

**Journal of Population Therapeutics
and Clinical Pharmacology**

INCORPORATING FETAL ALCOHOL RESEARCH

**Journal de la thérapeutique des populations
et de la pharmacologie clinique**

Official journal of the FACE Research Network

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2011 FACE POSTER COMPETITION ABSTRACTS

12TH ANNUAL FETAL ALCOHOL CANADIAN EXPERTISE (FACE) RESEARCH ROUNDTABLE

September 13, 2011
Prince Edward Island

The 12th FACE Research Roundtable was organized by the Motherisk Program of The Hospital for Sick Children and sponsored by the Brewers Association of Canada.

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2011 FACE RESEARCH ROUNDTABLE POSTER COMPETITION ABSTRACTS

1

Social perspective taking and social affective processing in children with Fetal Alcohol Spectrum Disorders

Stevens SA^{1,2}, Nash K^{2,3}, Koren G^{4,5}, Rovet J^{1,2,4}

¹Department of Psychology, University of Toronto; ²Neuroscience and Mental Health Program, The Hospital for Sick Children; ³Ontario Institute for Studies in Education of the University of Toronto; ⁴Department of Pediatrics, University of Toronto; ⁵Motherisk Program, The Hospital for Sick Children, Toronto, Ontario

Background: Children with Fetal Alcohol Spectrum Disorders (FASD) show severe behaviour and cognitive impairments. These children also display striking difficulties in adaptive and social functioning, which extend beyond their cognitive delays. However, the factors that contribute to these social impairments are not well understood. The objective of the present study was to investigate two areas of social cognition, namely social perspective taking and affect processing as potential contributing factors to the cognitive, behaviour and social impairments observed in FASD.

Methods: Studied at The Hospital for Sick Children were 42 children between 8 to 12 years of age; 25 had a diagnosis of a FASD (mean age 10.35) and 17 were typically developing control (TDC) children (mean age 10.25). The Theory of Mind and Affect Recognition subtests from the NEPSY-II (Korkman, Kirk & Kemp, 2009) were used to investigate social perspective taking and affect processing, respectively.

Results: Children with FASD had poorer social perspective taking skills compared to TDC, as observed by their lower percentiles on the Theory of Mind subtest. The FASD group also had poorer affect processing compared to the TDC group. Of note, the mean score on the Affect Recognition subtest for the FASD group remained in the average range, whereas the mean score for TDC approached the high average range. The FASD group, however, made more errors on happy, neutral and angry facial expressions.

Conclusions: Children with FASD displayed reduced social perspective taking and affect processing

abilities, compared to TDC. These social cognitive weaknesses may contribute to the poor behaviour regulation observed in children with FASD, and significantly impacts their daily social functioning.

Keywords: *Social cognition, theory of mind, affect processing*

Source of funding for the study: CFFAR; CIHR

Conflict of interest: None

Student/Trainee: Full-time PhD student

Corresponding author: sara.stevens4@gmail.com

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Evaluation of the neurobehavioral screening tool in children with Fetal Alcohol Spectrum Disorder (FASD) and Prenatal Alcohol Exposure (PAE)

La France M¹, Nash K², Koren G³, Andrew G⁴, Rasmussen C¹

¹Department of Pediatrics, University of Alberta; ²Psychology Department, The Hospital for Sick Children, Toronto, Ontario; ³Motherisk Program, The Hospital for Sick Children, Toronto, Ontario; ⁴Department of Pediatrics, University of Toronto; ⁴Glenrose Rehabilitation Hospital, Alberta

Background: The Neurobehavioral Screening Tool (NST) is a part of the National Screening Tool Kit for FASD. The NST is a parental rating scale consisting of ten questions identified by Nash et al. (2006) to be predictive of FASD based on the child's current behaviour. The purpose of this study was to field test the NST to determine how well it differentiates between children diagnosed with FASD, those with PAE but no FASD diagnosis, and typically developing controls.

Methods: We administered the NST to three groups of children (6 to 17 years): 36 with FASD and 21 PAE assessed at the Glenrose Hospital FASD clinic in Edmonton, and 21 controls recruited locally.

Results: Significantly more children with FASD screened positive on the NST compared to the PAE and the control group. The sensitivity of the NST in our

sample was 36.1%. Within the FASD group, there was a higher sensitivity among older participants (50%) 12-17 years, than younger participants (22%) 6-11 years. The NST had 100% specificity against the control group; none of the controls tested positively.

Discussion: Despite overall low sensitivity, the specificity in our sample is very high indicating that the selected items are specific to FASD. This supports the use of the NST as a national screening tool which would lead to large numbers of children being screened and undergoing the process of diagnosis.

Keywords: Neurobehavioral screening tool, fetal alcohol spectrum disorder, prenatal alcohol exposure

Source of funding: The Public Health Agency of Canada

Conflict of interest: None

Student/Trainee: Trainee

Corresponding author: mtl5@ualberta.ca

3 The patterns of sleep disorders and circadian rhythm disruptions in children and adolescents with Fetal Alcohol Spectrum Disorders

Goril S^{1,2}, Shapiro CM^{1,2,3}

¹Youthdale Child and Adolescent Sleep Centre, Toronto, Ontario; ²Collaborative Program in Neurosciences, University of Toronto, Toronto, Ontario; ³Department of Neuropsychiatry, Toronto Western Hospital, Toronto, Ontario

Background: Sleep disorders have been poorly described in children and adolescents diagnosed with Fetal Alcohol Spectrum Disorders (FASD). The objective of this study is to describe the sleep and circadian rhythm characteristics of children with FASD using overnight polysomnography, sleep questionnaires, and the Dim Light Melatonin Onset (DLMO) test. To our knowledge, no comprehensive studies of this nature have been conducted.

Methods: Thirty six children aged 6-18 years diagnosed with FASD were recruited from various FASD clinics to the Youthdale Child and Adolescent Sleep Centre in Toronto. After a medical consultation, each participant had one night of overnight polysomnography, as well as an additional night of DLMO. Participants completed various sleep and FASD questionnaires. Data was analyzed using SPSS 19.

Results: Significant differences were found when comparing the sleep architecture of FASD participants to normative data. There was a high prevalence of

sleep disorders in this sample. Most of the melatonin profiles of the FASD participants were found to be abnormal, suggesting HPA axis disturbances. The melatonin results are congruous with the brain pathology in studies of animals prenatally exposed to alcohol.

Conclusion: The high prevalence of sleep disorders and melatonin abnormalities in this population warrants sleep assessments, as part of and/or in conjunction with the diagnostic process.

Keywords: Fetal alcohol spectrum disorders, sleep disorders, circadian rhythm

Source of funding: Youthdale Foundation

Conflict of interest: None

Student/Trainee: Full-time

Corresponding author: s.goril@utoronto.ca

4 Executive functioning predicts adaptive functioning in children with FASD and PAE

Denys K¹, Zwaigenbaum L¹, Andrew G¹, Tough S², Rasmussen C¹

¹Department of Pediatrics, University of Alberta; ²Department of Pediatrics and Department of Community Health Sciences, University of Calgary

Background/Objectives: Adaptive functioning (AF) deficits are common in individuals with FASD. Although IQ is predictive of AF in other populations (Liss et al., 2001)¹, this is not the case in FASD. Schonfeld and colleagues (2006)² found that executive functioning (EF) predicted social skills in individuals with FASD, however, there are no studies linking EF with AF in FASD.

Methods: Participants (aged 6 - 16) were in two groups, FASD (N=41) and prenatal alcohol exposure, not meeting criteria for FASD (PAE; N=26). Parents rated their child's EF and AF, using the Behavioral Rating Inventory of Executive Function (BRIEF) and the Adaptive Behavior Assessment System Second Edition (ABAS-II), respectively. We also examined whether IQ, age of assessment, SES, and home stability were related to AF.

Results: Based on one-way ANOVAs, the PAE group performed significantly better than the FASD group on all of the ABAS composites, the BRIEF Metacognitive Index (MI), and approached significance on the BRIEF Global Executive Composite. Stepwise regression analyses revealed that the BRIEF MI was the best predictor of the General Adaptive, Conceptual, and

Practical ABAS-II composites, but nothing predicted the Social composite. IQ only predicted scores on the Conceptual composite.

Conclusions/Discussion: We found EF to be a better predictor of AF than IQ and other environmental factors. This has implications for policy and services: many disability services are not available to individuals with FASD and PAE because they have IQs above 70, whereas EF measures may be more informative for severe impairments in AF.

Keywords: *Executive functioning, adaptive functioning, IQ, FASD, PAE*

References:

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- 2) Schonfeld AM, Paley B, Frankel F, O'Connor MJ. Executive functioning predicts social skills following prenatal alcohol exposure. *Child Neuropsychology* 2006;12:439-452.

Sources of funding for the study: CIHR Operating Grant

Conflict of interest: None

Student/Trainee: Full-time MSc student

Corresponding author: kennedydenys@gmail.com

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Ecogenetic and epigenetic influences in FASD

Kapalanga J^{1,2,3,4}, Wong D^{1,2}, Gandy A^{1,2}, Avind Singh A², Bhoka G⁵

¹Pediatrics, Dalhousie University, Halifax, Nova Scotia;

²Summerside Medical Center, Summerside, PEI; ³Grey Bruce Health Services, Owen Sound, Ontario;

⁴Cambridge Memorial Hospital, Cambridge, Ontario;

⁵Obongi Hospital, W Moyo, Uganda

Background/Objectives: Alcohol is globally consumed, and its long term adverse effects on fetal development are well characterized. However, *in utero* alcohol exposure does not always result in Fetal Alcohol Spectrum Disorders (FASD). This study explores the role of ecogenetic and epigenetic factors in FASD. We hypothesize that ecogenetic and epigenetic factors determine susceptibility to FASD.

Methods: To test this hypothesis we examined the prevalence of FASD between two disparate population groups; one in Prince Edward Island (PEI), Canada and

the other in Moyo, Northern Uganda. Moyo was chosen because alcohol consumption in the district is especially widespread and yet alcohol-related behavioral and academic issues are rare. In PEI, FASD is a significant public health concern. Both Moyo and PEI are remote regions with stable populations of comparable size. Children aged 2 years to 10 years exposed *in utero* to alcohol as determined by maternal history were assessed by a dysmorphologist/paediatrician for clinical features of FASD.

Results: Fifty seven (57/87 or 65.5%) Moyo mothers and 49 (49/89 or 55%) PEI mothers admitted to consuming alcohol during pregnancy. Based on maternal history a total of 65 Moyo children and 52 PEI children were exposed *in utero* to alcohol. Twenty-six or 40% Moyo children and 42 or 80% PEI children were confirmed to have FASD.

Conclusions/Discussions: PEI children exposed *in utero* to alcohol are significantly more likely to have FASD than Moyo children ($p < 0.01$). Differences in ecogenetic and epigenetic factors are postulated to be the explanation for the two-fold difference in FASD prevalence between Moyo and PEI children. These findings are preliminary and warrant further and larger studies that analyze ecogenetic and epigenetic factors that could influence risk susceptibility to FASD.

Keywords: *Ecogenetics, epigenetics, risk susceptibility, clinic assessment*

Source of funding: None

Conflict of interest: None

Corresponding author: jkapalanmdphd@pol.net

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Available interventions and services used by Canadian adolescents with FASD and their families

Todorow M^{1,2}, Moore TE², Fantus E¹, Sorbara D¹, Nulman I¹

¹Hospital for Sick Children, Division of Clinical Pharmacology and Toxicology, University of Toronto, Toronto, Ontario; ²York University, Department of Psychology, Toronto, Ontario

Background/Objectives: FASD is a leading cause of developmental disability; however, little research has focused on management post-diagnosis. This study aims to describe the availability and usage of interventions and services by Canadian adolescents with FASD and their families.

Methods: This observational, follow-up study included

44 adolescents between 11.0 and 18.9 years of age (31 males, 13 females), diagnosed with a FASD at the Motherisk Clinic at The Hospital for Sick Children in Toronto, Canada. Quantitative and qualitative data were collected via semi-structured telephone interviews with primary caregivers. Qualitative data were analyzed using thematic analysis.

Results: Psychopharmacotherapy was the most common type of intervention utilized by adolescents (68%). Commonly used medications included stimulant/non-stimulant attention medications (52%), antipsychotics (43%) and antidepressants (16%). Thirty-two percent of adolescents were receiving individual counselling/therapy and 7% were participating in group interventions. Only 14% of adolescents had ever been referred to or received any FASD-specific interventions/services. Forty-three percent of caregivers had received FASD training and 16% attended caregiver support groups. The mean caregiver rating of accessibility of interventions/services for adolescents was 1.85 ($SD = 1.14$) on a numerical scale of 1 to 5 (1 = very difficult to get, 5 = very easy to get). The most common categories of interventions/services reported by caregivers as needed, yet difficult to obtain or unavailable, were FASD-specific interventions/services and social skills interventions.

Discussion/Conclusions: The majority of Canadian adolescents with FASD are not receiving interventions, other than psychopharmacotherapy. Despite significant advancements in the diagnosis of FASD, little progress has been made in supporting these individuals post-diagnosis.

Keywords: FASD, adolescents, observational

Source of funding for the study: CIHR and Glendon College

Conflict of interest: None

Student/Trainee: Full-time

Corresponding author: michelle.todorow@sickkids.ca

7

How to approach sleep problems in children with FASD: The first Canadian FASD & sleep consensus paper

Ipsiroglu OS¹, Andrew G², Carmichael-Olson H³, Chen M⁴, Collet JP⁵, Pei J⁶, Garden J¹, Hanlon-Dearman A⁷, Houben R⁸, Jan JE¹, Keivers K⁹, McNaughton D¹⁰, Looock C¹¹, Vitale-Cox L¹², Yo W¹³, Veer D¹, Weinberg J¹³, Witmans M¹⁴

¹FASD & Sleep Research Group BCCH/SHHCC, University of British Columbia, Vancouver; ²FASD

Clinical Services at Glenrose Rehabilitation Hospital, University of Alberta, Edmonton; ³Psychiatry and Behavioral Sciences, University of Washington, Seattle; ⁴University of Washington School of Medicine, Seattle; ⁵Child and Family Research Institute, British Columbia Children's Hospital, Vancouver; ⁶Canada NorthWest FASD Research Network/Intervention Team/ Educational Psychology, University of Alberta, Edmonton; ⁷Manitoba FASD Centre/ Children's Hospital of Winnipeg, Winnipeg; ⁸Health2Media, Vancouver; ⁹University of the Fraser Valley, Abbotsford; ¹⁰Social Work, Children's & Women's Health Centre of British Columbia, Vancouver; ¹¹British Columbia Children's Hospital, Sunny Hill Health Center for Children, Vancouver; ¹²Elispogtog First Nation Education Division EHCW Eastern Door Diagnostic Team New Brunswick, New Brunswick; ¹³Department of Cellular and Physiological Sciences, University of British Columbia, Vancouver; ¹⁴Stollery Children's Hospital, University of Alberta, Edmonton

Background: Although up to 85% of children with FASD experience sleep problems (SP) which significantly impact functioning and quality-of-life (QoL), SP are rarely addressed by health care providers (HCP). In order to develop a framework for a structured approach to SP and to determine skills and knowledge necessary to diagnose and treat children with SP and/or disorders, an interdisciplinary consensus meeting was organized prior to the 4th International FASD Conference in Vancouver (2011).

Methods: Existing published/submitted clinical research exploring the problem and trends of medical treatment (medication) were reviewed. Based on the Canadian Health Care Systems' universal services philosophy, a 3-Level-Curriculum was proposed and the needs for future research and knowledge dissemination were discussed as a guideline for screening/assessing SP.

Results: Level I includes screening with a focus on behaviour, day- and night time situations and a simple measure for quality-of-life. All HCPs should be "empowered to close the screening based sleep-service-related gaps at the level of care they offer". Level II includes assessments by HCPs (e.g. occupational/behavioural therapist or community paediatrician) using sleep-logs/-diaries as clinical monitoring/evaluation tools and validated sleep questionnaires, thus requires a formal training. Level III represents regional health care services and is the highest level of the curriculum in regards to structured knowledge dissemination.

Discussion: Knowledge dissemination is needed to enable this proposed approach and will be provided

through the collaboration of the Consensus Group with the Intervention Team of the Canada Northwest FASD Research Network. Evaluation of activities will guide and coordinate future research needs.

Keywords: *Sleep problems, 3-Level-Curriculum, knowledge dissemination*

Source of funding: Victoria Foundation FASD-Action-Fund

Conflict of interest: None

Corresponding author: oipsiroglu@cw.bc.ca

8

Restless Leg Syndrome (RLS) in children and youth with FASD and Prenatal Substance Exposure (PSE) – a clinically missed diagnosis aggravating the challenging behaviour?

Ipsiroglu OS¹, Black A², Garden J¹, Jan JE¹

¹FASD & Sleep Research Group, BCCH/SHHCC, University of British Columbia, Vancouver; ²Shriner's Gait Lab, Sunny Hill Health Centre for Children, Vancouver

Background: RLS can cause sleep problems (SP), and is a “disorder characterized by disagreeable leg sensations that usually occur prior to sleep onset and that cause an almost irresistible urge to move the legs.” Sleep related day- and night-time symptoms may not be recognized, or are missed in children with FASD/PSE, as FASD/PSE is usually associated with challenging behaviour including daytime hyperactivity and bedtime resistance. Results of clinical sleep assessments suggest that optimizing our understanding before triaging patients for further diagnostic/therapeutic care would be helpful.

Methods: We initially used an ethnographic approach adapted from medical anthropology to explore parent(s)/caregiver(s) perceptions of “challenging behaviour” and SP; then we developed and piloted home-based over-night-video-sleep-studies with which we could describe observable and reproducible SP; out of the video-reports we developed standardized descriptions.

Results: We are describing day- and night-time related RLS related clinical symptoms in 27 patients with FASD/PSE: “tossing and turning around”; “messy bed”; “fighting sleep”; “restless sleep” etc; 23/27 patients had insomnia, 22/27 parasomnias, 22/27 had symptoms strongly suggestive of RLS; 12 cases were diagnosed, 10 cases are re-evaluated.

Discussion: RLS related discomfort/urge-to-move/pain seems to be a main cause of SP. Children may develop movement based abilities and/or extreme adaptive movement strategies (trampoline jumping until collapsing) to overcome difficulties falling asleep, which are interpreted as part of their challenging behaviour. History and analysis of behavioural patterns *in conjunction* seems to be a key in patients with expressive language difficulties. Our observations open a new causality related diagnostic/therapeutic care option.

Keywords: *RLS, challenging behaviour, sleep*

Source of funding: Victoria Foundation FASD-Action-Fund

Conflict of interest: None

Corresponding author: oipsiroglu@cw.bc.ca

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Insomnia & Restless Legs Syndrome (RLS) in chronic care management of children and adolescents with neurodevelopmental conditions. An Applied Medical Anthropology and Developmental Pediatrics (AMA/DP) course for undergraduates

Ipsiroglu OS¹, McGuire M², McKellin W³

¹FASD & Sleep Research Group BCCH/SHHCC, University of British Columbia, Vancouver; ²FASD & Sleep Research Group BCCH/SHHCC, University of British Columbia, Vancouver/Department of Anthropology, University of British Columbia, Vancouver; ³Department of Anthropology, University of British Columbia, Vancouver

Background: The purpose of this course is to introduce undergraduate students to the concepts of AMA/DP in chronic care management of children with neurodevelopmental conditions (NDC), such as FASD. Special attention was given to the topics of insomnia due to RLS in MA/DP research context and to analytic matters as the nature of description, conceptualization, generalization and content analysis.

Methods: Selected methods from AMA/DP were applied to observe, describe, and interpret the medical phenomena of RLS in cultural and medical context and social organization in children and youth with NDC. The course covered participant observation, disease and life histories, narratives, ethnographic semantics, information gathering techniques, with a focus on narratives, videos, and questionnaires.

Results: Undergraduate students were able to develop 1) new perspectives to NDC and/or to patients who have different life experiences, and understand how to interpret and make use of obtained qualitative and quantitative information, and 2) their own focused research questions, and try new explorative research tools in their own areas of expertise (e.g. nutritional sciences, pharmacology, kinesiology, ...) presented at www.chroniccare4sleep.org.

Discussion: Sleep spans across various organ-specific conditions and as a theme has involved most disciplines and natural sciences, including humanities and arts, but in medicine the content has been 'medicalized' from subspecialty viewpoints. In applied AMA/DP the focus is more on the contextual relationship of the biological experience *sleep* with the concept of medicalization, along with cultural and social phenomena helping individuals broaden their understanding and overcome personal/professional prejudices.

Keywords: *Undergraduate training, applied medical anthropology, developmental paediatrics*

Source of funding: Victoria Foundation FASD-Action-Fund

Conflict of interest: None

Corresponding author: oipsiroglu@cw.bc.ca

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The bridge the gaps approach. Web-based help for parents and health care professionals (HCP) for sleep problems in children & adolescents with FASD and other neurodevelopmental conditions

Ipsiroglu OS¹, Pei J², McGuire M³, Houben R⁴, Loock C⁵

¹FASD & Sleep Research Group BCCH/SHHCC, University of British Columbia, Vancouver; ²Canada NorthWest FASD Research Network/Intervention Team/Educational Psychology, University of Alberta, Edmonton; ³FASD & Sleep Research Group BCCH/SHHCC, University of British Columbia, Vancouver/Department of Anthropology, University of British Columbia, Vancouver; ⁴Health2Media, Vancouver; ⁵British Columbia Children's Hospital, Sunny Hill Health Center for Children, Vancouver

Background: Individuals with FASD and other neurodevelopmental conditions are at high-risk for developing sleep problems (SP). In consequence the sleep deprivation leads to additional co-morbidity, e.g. hyperactive behaviour, and further

physical/cognitive/emotional impairments. Our research/clinical experience has shown that clinical symptoms and behaviour are often not recognized as sleep related, and that parents/caregivers' reports about SP are not given appropriate attention by HCPs. Children are often medicated with psychotropic substances without critical evaluation of the benefit to potential harm.

Methods: The website www.chroniccare4sleep.org is a portal and an information exchange source for SP experienced by children and adolescents with neurodevelopmental conditions; the FASD-pages are the first to go public (September 10, 2011). Published literature reviews, developed and existing screening questionnaires can be downloaded for parents'/HCPs' use. Presented information has been peer reviewed and/or is based on own experience accepted as "standard-of-care".

Results: We predict that providing parents/caregivers and HCP with different levels of information will help to bridge the current situation of neglect in regards to SP and give us valuable information about needs/requirements. The website-counter and received feedback enables us to monitor interest, acceptance and improve presentations on an ongoing-basis.

Discussion: Besides anxiety, behaviour, airway-obstructions, discomfort/pain, RLS and other organic causes paired with socio-cultural factors can contribute to SP. We believe that directly enabling target groups to focus more specifically on SP and use a shared language will help to gain new information regarding the causes of challenging behaviour, mainstream medication trends and SP.

Keywords: *www.chroniccare4sleep.org, medication, sleep problems*

Source of funding: Victoria Foundation FASD-Action-Fund

Conflict of interest: None

Corresponding author: oipsiroglu@cw.bc.ca

11

Body and head movements during sleep and wake episodes in a rat model of FASD: Pilot for proving the video monitoring concept

Hung A¹, Chan F¹, Yo W², Ipsiroglu OS¹, Weinberg J²

¹FASD & Sleep Research Group BCCH/SHHCC, University of British Columbia, Vancouver; ²Department of Cellular and Physiological Sciences, University of British Columbia, Vancouver

Background: Based on our clinical observation that Restless Legs Syndrome (RLS, a neurologic sensorimotor disorder characterized by a range of sensations from discomfort to pain) is a major cause of insomnia in children and adolescents with FASD, we hypothesized that rat offspring with prenatal alcohol exposure (PAE) may exhibit altered movements during sleep compared to those in control animals. To investigate this, we first had to test the applicability of our overnight-video-sleep-studies system for monitoring activities/sleep of rats.

Methods: 2 control, 2 PAE male Sprague Dawley rats were videotaped in specially equipped cages for a total of 8 hrs during the lights on period. Equipment included infrared-light camera, netbook with synchronized audio/video software, and live time-stamp, constant frame-rates.

Results: Awake/asleep states, defined as open/closed eyes and responsive/unresponsiveness were identified. Overall, the following were found: awake: 146/140 min, asleep: 267/278 min, transitioning between awake/sleep: 64/63 min. We were able to observe behavioural patterns including prolonged grooming/head shivering activities before falling asleep, arousals and some active limb movements while asleep, which appeared to differ between PAE and control animals. We were not able to count leg movements reliably as sleeping position hindered observation of lower/upper limbs.

Discussion: The video methodology opens a new option for behavioural-sleep-observations in the rat model. While head and body movement can clearly be described, the study must be repeated with a larger sample size, and the relationship of these observations to RLS must still be demonstrated.

Keywords: *Rat model of FASD, sleep, behavioural observations*

Source of funding: Victoria Foundation FASD-Action-Fund, NIH/NIAAA R37AA007789

Student/Trainee: Trainee

Conflict of interest: None

Corresponding author: oipsiroglu@cw.bc.ca