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Sleep Medicine

Brief Communication – R1

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- AB contributed to the conceptualisation of the study and to the revision of the manuscript
- HT contributed to the conceptualisation of the study, to provide statistical supervision and to the revision of the manuscript

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Highlights

- McGill score positive predictive value is significantly lower in children with co-morbidities
- Children with co-morbidities with an abnormal McGill score should not be assumed to have OSA
- More detailed sleep studies are needed in children with co-morbidities and suspected OSA

Abstract

Background The McGill score is used to stratify severity of oximetry in children referred for investigation of obstructive sleep apnoea (OSA) to identify those with more severe disease and prioritize treatment. We hypothesized that its Positive predictive value (PPV) and Negative predictive value (NPV) in detecting OSA differs significantly between children with medical conditions and otherwise healthy children.

Methods We performed a two-year retrospective analysis of children referred for investigation of OSA who underwent a cardiorespiratory (CR) polygraphy study. McGill score was calculated from the oximetry trace blinded to polygraphy results. We looked at 2 definitions of OSA: Obstructive Apnoea Hypopnoea Index (oAHI) ≥1 and ≥5. McGill sensitivity, specificity, PPV and NPV were calculated. McGill score=1 was considered normal or inconclusive, >1 abnormal.

Results We studied 312 children, 190 males (61%), median age 4.5 (2.4-7.9) years. 129 were otherwise healthy and 183 had associated medical conditions. The PPV of the McGill score was significantly lower in children with medical conditions than otherwise healthy children. The NPV was similar in both groups of children.

Conclusions The higher number of false positives in children with medical conditions may be due to non-obstructive causes such as central apnoeas. Children with underlying lung disease are also more likely to desaturate following a brief apnoea or hypopnoea. Children with co-morbidities who have an abnormal McGill score should not be assumed to have OSA and need more detailed sleep studies to determine the reason for the oxygen desaturations.

Key words

McGill score, oximetry, obstructive sleep disordered breathing

Introduction

The McGill score is used to stratify the severity of obstructive sleep apnoea (OSA) in children and thus to prioritize their treatment (1). It ranges from 1 (normal or inconclusive) to 4 (severely abnormal) according to the number of clusters and the depth of desaturation events seen in the overnight oximetry. Previous work on otherwise healthy children has shown that oximetry, when compared to full polysomnography (PSG), has a high positive predictive value (PPV) and a low negative predictive value (NPV)(2). The current European Respiratory Society and American Academy of Pediatrics guidelines therefore suggest that an abnormal overnight oximetry based on McGill criteria can accurately detect OSA of at least moderate severity but a negative result does not exclude OSA with certainty (3, 4). These conclusions, however, were based on studies performed in otherwise healthy children being investigated for OSA and there is little data on children with co-morbidities. Two studies have focused on children with Down syndrome, one found that the McGill score in children with Down syndrome had a sensitivity of 43%, specificity 93%, PPV 94%, NPV 39% which was comparable to otherwise healthy children (5), whilst the other found that it overestimated the number of children with OSA, due to central apnoeas causing desaturations (6). A recent paper compared the oximetry recordings from children with obliterative bronchiolitis or cystic fibrosis, called collectively “obstructive lung disease (OLD)”, otherwise healthy children with SDB due to adenotonsillar hypertrophy and controls. Children with OLD had significantly lower baseline SpO2 when compared to the latter two groups. Whilst they had a higher number of desaturations/h compared with controls, it was lower compared with the adenotonsillar hypertrophy group (7).

We aimed to determine the relationship between the McGill score and cardiorespiratory polygraphy (CR Polygraphy) in the diagnosis of OSA in children referred to a tertiary care paediatric sleep centre. We hypothesized that the positive and negative predictive values of the McGill score in

detecting OSA would be worse in children with associated comorbidities compared with otherwise healthy children.

Methods

We carried out a two-year retrospective analysis of children (aged<18years) who underwent a CR polygraphy at the Paediatric Sleep and Ventilation Unit at Royal Brompton Hospital for evaluation of OSA. Patients acutely unwell at the time of the overnight sleep study were excluded. The oximetry traces were obtained via TCM CombiM® monitor, Radiometer, Copenhagen, Denmark and a simultaneous CR Polygraphy montage (SOMNOScreen™ plus, SOMNOmedics, Germany) was used for overnight monitoring. FT calculated the McGill score (1), blinded to CR Polygraphy results. McGill score=1 was considered normal or inconclusive, while >1 abnormal. We then compared the McGill score results with two different diagnostic definitions of OSA, either an Obstructive Apnoea Hypopnoea Index (oAHI) ≥1(mild, moderate and severe OSA) or an oAHI ≥5 (moderate and severe OSA).

Data were analysed by GraphPad Prism® software version 7.02 and by IBM SPSS® version 24. Descriptive statistics were generated for each measure. For all the reported variables a test of normality was performed. For non-parametric data, median with interquartile range was reported. McGill sensitivity, specificity, Positive Predictive Value (PPV), Negative Predictive Value (NPV) and accuracy were calculated for the whole cohort, for the otherwise healthy children and those with associated medical conditions. For all subgroups we have reported the results and the 95% Confidence Interval (95% CI) in Table 1 a and b. Sensitivity was the proportion of all children with OSA (at CR Polygraphy) with an abnormal McGill score. Specificity was the proportion of all children without OSA with a normal McGill score of 1. Accuracy was the proportion of all the correct assessment out of all the assessments.

Results

We studied 315 children, three patients' data was discarded because of oximetry artefact, thus a total of 312 patients were considered for analysis. The median age was 4.5 (2.4-7.9) years, 190 (61%) were male. 129 (41%), median age 4.5 (2.4-7.8) years, were otherwise healthy, while 183 (59%), median age 4.5 (2.4-8.0) years, had associated medical conditions: neurological/neuromuscular syndromes (n=50), upper airway malformations/wheeze (n=75), interstitial lung disease/chronic lung disease (n=40), congenital heart disease (n=15), obesity (n=3). Median age was not significantly different across the diagnostic groups.

One hundred and one of 312 (32%) children had OSA defined as oAHI≥1, 45/129 (35%) otherwise healthy and 56/183 (31%) with medical conditions. Forty-seven of 312 (15%) had OSA defined as oAHI≥5, 23/129 (18%) otherwise healthy and 24/183 (13%) with medical conditions.

Median (IQR) SpO_2 was 97.6 % (96.6-98.4), median (IQR) minimum SpO_2 was 90% (86-93) and median (IQR) oxygen desaturation index (ODI>4%) was 3.0 (1.8-5.2)/h in the whole study population. Otherwise healthy children had median (IQR) SpO_2 97.9% (96.9-98.7) and median (IQR) minimum SpO_2 91% (88-93). The median (IQR) ODI in otherwise healthy children was 2.8 (1.6-4.2)/h. Children with medical conditions had median (IQR) SpO_2 97.5% (96.2-98.3) and median (IQR) minimum SpO2 90% (86-92). The median (IQR) ODI was 3.2 (1.9-6.4)/h.

Median (IQR) McGill score in the whole population was 1 (1-2). It was 1 (1-1) in otherwise healthy children and 1 (1-2) in children with medical conditions. Median (IQR) McGill in neurological/neuromuscular syndromes was 1 (1-3), in upper airway malformations/wheeze was 1 (1-1), in children with interstitial lung disease/chronic lung disease was 1 (1-2), in those with congenital heart disease was 2 (1-2) and in children with obesity was 1 (1-1).

For OSA defined as obstructive oAHI ≥1, McGill' score sensitivity was low in the whole cohort (47%) and was similarly poor in the two groups of otherwise healthy children and those with medical conditions (40% and 52% respectively). The specificity was lower in the group of children with medical conditions compared with the otherwise healthy group (79% vs 92%). The PPV was also lower in the children with medical conditions (52% vs 72%). The NPV was similar between the two groups. Out of all the subgroups of children with medical conditions, it was the group of children with neurological/neuromuscular disorders in whom the McGill score had the highest PPV and NPV (70% and 83% respectively). Conversely, children with congenital heart disease had the lowest PPV and NPV (30% and 60% respectively).

When OSA was defined as oAHI ≥5, McGill's sensitivity increased in the overall population (74%) and in the two groups of otherwise healthy children and those with medical conditions. Nevertheless, it was still too low to be satisfactory for a screening test. The specificity and PPV were once again lower in children with associated medical conditions than otherwise healthy children. There was little difference in the NPV between the two groups. Results are summarised in Table 1a and b.

Table 1. *Sensitivity, specificity, Positive and Negative predictive value (PPV and NPV respectively) for McGill score when compared to cardiorespiratory polygraphy in detecting Obstructive Sleep Apnoea in otherwise healthy and children with medical conditions.*

a. OSA defined as oAHI≥1

	Sensitivity 95% CI	Specificity 95% CI	PPV 95% CI	NPV 95% CI	Accuracy 95% CI
Total cohort (n=312)	47/101 (47%) (36.6-56.7)	177/211 (84%) (78.2-88.6)	47/81 (58%) (48.8-66.7)	177/231 (77%) (73.0-79.9)	224/312 (72%) (66.5-76.7)
Otherwise healthy (n=129)	18/45 (40%) (25.7-55.7)	77/84 (92%) (83.6-96.6)	18/25 (72%) (53.7-85.1)	77/104 (74%) (69.0-78.5)	95/129 (74%) (65.2-81.0)
Medical conditions (n=183)	29/56 (52%) (38.0-65.34)	100/127 (79%) (70.6-85.5)	29/56 (52%) (41.4-62.0)	100/127 (79%) (73.6-83.1)	129/183 (70%) (63.3-77.0)
- Neurological/neuromuscular syndromes (n=50)	14/19 (74%) (48.8-90.9)	25/31 (81%) (62.5-92.6)	14/20 (70%) (52.0-83.4)	25/30 (83%) (69.8-91.5)	39/50 (78%) (64.0-88.5)
- Upper airway diseases/wheeze (n=75)	6/19 (32%) (12.6-56.6)	48/56 (86%) (73.8-93.6)	6/14 (43%) (23.0-65.3)	48/61 (79%) (72.8-83.6)	54/75 (72%) (60.4-81.8)
- Lung diseases (n=40)	6/13 (46%) (19.2-74.9)	21/27 (78%) (57.7-91.4)	6/12 (50%) (28.5-71.5)	21/28 (75%) (63.6-83.8)	26/40 (67%) (50.9-81.4)
- Congenital heart disease (n=15)	3/5 (60%) (14.7-94.7)	3/10 (30%) (6.7-65.3)	3/10 (30%) (15.8-49.4)	3/5 (60%) (26.4-86.3)	6/15 (40%) (16.3-67.7)
- Obesity (n=3)	0/0 (0%) N/A	3/3 (100%) (29.2-100)	0/0 (0%) N/A	3/3 (100%) N/A	N/A N/A

b. OSA defined as oAHI≥5

	Sensitivity 95% CI	Specificity 95% CI	PPV 95% CI	NPV 95% CI	Accuracy 95% CI
Total cohort (n=312)	35/47 (74%) (59.7- 86.1)	219/265 (83%) (77.5 87.0)	35/81 (43%) (35.8-51.01)	219/231 (95%) (91.8- 96.8)	254/312 (81%) (76.6-85.6)
Otherwise healthy (n=129)	15/23 (65%) (42.7-83.6)	96/106 (91%) (83.3- 95.4)	15/25 (60%) (43.6- 74.4)	96/104 (92%) (87.2-95.5)	111/129 (86%) (78.9-91.5)
Medical conditions (n=183)	20/24 (83%) (62.6- 95.3)	123/159 (77%) (70.1-83.6)	20/56 (36%) (28.4- 43.8)	123/127 (97%) (92.6- 98.7)	143/183 (78 %) (71.5- 83.9)
- Neurological/neuromuscular syndromes (n=50)	12/14 (86%) (57.2- 98.2)	28/36 (78%) (60.9- 89.9)	12/20 (60%) (44.0-74.1)	28/30 (93%) (79.3-98.1)	40/50 (80%) (66.3-90.0)
- Upper airway diseases/wheeze (n=75)	4/5 (80%) (28.4- 99.5)	60/70 (86%) (75.3- 92.9)	4/14 (29%) (16.3- 45.2)	60/61 (98%) (91.2-99.7)	64/75 (85%) (75.3-92.4)
- Lung diseases (n=40)	1/2 (50%) (1.3- 98.7)	27/38 (71%) (54.1-84.6)	1/12 (8%) (2.0-28.4)	27/28 (96%) (86.9-99.1)	28/40 (70%) (53.5-83.4)
- Congenital heart disease (n=15)	3/3 (100%) (29.2-100.0)	5/12 (42%) (15.2-72.3)	3/10 (30%) (21.0-41)	5/5 (100%) N/A	8/15 (53%) (26.6-78.7)
- Obesity (n=3)	0/0 (0%) N/A	3/3 (100%) (29.2-100.0)	0/0 (0%) N/A	3/3 (100%) N/A	N/A N/A

Discussion

In our cohort of children referred for investigation of obstructive sleep disordered breathing, the McGill score had a lower PPV in the group of children with medical conditions than the group of otherwise healthy children due to a higher number of false positives. The NPV was similar between the two groups.

This low PPV, particularly in children with underlying lung diseases, may be because they are more likely to desaturate following a brief hypopnoea or apnoea than healthy children. Desaturations have also been shown to occur during periods of phasic partial respiratory muscle inhibition in rapid eye movement sleep, resulting in decrease in functional residual capacity, airway closure in the dependent lung regions and ventilation-perfusion mismatch (8). Moreover, children with neurological conditions or syndromes may have non-obstructive events such as central apnoeas resulting in desaturations (Figure 1) which may also result in a false positive McGill score. This is in keeping with the findings of Coverstone et al who also found that central apnoeas were the cause of some children with Down syndrome to have positive McGill scores (6). Our NPV was higher than that reported by Brouillette et al. which may be due to the different thresholds of referral and different patient populations – 60% of the children in that study had OSA confirmed on PSG, compared with 35% in our otherwise healthy group and 31% in our group of children with co-morbidities (2). Interestingly, whilst the majority of the patients in that study were otherwise healthy, they reported that the three false-positive oximetry results they had were all in the subgroup of children who had diagnoses other than adenotonsillar hypertrophy that might have affected breathing during sleep, which would be consistent with our results. The use of CR Polygraphy instead of PSG in the diagnosis of OSA could in part explain why in our work even in otherwise healthy children PPV was lower (72% for oAHI≥1 or 60% for oAHI≥5 vs 93%) than previously

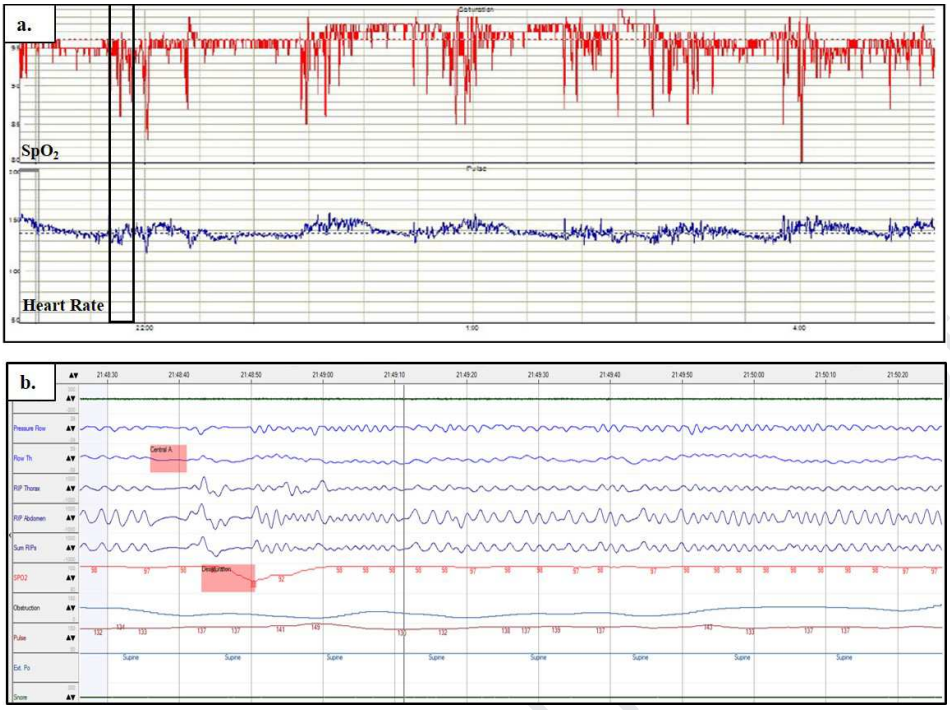
reported (4). CR Polygraphy is commonly used in the diagnosis of OSA in the UK but we acknowledge it can potentially underestimate OSA severity when compared to PSG.

To the best of our knowledge, this is the largest study to examine the use of the McGill score in children with co-morbidities. The main limitations of this work are its retrospective nature and the small size of some of the subgroups of medical conditions, such as obesity. We acknowledge that confirmatory prospective studies are needed.

Conclusion

In conclusion, our data have shown that the PPV of the McGill score is significantly lower in children with medical conditions than otherwise healthy children. Children with co-morbidities who have a positive McGill score must not be assumed to have OSA, and require more detailed sleep studies to determine the physiological reason for their desaturations.

Figure 1.



Overnight oximetry (a.) coupled with cardio-respiratory polygraphy (b.) of a 6 month old patient with Prader Willi Syndrome

a. Overnight oximetry, scored as McGill score 3
b. Example of a 2 minute epoch from cardiorespiratory polygraphy showing a central apnoea resulting in a desaturation to 88%

References

1. Nixon GM, Kermack AS, Davis GM, Manoukian JJ, Brown KA, Brouillette RT. Planning adenotonsillectomy in children with obstructive sleep apnea: the role of overnight oximetry. Pediatrics. 2004;113(1 Pt 1):e19-25.
2. Brouillette RT, Morielli A, Leimanis A, Waters KA, Luciano R, Ducharme FM. Nocturnal pulse oximetry as an abbreviated testing modality for pediatric obstructive sleep apnea. Pediatrics. 2000;105(2):405-12.
3. Kaditis AG, Alonso Alvarez ML, Boudewyns A, Alexopoulos EI, Ersu R, Joosten K, et al. Obstructive sleep disordered breathing in 2- to 18-year-old children: diagnosis and management. Eur Respir J. 2016;47(1):69-94.
4. Marcus CL, Brooks LJ, Draper KA, Gozal D, Halbower AC, Jones J, et al. Diagnosis and management of childhood obstructive sleep apnea syndrome. Pediatrics. 2012;130(3):e714-55.
5. Lin SC, Davey MJ, Horne RS, Nixon GM. Screening for obstructive sleep apnea in children with Down syndrome. J Pediatr. 2014;165(1):117-22.
6. Coverstone AM, Bird M, Sicard M, Tao Y, Grange DK, Cleveland C, et al. Overnight pulse oximetry for evaluation of sleep apnea among children with trisomy 21. J Clin Sleep Med. 2014;10(12):1309-15.
7. Katsouli G, Polytarchou A, Tsaoussoglou M, Loukou I, Chrousos G, Kaditis AG. Nocturnal oximetry in children with obstructive lung disease or sleep-disordered breathing. Pediatr Pulmonol. 2019;54(5):551-6.
8. Muller NL, Francis PW, Gurwitz D, Levison H, Bryan AC. Mechanism of Hemoglobin desaturation during rapid-eye-movement sleep in normal subjects and in patients with cystic fibrosis. Am Rev Respir Dis. 1980 Mar;121(3):463-9.

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- McGill score positive predictive value is significantly lower in children with co-morbidities
- Children with co-morbidities with an abnormal McGill score should not be assumed to have OSA
- More detailed sleep studies are needed in children with co-morbidities and suspected OSA