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The risk for paediatric obstructive sleep apnoea in rural Queensland

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Background: The importance of assessing patients for paediatric obstructive sleep apnoea (OSA) cannot be more highly stressed and orthodontists may play an essential role in risk screening. The Paediatric Sleep Questionnaire (PSQ) is a validated tool to identify whether a child is at risk for paediatric OSA.
Objectives: The likelihood of paediatric OSA in school-aged children residing in Far North Queensland (FNQ) will be assessed using the PSQ.
Methods: Parents of children aged between 4 and 18 years were invited to participate through schools and social media messaging to complete an online PSQ questionnaire to assess their OSA risk and demographics.
Results: The final sample consisted of 404 school-aged children of whom 62.5% were found to be at a high-risk for paediatric OSA. The high risk was significantly associated with males and those of overweight/obese BMI status ($p < 0.001$). Race and age were not significant associations ($p > 0.05$).
Conclusions: Within the contributing sample of school-aged children in FNQ, a significant number were found to be at high-risk of paediatric OSA. Males and overweight/obese children were measured risk factors.
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Introduction

Paediatric obstructive sleep apnoea (OSA) is a sleep-related breathing disorder (SRBD) characterised by partial or complete obstruction of the airway leading to disturbances in breathing while asleep. Fully diagnosed paediatric OSA has a reported prevalence of 1–4% in school-aged children, while many additional children, suspected of suffering from OSA and other forms of SRBD, remain undiagnosed.[1–3] If left untreated, paediatric OSA may result in negative consequences related to behavioural problems, irritability, learning difficulties, a lack of concentration, an inability to focus, poor academic performance, nocturnal enuresis, a failure to thrive, compromised immunity and cardiovascular disease.[4–7]

Although there is a varied success rate, there are many available options for the management paediatric OSA. The first line of treatment is usually adenotonsillectomy but weight-loss, intranasal corticosteroids, nasal surgery, orthodontic management, and continuous positive airway pressure (CPAP) may be considered.[8–15] The type of treatment prescribed is dependent on the aetiology and the severity of the OSA. While adenotonsillar hypertrophy remains the most common OSA risk factor, other factors include obesity, asthma, allergic, or non-allergic rhinitis, chronic sinusitis, a

deviated nasal septum and neuromuscular conditions (including both hypotonia and hypertonia).[16] Children with craniofacial morphology consistent with maxillary and mandibular retrognathia, reduced maxillary dentoalveolar transverse widths, a steep mandibular plane angle and a vertical direction of growth may also place a patient at risk for paediatric OSA. Habitual snoring, disturbed sleep, mouth breathing, excessive daytime sleepiness, and hyperactivity may also be present.[17–19]

Overnight sleep laboratory-based polysomnography (PSG) is widely recognised as the "gold standard" in the diagnosis of OSA and other types of SRBD in children.[8,20] However, limitations related to the high cost, the inconvenience and discomfort for the parent and child to be hospitalised overnight, and long waiting times between referral and testing have made PSG impractical for routine clinical use and research purposes.[21] Several screening questionnaires have been developed to improve OSA detection so that the health system is not unnecessarily strained.[22] The Paediatric Sleep Questionnaire (PSQ) introduced by Chervin et al. has been validated against the gold standard of PSG, with a sensitivity of 0.85 and specificity of 0.87. It has been identified as a reliable screening tool to identify children at high risk of OSA who might then be referred to a paediatric sleep specialist for further investigation.[23] As a more robust screening tool, it implies better use of high level but limited resources.

Screening, diagnosing, and managing paediatric OSA requires an interdisciplinary and co-operative approach between the orthodontist and other medical professionals to optimise OSA care.[24] As many children are referred to orthodontic practices, there is an opportunity for the advantageous and early screening of patients to detect paediatric OSA. The orthodontist may be required to screen for OSA, identify underlying dentofacial relationships, and assist the physician in managing the disease. Because adenotonsillar hypertrophy is the most common aetiology, the simple recognition and documentation of tonsillar size during a comprehensive clinical examination can precipitate a referral to the patient's primary care physician or ear, nose, and throat (ENT) surgeon or a sleep medicine specialist. Orthodontists, as experts in the science of facial growth and development, are well suited to collaborate with physicians and other allied health providers in the management of OSA.[24]

The PSQ is simple survey which takes only 5 min to complete and may easily be implemented as part of a routine medical history for all paediatric patients. After a child receives a definitive diagnosis and management plan from the paediatric sleep specialist, the orthodontist may be further involved in managing and monitoring the patient in co-ordination with the sleep specialist and other medical professionals.

To date, the prevalence of paediatric OSA in far North Queensland (FNQ) has not yet been reported. It is known that children living in regional, rural, and remote areas of Australia do not have the same access to sleep medicine as children living in metropolitan areas. A 2015 study conducted to determine the incidence of OSA in adults at Cairns and Alice Springs Hospitals concluded that the rates of access to diagnostic sleep study tests were significantly lower for regional, rural and remote populations than for those in metropolitan areas.[25] Furthermore, at Cairns Hospital, PSG is currently only available for people over 15 years of age and so children in Far North Queensland, who are under 15 years, are required to travel to Brisbane for OSA assessment.

Due to the lack of available PSG services for those under the age of 15 years in FNQ, it is suspected that there is a large number of undiagnosed children with OSA. Therefore, the aim of the present study was to assess the risk level of children in FNQ for OSA.

Method

Approval was obtained from the Queensland Government Research Inventory (QERI) and James Cook University's Human Research Ethics Committee (Ethics Approval Number H7699) to conduct this study. All of the schools in the Cairns region of FNQ (Table I) were emailed with information about the study and permission was sought from the principals to forward the study information sheet to the parents of the students at their respective school. The information sheet included a link that directed parents to complete the SurveyMonkey® website questionnaire. Advertisements through social media were also used to recruit FNQ participants Following the definition provided by the Queensland Government's Department of Education, the borders of FNQ were defined as the Torres Strait Islands in the North, Cardwell in the South and the Gulf of Carpentaria in the West.[26] Areas within this region are classified

Table I. All the schools in the Cairns region of Far North Queensland that were contacted for participation.

	Number of schools	Number of students
State Primary Schools	69	15,399
State High Schools	11	10,340
State Combined (Primary and High) Schools	16	11,597
Private Primary Schools	24	5,917
Private High Schools	8	3,793
Private Combined (Primary and High) Schools	9	5,579
Total	137	52,625

as outer regional Australia, remote Australia, and very remote Australia (Figure 1).

The inclusion criteria consisted of school-aged children between the ages of 4 and 18 years who were of good general health. Children with a medical history consistent with craniofacial anomalies such as cleft lip and/or palate, craniosynostosis, Pierre Robin sequence, and Down's syndrome were excluded from the sample.

An online questionnaire was created on SurveyMonkey® (Survey Monkey, San Mateo, CA). The first page of the questionnaire contained information about the study and asked the respondent for consent to participate. The second page included the Paediatric Sleep Questionnaire (PSQ) which was a series of questions defined by a validated sleep breathing disorder screening tool (Figure 2). The tool consisted of 22 questions related to night-time and sleep behaviour (snoring), sleepiness, and daytime behaviour (hyperactivity and inattentiveness). Each question required either a "yes", "no," or "don't know" answer. Each "yes" answer was assigned a score of 1, and each "no" or "do not know" answer was assigned a score of 0. The number of "yes" responses was divided by the total number of answered questions. The PSQ interpretation guideline indicated that positive replies over 33% of the total answers were suggestive of a high risk of OSA. Hence, children at a high risk of OSA were identified as having 33% or more of the answers as "yes", which was a score of 0.33 or higher. Additional questions were also included on the second page and provided data related to the child's age, gender, race, medical history, height, and weight. All data collected were strictly anonymous. A pop-up message appeared on the screen at the end of the

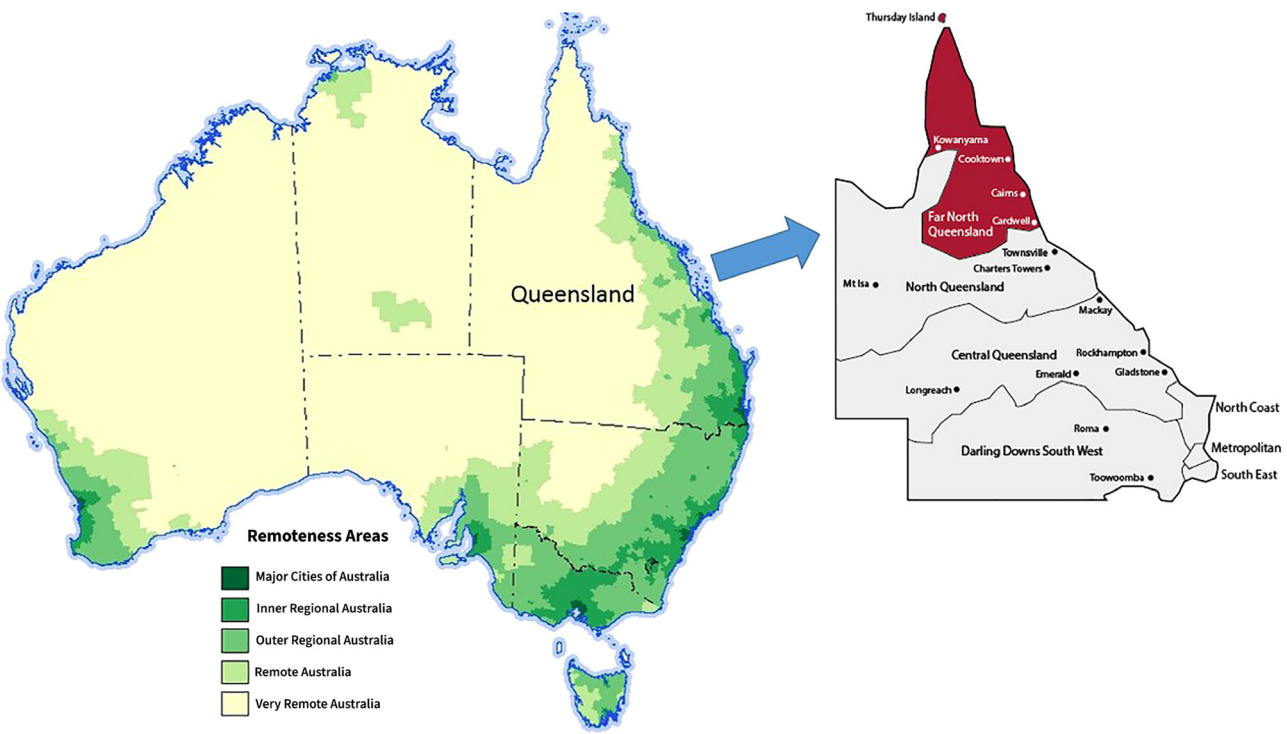

Figure 1. Map of the 2016 Remoteness for Australia. Blue arrow shows the Far North Queensland region. Retrieved from: https://www.abs.gov.au/websitedbs/D3310114.nsf/home/remoteness+structure. https://www.health.qld.gov.au/mass/subsidy-schemes/rural-remote.

Paediatric Sleep Questionnaire	
Please answer the following questions about your child for the past month.	
1. Please list any medical conditions your child has.	
2. What is your child's age?	
3. What is your child's gender?	
4. What is your child's current height?	
5. What is your child's current weight?	
6. Please describe your child's race/ethnicity.	
7. When sleeping, does your child snore more than half the time?	Yes / No / Don't know
8. When sleeping, does your child always snore?	Yes / No / Don't know
9. When sleeping, does your child snore loudly?	Yes / No / Don't know
10. When sleeping, does your child have "heavy" or loud breathing?	Yes / No / Don't know
11. When sleeping, does your child have trouble breathing or struggle to breathe?	Yes / No / Don't know
12. Have you ever seen your child stop breathing during the night?	Yes / No / Don't know
13. Does your child tend to breathe through the mouth during the day?	Yes / No / Don't know
14. Does your child have a dry mouth on waking up in the morning?	Yes / No / Don't know
15. Does your child occasionally wet the bed?	Yes / No / Don't know
16. Does your child wake up feeling unrefreshed in the morning?	Yes / No / Don't know
17. Does your child have a problem with sleepiness during day?	Yes / No / Don't know
18. Has a teacher commented that your child appears sleepy during the day?	Yes / No / Don't know
19. Is it hard to wake up your child up in the morning?	Yes / No / Don't know
20. Does your child wake up with headaches in the morning?	Yes / No / Don't know
21. Has your doctor or paediatrician advised you that your child stop growing at a normal rate at any time since birth?	Yes / No / Don't know
22. Is your child overweight?	Yes / No / Don't know
23. My child often does not seem to listen when spoken to directly.	Yes / No / Don't know
24. My child often has difficulty organising tasks and activities.	Yes / No / Don't know
25. My child often is easily distracted by extraneous stimuli.	Yes / No / Don't know
26. My child often fidgets with hands orfeet or squirms in seat.	Yes / No / Don't know
27. My child often is "on the go" or often acts as if "driven by a motor".	Yes / No / Don't know
28. My child often interrupts or intrudes on others (e.g. butts into conversations or games).	Yes / No / Don't know

Figure 2. Paediatric Sleep Questionnaire (PSQ).

online survey for children identified to possibly be at a high risk of OSA and recommending a visit their local health professionals for further assessment.

As reported by parents, the BMI score was calculated by dividing the child's weight in kilograms by the square of the child's height in metres. The BMI category was then determined by using the appropriate BMI score cut-off points according to the child's age and gender.[27]

Statistical analysis

All data were analysed using IBM SPSS Statistics for Windows (version 26.0; IBM Corp., Armonk, NY, USA). The Pearson Chi-Square test was applied to examine the risk categories by gender, age, and race. The risk categories were analysed by age and BMI by using the Mann–Whitney test. Logistic regression analysis was applied to predict risk based on gender, age, and race. Statistical significance was set at $p < 0.05$ for all analyses.

Results

A total of 697 replies were received from the online questionnaire. Of these, 290 replies were excluded due to incomplete data. Three additional entries were excluded after applying the exclusion criteria. A final sample size of 404 entries was analysed.

The subject sample consisted of 178 (44%) females and 222 (55%) males. Four subjects (1%) did not specify their gender. The largest racial category was Caucasian (79.0%), followed by Asians (6.9%), Aboriginal and Torres Strait Islanders (ATSI) (5.0%), not reported (4.2%), Pacific Islanders (3.5%), and other (1.5%).

A total of 251 (62.1%) subjects were at high risk for paediatric OSA as identified by the PSQ results.

A significant association was found between gender and risk category, as a higher proportion of males scored in the high-risk category compared with females ($\chi^2 = 15.323$, $p < 0.001$) (Table II). Significant differences were also found between multiple individual questions (i.e. inattention, snoring, breathing, and bed-wetting) based on gender, with males found to be consistently and significantly higher than females (Table III).

Table II. Association between gender and risk category.

Gender	Low risk	High risk
Male	65 (29%)	157 (71%)
Female	86 (48%)	92 (52%)

A Mann–Whitney test determined no significant difference in age between the low risk (Median = 11, IQR = 6) and high risk (Median = 9, IQR = 7) groups ($p = 0.08$). No significant associations were found between age and scores on any outcome variable groupings (all p values > 0.05).

No significant association was found between race and risk category ($\chi^2 = 8.94$, $p = 0.063$). A high proportion of ATSI and Pacific Islander children fell into the high-risk category (Table IV). There were no significant associations between race and the grouped outcome variables (all p's > 0.05).

A significant association was found between BMI and risk category ($\chi^2 = 25.91$, $p < 0.001$). A higher number of participants, who were classified as overweight/obese, were in the high-risk category (Table V). When age was categorised into ≤11 years and teenagers (12–18 years), there was a significant association between age and risk category, $\chi^2 = 4.81$, $p < 0.05$. A higher number of younger individuals were in the high-risk category (Table VI).

Logistic regression was used to predict paediatric OSA's risk based on gender, age, BMI, and race (Table VII). Gender was predictive, as males were significantly more likely to be classified as high risk compared with females. BMI category was also a significant predictor, with those in the overweight/obese category more likely to be at high risk than those who were underweight. Age and race were not found to be significant predictors of risk category ($p > 0.05$).

Discussion

The percentage of school-aged children in FNQ found to be at high risk of having OSA was 62.1%. Males were 1.37 times more likely than females to be at high risk, and overweight/obese children were 1.53 times more likely than children of normal BMI and 1.62 times more likely than underweight children to be at high risk. No significant association was found between age and paediatric OSA risk. The current findings agree with the paediatric OSA meta-analysis of Lumeng and Chervin,[3] which concluded that OSA was significantly associated with the male

Table III. Association between gender and risk category.

Variable	Male mean (SD)	Female mean (SD)	*t*-value	*p*-value
Inattention	4.05 (1.92)	2.91 (2.10)	−5.57	<0.01
Snoring frequency	0.57 (0.80)	0.42 (0.68)	−2.03	<0.05
Snoring quality	0.82 (0.73)	0.60 (0.70)	−2.96	<0.01
Breathing	0.34 (0.64)	0.22 (0.52)	−2.13	<0.05
Mouth breathing	0.84 (0.79)	0.88 (0.82)	0.54	0.57
Daytime sleepiness	1.87 (1.32)	1.89 (1.27)	0.05	0.96
Bed wetting	0.35 (0.48)	0.20 (0.40)	−3.49	<0.01
Total	9.22 (4.19)	7.52 (4.08)	−4.06	<0.01

Table IV. Association between race and risk category.

Race	Low Risk	High Risk
Caucasian	119 (37.3%)	200 (62.7%)
Asian	15 (54%)	13 (46%)
Indigenous	6 (30%)	14 (70%)
Pacific Islander	2 (14%)	12 (86%)
Other	4 (67%)	2 (33%)
Not reported	7 (41.2%)	10 (58.8%)

Table V. Association between BMI and risk category.

BMI category	Low risk	High risk
Underweight	26 (50%)	25 (50%)
Normal	88 (47%)	98 (53%)
Overweight/obese	21 (19%)	88 (81%)

Table VI. Association between age and risk category.

	Risk	
Age	Low	High
≤11 years	88 (58)	63 (42)
12+ years	172 (69)	77 (31)

gender and high BMI scores. However, there was insufficient evidence to correlate age with paediatric OSA. Although there was an expectation that many children in FNQ might be at OSA high risk, the result (62.1%) was much higher than anticipated. The prevalence of high-risk OSA varies considerably within the reported literature but recent studies indicated a prevalence between 7.3 and 33.3%.[28–33]

There are several possible reasons for the current high-risk prevalence. The first explanation is the large number of male (55%) and overweight/obese (27%) study respondents, as males and high BMI scores are significantly associated with OSA. A total of 58 subjects (14.4%) did not provide sufficient data to calculate their BMI, and so it is possible that the actual percentage could further change. The second and more likely reason for the inflated high-risk findings is perhaps due to selection bias related to the method of recruiting participants. Parents of children experiencing signs and symptoms of paediatric OSA may have had a vested interest in completing the questionnaire and were, therefore, more likely to participate in the study. Parents were informed about the importance of early diagnosis and paediatric OSA management before completing the questionnaire which may have led to biased responses. Most participants may have completed the questionnaire because they thought their child might be at risk of having paediatric OSA. It may also be argued that, due to the likelihood that the parents were initially unaware of SDB, the condition may have been brought to their attention by a comment in the survey which noted its importance. These possible biases were considered; however, disclosing the questionnaire's content is mandatory to achieve participant consent and is required by Human Ethics Committees.

The association between race and paediatric OSA risk was highlighted by a higher percentage of Pacific Islanders (86%) and ATSI children (70%) found to be at high-risk of having OSA compared to Caucasian children (62.7%). In comparison, a smaller percentage of Asian children (46%) were at high-risk. Although it would appear that Pacific Islanders and ATSI children were more likely to have paediatric OSA, a statistically significant association between

Table VII. Logistic regression coefficients.

Predictors	Levels	OR (95% CI)	*p*-values
Gender	Female	REF	
	Male	2.44 (1.51–3.94)	0.001
Age		0.95 (0.90–1.02)	0.097
BMI	Underweight	REF	
	Normal	1.19 (0.62–2.27)	0.121
	Overweight/obese	4.77 (2.23–10.22)	<0.001
Race	Caucasian	REF	
	Asian	0.58 (0.25–1.36)	0.234
	Indigenous	1.10 (0.33–3.65)	0.449
	Pacific Islander	3.49 (0.72–16.89)	0.118
	Other	0.37 (0.04–3.77)	0.577

race and OSA risk was not identified likely due to the small sample size of participating children from non-Caucasian self-declared race categories. In contrast, studies in the U.S. have established a correlation between race and paediatric OSA, with African American children at greater risk than Caucasian American children.[34–36] However, there is currently limited research on the relationship between race and paediatric OSA in Australia. In adults, a significant association has been found between ATSI status and OSA.[37] Obese ATSI children have also been found to be at a greater risk of having OSA than non-ATSI obese children.[38] However, a study on primary school children in the Northern Territory did not find a significant association between race and SRBD, with similar rates of sleep breathing problems identified in both ATSI (3%) and non-ATSI (2%) groups.[39]

As a possible implication of the present findings, it has been suggested that there is a significant association between OSA and poor school performance,[40] and OSA management is associated with improved school performance. A U.S. study conducted on children with OSA in the bottom ten percentile of their class found that school performance improved in children following adenotonsillectomy. In contrast, school performance did not improve in children who remained untreated.[41] In Australia, the National Assessment Program Literacy and Numeracy (NAPLAN) examinations assess the school performance of students across the country. Each year, students in regional, rural and remote areas attain lower average grades compared with students in metropolitan areas for all domains tested (writing, reading, spelling, grammar and punctuation, and numeracy) and across all year levels. It is possible that barriers to accessing appropriate medical services for paediatric OSA in regional, rural, and remote areas may contribute to the lesser school performance of children in these communities compared with their counterparts in major cities.

The limitations of the present study include inherent recall risk as reliance was entirely on parent-reported data. Understanding the specific question may have played a role in the type of recorded answer. There was no opportunity to request clarification if required, as the data were collected online. There is a further possibility that English literacy could have also been a barrier in some rural or remote areas where indigenous communities' first language is often not English. To the extent of current knowledge, the PSQ questionnaire has not been validated for this specific population. As school-aged children are likely to sleep in a separate bedroom from their parents, some signs and symptoms of sleep-disordered breathing may have been overlooked. Additionally, BMI categories calculated from parent-reported weight and height data have been found to have low accuracy as parents are prone to underestimating children's weight and height data.[42,43] The same limitation might apply to the information related to medical conditions. However, to comply with the Ethics Committee requirements, the questionnaire's content and purpose were disclosed before obtaining consent from study participants. The parents were first informed about the signs and symptoms of paediatric OSA and the importance

of early management before completing the PSQ questionnaire. Therefore, most participants may have completed the questionnaire because they believed that their child might be at risk of paediatric OSA. A final limitation is that the PSQ focuses exclusively on identifying the risk for OSA which is at one end of the SRBD spectrum. The current number of high-risk cases who have a specific type of SRBD is unknown.

In conclusion, within the current sample of school-aged children (4–18 years old) in FNQ, it was found that 62.5% of the respondents were at risk of having OSA. Males and overweight/obese children were at an increased risk. Further studies with a larger sample size of Pacific Islander and ATSI children are recommended to explore the relationship between race and paediatric OSA. The PSQ is a relatively easy and validated tool to screen children for OSA and be simply incorporated into general dental practice. The PSQ is a valuable first step in screening patients to determine those who require further investigations to confirm a diagnosis of OSA. Timely referral, a confirmatory diagnosis, and focused OSA management may improve the quality of life and enhance a child's academic performance.

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Conflict of Interest

The authors declare no conflict of interest.

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