous publications characterising normal oxygen saturations (2) and the predictive value of Pulse Oximetry (PO) (3) used an OD definition of 4%. Developments in PO technology have also occurred; advancements should improve diagnostic accuracy, but may alter the findings of previous studies as different proprietary recording technologies may produce significantly different results (4).

Southampton Children’s Hospital records PO using Masimo oximeters (Irvine, California) concurrent to integrated Nonin oximeters (Plymouth, Minnesota) in all cardiorespiratory polygraphy (CP) studies. In 2015 we presented data that showed Nonin oximeters consistently report higher 3% OD Indices (ODIs) than Masimo oximeters (5). This abstract explores how PO recordings from these oximeters correlate with AHI, as both Nonin (via the AHI, using 3% definition) and Masimo (using 4% definition) aid in clinical decision making. We also consider whether the 4% ODI remains clinically relevant.

Materials and methods: Data were collected retrospectively from CP admissions between 25/11/2014 and 31/01/2015 that had successful CP and PO studies. Periods of wake and gross body movement were removed from the CP studies using observations from the embedded video. Artefact and periods of 'Wake', based on patient observation, were removed from the Masimo PO studies. Nonin 3% ODIs, Masimo 3% and 4% ODIs, and AHIs from CP were tabulated. Statistical analyses were performed using SPSS.

Results:

Demographics:
N 82 Gender (Female) 40 Age (years) (median, range) 7.2 (3.3-20.2)

Table 1 - Correlation with AHI

<table>
<thead>
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<tbody>
<tr>
<td>Nonin 3% ODI</td>
<td>0.76</td>
<td>0.000*</td>
</tr>
<tr>
<td>Masimo 3% ODI</td>
<td>0.61</td>
<td>0.000*</td>
</tr>
<tr>
<td>Masimo 4% ODI</td>
<td>0.60</td>
<td>0.000*</td>
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*p<0.001

Conclusions: All four PO variables had statistically significant correlations with AHI, although, the internal Nonin 3% ODI was, unsurprisingly, most closely associated with AHI (Table 1). - Our dataset does not suggest the Masimo 3% ODI would improve diagnostic accuracy compared to the 4% ODI (Table 1).

A STUDY TO ASSESS THE PREVALENCE AND SEVERITY OF DIMS (DISORDERS OF INITIATING AND MAINTAINING SLEEP) IN A SAMPLE OF IRISH CHILDREN AGED 6-48 MONTHS

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Introduction: Disorders of initiating and maintaining sleep (DIMS) are relatively common in young children. International literature identifies a prevalence rate of 20-40%¹ amongst children 1-5 years. Currently there are no published studies which indicate the prevalence rate in an Irish population. Sleep disturbance can impact significantly on family life and can lead to neurocognitive and psychosocial impairment, but if identified early, interventions to address sleep difficulties have been shown to have consistently positive outcomes¹.

Materials and methods: The aim of this study was to establish the prevalence of sleep disorders in a sample of 6-48 month old Irish Children and the suitability of the Tayside Children’s Sleep Questionnaire² (TCSQ) for use in the primary care setting to identify children with behavioural sleep disorders.

Methodology
The study is a cross sectional study in two populations. The clinical sample comprised 50 children referred to a sleep clinic in County Donegal, Ireland. The control groups consisted of 100 children attending the Public Health Nurse (PHN) for statutory routine developmental assessment, in two sites in County Kildare and County Donegal. Parents were asked to fill out demographic details and the TCSQ.

Results: The prevalence rate of DIMS in the clinical sample was 96%. In the County Donegal control sample the prevalence rate of DIMS was 40% compared to a rate of 66% in the County Kildare control sample. There was a significant difference in scores on the TCSQ between the clinical vs. control groups. There was a significant relationship between scoring above the TSCQ cut-off point and parent's subjective perception of whether or not their children had sleep difficulties. The TSCQ was also identified by PHN's as a useful screening tool for DIMS.

Conclusions: The prevalence rate of DIMS in this study was slightly higher than previous studies. The results of this study indicate that the TCSQ is a suitable screening tool for use in a Primary Care Setting to identify DIMS in children 6-48 months.

Acknowledgements: The parents who agreed to take part in this study. Dr Frank Sullivan for permission to use the Tayside Children's Sleep Questionnaire. The Director and Assistant Directors of Public Health Nursing in County Donegal and County Kildare for their support with the study.
POST DISCHARGE EVALUATION OF PUBLIC HEALTH NURSE LED SLEEP CLINICS FOR BEHAVIOURAL SLEEP DIFFICULTIES IN CHILDREN

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Introduction: The Public Health Nursing service in County Donegal, Ireland now provides community based, Public Health Nurse (PHN) led sleep clinics for pre-school children with behavioural sleep difficulties in all five primary care networks in County Donegal. During the initial sleep assessment in the sleep clinic The Tayside children’s sleep questionnaire (McCreavey et al 2005) was completed to screen for behavioural sleep difficulties. The Tayside Children’s Sleep Questionnaire (TCSQ) was developed in Scotland for use in a primary care setting to assess for disorders of initiating and maintaining sleep (DIMS) in children aged 1-5 years (McCreavey et al 2005) and has been tested by the designers for reliability and validity.

Materials and methods: The aim of the project was to follow up children who had attended the phn led sleep clinic to find out if they had a clinically significant behavioural sleep difficulty six months following discharge from the clinic. The objective was to that improvement in sleep practices in the children who attended the clinic were maintained six months following discharge from the clinic and demographic information was gathered from the case notes by the phns.

Project Plan: 60 parents who attended the PHN led sleep clinics in three sites in County Donegal and who had children discharged from the clinic for at least six months, were contacted and asked to complete a repeat TCSQ. The notes were also reviewed and demographic information on the children recorded for comparison purposes.

The TCSQ is a ten item questionnaire with a maximum score of 36. A score of 8 or more is considered clinically significant when screening for Disorders of Initiating and Maintaining Sleep (DIMS) in children.

Results: At the initial sleep assessment in the clinic only two children had scores less than 8 with 58 children scoring eight or above which is clinically significant for DIMS (McCreavey et al 2005).

TCSQ at the follow up at least six months after discharge from the clinic:
- 43 children (72%) had a repeat Tayside in the normal range (below 8).
- 4 (6%) children had a repeat Tayside score of 8.
- All 13 children who scored between 9 and 25 in the repeat TCSQ had scores which had gone down from the initial assessment TCSQ score.

To ascertain parental perception, parents were asked as part of the study if they thought their child had a sleep problem.

Pre-Intervention: 59 said yes and one said “some”.
Post-Intervention 52 said No and 8 said yes

Conclusions: The result from this audit show that 72% of the children who had a clinically significant TCSQ score at the initial sleep assessment had a repeat score in the normal range six months following discharge from the clinic which indicates the sleep difficulty had remained resolved.

Acknowledgements: The parents who agreed to take part in this audit. Dr Frank Sullivan for permission to use the Tayside Children's Sleep Questionnaire. The Director and Assistant Directors of Public Health Nursing in County Donegal for their support in developing the sleep clinics.
PARENT/CHILD CRIB INTERACTIONS CAN BE OBJECTIVELY MEASURED BY NANIT’S COMPUTER VISION TECHNOLOGY

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Introduction: Nanit has developed a system for analyzing infant sleep using computer vision technology, where a camera mounted over a crib is able to analyze a baby’s sleep without the need for a wearable device. This system has been shown to be accurate in determining sleep/wake states when compared to actigraphy (Glazer, et. al., 2017). Highly specific algorithms are able to distinguish parental interactions as ‘out of crib visits’ (where the baby was taken out of the crib, presumably to be fed or soothed) or ‘check-in visits’ (where the baby stayed in the crib but was soothed by the parents). Previous reports of parental visits rely upon self-reporting by parent sleep diaries. These diaries can be inaccurate (Werner et.al., 2008) and cumbersome to implement and analyze. The Nanit system is able to record parental crib interactions without the need for manual reporting. The aim of this study was to provide objective normative data on parent/child crib interactions using a large data set.

Materials and methods: Participants were current Nanit camera users in the USA and Canada who had given permission for Nanit to analyze their data. Data were collected on 2365 unique babies, aged 1-24 months, over 175 nights from October 2017-March 2018. A total of 212,928 night sleep sessions were analyzed. Proprietary algorithms analyzed total parental interactions and the number of times out of the crib.

Results: At one month, the average number of total parental interactions was 3.6 times per night and the average number of times out of the crib was 1.8 times per night. By 5 months, the average number of parental interactions had dropped to 2.6 times per night and the average number of times out of the crib was once per night. After 5 months, the average number of parental interventions and times out of the crib decreased gradually so that by 24 months, the average number of parental interactions was 0.6 times per night and the average number of times out of the crib was 0.1 times per night. By 13 months, 50% of all babies had no parental interactions at night and 75% were not taken out of the crib at night. By 24 months, 67% of babies had no parental interactions at night and 90% were not taken out of the crib at night.

Conclusions: This is the first time that data have been presented on such a large cohort to objectively measure parental crib interactions. Parents are typically interacting with their babies several times per night when they are very young. We see a decrease in the number of parental interactions as babies start to consolidate their sleep. These results are similar to what have been reported previously (Galland, et. al. 2012, Mindell, et al, 2016). With the development of computer vision, we are able to clearly classify parental soothing behavior. Nanit has the potential to revolutionize the understanding of pediatric sleep as our dataset increases exponentially and by accurately analyzing parental and infant behavior.

References


Acknowledgements: This study was funded by Nanit, Inc. Assaf Glazer, Tor Ivry, Jesse Lax, Yanai Ankri, and Shaked Dovrat are employees of Nanit. Haviva Veler and Natalie Barnett served as consultants for Nanit.
CHALLENGING DIFFERENTIATION BETWEEN EPILEPTIC SEIZURES AND CATAPLECTIC ATTACKS IN A CHILD WITH NIEMANN-PICK DISEASE TYPE C

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Introduction: Niemann-Pick disease type C (NP-C) is a rare and progressive autosomal recessive neurovisceral lysosomal lipid storage disorder leading to disabling neurological manifestation and premature death. The disease is difficult to diagnose due to its highly heterogeneous presentation. One of its characteristic symptoms, particularly in the late infantile and juvenile forms, is cataplexy that is found in 5-30 % of the NP-C patient. Epileptic seizures accompany the clinical course of the disease in up to 50 % of the patients. We present a child with juvenile form of NP-C with pharmaco-resistant epilepsy and frequent anamnestically diverse epileptic and cataplectic attacks that are clinically not always easy to differentiate, having great impact on the clinical management.

Materials and methods: 13-years-old boy that was diagnosed with NP-C at the age of 7 years by molecular genetic testing of NPC1 gene. He was put on miglustat treatment. Video recording of paroxysmal events was followed with nocturnal polysomnography recording (PSG) and long-term video-EEG for differentiation of epileptic seizures and cataplectic attacks.

Results: Patient presented with partial epileptic seizures and gelastic cataplexy one year after the diagnosis despite miglustat treatment. He was treated with levetiracetam and lacosamide. Fluoxetine was introduced for cataplexy but reportedly aggravated epileptic seizures and was later omitted by the parents. Video recording disclosed clear gelastic cataplectic attacks with loss of facial and neck tone. Other type of paroxysmal events included atonic head drops followed by asymmetric tonic posturing and unilateral dystonia/dyskinesia with upper limbs grabbing to avoid fall. The paroxysm occurred without clear trigger and consciousness was not clearly disturbed. PSG revealed disrupted sleep with sleep efficiency of 54 % and sleep onset REM despite fluoxetine treatment. Altered sleep patters included sudden increases in muscle tone during slow wave sleep, atypical K-complexes and spindle activity and increased amount of REM sleep (39 % of TST). Long-term video-EEG captured many seizures in awake, clinically appearing as motor arrest and staring, followed by motor activity with right hand resembling circular hand automatisms or tonic right arm elevation, restlessness with tonic mouth grimace (»chapeau de gendarme «) and grabbing hands automatism, sometimes followed by left hand nose wiping. Patient was not able to report any aura feeling due to his disability, was able to obey motor commands during some episodes (without loss of consciousness) but was not able to speak. There were no clear deficits after these seizures. EEG disclosed atenuation of activity over left frontal leads and vertex at the beginning, followed by spread over left hemisphere and in the phase of right hand tonic component, rhythmic sharp theta activity over left frontocentral leads.

Conclusions: Cataplectic attacks and epileptic seizures can both occur in patients with NP-C. Their clinical differentiation is not always obvious, so additional extensive neurophysiological investigations provide understanding of episodes and influence the further pharmacological treatment decisions. In this case we were not able to clearly distinguish cataplectic from partial seizures without long term EEG because of similar clinical semiology.
MATERNAL SLEEP-RELATED COGNITIONS AND YOUNG CHILDREN’S SLEEP: A COMPARISON BETWEEN THREE DIFFERENT ARAB SOCIETIES IN ISRAEL

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Introduction: Cultural and parental factors (e.g., parental sleep-related behaviors and cognitions) have an important role in the development of children’s sleep. However, to the best of our knowledge, no study has yet investigated parental factors in relation to children's sleep among Arab families in Israel. Overall, there are three Arab ethnocultural groups living in Israel, namely Christian, Muslim and Bedouin. Although sharing the same general Arab background and language, the three groups differ from each other culturally, residually, and in their social and economic characteristics. While Bedouin families live in a very traditional and conservative society, Christians are more urban and have a lower fertility rate, higher educational attainment, and higher occupational status than that of the Muslims and Bedouin. Thus, this study provides an opportunity to examine similarities and differences between these groups regarding parenting, and specifically, maternal sleep-related cognitions and children’s sleep in an Arab sample of young children and their mothers, and to assess the relationship between maternal sleep-related cognitions and sleep in these young children.

Materials and methods: Mothers of 120 young children from three different Arab Societies in Israel participated in this study (38 Christian, 62 Muslim, and 20 Bedouin). The average age of the children was 18.2 months (range: birth to three years; SD= 10.36). Mothers completed the Brief Infant Sleep Questionnaire (BISQ), and the Infant Sleep Vignettes Interpretation Scale (ISVIS) aimed at assessing parental sleep-related cognitions.

Results: Analysis revealed significant group differences on two ISVIS subscales: Distress and Temperament (F(2,117)=8.82, p<.002; F(2,117)=3.72, p<.018). On the ISVIS Distress subscale, Christian mothers scored significantly higher than did Muslim and Bedouin mothers, indicating that they were more likely to interpret infant night-wakings as a sign of distress and need for help. On the other hand, Christian mothers scored significantly lower on the ISVIS Temperament subscale compared to Muslim and Bedouin mothers, indicating that they were less likely to attribute infant night-wakings to infant nature. The difference between the mothers on the Limits subscale approached significance (F(2,117)=2.44, p=.091) suggesting that Christian mothers were less likely than Muslim and Bedouin mothers to emphasize the importance of limiting parental nighttime involvement as a response to children's night-wakings.

Likewise, significant group differences were found with regard to children's sleep (F(2,117)=3.93, p<.023). Post-hoc (Tukey) analysis indicated that Christian mothers reported more children's night-wakings than did mothers of Muslim and Bedouin children.

Significant Pearson correlations were found between the Maternal Distress, Limits subscales and child sleep. These findings demonstrated that mothers who were more likely to attribute infant-night-wakings to infant distress and who were less likely to emphasize the importance of limiting parental nighttime involvement, reported that their children had more fragmented sleep.

Conclusions: The findings of this study support the literature on the importance of cross-cultural factors in the development of sleep in early childhood and demonstrate for the first time cross-cultural differences in sleep and maternal sleep-related cognitions in Israeli Arabs. Our results confirmed that, like in other societies, maternal sleep-related cognitions are related to poorer sleep in young children.
**INTERPRETATION OF SLEEP STUDY DATA; ARE GUIDELINES ENOUGH?**

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**Introduction:** The AASM scoring guidelines are used by many sleep services as the standard criteria by which to analyse sleep studies in both adult and paediatric patients. However we believe that interpretation of results, and therefore also treatment options, requires a contextual approach. Knowledge of the patient’s background and clinical history are vital when interpreting a sleep study. Using a case study we demonstrate the need to rely not just on the scoring alone to produce a reliable sleep study report; clinical knowledge and experience must also play a key role. We present the sleep study of a 14 year old boy with Larsen’s syndrome. The technical report using guideline-based scoring is compared to the clinical report on the same study by the lead respiratory physician. We then discuss possible treatment options that could arise from the differing reports.

**Materials and methods:** An in-patient limited sleep study using Emblettta MPR and Sentec TcCO2 monitor was performed overnight in air, not ventilated. Data was downloaded onto RemLogic software, v.3.4.1. Analysis was performed using the AASM 2015 guidelines. According to the AASM guidelines, a hypopnoea should be scored as obstructive if any of the following criteria are met: a. There is snoring during the event. b. There is increased inspiratory flattening of the nasal pressure or PAP device flow signal compared to baseline breathing. c. There is an associated thoracoabdominal paradox that occurs during the event but not during pre-event breathing. There is an additional “in-house” criterion which we add to these which is, d. There is associated thoracoabdominal phase-shift that occurs during the event. These criteria for obstructive hypopnoeas compared with central hypopnoeas (which is not a classification used by this sleep service) describes a central hypopnoea as such if NONE of the above criteria are met.

**Results:** The study was initially scored blind by an experienced locum sleep physiologist using the criteria described above. Summary: AHI 11.3 (75% obstructive 25% central or unclassified). ODI 11.6 Mean TcCO2 6.7kPa

Based on these figures the physiologist’s report described a largely obstructive picture. The same study was subsequently viewed by a paediatric respiratory consultant proficient in sleep studies who reported mainly central hypoventilation, having knowledge of the patient and that his syndrome was unlikely to result in obstructed breathing. This resulted in commencement of non-invasive bi-level ventilation. However, a mainly obstructive report without the experienced perspective may have resulted in CPAP therapy.

**Conclusions:** Often the patient’s clinical notes are not available when analysing sleep studies. Also, some sleep centres prefer to score “without prejudice” and to leave the interpretation to the patient’s lead clinician. However, there is some concern that there may be times when clinical decisions are made, based on the physiologist’s report, by more junior/ inexperienced clinicians or in the absence of a senior, sleep-experienced physician. Therefore, the physiologist’s report should include the patient’s clinical background and should reflect that context.

**Acknowledgements:** Nursing Staff Ward 2a Great North Childrens Hospital
IS A SLEEP INTERVENTION DESIGNED FOR CHILDREN WITH ADHD EFFECTIVE IN DAILY CLINICAL PRACTICE?

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Introduction: Up to 70% of children with Attention-Deficit/Hyperactivity Disorder (ADHD) experience sleep problems. Given our efficacy trial of a brief behavioural intervention for sleep in children with ADHD improved sleep and functional outcomes in children, we aimed to determine whether similar benefits could be replicated when the intervention is delivered by practising paediatricians and psychologists.

Materials and methods: Children aged 5–12 years with ADHD and a moderate/severe sleep problem (N=361) were recruited for this translational cluster RCT through paediatrician practices in Victoria and Queensland, Australia. Clinicians were randomly allocated at the level of the paediatrician to either receive the sleep training or not. The intervention comprised two consultations covering sleep hygiene and standardised behavioural strategies. Primary outcome: change in the proportion of children with moderate/severe sleep problems by parent report at 3 months post-intervention. Secondary outcome: child behaviour, academic functioning, quality of life and parent mental health. Linear mixed effect models adjusted for potential a priori confounding variables. Imputed results are reported.

Results: The proportion of children with moderate to severe sleep problems was lower in the intervention group (28.7%, 36.5%) compared with usual care group (55.5%, 60.0%) (p<0.001) at 3 and 6 months, respectively. Flow on benefits to functioning were not observed.

Conclusions: A brief behavioural sleep intervention is effective in improving sleep problems when delivered in daily clinical practice by paediatricians and psychologists. The lack of flow on benefit to other aspects of functioning may represent a dilution of treatment delivery.

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RHYTHMIC MOVEMENT DISORDER: A NOVEL SOLUTION?

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Introduction: Traditionally we have viewed rhythmic movement disorder (RMD) as a condition that children grow out of. However, there is now evidence that this is not always the case and there are a group of children who will continue to rock unless an intervention is made. Untreated RMD causes significant sleep interference, impaired daytime functioning and actual or potential physical injury. The secondary impact on the family can also be considerable.

Materials and methods: To date there have been no randomized controlled trials looking at treatment options for RMD. Current evidence is limited to clinical perspectives, case studies or small observational studies. Current options include; improving sleep hygiene, environmental/safety adaptations, stimulus substitution, aversive stimulation, temporary sleep restriction and pharmacological treatment. Each treatment strategy has its limitations. Due to the lack of high quality evidence, treatment choices are currently guided by clinical experience and family preference. The sleep team at University Hospital Southampton have developed a novel approach for the treatment of RMD: hammocks.

Results: This is a case series poster presentation of the use of hammocks in the treatment of children with RMD. There are currently 32 children with RMD under the care of University Hospital Southampton. The gender split is 20 male and 12 female. Of these 32 children, five have been treated with the use of a hammock. Parents are often quite sceptical about their child sleeping in a hammock and some are not willing to consider it. In the families that have been willing to substitute the usual bed for a hammock, there has been considerable improvement. Four out of five of the children have had complete resolution of their symptoms. One child relapsed after a period of time in their usual bed. The exact aetiology of RMD is not known but one theory is that the rocking causes vestibular stimulation, which aids development of the vestibulo-ocular reflex and therefore gross motor development. The small gentle swaying movements of the hammock cause vestibular stimulation and this may mitigate the child’s urge to rock. Hammocks also provide a practical barrier to RMD as there is no hard surface for the child to hit themselves against.

Conclusions: RMD has a significant impact on both the patient and their family. Treating RMD can be a challenge and often requires a combination of approaches which are tailored to the family’s requirements. Based on the successful use of hammocks in treating RMD at University Hospital Southampton, we propose that other sleep clinics may wish to consider the use of hammocks as a treatment option.

Acknowledgements: Southampton Children’s Hospital Children’s Sleep Disorder Service
P#01-Saturday
MOTHERS’ AND FATHERS’ SLEEP IN THE PRENATAL-AND POSTPARTUM PERIOD: LINKS WITH INFANT SLEEP
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¹Ben-Gurion University of the Negev

Introduction: The development of infant sleep is linked with a variety of parenting factors such as maternal emotional distress, parental cognitions and soothing behaviors. These links highlight the importance of studying sleep within the family context. However, only a few studies investigated the relationships between child and parental sleep, and most of them are based on self-reported measures of maternal and child sleep. Thus, this study was aimed at assessing the changes parents experience in their sleep from pregnancy to postpartum, and at examining the links between infant and both parents’ sleep while using objective sleep measures.

Materials and methods: The study included 50 couples that were recruited during pregnancy. Parents’ sleep was assessed twice – during the third trimester of pregnancy and at 4 months postpartum. Infant sleep was assessed at 4 months.
Sleep was measured by actigraphy for 7 nights. We used the micro motionlogger sleep watch (Ambulatory Monitoring Inc, NY). Scoring was based on the Sadeh algorithms for infants and adults.

Results: Repeated measures analysis of variance were used to test within-subjects effects of time and parent on actigraphic measures of sleep. Maternal sleep quality (i.e. number of night-wakings, minutes awake after sleep onset, longest sleep episode) was significantly lower than paternal sleep quality at both assessment points. For example, mothers were awake for more minutes (F[1,49] = 76.60, p < 0.001), both during pregnancy (fathers: 18.26 ± 16.25; mothers: 40.44 ± 35.39) and at 4 months (fathers: 24.49 ± 21.24; mothers: 55.52 ± 27.80). A main effect for time was also found (F[1,49] = 7.59, p = 0.008) as both parents showed a significant increase in nocturnal wake minutes. However, fathers’ sleep duration was significantly shorter than mothers (F[1,49] = 5.74, p = 0.02) both during pregnancy (fathers: 392.26 minutes ± 52.27; mothers: 400.29 minutes ± 51.22) and at 4 months (fathers: 382.36 ± 52.22; mothers: 403.76 ± 54.79).

Significant positive correlations were found between both parents sleep measures and infant sleep measures. Infants’ number of night-wakings was significantly correlated with the same measure for mothers (r = .44, p < .05) and fathers (r = .30, p < .05). Furthermore, infants’ longest sleep episode was positively correlated with the longest sleep episode both for mothers (r = .65, p < .05), and for fathers (r = .36, p < .05). Infant sleep minutes and nocturnal wake minutes were associated only with maternal sleep (r = .34, p < .05; r = .35, p < .05 respectively).

Conclusions: Our findings suggest that the sleep of both parents is disrupted in the postpartum period compared to pregnancy, though mothers seem to be more affected. These changes are probably a result of the infant’s nocturnal wakefulness, as the sleep of both parents was associated with infant sleep. The findings highlight the importance of considering the influences of infant sleep disruptions on both parents’ sleep and on family dynamics.
FUNCTIONAL ORTHODONTIC APPROACH WITH UPPER JAW EXPANSION & MFT HELPS FOR THE MOUTH BREATHER AND SLEEP APNEA

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Introduction: Recently we need to think the each patient’s function for the long term stability before, during and after orthodontic treatment. We always treat and care with MFT(Myofunctional therapy) and then we can keep the long stability for each patient. Now I am sure We need to think about morphology and function for the treatment from MFT concept. It helps our team approach based on Efforts=Result and good communication between us and patients for a long time. I would like to introduce our team approach and educations and motivations(M.I.H.O.=Motivation& International Health Organization)

Methods and materials: in the 344 cases(feburary 2015~may2017) 34 girls cases, digital exam data comparison before and after orthodontic treatment with MFT. we compare the functional exam digital data( Lips Tongue, Frenulum, Occlusal forces and so on).

Occlusal Force measuring instrument: total dental bite (occlusion gm10) Nagano Keiki Lip Power Tester: Lip-de-cum (LDC-110R) Cosmo Instrument Tongue Pressure Tester; lip-de-cum (ldc-110) Tongue Strength Tester

The COSMO Instrument MFT Lesson is ① for people with cleft lip insufficiency, muscular stretching and massage from inside and outside the mouth was performed.

Results:

<table>
<thead>
<tr>
<th></th>
<th>before</th>
<th>after</th>
<th>better rate</th>
</tr>
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<tbody>
<tr>
<td>lip</td>
<td>6.3N</td>
<td>9.5N</td>
<td>18/34</td>
</tr>
<tr>
<td>tip of tongue</td>
<td>1.8N</td>
<td>5.7N</td>
<td>15/34</td>
</tr>
<tr>
<td>lingual frenulum</td>
<td>15mm</td>
<td>21mm</td>
<td>26/34</td>
</tr>
<tr>
<td>mouth open with frenulum stretch</td>
<td>13mm</td>
<td>28mm</td>
<td>30/34</td>
</tr>
<tr>
<td>mouth open</td>
<td>42mm</td>
<td>47mm</td>
<td>25/34</td>
</tr>
<tr>
<td>occlusal force(left)</td>
<td>19kg</td>
<td>31kg</td>
<td>21/34</td>
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<tr>
<td>(right) 16kg</td>
<td>25kg</td>
<td>9kg</td>
<td>17/34</td>
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</table>

Conclusions: I would like to discuss about our Myofunctional orthodontic approach and long term stability with my orthodontic cases.
1: MFT approach can support the better occlusion and stability
2: Early treatment and observation control with MFT can effect for the good stability.
3: Orthodontic stable result needs to establish for the dental functional occlusion.
4: Digital education system for our staff, patients and their family and students.

Acknowledgements: Because the MFT has a lot of unseen changes, it is important to quantify each check item and motivate the patient during the MFT training.

In addition, it was suggested that the teaching side was able to be confident that the MFT lesson and the muscle approach were effective and played an important role. Conclusions can be evaluated by quantifying the functional measurement, it was understood that the decision to determine the muscle approach from inside and outside the mouth and training and oral environment is effective.
P#31-Saturday

PRECISION MEDICINE IN ASSESSMENT OF SLEEP/WAKE-BEHAVIOURS: TECHNICAL & CONFIDENTIALITY CHALLENGES IN VIDEO-BASED ASSESSMENTS

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Introduction: The seminal work by Hippocrates and his students (~300 in-depth case reports, BC 460-380) provided the foundation for the framework of modern medicine. 2500 years later, we are picking up the concept of wondering and the methodology of in-depth phenotyping, in developing a participatory research network and home-based video-somnology/recordings of the person of interest (videos) and applying it to disentangle complex behaviours. Modern video technology, which is now widely available, changes the paradigm of office-based abstract assessments, allowing a glimpse into otherwise concealed scenarios in the lives of patients and their families and have the potential to decipher complex diagnoses.

Methods and materials: There are three main obstacles to the augmented application of videos, to support screening, diagnosis and therapeutic monitoring of medical conditions affecting sleep/wake behaviours of children: (A) data base management; (B) data transfer and (C) confidentiality.

Results: (A) Data Base Management. (1) Videos are saved under a patient ID, which is assigned to the caregiver/patient when they sign in to the participatory research network. (2) The patient name and ID is only saved on one computer, which fulfills safety standards (anonymized information). (3) The clinical information using the same ID is stored separately. (4) The consent forms are stored separately. (5) At a later stage, when results are published parents will receive a link to their own results and a code to review their own data.

(B) Data transfer. Videos are sent encrypted from home (e.g. smart phone), via any Wi-Fi-system, and de-encrypted in the lab.

(C) Confidentiality. Main Levels of de-identification/confidentiality (as suggested in the international harmonized ethics applications). (1) Full Confidentiality. No facial and body features with details – de-identified at pictogram/skeleton level; allows to capture angles and postures, which underlie a pose. (2) Minimal Compromised Confidentiality. Facial features are blurred; therefore, no facial features will be recognizable: Level A: the rest of the video (i.e. surroundings) can be recognized; Level B: the rest of the video can be de-identified at the pictogram/skeleton level; or Level C: the rest of the video can be de-identified at the cartoon level. (3) Compromised Confidentiality. Facial features and the rest of the video can be de-identified at the cartoon level. (4) No Confidentiality. Facial features and the rest of the video are not de-identified.

Conclusions: In this position paper for review by the IPSA-Board, we propose that health professionals should first investigate and collect descriptive qualitative information, so they are able to clinically describe phenomena of interest before collecting neurophysiological or genetic data. Modern data analysis techniques enable the creation of machine learning algorithms and cluster analyses, which will support deciphering complex clinical presentations like ADHD and RLS and their interactions with daytime vigilance and sleep. Our long-term research goal is to develop an application, which allows for the creation of four levels of video anonymization and will be used as an epidemiological participatory research tool.

Acknowledgements: BC Children’s Hospital Research Institute and Foundation
P#23-Sunday

PRECISION MEDICINE IN ASSESSMENT OF SLEEP/WAKE-BEHAVIOURS: OBSERVING VIGILANCE—WHAT PEDIATRICIANS CAN LEARN FROM BEHAVIOURS OF NIGHTTIME DRIVERS

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Introduction: Sleepless or sleep-deprived individuals may show changes in vigilance. To standardize vigilance assessments, we reviewed videos of nighttime drivers with a structured rating system and investigated the ratings of untrained observers.

Materials and methods: Nighttime driving videos of 60 adult volunteers, recorded between 2 and 4 AM, were provided by the Institute for Sleep-Wake-Research (ISWF) and the Austrian Automobile, Motorcycle and Touring Club (OEAMTC). Two 4.5-minute videos of 14 participants, recorded after 30 and 90 minutes of driving, were analyzed. (A) Six observers rated participants using the Karolinska Sleepiness Scale (KSS); ratings were compared with drivers’ self-ratings and intraclass correlation coefficients (ICC) were calculated to measure inter-observer reliability. (B) Behaviours were annotated with open-ended descriptions and separated into (i) task-oriented (i.e. driving); (ii) non-task oriented (i.e. non-driving); and (iii) posture-oriented (e.g. stretching) behaviours; ICC values were also calculated. (C) Timing of (earlier versus later) videos were predicted. (D) Raters were asked if they noticed a significant difference between videos recorded earlier versus later in the night and described the perceived discrepancies using open-ended descriptions. (E) Four videos were reviewed with a Delphi consensus process, determining to what extent empirically developed pictograms could support analyses.

Results: (A) KSS participant and observer ratings for the earlier (means, 3.0 vs. 4.3) and later recordings (mean, 6.5 vs. 6.1) were comparable, inter-observer reliability was also high (ICC, 0.888). (B) Changes were observed between the behaviour categories as night progressed: task-oriented behaviours decreased (ICC, 0.585); non-task oriented behaviours increased (ICC, 0.847), however no differences for posture-oriented behaviours (ICC, 0.586) were observed. (C) Common differences noted between earlier and later videos included frequency of movement (53%), road attentiveness (53%), drooping eyes (47%), and posture rigidity (29%). However, (D) observers failed to predict the timing of the two videos. Finally, (E) characteristic vigilance behaviours were identified to inform future pictogram development.

Conclusions: The KSS ratings between drivers and observers corresponded and a change in task versus non-task oriented behaviours was detected. The inter-observer reliability for task- and posture-oriented behaviours was low, which may be related to how observers failed to correctly predict the timing of the videos. Causes of the discrepancies might be due to a systematic error (e.g. not being able to clearly observe posture-oriented behaviours due to camera positioning). The introduction of pictograms, and further investigation of self-stimulating behaviours and fluency of movements may address the error.

Acknowledgements: BC Children’s Hospital Research Institute and Foundation; Kids Brain Health Network; Austrian Automobile, Motorcycle and Touring Club; Institute for Sleep-Wake-Research, Medical University of Vienna
P#32-Saturday
SLEEP DISORDERED BREATHING IN ENURETIC CHILDREN AND CONTROLS

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³Medical Sciences, Lung, Allergy and Sleep Research, Uppsala University

Introduction: Snoring is the commonest clinical manifestation of sleep disordered breathing (SDB) and its presence is associated with increased risk of nocturnal enuresis (NE) in childhood. Earlier polysomnographic studies of enuretic children have shown fairly normal sleep stage distribution. The sleep is, however, not necessarily “deep” but disturbed. Subclinical signs of SDB have recently been described in uncontrolled studies of children with therapy-resistant NE, a finding which may contribute to their high arousal thresholds. This fits well into the known fact that some children with NE and sleep apneas may become dry when treated for the latter condition. Respiration during sleep in enuretic children without a history of snoring or sleep apnoeas has not been thoroughly studied.

Materials and methods: Twenty children (19 boys and 1 girl) who suffer from therapy resistant NE and 21 healthy controls (18 boys and 3 girls) underwent one night of polygraphic sleep registration focused on SDB variables. The registration was performed with a portable sleep device (NOX T3, Nox Medical) at home, comprising electroencephalography, respiratory movements, nasal airflow and oxygen saturation.

Results: The oxygen desaturation index was slightly higher for the children with NE compared with the healthy controls (p<0.05), although this was largely explained by two outliers. No other differences were found in the respiratory variables.

Conclusions: No major differences in respiration during sleep was found between enuretic children and controls in this study.
NEW START IN SLEEP DIAGNOSTICS: IMPLEMENTING ACTIGRAPHY AT CHILDREN’S HOSPITAL IN VILNIUS

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Introduction: Actigraphy is used in both research and clinical settings to evaluate sleep in the home environment. It is a less sophisticated tool than polysomnography and can provide weeks of data. Actigraphy recently became available in clinical practice in the sleep laboratory at Children’s Hospital in Vilnius.

Materials and methods: We reviewed study processes and scorable nights in all actigraphic records registered between December of 2016 and December of 2017. All actigrams were recorded in the in-patient setting due to compromised reimbursement of sleep studies. The population included 13 patients (median age 14 years, range 5-17 years, 6 boys), 3 of them were diagnosed with sleep disturbance attributed to psychiatric disorder, 3 with neuromuscular disorders, 6 with primary sleep disorder, 1 with epilepsy. ActiGraph GT9X was used for all recordings. Patient or his/her parents were asked to complete a sleep diary that included comments about bedtime, wake time, and periods when and why a child did not wear the device. Patients were asked to wear the device for 24 hours, although they were allowed to remove device for short periods when needed. Epoch length, or sampling rate, was set to 60 seconds. A sleep interval of 10 epochs for onset of sleep and an awake threshold setting of medium were utilized. Mann-Whitney U test was used for comparison, the result is significant at p < .05.

Results: Review of 13 patients actigrams resulted in 30 scorable nights. In 8 of 13 cases recording was 2 nights long. In two cases only one night was captured. Only 3 cases wore actigraph for longer than 2 days and only one of them had 5 nights recorded. Actigraph placement on the non-dominant wrist was documented in recordings of 10 nights, only 2 nights were recorded with device placed on the ankle (single patient, suspected periodic limb movement disorder). In recordings of 18 nights position of device was not mentioned. Total time in bed did not significantly differ between first and 2 night of recording (445 min vs. 444 min), total sleep time (400 min vs. 394 min), sleep efficacy (89% vs. 88%) slightly decreased and WASO (47 min vs. 50 min) slightly increased in second night recordings, but insignificantly.

Conclusions: Too few nights per recording were captured in our sleep laboratory. Recording periods should be prolonged at least to 72 hours in all cases where possible. First and second nights recording in hospital setting did not differ significantly so single night recording may be acceptable in some cases. Actigraphy should be made available in out-patient clinic to evaluate sleep in home environment and to lower cost of the procedure. Standardized user-friendly electronic actigraph log should be implemented to achieve and archive additional data which is necessary for reading actigram. Fortifying an actigraphy program with tested patient and family training methods and tools may increase integrity of data.

Acknowledgements: To all doctors, nurses and technicians working in the sleep laboratory for their shared experience, patience and life-long learning.
P#04-Sunday
SLEEP DISORDERS IN CHILDREN WITH EPILEPSY

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Introduction: Attention, learning problems in children with epilepsy can be attributed to their seizures as well as concomitant sleep problems. Thus studies on sleep quality in children with epilepsy may help to improve management strategies. The aim of the present study was to investigate the pattern of sleep disturbances among children with epilepsy.

Materials and methods: 200 children with epilepsy and their parents were asked to fill out the questionnaire for the sleep disorders among children (Bruni et al, 1996). The scale of sleep disturbances (SDSC) includes: difficulty in initiating and maintaining sleep (DIMS), sleep breathing disorders (SBD), arousal disorders (DA), sleep-wake transition disorders (SWDT), disorders of excessive somnolence (DOES), sleep hyperhydrosis (SHY).

Results: We asked 200 children with epilepsy. There were 111 boys and 89 girls aged from 5, 5 to 17, 9 years. The girls had higher scores in DA (65 vs 59), DOES (64 vs 68), t= -2.37, p<0.02). DA was higher in children with generalized epilepsy (t= -2.44, p=0.017). The patients with focal seizures compared with generalized seizures had: DA higher in general, 55 vs 64, t= -2.63, p=0.009. The “worst group” of children with focal seizures, vs generalized seizures have: SBD higher (58 vs 52), t= -2.72, p=0.007, SHY higher (51 vs 56), t= -2.47, p=0.014. Patients with Rolandic epilepsy had lower scores in DIMS (p=0.06) and DOES (p=0.07). Children with Temporal epilepsy had lower scores in SBD (p=0.04), and tendency in DA (p 0.09). In children with seizures at night was significantly higher scores on SBD, SWDT, (p<0.005), also DA and SHY (p<0.02). Children group with epilepsy in remission had less sleep disorders (total T 63 vs 71, p=0.0009).

Conclusions: The present study suggests that there is a close relation between seizures and sleep disorders and it is dependent on the seizure type and time, on the activity of the disease. The scale of sleep disturbances may be used to select children with epilepsy for the diagnostic overnight polysomnography test. Identification and treatment of these problems might improve quality of life of children with epilepsy.
CHALLENGES IN EVALUATION OF POLYSOMNOGRAPHIC STUDY (PSG) IN A GIRL WITH IRREGULAR SLEEP-WAKE RHYTHM DISORDER AND PHARMACORESISTANT EPILEPSY WITH MULTIPLE HIGH VOLTAGE GENERALIZED DISCHARGES DURING SLEEP

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¹The Children's Memorial Health Institute, Department of Neurology

Introduction: Sleep disturbances are frequently met in children with epilepsy and neurodevelopmental comorbidities. There is little literature on PSG findings and management in such patients. We would like to present a case of a 7 year old girl with pharmacoresistant epilepsy, mental and motor impairment of unknown etiology in whom PSG study has been performed due to sleep-wake rhythm dysregulation. In Poland specialists familiar with sleep problems in neurologically disabled children are lacking.

Materials and methods: A 7 year old girl has been admitted to the Department of Neurology due to irregular sleep-wake rhythm with excessive daytime sleepiness, unexplained behavioral symptoms during sleep and pharmacoresistant epilepsy with severe, non-progressing mental and moderate motor impairment of unknown etiology. Medical history up to the 6th month of age was unrevealing. At that time epileptic seizures in the form of infantile spasms occurred with hypsarrhythmia EEG pattern. Magnetic resonance imaging normal. She has started treatment with valproic acid, followed by vigabatrin and ACTH with seizure remission. Seizures relapsed the following years. The genetic testing for Angelman syndrome did not confirm this diagnosis. Since the age of 4 she started having symptoms of sleep-wake rhythm disorder, with no improvement on behavioral and pharmacological therapy with melatonin. During the first admission to our department she had been put on ketogenic diet with good effect on physical status and seizure control. The EEG study revealed multiple generalized high voltage discharges during sleep and weak sleep patterns. As irregular sleep-wake symptoms and unexplained night behavior persisted she has been sent to our EEG Lab for PSG study. No OSA suspected.

Results: After receiving a signed consent the in-laboratory overnight PSG study has been done. The total recording lasted 6 hours 40 minutes (TIB). The girl fell asleep on her mother's knees, hypnagogic hypersynchrony could be seen from the very first epoch (sleep latency 0). It then turned into runs of high amplitude generalized discharges composed of slow waves with superimposed sharp elements and high voltage sharp and slow wave complexes. Weak, but recognizable sleep spindles could be seen. The distinction between NREM2 and NREM3 was impossible to perform. No signs of REM could be found. In the middle and at the end of the study (lights off) episodes of strange behavior reported by the mother occurred, lasting for almost 1 hour the first and about 90 minutes the second, with stereotypic body movements forward and backward in a sitting position with hyperventilation. Artifacts and background with predominant theta activity of low to moderate amplitude could be seen in the recording. These wake stages made sleep efficiency in the study as low as 55%.

Conclusions: We would like to discuss with the audience the interpretation of PSG findings of this patient and discuss the challenges of PSG interpretation in patients in whom physiological sleep patterns are lacking or very hard to find.
AGE AT DIAGNOSIS OF NARCOLEPSY

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1Heart of England Hospitals NHS Foundation Trust

Introduction: Narcolepsy is a condition characterized by four symptoms. They are excessive day time sleepiness, sleep paralysis, cataplexy and hypnagogic hallucinations. The age at diagnosis is generally in the late teens or early adult hood. The lag time between onset of symptoms and establishment of diagnosis is often many years. The symptoms evolve over time and this often leads to a delay in diagnosis. The diagnostic delay causes delay in initiation of treatment which can potentially be harmful in children.

Materials and methods: Our sleep service was set up in 2011. A retrospective chart review of children who have followed up at a Secondary Care Sleep Service been diagnosed with Narcolepsy was undertaken.

Results: There were 17 children (5 were transitioned to the adult sleep service within two years of the clinic being set up). The data presented here relates to 12 patients. There were 9 male and 3 female patients. The commonest presenting symptom was excessive day time sleepiness. The median age at presentation was 8 years and 2 months (age range 6 yrs -15 years and 8 months). Median time to diagnosis after first clinic visit was 6 months (range 1 month to 15 months). At the first consult the median time of hypersomnolescence was 12 months (range 6 months to 96 months). There were no significant variations in the age at presentation amongst the sexes. Excessive day time sleepiness was the commonest presentation (100%). The diagnosis was established from history (12), MSLT (9) CSF hypocretin levels (6). 6 of the 12 had all three tests. Definitive treatment was commenced as soon as a definitive diagnosis was made at a median age of 8 years and 6 months (age range5 years and 11 months -14 years and 3 months).

Conclusions: In our cohort the median age at diagnosis of narcolepsy was 8 years and 8 months. This is earlier than described in current literature. This has led to earlier initiation of treatment. Increased awareness amongst the General Paediatricians has led to earlier referral to our Sleep Service.

Acknowledgements: Patients and families
UNUSUAL CASES PRESENTING TO SLEEP CLINIC

C. Kallappa1 Titus K Ninan†
1Heart of England Hospitals NHS Foundation Trust

Materials and methods: Case notes of patients referred to sleep clinic with history of excessive crying and poor sleep.

Results: Case 1: 2yr 6 month old afro-Caribbean child, with no previous sleep problems and good sleep hygiene started having crying episodes after 2 hours of going to sleep and would continue throughout the night, pulling his nappy. It was eventually was found to be “nocturnal priapism”, the cause of which was not found. Child was happy and active in the day time. Urological, haemoglobin, sickle test and sleep investigations were normal. Medications, including clonazepam did not make any difference. Child grew out of it at 4 years of age, with only parents needing support and counselling.

Case 2: 10 month child, born at term was cooled at birth for birth asphyxia. Had mild developmental delay. Attended with complaints of poor sleep since 5 months of age Would sleep better in the car seat but only for a couple of hours and would be crying all night and day. It was eventually found to be muscle spasms due to spasticity and improved with muscle relaxants to a great extent.

Case 3: 6 year old male child with known cerebral palsy, seizure disorder, nocturnal seizures, fragmented sleep presented acutely with worsening sleep and moaning all through the night. Changing posture, nocturnal EEG, oximetry, haemoglobin, iron studies were all normal. This child had hip dislocation giving rise to pain which was worse on lying in bed. His sleep improved after treating the hip dislocation.

Conclusions: Children often present with unusual events in sleep and it is important to think outside the usual. Sleep disturbance due to priapism is known and often in NREM stage. This patient was different as no local cause was found. It was eventually thought to be a self-limiting condition as he completely outgrew it. The other two children had expected problems due to their underlying condition that would cause sleep disturbance. Therefore, it is important to examine these children in detail when such symptoms are present.

Acknowledgements: Patients and families
EXAMINING THE MISSING LINK IN SUCCESSFUL UPPER AIRWAY RECONSTRUCTION, RSTORATION, AND RE-EDUCATION: INTRODUCING THE STANDFORD OROFACIAL MYOFUNCTIONAL ASSISTED MAXILLO-MANDIBULAR SURGICAL PROTOCOL (MAMMA)

E. Kliman\(^1\), S. Yung Chuan Liu\(^1\), A. J. Yoon\(^1\), C. J. Gouveia\(^1\)

\(^1\)Stanford University

**Introduction:** This study examines both the short- and long-term effects of an orofacial myofunctional therapy (OMT) sleep surgery protocol. Based on literature review and expert opinion, OMT protocol was designed to take into consideration both exercises that dilate and tone the oropharyngeal muscles directly, as well as those thought to reawaken/ re-pattern the stomatognathic neuromuscular system as a whole. The goals of research are to better understand whether this protocol could improve quality of life, rapid return to function, and relapse rate following MMA surgery. This would be the first study of its kind in validate OMT re-education in the context of MMA sleep surgery, and could directly benefit thousands of sleep apnea patients, as well as broaden the significance of OMT throughout the scientific community.

**Materials and methods:** This is a prospective cohort study with immediate OMT as the intervention in MMA surgical cases. All patients receive pre-operative restorative breathing coaching and instruction for nasal rinsing - minimum 1 week and maximum 4 weeks pre-op. Patients will also begin tone, mobility, and posture exercises pre-op. All patients have a brief, weekly post-operative check-in with a myofunctional therapist via Zoom, as well as customized exercise videos to support 12 weeks of post-up OMT. All objective clinical data is measured pre- and post-op in clinic using the Iowa Oral Performance Instrument (IOPI). Subjective data is measured using sleep and quality of life questionnaires. All patients are required to fill our compliance forms that indicate how often they perform exercises and nasal irrigation.

**Results:** Ongoing through January 2019.

**Conclusions:** Ongoing through January 2019.

**Acknowledgements:** We would like to acknowledge IOPI for generously donating the equipment required for this research.
**PRECISION MEDICINE IN ASSESSMENT OF SLEEP/WAKE-BEHAVIORS: IS VIDEO-ASSISTED METHYLPHENIDATE DOSE FINDING FOR CHILDREN WITH ADHD A SUITABLE PROCEDURE? A FOLLOW-UP STUDY**

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**Introduction:** ADHD is an important disorder which affects 5-10% of any population and causes impairment to at least a third of them for the whole life span. Video-assisted structured observation (VSO) of facial expression changes and performance during math tests allows titration of optimal methylphenidate doses for children with ADHD. We introduced the VSO methodology in order to individualize treatment concepts in a reproducible way (J Attention Disorders 10, 2007).

**Materials and methods:** In this study, patients were followed-up for four years in order to evaluate the long-term benefit of this diagnostic procedure. All patients with an ADHD diagnosis, who underwent the VSO-based dose finding procedure in 2008, were invited for a 4-year follow-up study. 19/49 patients fulfilled all eligibility criteria for statistical analysis. DuPaul's parents rating scales, school reports and physical condition at beginning, in-between the first year and four years after video-assisted dose determination were analysed.

**Results:** Friedman's two factor variance analysis and Wilcoxon's test for paired samples showed significant improvement of parent ratings of attention, impulsivity and hyperactivity during the first year of treatment, which remained stable during follow-up. Most importantly, none of the patients showed an adverse drug reaction: All children grew on the same growth percentile of the time before therapy; blood pressure remained normal. 15 patients maintained the dosage of the first determination. In three patients parents initiated lowering the single dose by 5, 7.5 and 10 mgs; two patients increased their single dose by 2.5 and 5 mgs. School reports improved in the first year but returned to pre-therapy-levels after four years.

**Conclusions:** We developed a VSO-procedure to determine the appropriate stimulant dose by observation of the variability of the smile of children and adolescents, allowing us to determine exact single doses of methylphenidate with 2.5 mg accuracy in collaboration with the patient and parents. Initial results individualized treatment and outcome positively with reproducible and sustainable long-term benefit. The missing effect in school reports is subject of further investigations. Most importantly, the methodology is applicable in community based clinical practice and improves patient/family and professional interaction, adherence and collaboration.

**Acknowledgements:** Dr. Fritz Jansen for teaching me to look exactly to what happens, Prof. Dr. Gerhard Neuhäuser for his continuous support for decades, Tanja Glaser for her effort in data collecting and to Nadia Beyzaei, BSc (H-Behaviours-Lab. BCCH Research Institute, UBC, Vancouver, Canada) for the poster design.
IMPACT OF EVENING CLINICAL PLACEMENT ON SLEEP AND COGNITIVE PERFORMANCE OF NURSING STUDENTS

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Introduction: In Quebec, college nursing students have courses on Thursday and Friday in the morning and afternoon after three days of evening (16:00 to 00:00) or daytime clinical placement (08:00 to 16:00) from Mondays to Wednesdays. Although it is largely recognized that the former schedule is more likely to lead to fatigue, no study has examined the impact of evening clinical placement on sleep and cognitive performance of nursing students.

Materials and methods: Twenty-three nursing students aged 18-19 years wore an actigraph for one week of evening clinical placement and one week of daytime clinical placement, or vice-versa. Also, the Ruff 2 and 7 visual selective attention test, the Trail Making Test part B, the Letter Number Sequencing subtest of the WAIS-R, and the Stroop test were completed twice on the Thursday following both the three days of evening and daytime clinical placement. Wilcoxon signed ranks exact tests were used. Finally, students completed the Epworth Sleepiness Scale (ESS) and the Chalder Fatigue Scale (CFS), ESS score ≥11 and CFS score ≥4 respectively indicating excessive daytime sleepiness (EDS) and excessive fatigue.

Results: From Monday to Wednesday, bedtimes (00:01 vs. 22:33, p<.001) and wake times (08:12 vs. 06:08, p<.01) were later on the week of evening clinical placement compared to the week of daytime clinical placement. Also, sleep duration was longer on Sunday (8.0 vs. 6.4 hours, p <.001) and shorter on Wednesday (5.7 vs. 7.1 hours, p<.001) during the week of evening clinical placement compared to the week of daytime clinical placement. On the other hand, cognitive function test results did not differ between weeks of evening and daytime clinical placement. Still, comparisons of Ruff 2 & 7 results from weeks of evening and daytime clinical placement taken together with normative data for age and education revealed a discrepancy between the speed and the accuracy of performance, namely a good level of speed and a high number of errors. Moreover, 77.4% and 67.6% of nursing students respectively exhibited EDS and excessive fatigue. In addition, higher ESS scores were associated with later wake up times on weeks of evening clinical placement (r =.55, p<.01). Finally, higher levels of fatigue were associated with lower performance in executive function tests assessing control and flexibility abilities (i.e., Ruff 2 & 7 accuracy and TMT-B (r≤-.0.55, p≤.05)).

Conclusions: Even though nursing students exhibited 1) a significant delay in the timing of their major sleep episode during the week of evening clinical placement, 2) a lower sleep duration on Wednesday due to an early morning awakening on Thursday to attend classes, and 3) high levels of daytime sleepiness and fatigue, their cognitive performance was similar following evening and daytime clinical placement. Cognitive function test results indicate a tendency for students to overemphasize the speed of their responses while minimizing the importance of attention accuracy. Further evaluation with larger samples is warranted, namely to better understand the tendency of students to make numerous selective attention errors as well as the association between fatigue and the decrease in executive performances.

Acknowledgements: This research was funded by the Programme d’aide à la recherche et à l’apprentissage (PAREA) from the Ministère de l’Éducation et de l’Enseignement supérieur du Gouvernement du Québec.
WHAT IS THE NEUROPSYCHOLOGICAL IMPACT OF KLEINE LEVIN SYNDROME? TWO CLINICAL CASE STUDIES

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Introduction: Kleine Levin Syndrome (KLS) is a rare sleep disorder with typical onset during adolescence which can profoundly affect cognitive functioning. It is characterised by persistent episodic hypersomnia (often sleeping 20 or more hours a day); impaired cognitive and executive functioning; impact on mood including a subjective experience of derealisation; variations in normal appetite and sexual behaviour. Identified cognitive changes during KLS episodes include confusion and deficits in concentration, attention and memory, though formal cognitive testing has rarely been used (Arnulf et al, 2005). Longer term impact of KLS highlights cognitive deficits in processing speed and verbal memory with greater impairment in those with more frequent and shorter episodes (Uguccioni et al, 2016). Aims of project: to generate detailed neuropsychological profiles of young people with KLS 'in' and 'out of' episode to better understand the impact of KLS on their functioning; to identify any specificity of impairments. Here we report the first two cases.

Materials and methods: Intellectual functioning assessed out of KLS episode using WASI All other neuropsychological tests used at assessment both 'in episode' and 'out of episode', with at least 6 months between assessments: Working memory - digit span from WAIS Processing speed - Coding and symbol search from WAIS Auditory memory - logical memory and verbal paired associates from WMS Executive function - colour word interference and trail making from D-KEFS Motor speed - pegboard from WRAMVA Sustained attention - Continuous performance test from Conners

Results: Data presented for two 18 year old men of average intellectual ability with KLS onset aged 14 and 15 respectively, typically experiencing multiple episodes a year and mean episode duration under two weeks. Comparison of 'out of episode' and 'in episode' performance: impairments identified 'in' KLS episode in immediate and delayed verbal memory; virtually across the board in tasks of executive functioning (particularly letter-number sequencing and inhibition of impulses); reduced processing speed and weaker sustained attention

Conclusions: Initial data from two case studies indicate compromised neuropsychological functioning during KLS episodes Performance most markedly affected in executive functioning, specifically with weaknesses in sequencing skills and inhibiting impulses Deficits also identified in auditory memory (recall and recognition), as well as reduced processing speed and sustained attention Variation in profiles in line with clinical report of heterogeneous experience of KLS Points to the value of a larger scale research project to corroborate these initial findings

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FRENCH VALIDATION OF A SCREENING TOOL FOR SLEEP DISORDERS IN PRESCHOOL CHILDREN

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Introduction: France does not dispose of pediatric instruments for evaluating sleep disorders in preschool children. The aim of this study is to validate the French version of the Sleep Disorders Scale for Children (Bruni et al., 1996; Putois et al., 2017) in a French population of preschool children (range 6 months to 4 years). In order to offer paediatricians a valid and standardized screening tool, it is necessary to determine the best factorial structure among those proposed in literature (Marriner et al., 2017; Putois et al., 2017; Romeo et al., 2006).

Materials and methods: Out of 780 questionnaires sent to nurseries, 316 questionnaires from healthy preschool French-speaking children (control group, mean age= 22 months, SD= ± 11 months) completed by parents, were collected. In addition, 105 questionnaires from children seen in pediatric sleep consultations (clinical group, mean age= 24 months, SD= ± 12 months) completed by parents, were also collected. The children of the clinical group also received a complete sleep assessment by sleep specialists in order to confront their sleep diagnosis with the results of the SDSC questionnaire.

Results: The main psychometric properties of the French version of the SDSC for preschool children are good. However, none of the three factorial structures proposed already in literature seem to suit the data we obtained in children from 6 months to 4 years concerning in particular items such as cataplexy, sleepwalking, bruxism and hyperhydrosis.

Conclusions: Our study pre-validates the use of the SDSC as an instrument for evaluating sleep disorders in children under 4 years old in France. Further discussion of the SDSC’s factorial structure is necessary for an optimized screening use.

Acknowledgements: Special thanks to all the teachers who supported our study as well as the parents who completed the questionnaires. We would also like to thank all the pediatricians that participated in our study.
DENTAL ARCH MORPHOLOGY ANALYSIS OF OBSTRUCTIVE SLEEP APNEA (OSA) CHILDREN IN PRIMARY DENTITION USING 3-DIMENSIONAL METHOD

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Introduction: Constricted maxilla, larger palatal height were reported in several adult OSA studies. However, limited information did we know about the dental arch morphology of OSA children in primary dentition. The aim of the study was to examine the relation between dental arch morphology and OSA using 3-dimensional analysis.

Materials and methods: The study group comprised of 18 children (4-6 y/o) diagnosed OSA (AHI>1) and age-matched non-obstructed children (AHI<1) as control group. Dental impressions of both upper and lower arch were taken and were scanned as digital model. Linear measurements of intercanine, intermolar width, arch length, and upper palatal height were measured using Simplant software. Palatal surface area and palatal volume were also calculated.

Results: Children with OSA had no significant difference in linear dental arch morphology comparing with children of control group. However, OSA children had smaller intercanine width, smaller palatal area, larger palatal height tendency.

Conclusions: The study used 3-D analysis for better accuracy. Unlike previous studies, present study focused on the children with primary dentition. The results showed that no significant difference in linear measurements, surface area and palatal volume between study group and control group.
SLEEPING WITH A RARE CONDITION: A CASE REPORT OF SLEEP PROBLEMS ASSOCIATED WITH A 5Q DUPLICATION AND 9P DELETION

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Introduction: Microdeletion and microduplication syndromes are described as entities with various phenotypic findings, such as cognitive impairment, abnormal sleep patterns, among others. Sleep problems have been underdiagnosed, sleep-related breathing disorders (SBD) in particular, being under a more intricate clinical picture of other symptoms. The early recognition and early clues of sleep problems is very important to a better intervention and prevention, improving quality of life.

Materials and methods: We describe a 12-year-old boy who attended a specialized sleep consultation at a neurodevelopmental centre). His mother complained of sleep problems "since ever", with current complaints of initial insomnia (already taking 2mg of oral melatonin), superficial sleep, frequent night awakenings, parasomnias, roncopathy, difficult awakenings and physical tiredness throughout the day. He needed 10-11 hours of daily sleep to avoid severe mood and behavioral dysregulation. Physical examination revealed a craniofacial dysmorphism (flat facies and nose, small mouth, micrognathia) and a mandibular expansion device. In clinical history, we found that he’s the second child of non-consanguineous, caucasian parents with normal karyotype. At 9 months old he was asked to do a cytogenetic analysis because he didn’t grew or gain weight accordingly - it revealed a

46.XY,add(9)(p24.1).ish der(9)t(5; 9)(q34;p24.1)(wcp5+; wcp9+)de novo karyotype, as determined by G-banding combined with sub telomere FISH. The rare cases described in literature point to respiratory obstruction due to craniofacial features and respiratory muscles weakness, among other physical abnormalities. He presented a developmental delay, with major impact on motricity and cognition, requiring interventions until nowadays. We asked his mother for a sleep diary and a polysomnography in a certified sleep lab. Actigraphy was proposed but denied for his hypersensitivity.

Results: Sleep diary was completed for 24 days, throughout school and vacation days. Sleep latency was normal, and the mean TST was 1 hour more on vacation (653.8m vs 616.5m). Tiredness at waking up and changes in behavior weren’t always associated with less hours of sleep, but the major emotional outburst happened the one-night bedtime was significantly delayed. He woke spontaneously on 7 nights, most of them to drink water, but once feeling refreshed after a 5-hours sleep. The polysomnography showed a slight diminish sleep efficiency (84.4%), with abundant sleep fragmentation, although maintaining sleep architecture. N2 was enlarged, SWS’s normal and REM’s diminish. AHI was 1.2/h, with a predominance of central events. He’s diagnosed with upper airway resistance syndrome due to flow limitations.

Conclusions: Sleep problems of this specific chromosomopathy have not been described. A high proportion of children with just 9p deletion take longer to settle or need a short sleep before waking refreshed. In this case, melatonin helped falling asleep, but his respiratory condition and emotional factors interfere in his sleep, perhaps masking an irregular sleep-wake rhythm. Prolonged sleep was felt needed to compensate the impact on global functioning. Sleep problems in genetic syndromes are challenging and studying them could provide clues to understand their pathogenesis and find new strategies to treat and prevent the underlying conditions.
TRANSCUTANEOUS PCO2 AND END TIDAL PCO2: CAN THEY BE USED AS ARTERIAL PCO2 SURROGATES IN AWAKE CHILDREN?

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Introduction: The gold standard test for measuring PaCO₂ is the arterial blood gas analysis (ABG). This method gives a cross-sectional measure of a variable and dynamic phenomenon. It is a painful procedure that might trigger anxiety in children, can develop complications, is expensive and requires a laboratory. There are alternative methods that allow non-invasive and continuous monitoring of PCO₂: end-tidal PCO₂(PetCO₂) and transcutaneous PCO₂ (PtcCO₂).

Objective: to examine the utility of PetCO₂ and PtcCO₂ as surrogates of PaCO₂ in an awake pediatric population.

Materials and methods: Prospective, observational, cross-sectional and comparative design study. We included children referred to the sleep unit of the Paediatric Hospital, J.P.Garrahan, Argentina, between June 2016 and April 2017 who required an awake ABG. After 20 minutes of simultaneous PetCO₂ (NONIN Life Sense LS1 9R) and PtcCO₂ (Sentec digital transcutaneous monitor with V-Sign sensor) monitoring, the arterial puncture was performed and blood sample immediately processed in a multiparameter gas analyzer (RadiometerABL 800 Flex). A sample size of 67 patients was calculated. Spearman Correlation Coefficient and Bland and Altman analysis were applied. A PCO₂ mean difference value of up to 3 mmHg was considered clinically acceptable.

Results: A total of 68 sets from 67 children were analysed. Male 35 (52%), age (median) 9.11 y (0.23-18.73). Their major diagnosis were neuromuscular disease 35 (52.24%), congenital central alveolar hypoventilation syndrome 8 (11.94%), myelomeningocele 7 (10.45%), craniofacial abnormalities 4 (5.97%), obesity 3 (4.48%), adeno-tonsylar hypertrophy 1 (1.49%), others 7 (10.45%). Sixty four (94%) were studied during spontaneous ventilation. Twenty patients (30%) needed more than 1 puncture, 63 (92%) felt pain during the puncture, 39 (58%) cried and 45 (66.6%) were tachypneic.

The median and range of PCO₂ (mmHg) was: PaCO₂ 36.7 (20.4-86.3), PetCO₂ 33 (20-68) and PtcCO₂ 38.8 (26-84). Spearman correlation index between PetCO₂/PaCO₂ was r = 0.60 and PtcCO₂/PaCO₂ was r = 0.9. Bland Altman analysis: mean gradient (±2SD): PaCO₂- etCO₂: 2.93 (-9.88; 15.76) and PtcCO₂- PaCO₂ -1.87 mmHg (-8.95; 5.20).

Conclusions: ABG is the gold standard method for CO₂ measurement but can not always be obtained. As expected it triggers pain and consequently tachypnea modifying the real respiratory stable condition. There was a trend to underestimate PCO₂ with PetCO₂ and to overestimate the measurement with PtcCO₂.

Correlation with ABG was excellent with PtcCO₂ and regular with PetCO₂. The systematic bias of both methods was clinically acceptable, however PaCO₂ values show important dispersion. PtcCO₂ seems to be a better non invasive alternative method to PCO₂ monitoring in this pediatric awake population.

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Comparing Sleep in Children with Attention-Deficit/Hyperactive Disorder (ADHD) Between Those with and Without Comorbidities

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Introduction: During the last years several studies have been published comparing sleep in children with ADHD versus controls with different purposes: (1) to detect differences in sleep architecture that can be used as specific biomarkers in differential diagnosis of ADHD from other psychiatric diseases, (2) to detect associated sleep disorders that could worsen symptoms of ADHD, (3) and to provide new theoretical frameworks to better understand the pathogenesis of ADHD.

There is a significant heterogeneity in both the methodology and the results of the studies mentioned above. About the first one, few studies have been made with drug-naïve and ADHD patients without any psychiatric comorbidity, particularly the Oppositional Defiant Disorder (ODD), being both important factors that can influence sleep parameters.

The objective of our study is to evaluate the existence of differences in sleep architecture measured by nocturnal video-polysomnography (vPSG) between ADHD patients with comorbidities and ADHD patients without comorbidities.

Materials and methods: Eight ADHD children (6 males; 2 females; mean age = 11.6y) with comorbidity (5 with ODD, 2 with anxiety and 1 with both) and eight without (7 males; 1 female; mean age = 11.4y) were included; thirteen were taking Methylphenidate, three Risperidone, one Lithium and three were drug-free. All participants underwent (1) a psychiatric semi-structured interview to obtain the diagnosis and to detect possible comorbidities, (2) a sleep questionnaire to detect primary sleep disorders, (3) and a nocturnal vPSG. Sleep parameters analyzed were Total Bedtime, Total Sleep Time, Sleep Latency, Sleep Efficiency, Wake After Sleep Onset (WASO), Sleep Time and percentage in N1, N2, N3 and REM states, REM Sleep Latency, duration and number of REM cycles and REM density values.

Results: The examined sleep parameters were similar between ADHD patient with comorbidities and those without. There is however a tendency in group without comorbidities to have shorter WASO than group with comorbidities. Moreover, we observed, compared with normal values, an increase in REM latency and a decrease in REM percentage in ADHD patients.

Conclusions: The absence of significant differences in sleep architecture between one group and another shows that comorbidities, such as ODD or anxiety, and/or drugs mentioned above do not have a relevant influence in the sleep of these children, although we observe a major tendency in the group with comorbidities to wake up during the sleep than ADHD group alone.

We hypothesize that (1) differences observed in previous studies comparing ADHD with controls may be attributed to ADHD alone, and not to their comorbidity and/or medication, and (2) that comorbidities could produce a more fragmented sleep in ADHD children.

These results have no significant differences due to the small number of participants. A further study with more patients should be done for more definite results.

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Enhancement of Parental Health & Emotional Regulation Competencies After Treatment of Young Children's Insomnia

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Introduction: sleep onset problems and night waking problems - in sum insomnia symptoms are associated with various impairments. Besides the child's health, parental impairments are often also insomnia symptoms and mental health problems as depression.

Materials and methods: Parental health was evaluated before and after a sleep training for infants up to four years suffering from insomnia. Various child and parental sleep instruments as well as mental health questionnaires were implemented. All in all, more than 43 parents with children took part in the study.

Results: Overall, parents' health showed significant increases concerning various parameters after the sleep training (Mini-KiSS). Especially the mothers reported health-oriented improvements after Mini-KiSS six session based structured sleep training. Besides depressive symptoms, also emotional regulation competences were enhanced. In addition, parental relationship was rated better after the short sleep training. More detailed results concerning those parameters will be reported.

Conclusions: Mini-KiSS sleep training with only six sessions is not only effective for children's sleep - it is also helpful for parental health and relationship. Therefore, sleep training should be implemented early if symptoms of insomnia are reported or parents need sleep information and guidance for their children's sleep.
Comparing Behavioural State Annotations and PSG Annotations in Infants

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Introduction: During the first year of life, infants spend more hours asleep than awake, making sleep one of the most important activities for their developing brains. Monitoring sleep, both in term and preterm infants, can offer valuable insights into their neuro-physiological development. The most common way to monitor infant sleep is by polysomnography (PSG), but this method requires obtrusive contact sensing on the body of the infant. This is undesirable in fragile NICU-infants, and impractical in a home situation. Therefore, we developed a framework for unobtrusively scoring infant behavioural states based on the work by Prechtl (1974), and we compared it to golden standard PSG annotations.

Materials and methods: Nine healthy infant participants (182 days ± 69 days) took their morning or daytime nap in the Tilburg University Babylab. PSG was recorded according to the guidelines set by the AASM (2007), and infants were continuously monitored by video and audio recordings. A non-obtrusive sensor was also used to collect respiratory data. Data were inspected in 30s epochs and scored by (1) an expert who rated the PSG data (“PSG-rater”), and (2) by 2 independent scorers (“behavioural raters”) who rated the infant behavioural states based on an adaptation of the scoring system developed by Prechtl (1974; state 1 to state 5). Audio and video recordings were inspected retrospectively. Instead of solely scoring observable behaviour, the respiration signal was inspected for (ir)regularity, too. Also, a number of movement behaviours were added to Prechtl’s system to come to the new framework used for this study.

Results: Preliminary results for this small sample showed that total agreement between the two behavioural raters was 86.2%, and the corresponding unweighted kappa (κ) was .81. Agreement between each of the behavioural raters on the one hand and the PSG rater on the other was 82.2% (unweighted κ = .73) and 85.8% (unweighted κ = .78), respectively. Agreement between both the behavioural raters between themselves, and between each of the behavioural raters and the PSG-rater was highest for the active awake state (80%, 93%, and 90%, respectively). Agreement between the behavioural raters was lowest for the quiet awake state (28%). Lowest agreement between behavioural rater 1 and the PSG-rater was found for the quiet sleep state (69%), and lowest agreement between behavioural rater 2 and the PSG-rater was found for the active asleep state (61%).

Conclusions: By adapting Prechtl’s system into a new scoring framework we were not only able to achieve good inter-rater reliability between the behavioural raters, but also between each of the behavioural raters and the PSG-rater. Thus, our scoring system may provide an infant-friendly way of monitoring behavioural states in infants. Future work on a larger sample is needed to further substantiate our findings, and to study whether our framework would mainly be useful in non-clinical or also in clinical settings.

COMPARING VIDEO ACTIGRAPHY ACROSS PREMATURE INFANT SLEEP STATES

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Introduction: Sleep plays an important role for the development of neonates, in particular premature infants. Premature infant sleep consists of mainly active sleep and quiet sleep stages and wake state. Continuous monitoring of sleep states is indicative of their neural development over time. However, current methods of sleep monitoring in clinical practice, such as polysomnography (PSG) and behavioural observations require either attachment of electrodes to the infant’s fragile skin or time-consuming effort through human scoring [1]. Therefore, there is a strong need for unobtrusive sleep state monitoring of premature infants. Video-based monitoring is considered a non-contact and unobtrusive method to capture infants’ activity during sleep. By analysing body motions, we aimed at distinguishing between different sleep states automatically. Thereby, we quantified the body motion (called “video actigraphy”) and compared it across sleep states.

Materials and methods: Data of 29.8 h from seven premature infants (gestational age: 29.9 ± 2.7 wk) was analysed, where near-infrared videos were recorded for each infant. Sleep states were manually scored by two trained raters individually for each non-overlapping 30-s epochs, based on an adaptation of the behavioural scoring system developed by Prechtl in 1974 [2]. Respiratory signals, collected using an unobtrusive sensor, were used to assist the behavioural scoring. A 3DRS motion estimation algorithm [3] was employed to quantify motions by characterising pixel changes of consecutive video frames. Estimated non-zero motion values were counted and then averaged over each 30 s, resulting in epoch-based video actigraphy measures. The video actigraphy measures were then compared and statistically examined over sleep states.

Results: Mean and standard deviation of video actigraphy during wake, active sleep, and quiet sleep were 0.21 ± 0.19, 0.10 ± 0.21, and 0.09 ± 0.25, respectively. During wake state, the video actigraphy was significantly larger compared with that during the two sleep stages (examined with an unpaired t-test, p < 0.0001). This indicates that premature infants, in general, showed remarkably more body movements in wake state than in sleep state. In addition, no significant difference (p = 0.26) in video actigraphy was found between active sleep and quiet sleep.

Conclusions: By analysing the body motions (quantified using video actigraphy) for premature infants during different sleep stages, we found that it is feasible to discriminate between sleep and wake states for this patient group. However, our approach seems not capable of identifying specific sleep stages (i.e. active sleep or quiet sleep) with video actigraphy. From an application perspective, future work should be focused on investigating and developing an automated sleep/wake detection algorithm based on video actigraphy.

BEDTIME ROUTINES AND PRESCHOOLER SLEEP PROBLEMS: ASSOCIATIONS WITH CHILD SLEEP LOCATION

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Introduction: Bedsharing in infancy and early childhood is controversial: whereas proponents indicate that bedsharing promotes breastfeeding and close relationships, opponents cite safety concerns and potential issues with establishing independence from parents (Goldberg & Keller, 2007). Research also suggests sleep-related issues with bedsharing as children age. For example, co-sleeping (featuring a combined group of bedsharers and roomsharers) was associated with less optimal bedtime routines (Hayes et al., 2007). In another sample, bedsharing preschoolers experienced more sleep problems relative to solitary sleepers (Hayes et al., 2001). The present study was conducted to determine whether preschooler sleep location (independent, bedsharer, or roomsharer) was associated with parent use of bedtime routines and child sleep problems in the same sample.

Materials and methods: One hundred sixty-two American mothers of singleton children aged 2 to 5 years were recruited from MTurk to participate in an online study. Mothers completed a demographic questionnaire and inventories that inquired about aspects of child sleep habits including the Sleep Practices Questionnaire (SPQ; Keller & Goldberg, 2013), the Bedtime Routines Questionnaire, and the Children’s Sleep Habits Questionnaire (CSHQ; Owens et al., 2000). Parents received $4 in appreciation of their participation.

Results: Children were classified as solitary sleepers (n = 117), bedsharers (n = 20), or roomsharers (n = 25) based on parent responses on the SPQ. Demographic differences amongst the groups were only found when considering child sex and race (Caucasian versus non-Caucasian). As such, these variables were included as categorical covariates in the subsequent analyses.

Results from the BRQ indicated that mothers used routine behaviors more commonly with solitary sleepers relative to bedsharers; group differences were not found when considering the use of routine environments. Mothers more frequently engaged in adaptive behaviors at bedtime with solitary sleepers relative to bedsharers and roomsharers. Conversely, mothers engaged in more maladaptive behaviors with bedsharers relative to solitary sleepers and roomsharers.

Results from the CSHQ revealed that, relative to solitary sleepers, bedsharers experienced more overall sleep problems, bedtime resistance, sleep onset delay, sleep anxiety, nightwakings, parasomnias, and sleep disordered breathing. Relative to roomsharers, bedsharers also experienced more overall sleep problems, bedtime resistance, sleep onset delay, sleep anxiety, parasomnias, and sleep disordered breathing. Solitary sleepers and roomsharers differed only on bedtime resistance and sleep anxiety, with roomsharers receiving more problematic scores relative to solitary sleepers.

Conclusions: These findings indicate that bedsharing preschoolers experienced less optimal bedtime routines and more sleep problems relative to solitary sleepers and roomsharers. These findings are meaningful to parents and practitioners: parents should be encouraged to transition their bedsharing children to solitary sleeping or roomsharing arrangements before preschool, and pediatricians should be made aware of preschooler sleep location when assessing and making recommendations to improve child sleep.

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SLEEP-DISORDERED BREATHING IN ACHONDROPLASIA: 15-YEAR EXPERIENCE

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Introduction: Children with achondroplasia are at risk of sleep-disordered breathing (SDB) including obstructive and central sleep apnoea, eventually exaggerated by restrictive hypoventilation. Risk factors for these anomalies include short cranial base, midface hypoplasia, stenosis of the foramen magnum and chest deformity. The purpose of our study is to assess SDB in children/adolescents with achondroplasia.

Materials and methods: Retrospective analysis of clinical records of patients with achondroplasia referred to Sleep Laboratory of Pneumology Unit in a tertiary pediatric center, between 2003 and 2018. Demographic and clinical variables, polysomnography (PSG) and brain magnetic resonance imaging (MRI) results, therapeutic interventions (ENT surgery, maxillofacial surgery, neurosurgery and non-invasive ventilation (NIV)) and evolution were analyzed. Obstructive sleep apnea syndrome (OSAS) was defined if apnoea/hypopnea index (AHI) > 1; mild if AHI 1-5, moderate if 5-10 and severe if AHI > 10. Upper airway resistance syndrome (UARS) was defined if respiratory event related index >1.

Results: Eleven patients (6 female) were included, median age at referral was 3 years (minimum 4 months, maximum 15 years) and median follow-up time was 20 months (minimum 2, maximum 180 months). At presentation, 9 patients had snoring, 3 had witnessed apneas. All performed PSG (median efficiency 83%, median TST 487 minutes).

Patients were divided in two groups depending on the moment of PSG: before ENT surgery (A) and after ENT surgery – adenoidectomy or adenotonsillectomy (B). Group A included 6 patients and PSG results were: normal in 2, UARS in 1 (RDI 5.5) and OSAS in 3 (IAH 2, 5.7 and 14). About these 3 with OSAS: median age was 10 months, the youngest was 4 months and didn’t snore and all are on NIV. Group B included 5 patient and PSG results were: normal in 2, UARS in 1 (RDI 2.5) and OSAS in 2 (IAH 2.7 and 13). About these 2 with OSAS: patient with mild OSAS had been previously submitted to adenoidectomy; patient with severe OSAS started and maintained NIV until maxillofacial surgery was performed. Ten patients performed brain MRI: 2 had hydrocephaly and narrowing of foramen magnum with brainstem compression and underwent neurosurgery, 7 patients had narrowing of the foramen magnum without neurological symptoms. In these 7 patients central apnea index was ≤ 1.

Conclusions: Although having a small number of patients, this study outlined the importance of performing PSG in all patients with achondroplasia, even in the first months of life and without snoring. As suggest by literature, we didn’t found correlation between narrowing of foramen magnum and abnormal central apnea index. In patients with achondroplasia, a multidisciplinary therapeutic approach may be needed, including adenotonsillectomy, NIV and maxillofacial surgery.
SLEEP-DISORDERED BREATHING IN CRANIOSYNOSTOSIS

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Introduction: Craniosynostosis patients are at risk of sleep-disordered breathing (SDB), including upper airway resistance syndrome (UARS) and obstructive sleep apnea syndrome (OSAS), due to abnormal skull and facial structure.

Materials and methods: Retrospective analysis of the clinical information of children/adolescents with craniosynostosis followed in a Sleep Laboratory of Pneumology Unit in a tertiary paediatric centre during 17 years. Demographic and clinical variables, polysomnography (PSG) and nocturnal oximetry results, therapeutic interventions and follow-up were analysed. Obstructive sleep apnea syndrome (OSAS) was defined if apnoea/hypopnea index (AHI) > 1; mild if AHI 1-5, moderate if 5-10 and severe if AHI > 10. Upper airway resistance syndrome (UARS) was defined if respiratory event related index > 1.

Results: Eight patients were included (4 females), with a median age at referral of 22 months (minimum 4 months, maximum 15 years) and the following specific diagnosis: Crouzon’s (2), Pfeiffer’s (2), Alpert’s (1), Saethre-Chotzen (1), Muenke’s (1) syndromes (S) and non-syndromic complex craniosynostosis (1). Seven patients presented with snoring, 5 with witnessed apnoeas. At referral, seven patients performed PSG, results were: normal (1), UARS (1), mild OSAS (1), severe OSAS (4, including Alpert adolescent without snoring). One child performed nocturnal oximetry showing OSAS (minimal SpO2 66%). Initial therapeutic intervention was ENT surgery in 4 patients. During follow-up (50 to 158 months), monitored by PSG, two patients remained asymptomatic, one patient needed ENT reintervention and other was submitted to mid-face advancement surgery after a period of non-invasive ventilation (NIV). Three patients started and maintain NIV after severe OSAS diagnosis (follow-up 2 to 198 months). Patient with normal PSG was an infant with Muenke S., presenting with snoring and witnessed apnoea; he is under surveillance with a follow-up of 22 months.

Conclusions: All but the youngest patients had SDB, highlighting the importance of sleep evaluation in craniosynostosis patients. Treating SDB in this patients may be difficult and require a multidisciplinary approach.
FRAGMENTED SLEEP AND ITS CONNECTION WITH EXECUTIVE FUNCTIONING IN INFANCY


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Introduction: Executive functioning (EF) begins to develop during infancy and continues to develop into early adulthood. In adulthood and adolescence, it has been extensively shown that sleep difficulties impair performance of executive function related tasks, but only a few studies have investigated the associations between sleep and EF in infancy. Due to the long course of maturation of EF and its associations with the frontal lobes, it is suggested that EF might be especially vulnerable to the effects of sleep problems. In the current study, EF was investigated longitudinally in infants with and without fragmented sleep.

Materials and methods: Infants with (n=75, ≥3 awakenings in a night) and without fragmented sleep (n=63, ≤1 awakening in a night) were studied within the CHILD-SLEEP birth cohort at 8 and 24 months of age. At both ages, two computerised tasks (an Overlap and a Switch task) were performed in order to measure executive functioning and regulation of attention to emotional cues. The Overlap task measured attentional disengagement from emotional stimuli and the Switch task measured the ability to learn predictable stimulus sequences and to inhibit attention shifts to a previously cued location. At 24 months of age, in addition to the computer tasks, a parent-rated measure of EF (BRIEF-P) was performed.

Results: The findings revealed that there were no differences in pre-executive functioning at 8 months of age whereas differences were evident at 24 months of age. Infants with fragmented sleep were less able to learn new stimulus sequences and to inhibit their responses to a previously cued location compared to infants without fragmented sleep. In addition, in the Overlap task, infants with fragmented sleep showed a smaller attentional bias toward happy and fearful stimuli than the infants without fragmented sleep at 24 months of age. Parent-ratings of executive functioning also gave support for the differences between the two groups.

Conclusions: According to our results, it seems that the development of EF follows somewhat distinctive pathways in infants with and without fragmented sleep. It is suggested that the differences are most pronounced on the domains of emotional and behavioral regulation of executive functioning. In future, the connections between night awakening and emotional self-regulation abilities should be studied.

Acknowledgements: We would like to thank all the families who have participated in the CHILD-SLEEP birth cohort. The project was funded by the Academy of Finland, Gyllenberg foundation, Yrjö Jahnsson Foundation, Foundation for Pediatric Research, Finnish Cultural Foundation, the Competitive Research Financing of the Expert Responsibility area of Tampere University Hospital, Arvo and Lea Ylppö Foundation, and the Doctors' Association in Tampere.
RISK OF NARCOLEPSY AFTER THE APPLICATION OF THE VACCINE PANDEMIC H1N1 2009: RESULTS OF SOMNIA'S ARGENTINE STUDY

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¹FLENI

Introduction: The increase of narcolepsy cases after the 2009 H1N1 vaccination with adjuvants observed in Europe, was considered a sign of alert. However, the association between this finding and the public media information-bias could not be ruled out. SOMNIA is an international investigation that observes the relationship or, lack thereof, between monovalent vaccines against flu pandemic H1N1 with AS03 adjuvant and MF-59, and narcolepsy. Objective for Argentina: to estimate the risk of narcolepsy for pandemic (H1N1) 2009 with MF-59 adjuvant vaccine.

Materials and methods: case-control study. Cases of narcolepsy (Brighton Collaboration criteria) were obtained from Buenos Aires Sleep Centers, and control cases from ambulatory care units (same region, age, sex, and date of consultation). The vaccination dates were obtained from vaccination certificates. To estimate the risk of narcolepsy in vaccinated cases, OR was used and adjusted for the number of controls per case. The diagnostic index-day was established when the Multiple Sleep Latency Test was positive. Children up to 18 years of age were included from April 2009 to December 2014.

Results: 11 pediatric cases of narcolepsy and 77 controls were included in the study. No association was observed between the vaccine pandemic (H1N1) 2009 and an elevated risk of narcolepsy after application of the vaccine the pandemic H1N1 2009 – (adjuvant MF-59).

Conclusions: Our results do not support an association between receipt of MF59-adjuvanted pH1N1 vaccines and narcolepsy in Argentina. They are consistent with those of other countries where the SOMNIA study took place.

Acknowledgements: The SOMNIA study was funded by the Centers for Disease Control and Prevention (CDC), Atlanta, USA, under CDC contract number 200-2012-53425_addendum 0001.
SLEEP FOR HEALTH IN HOSPITAL (SHH): IMPROVING THE SLEEP ENVIRONMENT OF CHILDREN’S WARDS

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Introduction: Poor sleep quality impairs immune function, pain sensitivity and emotional regulation – all important factors in the recovery of hospitalised children and the wellbeing of their co-sleeping parents. We have published qualitative data indicating that noise, light and ward routines significant disturb sleep in children’s wards with adverse daytime effects. We report the outcomes of a complex intervention designed to improve the ward sleep environment.

Materials and methods: The sleep for health in hospital programme addresses three domains: • Knowledge and empowerment: achieved through brief mandatory staff training and parent information leaflets • Environmental change: red torches for night observations and sound level meters at nursing stations • Behaviour change: adherence to curfews enforced by a flag erected at 8pm, bed signs indicated usual bedtime for each child. Continuous overnight sound level monitoring over 3 days recorded median dB sound pressure before and after intervention in our acute surgical ward. Staff completed the NOMAD questionnaire designed to measure the extent to which a complex healthcare intervention has been effectively integrated into usual care based on normalisation process theory.

Results: Over 300 medical and nursing staff have been trained. Of the first 96 surveyed, 100% were committed to promoting optimal sleep after their session compared to 66% prior to training. Comments included ‘Didn’t appreciate how much sleep deprivation affected children & their parents’ 100 NOMAD questionnaires were completed 6-12 months after the project launch by a range of health professionals. 87.6% saw the potential value of SHH in their work; 95.6% wanted to continue to support SHH 82.5% believed that participating in SHH was a legitimate part of their role Median sound levels in the ward overnight decreased from 59.6 to 46.4 dB.

Conclusions: A carefully planned complex intervention can alter staff and parent behaviour to promote respect for children’s sleep in hospital alongside safe care. Staff are willing to integrate intervention to promote sleep into the daily routine and this has impacted noise levels. Promoting optimal sleep in children’s wards has the potential to improve the hospital experience of both children and their co-sleeping parents.

Acknowledgements: We would like to acknowledge Southampton Hospital Charity who funded this project.
MATURITY AND PERIODIC BREATHING IN HEALTHY INFANTS

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\textbf{Introduction:} Agis is the most important factor determining sleep and breathing maturation. Periodic breathing (PB), a respiratory pattern characterized by regular cycles of short apneic pauses and normal breathes, is common in preterm infants, also seen in term babies, and usually resolved with age in healthy children.

\textbf{Materials and methods:} We have studied 61 healthy newborn, 30 term infants (TI), and 31 preterm infants (PTI). A diurnal polysomnography after food intake was performed in all cases. We have compared, in both groups (TI and PTI), sleep apneas, PB pattern and SatO2.

\textbf{Results:} In our study, subjects with PB (16.6\% vs 56.2\%, \(p = 0.018\)), percentage of PB in TST (2.6 \pm 0.08\% vs 10 \pm 0.17\%, \(p = 0.002\)) and central AHI contained in PB periods (8 \pm 18\% vs 22 \pm 37\%, \(p = 0.01\)) are significantly lower in TI compared with PTI, with higher SAT02 in PB (92 \pm 2\% vs 80 \pm 11\%, \(p = 0.08\)). PB was predominant in NREM sleep (formerly Quiet Sleep), in 93.4\% of TI vs 65.7\% of PTI, \(p = 0.3\). Extreme PB was seen after birth in one PTI (88\% of TST) but all children had normal PSG after 6-12 months.

\textbf{Conclusions:} PB is a frequent respiratory pattern in healthy infants, usually present in MREM sleep and related with maturation. Low gestational and postmenstrual age contribute to its persistence, with resolution after months of life. We postulate that this disappearance may be indicator of chemoreceptor sensitivity maturation over time.

\textbf{Acknowledgements:} Many thanks to all the families and babies who made possible this study.
COMPLEX PHYSIOTHERAPY: A NEW APPROACH IN THE TREATMENT OF CHILDREN WITH TONSILLAR HYPERTROPHY AND RELATED SLEEP-DISORDERED BREATHING

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Introduction: Tonsillar hypertrophy is one of the most common sufferings in childhood. Traditional treatment is medical and surgical. The relationship of tonsillar hypertrophy with sleep-disordered breathing in children is undeniable. Snoring, apneic breaks, restless sleep are the main reasons for visiting general practitioners and subsequently otorhinolaryngologists. This determines the great interest of the medical community in solving these problems and seeking the best therapeutic approaches.

Aim: To study the efficacy of a complex of physical factors for influencing tonsillar hypertrophy in children and associated symptoms of sleep-disordered breathing (SDB).

Materials and methods: 70 children aged 3-10 years with tonsillar hypertrophy graded 3+, 4+ and some of SDB symptoms and were examined at Diagnostic and Consultative Center “St. Marina”. An authors’ questionnaire of sleep-disordered breathing symptoms and clinical estimation of tonsillar’s size according standard scale were performed. All subjects had parental written informed consent. A complex physiotherapy with ultrasound and polarized, polychromatic, non-coherent, low-energy light was made.

Results: In 78% of the children a significant reduction in the size of the tonsillas to the extent of normal variation (p <0.0001) was seen, which correlated with a decrease in the symptoms of SDB (p <0.001). The therapeutic effect is maintained at the first month after the end of therapy, and at the beginning of the 3 months the effect is observed to be diminished.

Conclusions: The positive correlation observed in terms of the monitored parameters, gives us the reason to propose that the complex physiotherapy treatment is suitable as an alternative to drug therapy, where possible. It can be used for adjunctive to improve efficacy of medical therapy in children with tonsillar hypertrophy and SDB symptoms.
TONSILLAR HYPERTROPHY AND DAILY BEHAVIORAL SYMPTOMS IN CHILDHOOD: INFLUENCE OF APPLIED PHYSICAL FACTORS

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Introduction: Tonsillar hypertrophy is a common pathology in childhood. The relationship between it, sleep related breathing disorders (SBD) and changes in children’s behavior is proven in a number of studies. The most common symptoms are reduced attention, hyperactivity and drowsiness during the day. Therapeutic behavior is directed to tonsillar hypertrophy and requires individual treatment.

Aim: To evaluate the effects of a complex of physical factors on the influence of daily behavioral symptoms in children with tonsillar hypertrophy.

Materials and methods: The study included 70 children aged 3-10 years with tonsillar hypertrophy with grade 3+, 4+ and daily behavioral symptoms. All of them were treated in the physiotherapy and rehabilitation sector at Medical Center “St. Marina” in Varna. A questionnaire for daily behavioral symptoms developed by researchers was used and a clinical assessment of tonsillar size was made using a standard scale. Informed consent of parents was taken for participation of children under 18 years. Ultrasound and polarized, polychromatic, non-coherent, low-energy light therapy were performed.

Results: A statistically significant difference (p = 0.0001) was observed when comparing the mean values of the frequency of hyperactivity, reduced attention and sleepiness before and immediately after physiotherapy using the pairs test. The observed symptoms decrease as the therapeutic effect is maintained at 3 months post-therapy. There is a relatively strong positive correlation between the reduction in the rate of tonsillar hypertrophy and the rate of somnolence (r = 0.358, p = 0.002) following physiotherapy. There is a statistically significant medium-strong positive correlation between the reduction of the degree of tonsillar hypertrophy and the decrease of average frequency of hyperactivity one month after physiotherapy (r = 0.319; p = 0.005).

Conclusions: The positive trend we observed regarding the monitored symptoms gives us warrants the use of physical procedures as appropriate concomitant treatment of children with tonsillar hypertrophy associated with daily behavioral symptoms. The developed physiotherapeutic methodology can be used as except for treatment also as preventive in often ill children who are at risk for the development of tonsillar hypertrophy in order to prevent changes in their behavior.
P#13-Saturday
MELATONIN INGESTION AFTER EXHAUSTIVE LATE-EVENING EXERCISE IMPROVES SLEEP QUALITY AND QUANTITY AND SHORT-TERM PERFORMANCES IN TEENAGER ATHLETES

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Introduction: Among adolescents, sufficient amount of sleep is fundamental to growth and development as well as recovering from daily activities (Strasburger and Hogan, 2013). Nowadays, Teenagers are not getting enough sleep, and there are many reasons for this trend. It is accepted that nocturnal exercise is the cause of a sleep disorder (Postolache et al., 2005; Souissi et al., 2012). Nevertheless, evening intense physical exercise blunted MEL secretion due to exercise induced cortisol increase (Monteleone et al., 1992). Exogenous melatonin and through its hypothermic, hypnotic, antioxidant and anti-inflammatory effects, has proved useful for treating some sleep disorders (Nave et al., 1995). To the best of the authors’ knowledge, the interaction between intense nocturnal exercise, melatonin, sleep and short-term performances has not been studied. Therefore, the purpose of this study was to explore the effect of MEL-10mg ingestion after late evening intensive exercise on sleep quality and quantity, cognitive performance, and short-term physical performances the following morning in healthy trained teenagers.

Materials and methods: Ten male adolescent athletes (mean±SD, age=15.4±0.3 years, body mass=60.69±5.7 kg, height=167.9±6.9 cm and BMI=21.21±2.5) performed two test sessions separated by at least one week. During each session, participants completed the Yo-Yo intermittent-recovery-test level-1 (YYIRT-1) at ~20:00h. Then, sleep polysomnography (PSG) was recorded from 22:15min to 07:00h, after a double blind randomized order administration of a single 10-mg tablet of MEL (MEL-10mg) or Placebo (PLA). The following morning, Hooper wellness index was administered and the participants performed the Choice Reaction Time test (CRT), the Zazzo test, and some short-term physical exercises (YYIRT-1, vertical and horizontal Jumps (VJ; HJ), Hand grip strength (HG), and five-jump test (5-JT)).

Results: Evening total distance covered in the YYIRT-1 did not change during the two conditions (P>0.05). Total sleep time (TST) (Δ=24.55mn; P<0.001), sleep-efficiency (SE) (Δ=4.47%; P<0.001), stage-3 sleep (N3 sleep) (Δ=1.73%; P<0.05) and rapid-eye-movement (REM) sleep (Δ=2.15%; P<0.001) were significantly higher with MEL in comparison with PLA. Moreover, sleep-onset-latency (SOL) (Δ=8.45mn; P<0.001), total time of nocturnal awakenings after sleep-onset (NA) (Δ=11mn; P<0.001), stage-1 sleep (N1 sleep) (Δ=1.7%; P<0.001) and stage-2 sleep (N2 sleep) (Δ=1.9%; P<0.05) durations were lower with MEL. The Hooper index showed a better subjective sleep quality, a decrease of the subjective perception of fatigue and reduced level of muscle soreness with MEL. Moreover, MEL improved speed and performance but not inaccuracy during the zazzo test. Choice Reaction Time (CRT) was faster with MEL. Morning YYIRT-1 (Δ=82m; p<0.001) and 5-JT (Δ=0.08m; p<0.05) performances were significantly higher with MEL in comparison with PLA. In contrast, HG, VJ, and HJ performances were not changed during the two conditions (p>0.05).

Conclusions: MEL-10mg administration after strenuous late-evening exercise improved sleep quality and quantity, sustained attention, subjective assessment of the general wellness state, and some short-term physical performances the following morning in healthy teenagers.

Acknowledgements: The authors wish to thank all participants for their voluntary efforts and participation in this study. The authors wish to express their sincere gratitude to all the managers, specialists, and technicians in the department of Functional Exploration of the Nervous system, CHU Sahloul, Sousse, Tunisia.
Introduction: Sleep problems pose a major public health issue, making 25% of our paediatric population sick and tired. The health and developmental sequelae for children are both short and long term, including: learning, mood, behavioural and social problems, brain damage, lowered IQ, systemic inflammation, lowered immunity, depression, obesity, endothelial dysfunction and eventually heart disease, cancer and metabolic disorders. Of the 90+ diagnosable sleep disorders, the second most common are the variants of sleep disordered breathing (SDB) that deprive the brain of oxygen. Two to four % of children have obstructive sleep apnoea (OSA), and more than 90% of those children are missed or misdiagnosed. Furthermore, snoring without OSA, has been shown to have similar detrimental effects as OSA, on children’s health, behaviour and development. That a large portion of children with SDB goes undiagnosed, poses a major public health and education issue. This author’s proposal is to develop an education method for parents to assist their understanding of the urgency and gravity of sleep problems, give them the knowledge they need to identify SDB and other sleep problems, as early as possible in their children. The author proposes an education method for parents which starts with a simple ‘Yes/No’ questionnaire designed to raise parental awareness of children’s sleep patterns, night time breathing, and recognise the ‘Red Flags’ signalling sleep problems. The second educational tool for parents, is a book that outlines a ‘sleep formula’ for kids and a ‘red flag’ system for understanding when their children need help. The aim of the public health campaign in Australia is ‘for every child to get the sleep they need every night.’

Materials and methods: Literature search on prevalence data for sleep disorders and their sequelae has been used to develop the case for early intervention and treatment. A Sleep Screening questionnaire called the SSS Disturbed Rest has been developed for use by Allied Health Practitioners as part of routine initial case history. The Sleep Screening is used routinely for all initial paediatric consultations presenting at our clinic for Speech Pathology assessment by way of medical or dental referral.

Results: Simple questionnaires probing features of sleep, demonstrate that at least 90% of the participating parent population missed key Red Flags for sleep problems and SDB.

Conclusions: Children’s sleep issues are related to behaviour, environment, and SDB and were modifiable by parent education and support. The screening questionnaire was successful with 70% parents, educating on the Red Flags for sleep problems. It demonstrated significant changes in children’s sleep after medical, dental and /or myofunctional intervention, and showed good potential as a tool for wider use. It would lend itself to further study and validation.

The author proposes that the questionnaire may assist with public health and education campaign for parents, and if supported by medical, dental and allied health professionals who treat paediatric populations, and professionals responsible for care and education of pre-adolescent children, has potential to improve parents’ sleep literacy, and children’s sleep.

Acknowledgements: Dr Jim Papadopoulos for permission to use the sleep screening acronym, SSS Disturbed Rest, for further development and use in my clinic.
DIFFERENCE OF SLEEP HABIT BETWEEN PRESCHOOLERS ATTENDING NURSERY SCHOOLS AND KINDERGARTENS

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Introduction: Sleep habit during childhood is affected by daytime activity and school schedule. The aim of the study was to elucidate the difference of sleep habits among preschoolers in different daytime conditions.

Materials and methods: All public kindergartens and nursery schools in Yamaguchi City, Japan participate in the study. Child and Adolescent Sleep Checklist was distributed to all caregivers of children between 4-5 years of age. Five hundred and seventeen responses (response rate: 62.9%) were included in the analysis. Sleep habits were compared between children attending kindergartens (n=239) and nursery schools (n=278).

Results: More than half (56.9%) of the children attending nursery schools take regular nap after lunchtime, but 50.2% of children attending kindergartens do not take a nap. Mean duration of nap was 39.2 minutes/day among children attending nursery schools, which was significantly longer than children attending kindergartens. Mean time returning home was significantly later in children attending nursery schools (5:42pm) than in kindergarteners (3:03pm). Bedtime on weekdays and weekends were significantly later, and wake time on weekdays was significantly earlier in children attending nursery schools. Time to fall asleep was significantly longer, and bedtime resistance and unrefreshed feeling in the morning was significantly prevalent among children attending nursery schools than kindergarteners.

Conclusions: Children attending nursery schools take regular nap and returning home later, and they showed later bedtime and had more problems before and after bedtime. Regular nap at nursery schools was associated with sleep problems of preschoolers and may require some modification of daytime school schedule.

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**SLEEP AND SLEEP LOCATION**

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**Introduction:** Healthy sleep duration and sleep efficiency are linked to better cognitive, physical, and socioemotional health in infancy and childhood. The recent change in Sudden Infant Death Syndrome (SIDS) guidelines by the American Academy of Pediatrics to include rooming in until 12 months of age was based on three studies that have been debated as insufficient to support the new guidelines and present concern regarding the potential negative impact of rooming in on infants’ sleep duration and efficiency. Secondary data analysis from a larger study examining sleep characteristics and stress among a diverse group of 12-month-old toddlers living with socioeconomic adversity will be presented to describe the associations between toddlers’ sleep location and their sleep characteristics.

**Materials and methods:** In the primary cross-sectional study, we recruited healthy toddlers living with socioeconomic adversity from Early Head Start programs and a community primary care clinic in the Northeastern United States. The cohort of toddlers in this secondary analysis were racially diverse with 41.9% black, 34.9% Hispanic, and 18.2% non-Hispanic White. More than half (55.8%) of the families were at or below the federal poverty level (income reported to be

**Results:** The sample included 44 toddlers (Mean age=15.4 months, SD=3.2; 65.9% female). The toddlers’ nighttime sleep duration averaged 487.6 minutes (SD=54.2) with a sleep efficiency of 79.9% (SD=5.1) and mean sleep fragmentation index of 30.6 (SD=6.4). The majority of the toddlers were either co-sleeping [29.5%(13/44)] or rooming in with their caregiver(s) [47.7%(21/44)] and 22.7%(10/44) were independent sleepers. Sleep duration and sleep efficiency were highest among independent sleepers compared to rooming in and co-sleeping toddlers (F=7.21, Prob > F = .002 and F = 3.32, Prob > F = .05, respectively). Variation in night-to-night sleep efficiency was significantly less among independent sleepers (F=4.83, Prob > F = .01).

**Conclusions:** While great improvements in the rate of infants dying from SIDS has been attributed to the guidelines set by the American Academy of Pediatrics, the recent guideline change to include rooming-in until 12 months of age is incongruent with socioemotional development at this age and may impact sleep health at this age. Separation anxiety peaks between 6-18 months, making the AAP rooming-in recommendation until 12 months a concern regarding the quality of sleep among 12-month old who are rooming in. While our primary aim was not to examine the effects of the AAP recommendation, our findings of associations between rooming-in and co-sleeping and sleep warrant further examination. Experts have questioned the strength of the data used to support the new recommendation of rooming-in until age 12 months. This study provides objective data to suggest that the AAP recommendation should be reconsidered due to the potential impact on sleep health.

**Acknowledgements:** We are grateful to all of the community families who graciously allowed us into their homes and participated in our research study.
DO CHILDREN SLEEP LONGER IN HOME OR INPATIENT CARDIORESPIRATORY POLYGRAPHY STUDIES?

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Introduction: Southampton Children's Hospital developed an innovative home sleep service enabling cardiorespiratory polygraphy studies (CP) to be performed at home (1). Inpatient CP studies are performed using the SomnoSCREEN device (respiratory effort, pulse oximetry, nasal pressure airflow, oronasal thermistry, snore, ECG, body position, actimetry, video); home CP studies are performed using the SomnoTOUCH device (respiratory effort, pulse oximetry, nasal pressure airflow, snore, body position, actimetry, video) (SMed, Germany). Parents are given the choice of home or inpatient CP. Those who choose to do the study at home attend a training session to be taught how to set up the equipment on their child; written and photographic instructions are also provided. Those who choose to attend an inpatient CP are admitted to a ward cubicle (patients <18 months in age are occasionally admitted to a bed in multi-patient bay) and have the study set up and observed overnight by a trained member of the Sleep team. Home CP provides a myriad benefits to the Sleep service and patients:
- Parents report that avoiding a hospital stay is appealing as it lessens disruption to the family routine
- Children may have anxiety related to hospital admissions
- Home CP is more cost effective as it does not require a bedspace and associated staffing
- Different surroundings and noise/light in the ward environment (2), may lead to an unsettled night and insufficient results from the sleep study This abstract explores whether longer sleep time is obtained from home CP when compared to inpatient CP.

Materials and Methods: Data was collected retrospectively from all successful home and inpatient CP between 1/6/17 and 30/8/17. Periods of wake and gross body movement were excluded using observations from the embedded video, and parental (home) or attendant staff's (inpatient) observations. Estimated Total Sleep Time (TST) was calculated using Somno software, statistical analyses were performed using SPSS.

Results: Table 1: Age and TST

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<th></th>
<th>Home</th>
<th>Inpatient</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>N</td>
<td>61</td>
<td>34</td>
<td>--</td>
</tr>
<tr>
<td>Age (years) (mean, IQR)</td>
<td>6.5 (1.8-7.2)</td>
<td>5.1 (1.9-7.2)</td>
<td>0.771</td>
</tr>
<tr>
<td>TST (hours)</td>
<td>8.4 (7.6-9.8)</td>
<td>7.5 (6.9-8.1)</td>
<td>0.003*</td>
</tr>
</tbody>
</table>

*p<0.01

Conclusions: Patients opting for the home CP service tended to be slightly older (group difference not statistically significant) than inpatients, but TST was higher in this group (Table 1). This previously undocumented benefit provides additional evidence of the value of the service and suggests its utilisation should be expanded where possible. Research into the subgroups that chose each type of CP study should be undertaken to evaluate if improvements to the home CP service can be made to expand service capacity and increase uptake of this option.

References:
HIGH PREVALENCE OF OSA IN CHILDREN REFERRED TO ADENOTONSILLECTOMY

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Introduction: Pediatric obstructive sleep apnea (OSA) is a common condition estimated to affect 1 - 4 % of the pediatric population. The main reason for OSA in children is adenotonsilar hypertrophy. There is disagreement regarding the need for sleep studies before adenotonsillectomy to confirm a diagnosis of OSA. Several studies have evaluated questionnaires and physical examination as tools to predict OSA with conflicting results. The aim of this study was to evaluate the prevalence of OSA among children referred to adenotonsillectomy and whether questionnaires or physical examination can help predict OSA.

Materials and methods: This is a prospective cohort study of children aged 2 - 6 years, referred for adenotonsillectomy. The patients selected are a random sample of all the referrals. PSG with video recordings and an otorhinological examination was performed. OSA was defined as an obstructive apnea hypopnea index (OAHI) ≥ 2 (mild: OAHI 2 - 5, moderate: OAHI 5 - 10, severe: OAHI ≥ 10). Tonsillar size and the oral cavity was graded from 1-4. The children’s parents completed the Pediatric Sleep Questionnaire (PSQ) and the OSA-18.

Results: 100 children were included in the study. The prevalence of OSA was 73 %, with 50 % having moderate to severe OSA. Their mean age were 3.6 years and 42 % were girls. The mean BMI z-score was 0.45 and 91.3 % of the children were in the normal weight range. Most of the children were of Caucasian ethnicity (84 %). None of the children were exposed to tobacco smoke at home and most of the children (84 %) had been breast feeding. 8 % had a premature birth and 27.8 % had a family history of OSA. We found no significant differences in the clinical characteristics for the different severity levels of OSA. When comparing OAHI in the different group of tonsil size, we found a significant difference (p < 0.01). Sensitivity and specificity for PSQ and OSA-18 were calculated using multiple cut off points, and the range was from 22.4 % to 96 %. In multivariate logistic regression analyses for the different levels of OSA severity, age was significantly associated with OAHI ≥ 2 and tonsil size with OAHI ≥ 5.

Conclusions: We found a high prevalence of OSA, and many had a moderate to severe OSA, but neither questionnaires nor clinical characteristics were sensitive enough to predict that. There was a tendency for the youngest children to have more severe OSA, and for the children with larger tonsils to have more severe OSA. In this population of children referred directly to surgery, it seems to be a good selection process in advance of the referral and PSG is not strictly demanding in advance of surgery. Because of the high prevalence of moderate to severe OSA, and the increased risk of complications, it is important to underline that one should give special attention to these children during and after surgery. There is also an increased risk for having a rest OSA after surgery, so the threshold for doing a PSG after surgery should be low.
NONINVASIVE VENTILATION EXPERIENCE IN THREE RUSSIAN INFANTS WITH CONGENITAL CENTRAL HYPOVENTILATION SYNDROME

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Introduction: Congenital central hypoventilation syndrome (CCHS) is rarely diagnosed in Russian Federation, only 7 case reports have been published in local journals, approximately 20 children and one adult are known to have been diagnosed with CCHS and 2 – with ROHHAD. Most of the patients are on positive pressure ventilation via tracheostomy or had been decannulated, four have diahpragm pacers. We report our experience with noninvasive positive pressure ventilation (NIPPV) in three CCHS infants.

Materials and methods: We analyzed medical history of three patients who are coordinated in Almazov National Medical Research Centre and City Hospital for Children No.17, St Petersburg, Russia. Girl V. is now 5.5 years old (NPARM 20/25), girl E. is 3 y.o. (20/27), boy Z. is 6 months old (20/26).

Results: Two babies were born preterm with gestational ages (GA) of 30 (girl V., a twin pregnancy, placental bleeding, moderate asphyxia), and 36 weeks (boy Z.), both appropriate for GA, both after IVF procedure. Girl E. was term, after normal pregnancy. Both term and near-term infants presented with apnea during first hours of life, were intubated and received SIMV. Further attempts of extubation led to hypoventilation or apnea, emergency intubations. Both had muscle hypotonia; boy Z got repeated ECG monitoring because of episodes of sinus bradycardia with HR falls to 75 bpm. They were diagnosed with CCHS at the ages of 10-14 weeks. Boy Z. was transferred to our hospital from another town at 17 weeks of age. Girl V. received surfactant and was extubated after 40 hours of SIMV after birth. In this girl, CCHS was mimicking residual RDS and cerebral ischemia, with compensated respiratory acidosis (pCO2 55-65 mm Hg), muscle hypotonia during first two weeks. In the third week, pCO2 raised to 146 mm Hg, NIPPV was started with gas exchange normalization in 5 hours. After 3 cycles of spontaneous breathing and ventilation (NIPPV or SIMV), CCHS was suspected and genetically approved at 7 weeks of age. In all cases other morbidities were ruled out. Ventilation strategies were discussed with the parents, and all of them refused tracheostomy. NIPPV was started at the ages of 7 weeks in girl V., 11 weeks in girl E and 18 weeks in boy Z. The last two infants needed NIPPV only during sleep, and were discharged home three weeks after. A preterm girl had to stabilize her sleep-wakfullness pattern and stayed inpatient till 4.5 months old. Both girls have good compliance with mask ventilation, good cardio-respiratory monitoring, ECHO, 24-ECG, abdominal US results, appropriate growth and neurocognitive development (speech is delayed in a 3 y.o. with 20/27 mutation). They use to change nasal, oro-nasal and facial masks. Both families took flights or car trips for up 2500 km distance. None had lower respiratory tract infection.

Conclusions: NIPPV started from infancy can be successful in patients with low length of the repeat mutation. It reduces respiratory tract infections risk, improves life quality, and is preferred by the families.

Acknowledgements: To Dr. Martin Samuels, Great Ormond Street Hospital, London To Dr. Johen Peters, Krankenfürsorge des Dritten Ordens, Munich who helped us with the first steps with CCHS and are always opened for discussion.
A DOUBLE-BLIND, PLACEBO-CONTROLLED, RANDOMIZED-WITHDRAWAL, MULTICENTER STUDY OF THE EFFICACY & SAFETY OF SODIUM OXYBATE IN PEDIATRIC SUBJECTS WITH NARCOLEPSY WITH CATAPLEXY


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Introduction: Narcolepsy commonly begins in childhood or adolescence. Sodium oxybate (SXB) is a standard of care for cataplexy and excessive sleepiness (ES) in adults with narcolepsy. However, there were no randomized, placebo-controlled prospective studies evaluating SXB in children and adolescents with narcolepsy. The aim of this study was to evaluate the efficacy and safety of SXB in children and adolescents with narcolepsy with cataplexy.

Materials and methods: Children and adolescents (7-16 years) diagnosed with narcolepsy with cataplexy who were either SXB-treated or SXB-naive at study entry were included. SXB-naive participants were individually titrated to a stable dose of SXB; SXB-treated participants remained on their current dose. After the stable dose period, participants entered a 2-week double-blind, placebo-controlled withdrawal period (DB) and were randomized 1:1 to continue SXB or be switched to placebo, after which participants entered an open-label safety period for a total study duration of 1 year. Efficacy measures compared the last 2 weeks of the stable-dose period to the DB period. Safety and tolerability also were assessed. The safety population included all participants who were dispensed the study drug.

Results: Among the 63 randomized participants, 41% were 7-11 years, 44% were female, and 38% were SXB-treated at study entry. A pre-planned interim analysis of 35 participants showed that efficacy was achieved (P=0.0002) based on the primary endpoint (change in weekly cataplexy attacks). Therefore, randomization to placebo during the DB period was terminated early. For the 63 randomized participants, weekly cataplexy attacks were significantly decreased in the placebo group (median 12.7/week) compared to participants continuing SXB (median 0.3/week; P<0.0001). Cataplexy severity as assessed by Clinical Global Impression of Change (key secondary endpoint) was worse in the placebo group compared to participants continuing SXB (placebo 65.6% vs. SXB 17.2% rated as “much worse” or “very much worse”; overall difference P=0.0006). ES was significantly worse in the placebo group with a median increase of 3.0 points on the Epworth Sleepiness Scale for Children and Adolescents (second key secondary endpoint) compared to no change in participants continuing SXB (P=0.0004). Adverse events >10% in the safety population (n=104) were enuresis, nausea, vomiting, headache, and weight decrease.

Conclusions: These results support the efficacy and safety of SXB for the treatment of cataplexy and ES in children and adolescents with narcolepsy with cataplexy. The safety profile of SXB in this study was consistent with previous studies in both adult and pediatric patients with narcolepsy.

Acknowledgements: This study was supported by Jazz Pharmaceuticals. Under the direction of the authors, Teresa Steininger, PhD, at Jazz Pharmaceuticals, and Kirsty Nahm, MD, of The Curry Rockefeller Group, LLC (CRG) provided medical writing assistance. Editorial assistance in formatting, proofreading, copyediting, and fact checking was also provided by CRG. Jazz Pharmaceutical provided funding to CRG for writing and editorial support. Dr. Rosen acknowledges the following support: “This publication was made possible by the Clinical and Translational Science Collaborative of Cleveland, 4UL1TR000439 from the National Center for Advancing Translational Sciences (NCATS) component of the National Institutes of Health and NIH roadmap for Medical Research. Its contents are solely the responsibility of the authors and do not necessarily represent the official views of the NIH.” Disclosures: Dr. Plazzi has participated in advisory boards for UCB and Jazz Pharmaceuticals; Dr. Rosen has been a consultant for Jazz Pharmaceuticals, Advance-Medical and Natus Medical; Dr. Ruoff has served as an advisory board member and unpaid consultant for Jazz Pharmaceuticals; Dr. Lecendreux has received honoraria from Shire and grant support from UCB; Dr. Dauvilliers is a consultant for and has participated in advisory boards for Jazz Pharmaceuticals, UCB Pharma, Flamel Technologies, Theranexus, and Bioprojet; Dr. Black is a part-time employee of Jazz Pharmaceuticals and shareholder of Jazz Pharmaceuticals plc.; Ms. Parvataneni, and Dr. Wang are, and Dr. Guinta was, full-time employees of Jazz Pharmaceuticals, who, in the course of this employment, has received stock options exercisable for, and other stock awards of, ordinary shares of Jazz Pharmaceuticals, plc; Dr. Mignot has received research support from Jazz Pharmaceuticals, Merck, and Glaxo Smith Kline (GSK), has consulted for Novo Nordisk and Reset Pharmaceuticals, and is on the speakers' bureau for Vox Media.
THE DIAGNOSTIC ACCURACY OF TYPE III PORTABLE SLEEP MONITORS VERSUS POLYSOMNOGRAPHY FOR OBSTRUCTIVE SLEEP APNEA IN CHILDREN: A SYSTEMATIC REVIEW

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Introduction: Obstructive sleep apnea syndrome (OSAS) is defined by reduction or cessation of the respiratory flow because of partial or total obstruction of the upper airway. It is highly prevalent in children (1 to 4%) and if not diagnosed and treated it can cause metabolic, cardiovascular and behavioral complications. Its diagnosis is performed via clinical evaluation and sleep study (polysomnography [PSG]). A diagnosed based only on clinical assessment is not considered reliable. The gold standard is polysomnography in the sleep laboratory; but it is complicated and expensive, due to the need of not easily available human and material resources. Another option for diagnosis are portable sleep monitors, either in the laboratory or at home. Among them type 3 monitors count with 4 to 7 channels and can be used at home. There is no systematic review analyzing the diagnostic accuracy only of type 3 monitors to diagnose OSAS in children.

Materials and methods: Searches were performed in performed in MEDLINE, EMBASE, PsycINFO, CINAHL, LILACS, African Index Medicus, Web of Science, Scopus, KoreaMed, China Knowledge Resource Integrated Database and the Cochrane CENTRAL and ProQuest using the appropriate keywords according to the PICO structure, such as child, obstructive sleep apnea, polysomnography, type 3 sleep monitor, unattended sleep study. The last searches were performed on March 28, 2018. The results were purged of duplicates using Mendeley Desktop and JavRef and evaluated by two reviewers individually to assess their potential inclusion, with differences being resolved by discussion or if persistent by a third reviewer. Quality of evidence was evaluated using QUADAS-2. PROSPERO register CRD42017079437.

Results: 1863 registers were found after the search, with 23 included from other sources, for a total of 1886. After removing duplicates 1145 remained, which were reviewed in title and abstract. 25 met criteria and were selected for review in full text. 15 weren’t included in the review due to several reasons, most common type 4 monitor. The 10 articles included evaluated 492 children. A meta-analysis was not possible due to the different monitors evaluated and the diverse cut points and indexes used for the diagnostic test. 7 were performed in the sleep laboratory, 2 at home and laboratory and 1 at home; 7 evaluated commercially available monitors, and 3 PSG channels. The mean age was 7.17 years, with ages from 2 to 18 years old, 59.44% male. All included oximetry and pulse rate, only one carbon dioxide or pulse waveform. With 1 event per hour cut point in the PSG sensibility ranged from 0.63 to 0.82, and specificity from 0.6 to 0.91 in the laboratory, and at home 0.7 to 1 and 0.43 to 0.66 respectively. The evidence was of moderate quality, worst at patient selection.

Conclusions: Diagnostic accuracy of type III monitors is low. Clinicians must be cautious with their use, especially if they are performed at home. It is necessary to explore other ways of simplified evaluation for OSAS in children.

Acknowledgements: Dr. Mario Orellana
**Introdution:** The use of modern information technologies (IT) can affect children's physical and mental health. The aim of the present study was to find out how long young children use different IT devices (screens) and whether the IT usage is associated with sleep habits, routines, sleep duration and sleep problems.

**Materials and methods:** We have analyzed data of 962 children, aged between 18-71 months (M = 42.57, SD = 15.23). Parents of toddlers and preschool children from different regions of Lithuania were asked to fill in questionnaires about their children's mental and physical health and the use of information technologies, screen time on workdays and weekends. The study was carried out in April-December 2017. The survey questionnaire contained the questions about the child's development and the social environment, usage of IT devices. Children's sleep problems were assessed using the Child's Behavior Checklist (CBCL /1½-5).

**Results:** The results of the study have revealed that most children's sleep duration is 9-10 hours per night. 96.2% of children, who slept 8 hours, and all children, who slept 7 hours during night, slept also during the day time. 75.8% of children at this age slept during the day. Younger children have the habits and routine before sleep time, but they also have more awakenings during the night time. About half of the children had at least one IT device in their bedroom. Children of parents with lower education are more likely to have an IT device in the bedroom, and they are permitted to watch movies before bed time. Also children who had an IT device in their bedroom had more difficulties initiating sleep, and they had more sleep problems (reaching the borderline or clinical level). Children who were sleeping shorter than 8 hours per night were using computers longer during the week and on weekends, also they spent more time on other screens during the weekends.

**Conclusions:** This study supports the recent findings in other countries that the use of IT in the early childhood before going to sleep and having devices in bedroom are associated with sleep problems, difficulties initiating sleep and shorter sleep duration.

**Acknowledgements:** This study is funded by Research Council of Lithuania (Agreement no. GER 2017/006).
A SIMULTANEOUS MANDIBULAR ADVANCEMENT AND PALATAL EXPANSION FOR THE MANAGEMENT OF PEDIATRIC OSAHS: PRELIMINARY RESULTS

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Introduction: Generally, the superior airways obstruction observed during sleep in pediatric OSAHS is associated to some anomaly of the maxilla-mandibular complex morphology. Hence, the current practice includes orthognathic therapies as management possibility. The aim of this study was to evaluate the efficiency of an innovative orthognathic device which simultaneously carries out palatal expansion and mandibular advancement on the evolution of the OSAHS.

Materials and methods: Children with an Apnea/Hypopnea Index (AHI) superior to the pathologic threshold of 1 event per hour of sleep were included to this study to be treated using this innovative orthognathic device. The evolution of the respiratory status observed during sleep was assessed with a polysomnography analysis. These data were confronted to the modification of the maxilla-mandibular complex anatomy, supposedly induced by the device, and evaluated through the analysis of lateral cephalograms and 3D impression of the maxillary and mandibular arches. T test of Students were applied to attest the significance of these evolutions.

Results: After several months of treatment, we have observed that the tested device induced an infero-anterior rotation of the mandible and a significant maxillary widening after several months of treatment. This increasing on the oral space was combined to a significant improvement of the airway obstruction particularly highlighted by the systematic reduction of the AHI under the severity threshold of 5 events per hour of sleep.

Conclusions: Finally, these preliminary results attest that the airway obstruction observed during sleep may be managed by the simultaneous palatal expansion and mandibular advancement induced by this innovative orthognathic device. Thus, we suggest this new therapeutic strategy as a valuable alternative treatment for children with OSAHS related to craniofacial anomalies.

Acknowledgements: We would like to thanks the Dental Office of Dr. Bonnaure (Rennes, France) for sharing of the tested orthognathic device and performing the imaging, as well as the Sleep Clinic of Dr. Moisdon (Rennes, France) for performing the polysomnography analyses.
SLEEP-DISORDERED BREATHING IN TWO CHILDREN WITH PANTOTHENATE KINASE-ASSOCIATED NEURODEGENERATION UNDER BACLOFEN THERAPY

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Introduction: Pantothenate Kinase-Associated Neurodegeneration (PKAN) is a rare neurodegenerative genetic disorder characterized by progressive extrapyramidal signs (rigidity, dystonia and choreoathetosis) due to iron accumulation in the basal ganglia. A mutation in the pantothenate kinase gene (PANK2) on chromosome 20p13 is implied. There are few reports on sleep in this disease, none referring to sleep-disordered breathing (SDB). Baclofen is a centrally acting gamma-aminobutyric acid receptor B (GABA-B) agonist used to treat severe spasticity associated with PKAN, that might depress central ventilatory drive inducing central sleep apnea (CSA) along with its myorelaxant effect.

Materials and methods: Two cases of PKAN under baclofen therapy with CSA diagnosis after level 1 polysomnography are described. Scoring of sleep stages and respiratory events was performed according to the updated 2017 Scoring Manual of the American Academy of Sleep Medicine.

Results: Case 1: 9 year-old boy with PKAN diagnosis and onset of symptoms at age 3, referred for evaluation of nocturnal apnea. He was medicated with oral baclofen, trihexyphenidyl, tetrabenazine and levopromazine. Parents reported snoring and breathing pauses not always related to snoring (more evident in the last 1,5 years) and multiple nighttime awakenings with dystonic movements (in the last 2 months). Overnight polysomnography revealed severe CSA with an overall apnea-hypopnea index (AHI) of 36/h and a central apnea index (CAI) of 34/h, bradipnea and prolonged expiration. The patient responded to bilevel positive airway pressure in spontaneous-timed mode (BPAP-ST) with good compliance (9h/night) and resolution of nighttime awakenings and dystonia during the night.

Case 2: 13 year-old boy with PKAN diagnosis and symptoms since 18 months, under oral baclofen, trihexyphenidyl, tetrabenazine, morphine, diazepam, chloral hydrate, clonidine and transdermal fentanyl therapy. He started having hypoxia episodes during sleep, with minimum oxyhemoglobin saturation of 80%, intermittent snoring, breathing pauses and multiple nighttime awakenings. Overnight polysomnography showed CSA with an overall AHI of 6/h and a CAI of 5/h, bradypnea, prolonged expiration with a brief respiratory effort and stertor at the end of each respiratory cycle. Nocturnal noninvasive ventilation (NIV) will be started.

Conclusions: Both patients presented obstructive symptoms but central respiratory events were predominant. Breathing pauses were reported, despite being a symptom not always mentioned by parents. Insomnia with nighttime awakenings is suggestive of SDB as noted in these cases. PKAN in pediatric patients presents with severe dystonia and spasticity, therefore baclofen is usually prescribed in high doses and should not be stopped. SDB should be considered and evaluated, especially if baclofen is combined with other respiratory depressants.

Severity of CSA was greater in case 1, though he was medicated with fewer respiratory depressant drugs. We postulate that PKAN itself may favor the occurrence of SDB. More studies regarding sleep in these patients are necessary.

PKAN is a very challenging disease for caregivers. Treatment of SDB with NIV, with consequent reduction of awakenings, results in significant improvement in the quality of life of patients and their families, as seen in case 1.
SLEEP RESPIRATORY FINDINGS IN ROHHAD SYNDROME

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Introduction: Rapid-onset obesity with hypothalamic dysfunction, hypoventilation and autonomic dysregulation (ROHHAD) syndrome is a very rare disorder occurring in the first years of life with a later onset of central hypoventilation, responsible for a high mortality.

Materials and methods: The authors describe the respiratory findings and management in a child with ROHHAD syndrome followed in a sleep laboratory of pulmonology unit in a tertiary pediatric hospital.

Results: A 2 years old girl presented with hyperphagia and weight gain (10th to >95th percentile), lowered activity, excessive sweating, thermal dysregulation, constipation and snoring in the last 2 months. She was born at term, with normal growth and development until then. Cerebral MRI was normal, and laboratory studies revealed hyperlipidemia, insulin resistance, hyperprolactinemia and high follicle-stimulating hormone, leptine and lactate dehydrogenase serum levels. Abdominal MRI showed a small right adrenal nodular image but positron emission tomographic scan excluded hypermetabolic neoplastic lesion. Overnight polysomnography (PSG) revealed mild obstructive sleep apnea syndrome (OSAS) – apnea-hypopnea index 2.3, mean SpO2 96%, minimum SpO2 93%, TcCO2 47 - 50 mmHg – and no cardiorespiratory response to hypoxemia and hypercapnia. Non-invasive ventilation (NIV) during sleep was started; difficult in the beginning due to behavioral problems. Three months later, she was admitted to PICU for hypercapnic coma (pH 7.1, pCO2 119mmHg); staying on invasive ventilation for 10 days and extubation being difficult because of central bradypnea. She was discharged on NIV with pulse oximetry and end tidal CO2 monitoring. PSG 6 months after diagnosis showed absence of cardiorespiratory response to hypoxemia or hypercapnia but no central apneas or hypopneas. During follow-up (thirty months) she was admitted two times for acute respiratory infections with hypercapnia due to central hypoventilation. Invasive mechanical ventilation was not needed.

Conclusions: Respiratory manifestations in ROHHAD syndrome can be seen months after the onset of obesity and OSAS can precede central hypoventilation. Development of intermittent and unrecognized central hypoventilation is the largest concern because of risk for respiratory arrest. NIV and close monitoring of hypoxemia and hypercapnia are essential to avoid fatal outcome.
Introduction: Sleep disorders, particularly abnormal circadian rhythms and sleep deprivation, are one of the risk factors for suicide. Moreover, we know that during adolescence, the developmental process frequently leads to a delayed sleep phase with a possible desynchronization of biological rhythms. We were interested in the sleep of suicidal teenagers, in order to study the basic chronotype.

Materials and methods: We recruited 58 suicidal adolescents and 225 controls, and then collected various components of the sleep and wake rhythm, dissociating sleep during school time and out of school time (Munich Chronotype Questionnaire = MCQT). We also collected evidence regarding energy, focus, quality of life and social support from validated scales (Life and Perceived Health-Ado = VSP-A, Multidimensional Scale of Perceived Social Support = MSPSS and Kidscreen-27).

Results: The suicides were on average 14.3 years old (1.5) (versus 14.2 (1.1), p = 0.8). We obtained significant results concerning total sleep time during the school time period (6h30 (2:12) versus 7:20 (1:09), p = 0.01), which reflects a greater sleep debt for suicidal teenagers. They also have a longer sleep latency (out of school period) (1:24 (1:20) versus 54 (1:12), p = 0.04), which contributes to increase their sleep debt. The results for the social Jet Lag were not significant, probably due to too much deterioration of sleep quality in suicidal patients.

Conclusions: These results illustrate that 4 weeks before the suicidal act, there are major changes in sleep that signify a degraded psychic state. For these adolescents, the hereditary and developmental components have an impact on the physiology of sleep, especially in terms of disinhibition, and those especially since this public frequently have a high impulsivity. These results encourage further research on these sleep disorders: are they primary, representing a prodrome vulnerability trait of a psychic disorder, or are they secondary, and therefore a marker of a more recent risk state?
RELATIONSHIP BETWEEN SLEEP SPINDLES AND COGNITIVE PERFORMANCE: ARE THERE ANY DIFFERENCES BETWEEN ADHD SUBTYPES?

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Introduction: Attention Deficit Hyperactivity Disorder (ADHD) is characterized by symptoms of hyperactivity, impulsivity, and/or inattention in childhood. ADHD is one of the most prevalent disorders and it is associated with difficulties in cognitive functioning and, therefore, with academic performance, being able to influence in not advancing academic courses or even in abandoning the studies. During childhood and adolescence, sleep undergoes very significant evolutionary changes that include variations in the quantity, distribution, and characteristics of sleep architecture and microstructure. When problems occur in some of these elements, sleep disorders may appear more so if there is psychiatric comorbidity of any kind and, especially, in ADHD. Sleep spindle properties index cognitive faculties such as memory consolidation and intellectual abilities. For this reason, scoring sleep spindle properties in polysomnographic recordings has become an important activity in both research and clinical settings. The relationship between spindles and daytime activity and cognitive performance could be of special interest in the context of ADHD.

The goal of the current investigation was to assess the possible differences in sleep patterns in children with ADHD, also carrying out the analysis of spindles and exploring the relationship between cognitive performance and sleep spindles.

Materials and methods: 60 ADHD children (15 ADHD predominantly inattentive type, 15 ADHD predominantly hyperactive/impulsive type, 30 ADHD combined type) aged between 7-11 years were assessed with polysomnography. The identification of spindles was the preliminary processing steps for the scoring of sleep stages, according both AASM and Rechtschaffen and Kales guidelines. For the assessment of cognitive performance, the Wechsler Intelligence Scale for Children-Revised (WISC IV) was used.

Results: Phase latencies, means of percentages of each phase, sleep efficiency as well as spindle analyses will be presented. Moreover, this data will be related with cognitive performance, distinguishing by higher or lower spindle presence.

Conclusions: This study provides clarity to the relationship of the presence of spindles and cognitive performance in ADHD children. Despite this, these results could be generalizable to understand the relationship between spindle analyses and cognitive performance in children.

Acknowledgements: This study was funded by the Ministry of Economy and Competitiveness and by the European Regional Development Fund (ERDF) in the Call for Projects of Excellence (REF: PSI2014-58046-P).
SPINDLE ANALYSIS IN ADHD SUBTYPES

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¹University of Granada

Introduction: Attention Deficit Hyperactivity Disorder (ADHD) is a neurodevelopmental disorder characterized by symptoms of hyperactivity, impulsivity, and/or inattention in childhood. Following the Diagnostic and Statistical Manual of Mental Disorders in its revised version DSM 5, ADHD can be divided into three subtypes: predominantly inattentive ADHD (ADHD-I), predominantly hyperactive/impulsive ADHD (ADHD), and ADHD of the combined type (ADHD-C). Up to 70% of children with attention-deficit/hyperactivity disorder (ADHD) experience sleep problems, and these are associated with poorer child and family well-being. In studies based on subjective assessment of sleep problems (parents report on their children's problems), there is widespread agreement about the existence of more problems in these children; while in studies using objective measures (polysomnography, actigraphy), these differences seem more subtle. The inconsistency of these results may be due to the lack of studies that explore the presence of markers of sleep microstructure like spindles differentiating the ADHD subtypes. Sleep spindles are distinctive electroencephalographic (EEG) oscillations in the frequency range of 9–15 Hz occurring in non-rapid eye movement (NREM) sleep and are a defining feature of stage 2 sleep. Sleep spindles have been repeatedly linked with general cognitive and memory abilities. As ADHD children show differences in cognitive performance between subtypes, it is important to examine the presence of spindles to better characterize sleep characteristics of these children. The aim of the present study was to do a spindle analysis to assess the possible differences between the three ADHD subtypes.

Results: Spindle analyses will be presented for each ADHD subtype, and related to sleep quality. ANOVA's tests will be performed to assess the differences between groups.

Materials and methods: 60 ADHD children (15 ADHD predominantly inattentive type, 15 ADHD predominantly hyperactive/impulsive type, 30 ADHD combined type) aged between 7-11 years were included in the study. All of them had a clinical diagnosis, but deficit attention and hyperactivity scale was administered for corroborate the distinction between subtypes (EDAH).

To evaluate the sleep quality all of children were assessed with polysomnography.

The identification of spindles was carried out according both AASM and Rechtschaffen and Kales guidelines.

Conclusions: In light of the results obtained, evidence is provided on the importance of the assessment of spindles for a better knowledge of ADHD profiles. These analyses are clearly important to differentiate sleep patterns in ADHD subtypes. It is also important to explain the contradictory results in cognitive performance between ADHD subtypes.

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USE OF TRACHEAL SOUND AND SUPRA-STERNAL PRESSURE SIGNALS IMPROVES RESPIRATORY ANALYSIS OF AMBULATORY POLYSOMNOGRAPHY IN CHILDREN

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Introduction: To assess the contribution of tracheal sounds (TS) and supra-sternal pressure (SSP) to optimize respiratory analysis of ambulatory PSG in the diagnosis of pediatric obstructive sleep apnea (OSA) syndrome. The evaluation criteria were: signal quality (percentage of time spent in the absence of artefact for each signal) and overall recording quality without, then with, the contribution of TS and SSP (total sleep time with satisfactory quality observed simultaneously on all signals).

Materials and methods: 119 children aged between 2 and 18 years, referred to the sleep medicine unit for snoring and/or suspicion of OSA. An ambulatory PSG using the PneaVox sensor (CIDELEC, France) was installed in the sleep laboratory. The quality of respiratory signals, TS, SSP, nasal pressure (Pnas), thoracic and abdominal bands (RIP) and oximetry sensor, was evaluated.

Results: 113 recordings were analyzed. The average recording time was 556 minutes. Without the use of the TS and SSP signals, 62.8% of the recordings were considered "excellent quality" and 74.3% "very good quality". Adding just the SSP signal, 77% of the recordings were of "excellent" quality and 77.9% of "very good". Finally, including both TS and SSP signals, 95.6% of the recordings were of "excellent" quality, and 95.6% of the recordings were of "very good" quality.

Conclusions: The combination of TS signal with Pnas and SSP signal to RIP significantly improves the analysis quality of flow rate and respiratory effort.
Introduction: Tongue functional re-education is an alternative treatment to increase pharyngeal and nasal patency to cure sleep disorder breathing. However, its success is dependent upon patients’ adherence to daily orofacial exercises. The Tongue Right Positioner (TRP) is an oral medical device, aiming at establishing mature tongue functions and increasing oro-pharyngeal muscles tone. The goal of this study is to assess the effectiveness of TRP treatments on children's upper airways patency almost one year after its end.

Materials and methods: 70 orthodontic patients (age: 11.4±2.7) were retrospectively included in this study based on full availability of lateral teleradiographs taken before (baseline), at the end of the active orthodontic treatment and at the end of retention period. 49 patients had orthodontic treatments associated with TRP (TRP group) while 21 patients had only classic orthodontic treatment (control group). Antero-posterior pharynx diameters were measure on radiographs at the level of the tongue base and the velopharynx. In parallel, the Peak Nasal Inspiratory Flow (PNIF) was measured on patients of the TRP group before and after TRP setting, when TRP is removed at the end of orthodontic treatment, and in average 11 months after TRP removal.

Results: Statistical analysis showed that compared to baseline, the pharyngeal diameters at the end of treatment period increased by 15.5% (p=0.005) for TRP group versus 3.0% (p=0.41) for control group at tongue base level and 13.2% (p=0.03) for TRP group versus -1.2% (p=0.85) for control group at velopharynx level. At the end of retention period, pharynx diameters compared to baseline increased by 12.2% (p=0.07) for TRP group versus 8.3% (p=0.06) at tongue base level and 15.9% (p=0.02) for TRP group versus 4.0% (p=0.41) for control group at velopharynx level. Additionally, compared to baseline in the TRP group, PNIF increased 9% (p<10^{-4}) just after TRP setting and 38.5% (p<10^{-4}) at the end of active orthodontic treatment. PNIF remained significantly stable +38% (p<10^{-4}) 11 months after TRP removal.

Conclusions: The TRP device constrains the tongue to adopt physiological functions. These data show that functional changes probably improving oropharyngeal muscles tone have led to significant increases in upper airways diameter and patency, independently from orthodontic treatment. Further the benefits of re-education with TRP on oropharyngeal functions and upper airways are sustainable. These results suggest that oro-pharyngeal re-education with the TRP should be consider as potentially preventive or curative treatment of mouth breathing and sleep disordered breathing for young patients.
OVERCOMING EARLY REGULATION PROBLEMS WITH MINI-KISS-SLEEPTRAINING

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Introduction: Regulation problems are one of the earliest indicators of mental health difficulties in childhood. They have long-time effects on self-regulatory competence, behaviour problems, social-skills and parent-child-relationship. Therefore early intervention in young age is necessary. The Mini-KiSS Sleep training is addressing parents of young children up to 4 years of age suffering from sleep problems and disorders - mainly insomnia.

Materials and methods: In a pilot study (n = 15 families) with children suffering from regulation and sleep problems participated. Of those children, 40% were female, and 60% were male. They were between 11 and 51 months old (M = 24.6, SD = 13.03). All suffered from insomnia according to ICSD-3 criteria.

Results: Sleeping problems, bedtime resistance, sleep anxiety, night waking, daytime sleepiness decreased significantly. Furthermore, duration of going to bed ritual, use of external support to maintain sleep and sleeping in the parents bed decreased significantly. Besides those sleep related effects, regulation problems as excessive crying decreased on a clinical relevance basis (Wessel-Criteria). However, feeding problems were not significantly reduced after the Mini-KiSS sleep training. In addition, self-regulation ability significant improved and less parental co-regulation was necessary after treatment.

Conclusions: First effects of the Mini-KiSS sleep training showed significant results not only concerning sleep but also for further regulation problems. Mini-KiSS with its six sessions might therefore be helpful in early intervention and prevention of regulation problems.
ADOLESCENTS’ MOTIVATIONS FOR BEDTIME SOCIAL MEDIA USE: FROM QUALITATIVE INSIGHTS TO QUANTITATIVE MEASUREMENT

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Introduction: There is now a well-established link between poor sleep outcomes in adolescence and heavy social media use, particularly around bedtime. However, little is known about what drives adolescents’ social media habits, including late night social media engagement, despite negative consequences for sleep. The purpose of Study 1 was to gain an in-depth understanding of the motivating factors for adolescents’ social media use and how these impact on bedtime behaviours and sleep. Study 2 then aimed to use this qualitative insight to develop a quantitative self-report measure that captures not simply duration of nighttime social media use, but underlying cognitive and emotional difficulties experienced when disconnecting at night.

Materials and methods: In Study 1, 24 adolescents (age range: 11-17) participated in focus groups. Semi-structured discussions focused on motivators for social media use and perceived impact on bedtime behaviours and sleep. Thematic analysis was used to identify themes across focus groups. Study 2 generated and refined an experimental pool of candidate self-report items based on the identified theme structure. Pilot validation data has been collected from older adolescents and young adults, who rated candidate items from “not at all true of me” to “extremely true of me”, and completed measures of social media engagement and sleep duration, timing and quality (Sleep Condition Indicator).

Results: In Study 1, inductive thematic analysis identified two overarching themes - ‘Missing Out’ and ‘Norms and Expectations’ - each with three sub-themes. Fear of the offline costs of missing out on online content created anxiety around disconnecting at night. Participants also continued late night online interactions, despite tiredness and lack of enjoyment, in order to meet perceived social expectations and etiquette.

In Study 2, candidate items were generated for each of the six sub-themes. Exploratory Factor Analysis (EFA) of the pilot validation data supported a 3-factor solution, which retains items from the three most salient sub-themes. Iterative EFA and reliability analyses resulted in a final 12-item measure (Cronbach’s alpha = .94), with 3 subscales: Vigilance, Obligation and Fear of Exclusion (alpha = .87, .90, .91, respectively). The final measure showed convergent validity with measures of social media investment (rs = .50, p<.001) and fear of missing out (rs = .71, p<.001), but was a better predictor of social media use in bed, perceived sleep impact and poorer sleep quality than general measures of social media engagement in multiple regression models.

Conclusions: This study provides novel in-depth understanding of adolescents’ motivations for bedtime social media engagement. Our findings highlight possible barriers to encouraging healthier social media habits for sleep. Interventions aimed at improving adolescent sleep by targeting social media use therefore need to address not only behaviours but also underlying motivations. To support the ongoing development of understanding and evidence in this area, our study provides a validated 12-item self-report measure of difficulty disengaging from social media at night, that is data-driven and based on adolescents’ own perspectives.

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Introduction: There is increasing evidence highlighting links between adolescent sleep outcomes and social media use, which has become a “hot topic” of interest to the public, researchers and practitioners. In the UK, there remains a need for evidence from a large-scale, nationally representative sample of adolescents. This study aims to address this gap in current evidence by quantifying the relationship between daily social media use and sleep outcomes in adolescents in the UK Millennium Cohort Study.

Materials and methods: 11,714 adolescents (aged 13-15) reported their typical daily social media usage and sleep habits as part of the UK Millennium Cohort Study (Sweep 5). Binomial logistic regression models predicted the odds of binary sleep outcomes for low (<1 h), high (3-5 h) and very high (5+ h) social media users, compared to average users (1-3 h). Sleep outcomes were: later than average bedtimes and risetimes on school days and free days, sleep onset latency (SOL) over 30 minutes, and frequent nighttime awakenings. Models included comprehensive control variables: including demographics, household characteristics, general health and psychological wellbeing. Multiple imputation was performed to account for missing data. All analyses accounted for the complex sample design and longitudinal attrition.

Results: Late bedtimes on school days and free days respectively were more likely amongst high (OR = 1.24, p <.05; OR = 1.31, p <.01) and very high (OR = 2.12, p <.001; OR = 2.43, p <.001) social media users, and less likely amongst low users (OR = .61, p <.001; OR = .57, p <.001), compared to average users. High and very high users were more likely to have late school day risetimes (OR = 1.61, p <.05; OR = 1.88, p <.01, respectively). Very high users were also more likely to have late free day risetimes (OR = 1.57, p <.01), whereas low users were less likely (OR = .78, p <.01). Although high (OR = 1.24, p <.05) and very high (OR = 1.46, p <.001) users were more likely to report SOL longer than 30 minutes in basic models (controlling for age and sex), these relationships were no longer significant in comprehensively controlled models. Similarly, in basic models, high and very high users were more likely to report frequent nighttime awakenings (OR = 1.72, p <.05, OR = 2.40, p <.001, respectively). However, after including all control variables, this effect was non-significant for high users (OR = 1.09, p =.44) and notably reduced in very high users (OR = 1.35, p <.01).

Conclusions: This study confirms links between adolescents' daily social media use and sleep outcomes using a large, nationally representative UK sample and comprehensive control variables. It highlights a dose-response relationship whereby the heaviest social media users are most likely to have later bed- and rise- times and frequent nighttime awakenings. The strongest effects were for later bedtimes, resulting in restricted sleep opportunity for adolescents with relatively fixed school day risetimes. This evidence provides a starting point for more in-depth exploration of adolescents' social media and sleep habits.

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SUCCESSFUL MULTIDIMENSIONAL INTERVENTION COMBINING COGNITIVE BEHAVIOURAL THERAPY, ORTHODONTIC INTERVENTION AND MYOFUNCTIONAL THERAPY, IN A CHILD WITH SYMPTOMATIC MILD OSAHS AND SLEEP DEPRIVATION: A CASE REPORT

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Introduction: Pediatric Obstructive Sleep Apnoea-Hypopnea Syndrome (OSAHS) has a complex etiology, including anatomical and neuromuscular factors, adenotonsillar hypertrophy and obesity, and presents clinically with a variety of complains, such as disturbed nocturnal sleep, different parasomnias, hyperactivity, behavioral and school performance problems, depression, insomnia, and psychiatric problems (Huang YS, 2017). Sleep deprivation is known to worsen most sleep disorders, including OSAHS and parasomnias, and it negatively impacts daytime neurobehavioral functioning in children, possibly leading to impairments in executive functioning, emotional reactivity and eventually academic difficulties (Maski, 2013). Adenotonsillectomy has been the recommended treatment for pediatric OSAHS, but this recommendation has been questioned because of variable results and evidence of progressive recurrence in long term studies (Huang YS, 2014). Emerging research suggests that orthodontic interventions as rapid maxillary expansion for treatment of malocclusion, may help with the symptoms of these patients. (Altalibi, M. 2014). Current literature demonstrates that myofunctional therapy (MFT) can lead to a return to a normal oral-facial anatomy (Chauvois A, 1999) and decrease apnea-hypopnea index (AHI) by approximately 50% in adults and 62% in children, so it could serve as an adjunct to other obstructive sleep apnea treatments. (Camacho, M.).

Materials and methods: We report the case of a seven years’ old child reported for sleep evaluation because of snoring, insomnia, disturbed sleep, sleep terrors and daytime functioning impairment, characterized by unstable mood, irritability and poor school performance attributed to concentration difficulties; at observation the child was found to have mouth breathing, atypical swallowing, severe malocclusion and compromised speech. A CBT programme was first initiated to improve insomnia, parasomnia and sleep deprivation, after which there was some clinical improvement, with less perceived sleep fragmentation, remission of sleep terrors and better mood and school performance; an overnight polysomnography (PSG) was then done, which confirmed mild OAHs and sleep fragmentation related to the respiratory events. A complete orthodontic study including cephalometric and facial analyses and cast models study was performed, preceding rapid maxillary expansion (RME). Results Evaluation after RME revealed critical improvement in sleep and daytime symptoms, school performance, correction of malocclusion and mouth breathing, and partial improvement in atypical swallowing and speech. The control PSG showed improvement in the overnight oxygen saturation, oxygen desaturation index, but not in the AHI. The child is now starting MFT as an adjunct therapy for OSA and further correction of atypical swallowing and speech issues.

Conclusions: This case report highlights the importance of a multidimensional approach to pediatric OSA. In this child, gains in total sleep time, sleep continuity and a significant improvement in oxygen saturation and oxygen desaturation index during sleep, as a result of combined orthodontic treatment and CBT, were responsible for a striking improvement in daytime functioning and school performance. The addition of MFT may lead to further improvement.
P#47-Sunday

STUDY OF THE FAMILIAL AGGREGATION OF OBSTRUCTIVE SLEEP APNEA SYNDROME BASED ON A PEDIATRIC INDEX CASE

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Introduction: Studies on first-degree relative of nonobese patients suggest that genetics may play a role in Obstructive Sleep Apnea Syndrome (OSAS) (Redline et al., 1995), and an almost two fold in increase of tonsillar hypertrophy in children with a parent affected by OSAS (Lundkvist et al., 2012). However, these are database and questionnaire based studies and the absence of sleep recordings to ascertain the diagnosis of OSAS can be criticized. Aims Our aim was to study familial aggregation of OSAS and tonsillar hypertrophy in a population of non-obese children.

Materials and methods: Prospective monocentric cohort study including children with suspected OSAS. All patients underwent clinical examination to determine whether or not they met the criteria for clinical OSAS according HAS criteria and to quantify tonsillar hypertrophy. At inclusion, the Berlin questionnaire was completed by at least one parent, and familial history of tonsillectomy was collected. Polysomnographic recordings were performed when indicated. A telephone followup for clinical OSAS persistence and history of ENT surgery was performed.

Results: Seventy three nonobese children (25 girls) median [Q1;Q3] 5.1 [3.3;7.0] years old, were included. Population characteristics are presented in table 1. Clinical OSAS was present in 62 (85%) of them, 27 (38%) had a familial history of tonsillectomy, and 21 (29%) had at least one parent with a positive Berlin questionnaire. Fifty six children underwent PSG recording, with an Apnea Hypopnea Index, available in 52 children, of 2.9 [1.3;7.5]/h. Children were divided into two groups: no or mild OSAS (n=34 (65%)), and moderate to severe OSAS (n=18 (35%)). There was no difference between groups for the familial history of tonsillectomy (p=0.14) or the frequency of positive Berlin questionnaire (p=0.51). The size of the children’s tonsils was not related to the Berlin questionnaire result (p=1). Residual clinical OSAS was detected in 17/37 (46%) children followedup. It was more frequent in children without tonsillectomy after inclusion compared to these with tonsillectomy (p=0.001) but was not influenced by the Berlin questionnaire (p=0.72).

Conclusions: This study did not evidence familial aggregation of OSAS and tonsil hypertrophy. However, there is a tendency of more history of adenotonsillectomy in parents of children with moderate to severe OSAS (p=0.14). Studies on a larger number of patients might confirm this tendency.
SLEEP AND MATERNAL EMOTIONAL DISTRESS IN SOLO MOTHER FAMILIES COMPARED TO TWO-PARENT FAMILIES

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Introduction: families by choice), yet the research regarding this family structure has been limited. Few previous studies demonstrated that solo mothers are at risk of emotional distress, which is associated with maternal and infant sleep problems. In the current study, we were interested in examining for the first time the characteristics of infant and maternal sleep patterns in solo mother families. The specific aims of the current study were: (1) to investigate maternal and infant sleep patterns in solo mother families compared to two-parent families; (2) to assess whether maternal emotional distress (i.e. anxiety and depressive symptoms) will be higher in solo mother families compared to two-parent families and (3) to examine whether family structure (solo mothers versus two-parent families) moderates the link between maternal emotional distress and between maternal and infant sleep problems.

Materials and methods: The study included 48 solo mother families and 48 two-parent families, with infants of 6 – 18 months age-old. Families were recruited mainly through announcements on social media, and childbirth preparation courses. All families were visited at home by a research assistant. Maternal and infant sleep was assessed using actigraphy and sleep logs for 5 days. In addition, mothers completed questionnaires related to maternal emotional distress.

Results: The results demonstrated that maternal sleep problems were elevated in solo mother families, as reflected in higher number of actigraphic night-wakings (t(94)=-2.29, p<.05) and lower self-reported sleep percentage (t(94)=2.17, p<.05). For infants, while there were no significant differences in actigraphic sleep measures, solo mothers reported more infant night-wakings (t(94)=-2.3, p<.05). Our findings indicated that solo mothers experience higher levels of emotional distress (depression: t(93)=2.71, p<.01; anxiety: t(94)=-3.91, p<.001), and that the associations between maternal emotional distress and actigraphic and reported sleep measures were moderated by family structure. For solo mothers, but not for married mothers, higher anxiety scores were associated with higher number of maternal actigraphic night-wakings (F(94)=3.80, p<.05) and with lower maternal reported sleep percent (F(94)=3.59, p<.05). Moreover, only for solo mothers, higher anxiety scores were associated with higher number of infant reported night-wakings (F(94)=5.52, p<.01). However, for married mothers, but not for solo mothers, higher emotional distress was associated with shorter maternal night duration (actigraphic and reported). For example, higher depression scores were associated with shorter actigraphic sleep duration (F(94)=4.12, p<.01).

Conclusions: These findings provide preliminary evidence that emphasizes the importance of considering family structure as a potential risk factor for sleep problems in mothers and possibly in infants as well. Our findings suggest that in this population of solo mother families, maternal emotional distress may be linked with maternal and infant sleep quality more strongly than in two-parent families. A deeper understanding of the specific characteristics of maternal and infant sleep in solo mother families, may lead to the development of sleep intervention programs that are especially designed to answer the needs of these families.
CONCOMITANT & LONGITUDINAL CHILDHOOD SLEEP CHARACTERISTICS ASSOCIATED WITH SUICIDAL THOUGHTS AT AGE 15

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Introduction: Suicidal thoughts have been linked to sleep problems during adolescence. There is limited knowledge related to the temporal association between sleep problems and subsequent adolescent suicidal thoughts. Our aims were 2-fold: 1) to determine which sleep problems are concomitantly associated with suicidal thoughts at age 15, and 2) to investigate whether childhood sleep characteristics predict suicidal thoughts at age 15.

Materials and methods: Longitudinal data of 1441 children were collected by questionnaires and interviews from 5 months to 15 years (13 waves). At 15 years, sleep characteristics (e.g., sleep need, average total sleep time, daytime drowsiness in class or while doing homeworks and trouble waking up) were assessed according to adolescent’s opinions. Suicidal thoughts in the past 12 months were assessed according to the Mental Health and Social Inadaptation Assessment for Adolescents (MIA). Childhood sleep characteristics (e.g., sleep latency, nighttime sleep, and nocturnal awakening duration) were completed according to maternal perception (2.5 months to 10 years). Group-based trajectory analyses were done according to each childhood sleep characteristic. Logistic regressions were performed to determine: 1) links between sleep problems and suicidal thoughts at 15 years, and 2) the predicting role of childhood sleep characteristics on the occurrence of suicidal thoughts at 15 years, after adjusting on different confounding variables.

Results: Around 23.2% of adolescents reported having suicidal thoughts in the past 12 months at age 15. We found a higher prevalence among girls compared to boys (31.1% Vs 14.7%, p<.001). At 15 years, adolescents with suicidal thoughts had more concomitant daytime drowsiness (p=0.03) and less average total sleep time (p=0.004). Children who followed a “long sleep latency trajectory” during childhood present significantly more suicidal thoughts at age 15 compared to children who followed a “short sleep latency trajectory” (35.5% Vs 20.1%), after adjusting on sex of the child, child age, child prematurity, low birth weight, internalizing/externalizing problems at age 1.5 and depression/anxious symptoms at age 15.

Conclusions: Daytime drowsiness and shorter average total sleep time are concomitantly related to suicidal thoughts at age 15. Importantly, a long childhood sleep latency predicted the emergence of suicidal thoughts at age 15, emphasizing the importance of addressing difficulty of sleep initiation in childhood.

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SLEEP AND BEHAVIOR PROBLEMS IN CHILDREN WITH EPILEPSY

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Introduction: Cross-cultural and regional research suggest that the prevalence rates of sleep problems in early childhood are high, with sleep problems more common and severe in children with epilepsy when compared to their healthy counterparts. Sleep disturbances in early childhood have been reported to predict later behavioral and psychiatric problems in children without medical conditions. However, no study has explored such a relation including prospective and objective sleep parameters in toddlers and preschool-age children with epilepsy.

Materials and methods: Sleep in 90 children with epilepsy was objectively assessed with a wrist actigraphy for 7 days and subjectively assessed with the parent-report Children’s Sleep Habits Questionnaire (CSHQ). Children’s behaviors were assessed through parental responses on the Child Behavior Checklist. Univariate and multivariate linear regression models were applied to investigate whether various aspects of sleep were associated with children’s internalizing and externalizing problems.

Results: Seventy-one (78.9%) children slept < 10 hours in a 24-hour period according to the actigraphy, with 75 (83.3%) children waking at least an hour during nocturnal sleep. Twenty-five (27.8%) children usually or sometimes had an inconsistent bedtime and 24 (26.7%) did not sleep the same amount each day. Twenty-nine (32.2%) and 18 (20.0%) children had an internalizing and externalizing problem in clinical range, respectively. Bedtime resistance, sleep anxiety, parasomnia, and elevated CSHQ total scores were significantly associated with increased internalizing and externalizing problems even after controlling for the child’s age, sex, seizure frequency, and the number of epileptic drugs used (p < 0.05).

Conclusions: Toddlers and preschool-age children with epilepsy experience both sleep and behavior problems and their problematic sleep is associated with behavioral difficulties. Findings suggest that screening of sleep and behavior problems as part of routine epilepsy care may be helpful to identify children with problematic sleep and unrecognized sleep disorders as well as those at risk of behavioral dysfunction.

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Introduction: Orofacial and pharyngeal muscles are involved in important functions including breathing, with the vital role of maintaining airflow. Because changes in these muscles can impair airflow, orofacial myofunctional therapy (OMT) is recommended to treat residual apnea after adenotonsillectomy (AT). AT is the first-line treatment for Obstructive Sleep Apnea (OSA). However, a literature review shows that there are no clear parameters for the indication of OMT neither outcomes measures. In the present study, we investigated whether Orofacial Myofunctional Evaluation with Scores protocol (OMES-protocol) is a valid measure of orofacial myofunctional disorder (OMD) in children with OSA.

Materials and methods: Validation study approved by the local institutional ethics committee (process number 1266/2017). Ninety-six children were evaluated, of which 52 were consecutive patients with a respiratory and snoring complaint. After the full-polysonomography, 36 children received the diagnosis of OSA (OAHI ≥ 1) and were included in the study (OSA group). Sixteen children with primary snoring were excluded. Among the children without respiratory complaint (n=44), 33 had nasal breathing and were included in the control group (C group).

The myofunctional assessment was performed using the OMES-protocol that comprise predetermined scores. OMES scores may be summarized in score by category appearance/posture, mobility or stomatognathic functions and, thus, in a total score that range of 36 to 104. The higher the OMES score, the better the orofacial functional status. Patient's scores were transformed in the percentage of the maximal score of each category and the total score of the protocol to determinate the myofunctional performance. A speech-language pathologist, previously trained and with good reliability performed all the evaluations.

The internal consistency of OMES-protocol was assessed by Cronbach's alpha coefficient. The non-parametric Mann-Whitney test was used for group comparisons (Criterion validity). Logistic regression analysis was used for to verify the probability of using OMES scores categories (%) to discriminate OSA group (1) of C group (0). The accuracy, sensitivity, and specificity values were determined by ROC curve analysis of OMES total score (%).

Results: The internal consistency of the OMES categories was adequate (Cronbach's α = 0.792). Criterion validity: Children with OSA had significantly lower scores for all categories and OMES total score than C group (P<0.0001).

Logistic regression showed that the percent of cases correctly classified by categories scores of OMES protocol was of 88.41%, (C group=31 cases: 90.91% and OSA group=31 cases: 86.11%). The ROC analysis showed that the OMES total score (%) was significantly different than the chance for the detection of the presence of OMD [AUC=0.983, P<0.001, CI:0.918-0.999], with the performance score of 84.627% as the cut-off point. The sensitivity and specificity values were, respectively, 88.89% (CI:73.9-96.9%) and 100% (CI:89.4-100.0%).

Conclusions: The OMES-protocol is valid to quantitative evaluation of OMD in children with OSA. It may be contributing to appropriately define parameters for the indication of OMT, as well as to verify the effects of the therapeutic strategies.
SCN8A MUTATION IN A CHILD PRESENTING WITH SEIZURES, SLEEP DISORDER AND DEVELOPMENTAL DELAY

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Introduction: Mutations in SCN8A are associated with cognitive impairment with or without cerebellar ataxia and with early infantile epileptic encephalopathy-13. Loss-of-function mutations can be associated with cerebellar ataxia and cognitive issues, whereas gain-of-function mutations can underlie epileptic encephalopathy. Abnormal sleep architecture has been described in animal models harboring SCN8A mutations.

Materials and methods: Clinical, laboratory and neuroimaging retrospective data review of a patient harboring mutation in the SCN8A gene identified through exome sequencing.

Results: The proband is a 4-yr-old female, born to non-consanguineous parents, after uneventful pregnancy and full term birth, presenting with “idiopathic” epilepsy (10 to 15 seizures per day) in her first month of life. An EEG showed multifocal epileptiform activity with a normal background and the MRI was normal. An extensive genetic and metabolic workup was negative. Whole exome sequencing (WES) was performed in order to unravel the cause of her disease and de novo pathogenic mutation in SCN8A gene was identified.

Conclusions: Our findings extend the spectrum of SCN8A mutations and the clinical features of patients with SCN8A mutations. Most affected patients manifested as refractory epilepsy and severe intellectual disability, only a small number of patients presented with milder clinical patterns. Additionally, sleep disorders were only described - so far - in animal studies. Our study confirmed that SCN8A mutations can lead to sleep disorder in some patients and treatment with melatonin can be helpful in its management.

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HURLER SYNDROME: SEVERE SLEEP APNEA AS INITIAL PRESENTATION IN A 10 MONTHS OLD CHILD.

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Introduction: The mucopolysaccharidoses (MPS) are a heterogeneous group of inherited metabolic disorders, each associated with a deficiency in one of the enzymes involved in glycosaminoglycan (GAG) catabolism. Over time, GAGs accumulate in cells and tissues, causing progressive damage, a variety of multi-organ clinical manifestations, and premature death. Sleep abnormalities have been described for all MPS disorders. They can be very debilitating and affect the quality of life of both patients and their families, not only at night, but also during daytime, causing excessive daytime fatigue, lack of sleep, affecting memory, cognition, behavior, and activity level.

Materials and methods: Clinical, laboratory and polysomnographic retrospective data review of a patient with MPS I (Hurler syndrome).

Results: Female patient, 10 months old, born to a young, healthy and non-consanguineous couple, after uneventful pregnancy, was referred to evaluation for investigation of respiratory and sleep problems. According to family, patient showed many episodes of apnea and snoring since 6 months of age that were getting worse. During the day, patient had some daytime sleepiness and could take several “naps”. Initial laboratory screening was normal but PSG studies showed severe sleep obstructive apnea. Since patient showed some coarse facial features and motor delay at 12 months, patient was seen by a clinical genetics team and a lysosomal storage disorder was suspected. Biochemical testing pointed out to increased GAG excretion in urine and enzyme assays for MPS disorders were performed. Patient showed very decreased activity of alpha-L-iduronidase, confirming the biochemical diagnosis of Hurler syndrome (MPS I). Specific therapeutic options in this disease are very few, being hematopoietic stem cell transplantation (HSCT) and enzyme replacement therapy (ERT) the main ones. Family declined HSCT and child was placed in compassionate program to early ERT access.

Conclusions: In MPS I patients with upper airway abnormalities, sleep-induced loss of airway muscle tone can lead to airway collapse and obstructive sleep apnea (OSA), causing loud and disruptive snoring. OSA is a frequent observation in Hurler patients, being initially associated with short recurrent events of absent or reduced ventilation due to upper airway obstruction, alternating with periods of increased ventilation between periods of obstruction. In later stages, Hurler patients may become unable to increase ventilation between periods of obstruction due to weakness or chest wall abnormalities like kyphoscoliosis; losing this compensation for the obstructive events leads to nocturnal hypoventilation. Long term follow-up with systematic screening for OSA is mandatory in all MPS I patients and should be performed on yearly basis and maybe earlier than 2 years of age since many Hurler patients can show severe symptoms of OSA. ERT reduces GAG storage and is associated with moderate improvements in some clinical manifestations and in particular respiratory function and physical capacity. Positive results have also been observed in clinical trials with ERT with regard to sleep apnea and hypopnea. Early treatment can improve the outcome of sleep disorders in MPS patients and help to prevent complications and irreversible complications of this devastating disease.

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P#08-Saturday
INITIATING AND TITRATING VENTILATION IN CHILDREN: DO DIFFERENT MODELS OF DELIVERY AFFECT ADHERENCE AND HEALTH ECONOMIC OUTCOMES?

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Introduction: Home mechanical ventilation is offered to increasing numbers of children¹. Due to increased service demand two models for ventilation initiation and titration (VIT) have developed in Southampton. This service evaluation explored model outcomes.

Materials and methods: Over eight months children underwent VIT either 1. By admission to a sleep laboratory with live adjustment of ventilator settings according to the American Academy of Sleep Medicine Titration Algorithm (Group 1) or 2. By ward admission with daily adjustment of ventilator settings according to unattended sleep study results from the previous night. (Group 2). All children underwent pre-admission mask orientation. Follow up at 3-6 months was by telephone, home and clinic visits to determine adherence defined by four hours of ventilator tolerance.² Statistical analysis was performed using SPSS.

Results: Thirty four admissions (17 boys, 17 girls) were evaluated. Median age (interquartile ranges (IQR)) for Group 1 (n=24) were 12.8 (8.2-15.2 years), and for group 2 (n=10) were 5.4 (2.4-16.2 years). Median (IQR) length of stay was significantly less for group 1 (1 night, 1-1) versus group 2 (3 nights 2-3) (p = 0.000). Follow up was available on 29 children (19 group 1, 10 group 2). There was no significant difference in adherence with respect to preset criteria (p = 1.000).

Conclusions: Single night admission with live adjustment of ventilator settings achieved significantly reduced lengths of stay with no impact on long term adherence to ventilation demonstrating health economic benefits.

AN ARTS-NEUROSCIENCE APPROACH TO ENGAGING WITH ADOLESCENTS ABOUT SLEEP AND SLEEPLESSNESS

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Introduction: Sleep disorders can be associated with impaired quality of life, depression and anxiety. Some disorders – such as insomnia – are more prevalent in older adults, and are partly conditioned by cultural and societal factors. Promoting awareness about the importance of sleep in adolescents and young adults could encourage good sleep hygiene, which may be protective against later sleep disturbance. We hypothesised that engagement with adolescents and young adults using an arts-neuroscience approach may particularly capture their attention. Therefore, we established an inter-disciplinary collaboration between clinical and basic neuroscientists, artists, and an expert on cultural aspects of sleeplessness, and piloted two engagement events.

Materials and methods: Art was created in three media - watercolour, leather, and a light installation - which referenced sleep electroencephalograph patterns. Photographs of these works were used during a patient-engagement art workshop held at the Macmillan Teenage Cancer unit. Adolescents were invited to respond artistically to the theme of sleep, and share their thoughts on the topic. The Teenage Cancer unit routinely offers art activities to patients, and this workshop was run according to standard practice and therefore did not require additional permissions. Patient participation in the art workshop was voluntary, and no demographic data was collected during this pilot study. A lecture about this event is available (https://www.youtube.com/watch?v=CRbAdSkqpmw).

The second event was a public-engagement evening event at the Grant Museum of Zoology in which sleep science was presented with a unique slant: how does sleep differ across the animal kingdom, from flies to humans?

Results: The adolescents who took part in the art workshop were often most enthused talking about and artistically representing their pre-sleep and post-sleep, i.e. late evening rituals and the beginning of the next day. They mentioned habits including listening to music, looking at the dark outside, spending time with pets, recurrent thoughts, sleep position, exercise, and the beginning of the school day. They also navigated their chemotherapy by making predictions about its effect on their sleep, e.g. based on their experience during their last chemotherapy treatment they were expecting to sleep more or less, or sleep badly the night before the first dose.

The event at the Museum of Zoology was well-attended (fully booked), was followed by many questions from the audience, and several attendees stayed behind to chat to the speakers.

Conclusions: An inter-disciplinary approach, in which sleep and sleeplessness are approached from multiple angles, can be successfully used to engage with adolescents and adults. Future longitudinal studies should investigate whether promoting awareness about the importance of sleep during adolescence could instil resilience towards later sleep disturbance.

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PROSPECTIVE EVALUATION OF PULSE TRANSIT TIME IN CHILDREN WITH SLEEP DISORDERED BREATHING


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Introduction: Pulse transit time (PTT) has been identified as a sensitive non-invasive measure of arousals and respiratory effort in children with sleep disordered breathing (SDB). However, its use is currently hampered by insufficient data to indicate what values of PTT are likely to be associated with SDB and by concern that the measurement may not discriminate sufficiently between disease states.

Materials and methods: We have prospectively evaluated the PTT values obtained on 267 children admitted for a multi-channel sleep study in a UK based district general hospital over a 2 year period. The sleep studies were carried out using Stowood Scientific Instruments Visi-3 sleep systems. Sleep study findings were categorised based on oximetry, audio and video findings as normal; primary snoring; upper airway resistance syndrome (UARS); obstructive sleep apnoea (OSA) and ‘abnormal other’. We have also categorised the findings using standard oximetry criteria. The oximetry and pulse transit time data (PTT arousals, respiratory swing) were evaluated by a sleep physiologist and artefactual data were excluded. Analysis was performed if there was a minimum of 4 hours (oximetry) and 3 hours (PTT) artefact free data. Findings were recorded in a sleep study database. Sleep study categories were determined by a clinician (MY) using oximetry, audio and video criteria, without reference to the PTT values. Receiver Operator Characteristic Curves (ROC) for different thresholds of PTT Swing were constructed for a random sample of 50% of the data (the ‘training set’), and the calculated thresholds of interest were validated against the other 50% (the ‘test set’).

Results:

<table>
<thead>
<tr>
<th>Categories</th>
<th>Number of children</th>
</tr>
</thead>
<tbody>
<tr>
<td>Normal</td>
<td>101</td>
</tr>
<tr>
<td>Borderline</td>
<td>119</td>
</tr>
<tr>
<td>Abnormal low risk</td>
<td>16</td>
</tr>
<tr>
<td>Abnormal high risk</td>
<td>20</td>
</tr>
</tbody>
</table>

We found a PTT respiratory swing value of 17.82 detected children with sleep disordered breathing (UARS and OSA) with a sensitivity of 0.83 and a specificity of 0.76. A respiratory swing value of 21.52 had a sensitivity of 0.56 and a specificity of 0.90. We replicated the ROC with the testing set, and the sensitivity and specificity findings were similar in the test data with thresholds of 17.84 and 21.50, thereby validating the role of PTT. Of the 119 children with borderline oximetry values, 83 were categorised as having a normal study or primary snoring based on oximetry, audio and video criteria; 64 of the 83 had a PTT respiratory swing below the threshold of 17.82. A further 36 children were categorised as having UARS and 28 of these had a PTT respiratory swing above the threshold of 17.82; χ² (p < 0.001).

VALIDITY OF ACTIGRAPHY COMPARED TO POLYSOMNOGRAPHY FOR THE STUDY OF SLEEP IN CHILDREN WITH AUTISM SPECTRUM DISORDER

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Introduction: Actigraphy (ACT) is a non-invasive objective assessment tool for the study of rest-activity cycles as a proxy to sleep-wake rhythms (approved by the American Sleep Disorders Association). It is of particular interest in the assessment of sleep in vulnerable populations for whom carrying out a polysomnography (PSG) is complicated. The majority of young children with Autism Spectrum Disorder (ASD), up to 80 percent, have sleep disturbances. It is important to evaluate sleep parameters in this population since sleep difficulties can have a considerable impact on both cognitive and behavioral functioning. As PSG recordings are not commonly available, or can be difficult to perform in children with ASD, ACT has become a tool of choice to assess sleep quality in children with ASD. However, to date, no study has investigated the validity of ACT compared to PSG, the consensus gold standard, for the measurement of sleep quality parameters in ASD children - which was the main objective of our study.

Materials and methods: During the same night of hospitalisation, PSG and ACT were conducted in 21 children (5 girls and 16 boys; mean age 4.8 years ± 1.4) diagnosed with ASD according to DSM-5 criteria and standardised diagnostic scales (ADI-R and ADOS). Sleep parameters were total sleep time (TST), sleep latency (SL), wake after sleep onset (WASO) and sleep efficiency (SE) defined as TST divided by time in bed. To compare PSG and ACT, recommended agreement analysis methods were conducted, including Intraclass Correlation Coefficient (ICC), Bland-Altman plots and tests of equivalence. For the latter, the clinically significant range of acceptability was set to ±30 min for TST, ±15min for SL, ±15min for WASO and 5% for SE.

Results: Compared to PSG, ACT overestimated TST by 17.8 min (106.4 SD), and SE by 0.6 % (10.8 SD). On the contrary, ACT underestimated SL by 4.6 min (15.0 SD) and WASO by 4.4 min (42.1 SD). According to the tests of equivalence (significant p-values indicate equivalence), the difference between ACT and PSG measures was clinically acceptable for: TST (p=0.022), SL (p<0.001) and SE (p=0.048), but not for WASO (p=0.19), due to an important inter-individual variance. There was a good correlation between methods for SL (ICC=0.89) and WASO (ICC=0.87), and a fair correlation for TST (ICC=0.53) and SE (ICC=0.50).

Conclusions: Results suggest that ACT shows fair to strong intraclass correlation with PSG in children with ASD. Tests of equivalence estimated that the differences between methods were clinically acceptable for most sleep parameters (TST, SL, SE) but not for WASO. Those results strongly support the use of actigraphy as a non-invasive objective method to assess sleep quality, which is particularly interesting in this population where PSG can be difficult to carry-out.
P#60-Sunday

INTERVENTION FOR CHILDREN’S SLEEP HABITS USING AN INTERACTIVE SMARTPHONE APPLICATION


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Introduction: Recent studies have revealed that sleep problems in young children irreversibly affect brain development. In addition, children’s sleep problems increase the parenting stress of their caregivers. Furthermore, problems and/or limitations of traditional face-to-face sleep educational activities occur because it is not easy to deliver the appropriate sleep health literacy to the caregivers who really need such a guidance. Given a background of shorter sleep and later bedtime in Japanese children, there is an urgent need to develop an intervention method for infants’ sleep problems that fits current situations and Japanese culture. We developed a smartphone application for interaction between caregivers and pediatric sleep experts to improve infants’ sleep habits in Japan. The aim of this study was to present its functions, case examples, and user feedback. Consideration of its possible usage in the field of sleep medicine and sleep literacy education for caregivers of children is also discussed.

Materials and methods: In this application, sleep health literacy education was primarily delivered through animation. The caregivers inputted the sleep habits of their infants and themselves for 8 days of each month. Pediatric sleep experts analyzed the information entered by the caregivers and sent multiple suggestions to each caregiver, of which the caregiver selected one to try. Ten caregiver-infant (1-2 years old) pairs used the application for a trial period of 2 months with actigraphy connected to the smartphone via Bluetooth for checking compliance and evaluating the comfort level of the users.

Results: After the first intervention, the nighttime sleep hours of infants were extended in the weekends. In addition, in most cases, caregivers reported behavioral improvement in themselves as well as their children. The caregivers reported improvement in the ease of parenting after the intervention.

Conclusions: The trial use of this application showed its effectiveness for improving the sleep habits of Japanese infants. Furthermore, the application would be effective for alleviating the parenting burden of Japanese caregivers. We report only several cases here, and further study is needed to include more participants.

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Correlates of Nocturnal Sleep Duration, Nocturnal Sleep Variability and Nocturnal Sleep Problems in Toddlers: Results from the GET UP! Study

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Introduction: Sleep in early years is important for health and development. However, correlates of nocturnal sleep characteristics in young children are not fully understood. The present study aims to explore the correlates of nocturnal sleep duration, nocturnal sleep variability and nocturnal sleep problems in a sample of Australian toddlers.

Materials and methods: Participants were 173 toddlers (average age 19.7±4.1 months) from the GET UP! Study. Nocturnal sleep duration, nocturnal sleep variability, naps and physical activity were measured using 24-h accelerometry (Actigraph GT3X+) over 7 consecutive days. Nocturnal sleep problems were assessed using the Tayside Children’s Sleep Questionnaire. Screen time was reported by the parents. Multiple logistic regression models were used to examine the association between potential correlates (i.e., age, sex, socio-economic status, body mass index, physical activity, screen time, nap(s), bedtimes, and wake-up times) and nocturnal sleep characteristics.

Results: Older children were more likely to have greater sleep variability (OR: 1.97; 95%CI: 1.08-3.61). Less physical activity (OR: 2.38; 95%CI: 1.27-4.45), shorter nap(s) (OR: 2.42, 95%CI: 1.29-4.55) and later wake-up times (OR: 4.42; 95%CI: 2.32-8.42) were associated with higher odds of having longer nocturnal sleep duration. Late bedtimes were associated with shorter nocturnal sleep duration (OR: 0.09; 95%CI: 0.04-0.18) and with greater nocturnal sleep variability (OR: 1.97; 95%CI: 1.06-3.68). None of the potential correlates were associated with nocturnal sleep problems.

Conclusions: The present study identifies several correlates of nocturnal sleep duration (total physical activity, nap, bedtime and wake-up time) and nocturnal sleep variability (age and bedtime), whereas no correlates were identified for nocturnal sleep problems. The association between late bedtime and unfavourable nocturnal sleep outcomes suggests this may be a modifiable target for future sleep intervention in early childhood.

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P#61-Saturday

AN EXPLORATORY STUDY OF PULSE OXYGEN SATURATION MONITORING IN GENERAL HOSPITALIZATION CHILDREN

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Introduction: To explore the clinical characteristics of pulse oxygen saturation monitoring in general hospitalized children, and to provide reference for further clinical study.

Materials and methods: We enrolled the children hospitalized in Shanghai Children's Hospital affiliated to Shanghai Jiao Tong University as the research object. From July 2017 to December 2017, who were received the dynamic monitoring of pulse oximetry for their children. An analysis was conducted for the relationships of blood oxygen saturation (SpO2) with age, disease type and body mass index (BMI).

Results: A total of 75 children were included in this study. The monitoring results of SpO2 showed that gender had no effect on SpO2 value, but there were notable differences in the maximum, minimum, extreme value, standard deviation and coefficient of variation of SpO2 value in different age groups. The maximum and the average value of SpO2 increase with the increase of BMI value (p<0.001), and the extreme value, standard deviation and coefficient of variation of SpO2 decrease with the increase of BMI value (p<0.05). The maximum, the extreme value, the standard deviation and the coefficient of variation of SpO2 increase with the increase of age (p<0.001), and the minimum and average value of SpO2 decrease with age (p<0.001).

Conclusions: Pulse oximetry monitoring for general hospitalized children has certain clinical significance, it is necessary to carry out further prospective studies to determine its clinical value.