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PRS GLOBAL OPEN

ORIGINAL ARTICLE
Pediatric/Craniofacial

Improvement in Sleep Architecture is associated with the Indication of Surgery in Syndromic Craniosynostosis

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Background: Children with syndromic craniosynostosis (sCS) often suffer from obstructive sleep apnea (OSA) and intracranial hypertension (ICH). Both OSA and ICH might disrupt sleep architecture. However, it is unclear how surgically treating OSA or ICH affects sleep architecture. The aim of this study was twofold: to explore the usefulness of sleep architecture analysis in detecting disturbed sleep and to determine whether surgical treatment can improve it.

Methods: Eighty-three children with sCS and 35 control subjects, who had undergone a polysomnography (PSG), were included. Linear-mixed models showed the effects of OSA and ICH on sleep architecture parameters. In a subset of 19 patients, linear regression models illustrated the effects of OSA-indicated and ICH-indicated surgery on pre-to-postoperative changes.

Results: An increase in obstructive-apnea/hypopnea index (oAHI) was significantly associated with an increase in N2-sleep, arousal index, and respiratory-arousal index and a decrease in REM-sleep, N3-sleep, sleep efficiency, and sleep quality. ICH and having sCS were not related to any change in sleep architecture. OSA-indicated surgery significantly increased the total sleep time and sleep efficiency and decreased the arousal index and respiratory-arousal index. ICH-indicated surgery significantly decreased REM-sleep, N1-sleep, sleep efficiency, and sleep quality.

Conclusions: For routine detection of disturbed sleep in individual subjects, PSG-assessed sleep architecture is currently not useful. OSA does disrupt sleep architecture, but ICH does not. OSA-indicated surgery improves sleep architecture, which stresses the importance of treating OSA to assure adequate sleep. ICH-indicated surgery affects sleep architecture, although it is not clear whether this is a positive or negative effect. *(Plast Reconstr Surg Glob Open 2019;7:e2419; doi: 10.1097/GOX.0000000000002419; Published online 10 September 2019.)*

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Presented at the International Society for Craniofacial Surgery meeting 2017, Cancun, Mexico.
Received for publication June 3, 2019; accepted July 1, 2019.
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DOI: 10.1097/GOX.0000000000002419

INTRODUCTION

Many children with syndromic craniosynostosis (sCS) are at risk of abnormally disturbed sleep. For example, there is the problem of upper airway obstruction (UAO), potentially of multiple etiologies, caused by hypoplasia of several facial structures[1]. In Apert or Crouzon syndrome, during early life, these abnormalities translate in to the problem of obstructive sleep apnea (OSA) in as many as two-thirds of the patients.[2] Then, there is also the problem of intracranial hypertension (ICH), which may be a major problem on its own, or a factor that during sleep interacts with the respiratory physiology of UAO and OSA.[3,4] In sCS, ICH has a number of potential

Disclosures: *The authors have no financial interest to declare in relation to the content of this article.*

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causes including OSA, hydrocephalus, cerebral venous hypertension, and craniocerebral disproportion.[5–7]

We have previously shown that children with sCS with no OSA or ICH have normal sleep pattern, which was determined by the presence of normal total sleep time (TST), normal number of arousals, and normal sleep architecture using electroencephalography (EEG) derived hypnograms.[8] In contrast, sCS children with moderate-to-severe OSA had higher arousal index, higher respiratory effort-related arousal (RERA) index, lower sleep efficiency, less rapid-eye movement (REM) sleep, and more non-REM stage 1 (N1) sleep. ICH, on the other hand, was only related to higher RERA index; however, this was probably related to the mild OSA that those patients had. Of further interest, we found that 5 of these cases undergoing monobloc surgery showed some improvement in sleep architecture. As it was not clear whether this improvement was the result of treating the OSA, or the ICH, in this report we have extended the number of sCS children with PSG, as well as the number of pre-to-postoperative observations. The aim of this study was twofold: to evaluate the role of sleep architecture in the diagnostic work-up of children with sCS and to investigate the consequences of elective surgery for OSA and ICH on sleep architecture.

PATIENTS AND METHODS

This report comes from ongoing prospective work in a national sCS cohort evaluated and managed at the Dutch Craniofacial Center (Sophia Children's Hospital – Erasmus University Medical Center, Rotterdam, The Netherlands). As such, all clinical care follows protocolized management,[9] and studies are approved by the institutional research ethics board for human studies (MEC-2005–273 and MEC-2017-1143).

The inclusion criteria for this report were: age ≤18 years; diagnosis of sCS; and, performance of level 1 polysomnography (PSG). Eighty-three cases were compared with 35 control subjects. The controls did not have OSA on PSG, and there was no likelihood of ICH on clinical examination. These subjects were selected from four groups of referrals: cases undergoing PSG, but otherwise well; or cases of non-syndromic unicoronal synostosis (cases without a *TCF12* mutation; *TWIST 1* mutation; or *FGFR 1, 2, or 3* mutation); or cases being investigated for brief resolved unexplained event (BRUE)[10]; or cases with unexplained daytime sleepiness.

Polysomnography

All children underwent one or more video-assisted PSG (Brain RT, OSG, Rumst, Belgium). During the PSG, several cardiorespiratory parameters were assessed: electrocardiography, nasal airflow (thermistor), chest and abdominal wall motion, a capillary blood gas test, arterial blood oxygen-hemoglobin saturation using pulse oximetry (SpO_2), and transcutaneous partial pressure of carbon dioxide ($tcpCO_2$). EEG was recorded continuously and used for sleep architecture analysis. The PSG-derived variables were used in the analysis if the TST was at least 360 minutes, and free from artifacts.

Respiratory

PSG studies were assessed according to the American Academy of Sleep Medicine (AASM) 2012 updated guidance for scoring pediatric respiratory events.[11] A physician first scored all respiratory events for clinical purposes. For this report, all PSGs were revalidated (RdG) and scored using the criteria we have previously published (8).

The obstructive-apnea/hypopnea index (oAHI) was calculated by dividing the sum of all obstructive apneas and hypopneas by the TST. The severity of OSA was categorized as: ***Mild***, an oAHI ≥1 and <5; ***Moderate***, an oAHI ≥5 and <10; and ***Severe***, an oAHI ≥10.

Sleep architecture

Hypnograms were generated using the 2012 AASM guidance on scoring.[11] The EEG and chin-EEG signals were assessed in 30-second epochs according to the same criteria as in our previous work.[8]

Sleep efficiency was defined as the TST divided by the total time in bed. To calculate sleep quality, the sum of the amount of REM-sleep and N3-sleep was divided by TST. Wake time After Sleep Onset (WASO) was defined as the total time (in minutes) awake between the first moment of falling asleep until the last moment waking up.

Arousals were also scored according to the criteria we previously published.[8] If an arousal lasted longer than 30 s, it was considered an awakening. Since the 2012 AASM update, respiratory effort-related arousals (RERAs) are often scored as hypopnea, which may lead to an apparent reduction in RERA frequency. Therefore, in this study, a respiratory arousal was included and defined as an arousal that followed a respiratory event, such as an apnea or hypopnea. The arousal index was calculated by dividing the number of arousals by the TST. The same was true for the respiratory arousal index.

Intracranial hypertension

The presence or absence of ICH at the time of each PSG was established by using information from invasive ICP measurements, optical coherence tomography (OCT) scans, or fundoscopy.

An invasive ICP measurement was considered normal if baseline pressure during the day and night was below or equal to 10 mmHg. Baseline pressure between 10 and 15 mmHg was considered normal or borderline abnormal based on the height and duration of plateau waves. Plateau waves were considered normal if the pressure stayed below 25 mmHg, and borderline abnormal when the pressure was between 25 and 35 mmHg. Plateau waves above 35 mmHg were considered abnormal. The duration of plateau waves was considered normal if it was shorter than 10 minutes, borderline abnormal between 10 and 20 minutes, and abnormal if longer than 20 minutes. If the baseline pressure during ICP monitoring was greater than 15 mmHg, it was considered abnormal (12).

OCT imaging (Spectralis OCT scanner, Heidelberg Engineering, Heidelberg, Germany) was used to assess total retinal thickness (TRT). In our clinic, the normal range of TRT has been derived from 67 healthy 4-to-12-

year-old children (*unpublished data*), and we use values above the 97.5th percentile to indicate abnormality (i.e., TRT > 503 µm).

ICH was defined as being present when one or more of the following was true: a positive fundoscopy with pseudopapilledema ruled out, a positive invasive ICP measurement, or an increased TRT.

Surgical treatment

Our surgical treatment for children with sCS includes cranial vault expansion within the first year of life: occipital distraction with springs for Apert and Crouzon syndrome; and, a fronto-orbital advancement for Saethre-Chotzen and Muenke syndrome.

If a child develops OSA within the first year, the initial treatment is based on the severity of the OSA: prone positioning, oxygen support, continuous positive airway pressure (CPAP), or the insertion of a tracheal cannula. Endoscopy of the upper airway is performed to identify the levels of obstruction in cases with moderate to severe OSA. This mainly concerns patients with Apert and Crouzon syndrome. Based on the results from the endoscopy, a monobloc distraction with or without mandibular distraction is performed from 2 years of age and above. Otherwise, such surgery is delayed until the age of 7 to 9 years of age.

In complex craniosynostosis (children with multiple fused sutures, but without a known genetic mutation), the choice of treatment depends on the skull deformity and associated OSA and/or ICH. If a child develops ICH during follow-up, a subsequent cranial vault expansion is considered depending on the cause and severity, unless obvious hydrocephalus is detected for which a third ventriculostomy is performed or a ventriculoperitoneal shunt is inserted.

Statistical Analysis

Statistical analysis was performed in the statistical programming language R (R Core Team, 2013, Vienna, Austria). To determine disrupted sleep architecture, we performed a linear-mixed model, and investigated the effects of OSA, ICH and the presence of sCS versus control status. All subjects (sCS and control group) were included in the model, and all PSGs were included in the model to account for the correlation between repeated measurements over time in each patient. For each sleep architecture parameter (dependent variable) the model was adjusted to achieve the best fit. The independent variables were 'ICH', 'oAHI', 'patient vs. control', 'age' and 'gender'. Effects of the variables were checked for linearity and adjusted. Outliers were excluded from analysis. Spline interpolation was used in case of non-linearity and if it improved the fit of the model. The appropriate random-effects structure that best fitted the data was selected based on likelihood ratio tests. The appropriate fixed-effects structure was selected using F and likelihood ratio tests. Residual plots were used to validate the models' assumptions.

To evaluate the effect of surgical treatment on sleep architecture, analysis was performed in a subset of 19 patients who underwent a PSG preoperatively and postoperatively. The different types of surgery performed were categorized based on their indication, i.e., correction of OSA (oAHI ≥5, moderate-to-severe OSA) and/or ICH. Surgeries for OSA included adenotonsillectomy, nasal septum corrections and mandibular distraction osteotomy. Surgery performed for OSA when the preoperative oAHI was below 5, was not considered as OSA-indicated. Surgeries for ICH included all calvarial expansions. Monobloc surgery can be indicated for OSA, or ICH, or both. Cranial vault surgery carried out as part of the standard protocol, but in a patient without signs of ICH, was not scored as being an ICH-indicated procedure.

In the pre-to-postoperative group, change in scores (delta-scores) in sleep architecture parameters were calculated. Then the linear regression models with the delta-scores as dependent variables were created. The independent variables were age at the time of surgery, whether the indication of surgery was OSA or not, and if the indication of surgery was ICH or not. Afterwards, we performed a post-hoc power analysis for the linear regression models; a power of 0.80 and above was considered sufficient.

RESULTS

Patients

Eighty-three patients (43 males, 51.8%) with sCS underwent PSG and were screened for ICH (Table 1). Forty-nine patients underwent only one PSG, 21 patients two PSGs, eight patients three PSGs, three patients four PSGs, and two patients five PSGs.

Effects of OSA and ICH on sleep architecture

Scatterplots of all sleep architecture parameters of children with sCS and OSA or ICH against a locally estimated scatterplot smoothing (LOESS) curve of control subjects and children with sCS without OSA or ICH are presented in Supplemental Digital Content 1. (**See figure, Supplemental Digital Content 1**, which displays scatterplots of sleep architecture parameters of the total population of children with syndromic craniosynostosis (sCS) against a locally estimated scatterplot smoothing (LOESS) curve and its standard error of 99 polysomnographies of control subjects and children with sCS with an obstructive-apnea/hypopnea index (oAHI) <1 and without intracranial hypertension (ICH). **http://links.lww.com/PRSGO/B195**) Table 2 shows the effects of oAHI, the presence of ICH, and having a sCS on sleep architecture parameters. A one-point increase in oAHI was associated with an increase in both the arousal index and the respiratory arousal index, with 0.13 (p<0.001) and 0.15 (p<0.001) events/hour, respectively. Every point increase in oAHI was also associated with a decrease in sleep quality of -0.28% (p=0.010), decrease in sleep efficiency of -0.19% (p=0.038), and decrease in the amount of N3-sleep of -0.21% (p=0.039). In addition, in regard to the amount of N2-sleep, every point increase in oAHI was associated with increase in amount of N2-sleep 0.25% (p=0.004). After the exclusion of out-

Table 1. Patient Characteristics

	Total population sCS n=83		Pre-to-postoperative population sCS n=19		Control population n=35	
Age at first (or pre-op) PSG, yr (median, IQR)	3.08	(0.58 – 8.89)	2.06	0.56 – 5.00	4.41	(1.49 – 7.82)
Age post-op PSG yr (median, IQR)	-	-	3.64	1.93 – 5.99	-	-
Age at surgery yr (median, IQR)	-	-	2.90	0.87 – 5.15	-	-
Male (n, %)	43	51.8%	13	68.4%	15	42.9%
ICH (n, %)	25*	30.1%	8†	42.1%	0	0%
OSA (n, %)						
No	48*	57.8%	4†	21.1%	35	100%
Mild	18*	21.7%	9†	47.4%	0	0%
Moderate	7*	8.4%	3†	15.8%	0	0%
Severe	10*	12.0%	3†	15.8%	0	0%
Diagnoses (n, %)						
Apert	20	24.1%	8	42.1%	-	-
Crouzon	31	37.3%	8	42.1%	-	-
Muenke	9	10.8%	1	5.3%	-	-
Saethre-Chotzen	10	12.0%	1	5.3%	-	-
TCF12	2	2.4%	0	0%	-	-
IL11RA	1	1.2%	0	0%	-	-
Complex craniosynostosis	10	12.0%	1	5.3%	-	-
ICH-indicated surgery (n, %)	-	-	4	21.1%	-	-
OSA-indicated surgery (n, %)	-	-	2	10.5%	-	-
Both ICH-indicated and OSA-indicated surgery (n, %)	-	-	4	21.1%	-	-
No ICH-indicated nor OSA-indicated surgery (n, %)	-	-	9	47.4%	-	-

Patient characteristics of the total group of children with craniosynostosis (sCS), the subgroup of children with sCS with preoperative and postoperative measurements, and the healthy control population. ICH=intracranial hypertension, OSA = obstructive sleep apnea.
*Maximum OSA stage measured in one of the polysomnographies, and ICH scored if present during one of the PSGs (this table only).
†OSA stage or presence of ICH at the time of the first PSG.

liers, the presence of ICH was not associated with any change in sleep architecture. However, in two very young infants with multiple bone defects due to hydrocephalus, the wake time after sleep onset (WASO) was greatly increased at 479.5 and 471.0 minutes. Having sCS was not associated with any changes in sleep architecture.

Effect of surgery on sleep architecture

A subset of 19 patients underwent PSG before and after surgery. The characteristics of this subset of patients are presented in Table 1. The median interval between the preoperative PSG and surgery was 0.20 years (IQR: 0.13 – 0.54), the median interval between the surgery and the postoperative PSG was 0.74 years (IQR: 0.31 – 1.04), and the median interval between the preoperative and postoperative PSG was 1.10 years (IQR: 0.69 – 1.45). Scatterplots of all pre-to-postoperative changes in sleep architecture parameters of this subgroup of children with sCS against a LOESS curve of control subjects and children with sCS without OSA or ICH are presented in Supplemental Digital Content 2. (**See figure, Supplemental Digital Content 2**, which displays scatterplots of all pre-to-postoperative changes in sleep architecture parameters of the subgroup of 19 children with syndromic craniosynostosis (sCS) with a preoperative and postoperative polysomnography, against a locally estimated scatterplot smoothing (LOESS) curve and its standard error of 99 polysomnographies of control subjects and children with sCS with an obstructive-apnea/hypopnea index (oAHI) <1 and without intracranial hypertension (ICH). **http://links.lww.com/PRSGO/B196**) The results of the linear regression models of the delta-scores of the different sleep architecture parameters are presented in Table 3. The results show that surgery with an OSA indication is associated with decrease in the arousal index (-6.89, p=0.030) and the respiratory arousal index (-5.49, p=0.013). OSA-indicated surgery was also associated with increase in TST of 96.26 minutes (p=0.025), and with increase in sleep efficiency of 13.55% (p=0.017). Surgery with an ICH indication was associated with decrease in sleep efficiency of -15.37% (p=0.006) and with decrease in sleep quality of -11.27% (p=0.032). The latter being explained by significant decrease in the amount of REM-sleep of -11.23% (p=0.001) and by an increase in the amount of N1-sleep of 12.36% (p=0.034).

DISCUSSION

In this study of pediatric patients with sCS we have three main observations. First, OSA does, and ICH does not, affect sleep architecture. Second, surgery for moderate-to-severe OSA improves sleep architecture. Third, surgery for ICH affects sleep architecture in a way that is different to the way it is after OSA-indicated surgery. Taken together, we have extended our previously-reported preliminary observations (8) in sCS, and now affirm that in our practice, sleep architecture analysis is used as valuable information in the assessment of children with sCS.

The scatterplots in Supplemental Digital Content 1 show that sleep architecture parameters vary greatly between subjects, both with and without sCS. Hence, it was difficult to establish certain cut-off values for normal or abnormal sleep architecture. We found that OSA on its

Table 2. OSA and ICH in relation to sleep architecture.

	Mean	Regression coefficient		p-value
TST	513.6	oAHI	-0.91	0.108
(min)		ICH	8.15	0.780
		Patient vs. control	0.13	0.993
Arousal	6.86	oAHI	0.13	<0.001*
(events/h)		ICH	-0.87	0.625
		Patient vs. control	0.69	0.362
Resp. Arousal	0.96	oAHI	0.15	<0.001*
(events/h)		ICH	-0.71	0.466
		Patient vs. control	0.34	0.405
WASO	82.97	oAHI	0.61	0.219
(min)		ICH	14.94	0.608
		Patient vs. control	-0.12	0.991
Efficiency	78.47	oAHI	-0.19	0.038*
(%)		ICH	-6.97	0.157
		Patient vs. control	3.17	0.114
Quality	59.1	oAHI	-0.28	0.010*
(%)		ICH	4.14	0.501
		Patient vs. control	-2.33	0.335
REM	21.59	oAHI	-0.11	0.036*
(%)		ICH	-3.37	0.266
		Patient vs. control	0.20	0.865
N1	13.41	oAHI	0.05	0.412
(%)		ICH	6.50	0.067
		Patient vs. control	-0.10	0.946
N2	27.06	oAHI	0.25	0.004*
(%)		ICH	-6.38	0.170
		Patient vs. control	1.19	0.541
N3	38.54	oAHI	-0.21	0.039*
(%)		ICH	0.32	0.951
		Patient vs. control	-1.58	0.493

Linear-mixed model of 118 subjects with a total of 177 measurements of sleep architecture using polysomnography, of which 83 subjects with 138 measurements are children with syndromic craniosynostosis (sCS) and 35 healthy control subjects with 39 measurements. The effect of the obstructive-apnea/hypopnea index (oAHI), intracranial hypertension (ICH) and the effect of having a sCS compared to being a control subject is being presented. Data are corrected for gender and age at the time of the polysomnography. TST = Total Sleep Time, WASO = Wake time After Sleep Onset, REM = rapid eye movement sleep, N1 = non-REM stage 1 sleep, N2 = non-REM stage 2 sleep, N3 = non-REM stage 3 sleep or deep sleep.

own had a disruptive effect on sleep architecture, which is generally similar to findings in other reports.[13–15] In addition, a few studies show that surgical treatment does not improve sleep architecture,[16–19] but this literature contrasted with our finding that there was a small improvement with surgical treatment for OSA in patients with sCS. This finding is supported by an increase in sleep efficiency and TST, and a decrease in the number of arousals and respiratory arousals. However, there was no improvement in the percentage of REM-sleep or in sleep quality in the linear regression model.

Not much is known about the effect of ICH on sleep architecture. Our findings suggest that sCS children with ICH have normal sleep architecture. However, based on our own clinical experience, parents of children with ICH do report that their child sleeps more restlessly than other children. It is possible that this effect is too subtle or irregular to detect using only a single night PSG. However, we did find a disturbed sleeping pattern in two young infants with pronounced hydrocephalus. We cannot be sure if the increased pressure is related to the disturbed sleeping pattern, but it remains an interesting topic of research. In the present study, we found that cranial vault surgery for ICH did have an effect on sleep architecture. In the pre-to-postoperative analysis, ICH-indicated surgery decreased sleep efficiency, sleep quality and the percentage of REM-sleep, while increasing the percentage of N1-sleep. The plots of Supplemental Digital Content 2 show that in all but one subject who underwent ICH-indicated surgery there was a decrease in the amount of REM-sleep.

The pathophysiological processes underlying our findings are unknown. In a study of 28 adults with normal-pressure hydrocephalus, wakefulness and cerebral blood flow (CBF) were investigated before and after

Table 3. Effect of OSA-indicated and ICH-indicated Surgery on Changes in Sleep Architecture

	Median Pre-op (IQR) n=19	Median Post-op (IQR) n=19	Surgical indication	Regression coefficient	R^2	p-value	Post-hoc power
TST	539.0	509.5	OSA	96.26	0.35	0.025*	0.85
(min)	(494.5 – 614.5)	(500.8 – 569.0)	ICH	-39.07		0.303	
Arousal	8.0	4.9	OSA	-6.89	0.32	0.030*	0.80
(events/h)	(6.6 – 11.5)	(4.1 – 6.7)	ICH	-0.79		0.776	
Resp. Arousal	1.0	0.4	OSA	-5.49	0.42	0.013*	0.93
(events/h)	(0.7 – 1.3)	(0.1 – 0.7)	ICH	1.70		0.371	
WASO	89.0	69.0	OSA	-55.86	0.26	0.277	0.68
(min)	(31.0 – 166.0)	(47.0 – 102.2)	ICH	60.99		0.212	
Efficiency	82.3	80.0	OSA	13.55	0.63	0.017*	0.99
(%)	(70.5 – 85.7)	(71.4 – 85.5)	ICH	-15.37		0.006*	
Quality	59.4	56.3	OSA	7.80	0.38	0.137	0.89
(%)	(57.5 – 67.4)	(51.4 – 63.4)	ICH	-11.27		0.032*	
REM	19.8	17.7	OSA	1.24	0.55	0.690	0.99
(%)	(16.0 – 26.0)	(15.8 – 21.2)	ICH	-11.23		0.001*	
N1	13.6	16.7	OSA	-4.25	0.31	0.453	0.78
(%)	(10.4 – 17.1)	(11.3 – 26.3)	ICH	12.36		0.034*	
N2	24.3	26.1	OSA	-3.58	0.05	0.570	0.16
(%)	(16.0 – 28.8)	(18.6 – 29.4)	ICH	-1.09		0.856	
N3	40.9	38.8	OSA	6.19	0.10	0.320	0.27
(%)	(35.9 – 45.0)	(35.1 – 41.1)	ICH	-0.58		0.921	

Linear regression models of the delta-scores of sleep architecture parameters. The effect of surgical treatment, based on its indication for obstructive sleep apnea (OSA) and/or intracranial hypertension (ICH), corrected for age at the time of surgery. TST = Total Sleep Time, WASO = Wake time After Sleep Onset, REM = rapid eye movement sleep, N1 = non-REM stage 1 sleep, N2 = non-REM stage 2 sleep, N3 = non-REM stage 3 sleep or deep sleep.

ventricular shunting.[20] Postoperatively, patient wakefulness increased and CBF increased in the hippocampal regions. Spruijt et al.[21] showed that in 12 sCS children with ICH and abnormal CBF-indices – as shown by transcranial Doppler with increased peak systolic velocities and higher resistance indices – that cranial vault surgery normalized these parameters, suggesting a change in CBF in children with ICH. In the present study, if we assume that there is a similar change in CBF (we did not measure it), then this change might be the cause of changes in REM-sleep and N1-sleep, sleep quality and sleep efficiency. Further multimodel monitoring research is needed to determine whether the changes in sleep efficiency and quality are beneficial or not and if they are permanent or temporary. Nevertheless, we still consider surgical treatment for ICH in sCS an essential part of the treatment protocol.

Our study does have three main limitations. The first limitation is the small sample size. Since sCS is a very rare condition, and because we only started PSG in this population in 2012, it is a challenge to quickly increase the number of patients. Between our preliminary report in 2016 [8] and now, we have added 14 patients (almost threefold increase) to the pre-to-postoperative analysis, and 44 patients to the total population (more than doubled). The small sample size also means that only large changes in sleep architecture parameters are detected, and we may have missed subtle changes in sleep architecture parameters we did not have enough power for in the pre-to-postoperative analysis (i.e. WASO, N1, N2, and N3). Nonetheless, we are able to draw some conclusions and insights into the effects of surgery on sleep architecture in children sCS. Second, it was not possible to deal with any first night effects in the PSG findings because we did not have the capacity to perform studies on two or three consecutive nights. However, as all patients were exposed to the first night effect, it was still possible to compare measurements between subjects. Third, we have a pragmatic method for defining ICH. The gold standard approach is to diagnose ICH using invasive ICP measurements. We consider this method too invasive in this population – the risks of invasive monitoring outweigh the benefits of making the diagnosis invasively rather than noninvasively – and so we use OCT, fundoscopy, magnetic resonance imaging, and the head circumference for our diagnosis. Although not as accurate as an invasive measurement, we think that our methodology is sensitive enough to establish the presence of ICH.

In children with sCS, PSG-assessed sleep architecture adds valuable information in the diagnostic work-up of OSA. For example, it allows for the detection of arousals, assessment of sleep quality and sleep efficiency, and is useful to more adequately calculate the TST. The results of this study show that OSA disrupts sleep architecture but ICH does not. Surgery for OSA improves sleep architecture, stressing the importance of treating OSA to assure adequate sleep. Surgery for ICH affects sleep architecture, although it is still not clear whether this is a positive or negative effect.

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