The sound of sleeping soundly? A review on the natural history and impairments of paediatric primary snoring

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Abstract

Background: Primary snoring (PS) is a common form of sleep disorder breathing among Hong Kong children. Often deemed as harmless, paediatric PS is underreported and undermanaged. This study aims to review the natural history and consequences of paediatric primary snoring. Method: After selecting studies that compare PS children with healthy controls, 56 publications and other related papers were retrieved from PubMed, and analysed. Results: Pediatric PS has severe detrimental effect on cardiovascular, neurocognitive, and neurobehavioural health, as well as quality of life of the family. It does not remit naturally and may progress into obstructive sleep apnoea.

Keywords: Children, Impairment, Natural history, Primary snoring

Introduction

In Hong Kong, the sight of a child snoring and purring gently in slumber is often seen as an endearingly cute view – after all, snoring is probably an indicator of the child being soundly asleep, frolicking in some faraway dreamy wonderland, isn’t it? The answer is a definite no; the public often underestimates the gravity and the implicit health consequences related to the seemingly innocent act of snoring, as suggested by studies demonstrating parents’ general attitude of indifference towards snoring, as well as an underreporting of the symptom by parents under clinical setting.1,2

Snoring, which is the sound produced by vibration of upper airway soft tissue during sleep,3 is indicative of a spectrum of respiratory disorders known as sleep-disordered breathing (SDB). Based on the ascending severity of related apnoeas (cessation of airflow) and hypopnoeas (reduction of airflow), SDB can be classified into primary snoring (PS), upper airway resistance syndrome (UARS) and obstructive sleep apnoea (OSA), where PS is characterised by snoring without episodes of apnoea or hypoventilation.3 Literature has extensively shown that OSA and UARS in childhood can lead to disturbances in sleep architecture,4,5 cardiovascular complications,6,7 psychosocial problems,8 neurocognitive deficits,9-13 behavioural issues14-16 and lower quality of life.17-19 These health impacts are mostly thought to be consequences of the apnoea-induced intermittent hypoxia in these more serious forms of SDB. However what we must ask is whether paediatric primary snoring, without the obviously debilitating apnoea and hypopnoea, has been empirically proved to have similar insidious sequelae – as even physicians commonly pass paediatric primary snoring as something benign and unremarkable.1

The prevalence of habitual snoring reported around the world differs to some extent, due to the varying definitions of habitual snoring, the methodology (parental report as opposed to overnight ambulatory monitoring), as well as the heterogeneity of sample groups in terms of age, body size, et cetera. However, the general reported figure is generally around 3 to 10 percent in most reports.1,20-25 One telephone survey study covering 3047 school children performed in Hong Kong demonstrated that our city’s children are particularly affected by habitual snoring, quoting a startling figure of 10.9%,24 evidently on the higher spectrum when compared to similar studies in other communities. It has been suggested that the pathogenesis of SDB and snoring is related to sleep deprivation,26 anxiety, and stress level.27 Incidentally, Hong Kong school children are notoriously known for their shorter sleeping hours when compared to that of their foreign counterparts,24 as well as heightened anxiety level, parental stress and academic workload28 as a result of the highly competitive educational system, which imposes extended hours of
school-related work, supplementary classes, and extracurricular requirements. These eventually render Hong Kong children more prone to snoring, and more vulnerable to the health hazards of various forms of SDB, including PS.

Another notable point is that among children who experience habitual snoring, more are affected by PS instead of the more serious forms of SDB. Should PS indeed carry detrimental paediatric health consequences—and given the fact that within the already prevalent Hong Kong paediatric snoring population, most children are PS patients— it would imply a tremendous gravity of the issue, which is further made worse by physicians’ common attitude of considering PS as a benign condition.

In an effort to investigate the potential consequences of paediatric PS, articles covering natural history and impairments of the condition were retrieved and reviewed from PubMed, using the keywords “primary snoring” and “child”, with the keyword “snoring/aetiology” excluded to limit the results, and with the limit “humans” activated. Out of the 107 suggested articles, studies that do not differentiate between different classes of SDB were excluded, ultimately yielding 56 papers. These papers and other related studies were reviewed and analysed. The analysis are summarised and described as follows.

Natural history of paediatric primary snoring

While multitudes of studies have discussed the natural history of OSA if left untreated, number of papers looking into the natural course of unmanaged PS remains scarce, as PS is often deemed unwarranted for treatment. A total of 6 cohort studies were identified that contain data on the natural progression of PS in children without submitting for treatment, with comparable scoring criteria for the condition, using either the apnoea hypopnoea index (AHI), apnoea index (AI), or the obstructive apnoea hypopnoea index (OAHI) as measured by overnight polysomnography (PSG) in the follow-up visits. AHI measures the number of obstructive apnoeas, mixed apnoeas, and hypopnoeas per hour of sleep; AI measures the number of obstructive apnoeas and mixed apnoeas per hour of sleep; while OAHI measures obstructive apnoeas and hypopnoeas. PS is identically defined as an index <1 in snorers, while some of the study include the oxygen saturation (\( \text{SpO}_2 \)) as another parameter. The findings are summarised in Table 1.

Regardless of age, all of these cohort studies indicate that majority of PS does not remit naturally without treatment, as many of the studies highlighted that there are no significant change in the PS group as a whole. This result resonates with some other SDB cohort studies that demonstrate paediatric habitual snoring does not naturally remit without treatment in 1 year and 5 years. As majority of SDB cases are PS, the two studies can further support the continuous nature of PS. This fact justifies the necessity of greater clinical attention on PS, because if not managed and treated, majority of PS cases will persist, leading to impairments discussed in the later sections of this review.

Cardiovascular effects

Based on a clinical review published in 2013, children with SDB exhibit autonomic dysfunction very much like that seen in adult SDB patients; these children have elevated systolic and/or diastolic blood pressure (BP), impaired baroreflex sensitivity, and elevated generalised sympathetic activity. It is most important to determine whether children with PS are also exposed to these cardiovascular conditions; a predisposition at an early age like this is highly unfavourable as it suggests earlier and higher occurrence of cardiovascular and cerebrovascular events in adulthood. Should the correlation be positive, then PS should be actively managed in order to promote cardiovascular health of the children.

A total of 6 researches comparing BP of a group of PS children and a group of healthy controls were retrieved. Their findings are summarised in Table 2.
Table 1. Summary of literature on natural history of primary snoring

<table>
<thead>
<tr>
<th>Author</th>
<th>Region</th>
<th>Age at PS subjects</th>
<th>Number of subjects</th>
<th>Study design</th>
<th>Scoring criterion</th>
<th>Natural History</th>
</tr>
</thead>
<tbody>
<tr>
<td>Barros et al. (2014)²⁰</td>
<td>Brazil</td>
<td>2-12</td>
<td>10</td>
<td>Cohort; Follow up after 6 months</td>
<td>AHI based on overnight PSG assessment</td>
<td>No significant change</td>
</tr>
<tr>
<td>Vlahandonis et al. (2013)²³</td>
<td>Australia</td>
<td>7-13</td>
<td>22</td>
<td>Cohort; Follow up after 4 years</td>
<td>AHI based on overnight PSG assessment</td>
<td>55% of PS children remained in PS group; 9% of PS children progressed to OSA</td>
</tr>
<tr>
<td>Topol et al. (2013)²¹</td>
<td>USA</td>
<td>1-12</td>
<td>19</td>
<td>Cohort; Follow up after 3 years</td>
<td>9 out of 19 underwent overnight PSG assessment</td>
<td>No significant change in group; 1 in 9 of the children undergoing PSG assessment developed into OSA</td>
</tr>
<tr>
<td>Li et al. (2013)²²</td>
<td>Hong Kong</td>
<td>6-13</td>
<td>70</td>
<td>Cohort; Follow up after 4 years</td>
<td>OAHİ based on overnight PSG assessment</td>
<td>31.4% remained in PS group; 7.1% developed moderate to severe OSAS; 37.1% developed OSA</td>
</tr>
<tr>
<td>Anuntaseree et al. (2005)²⁶</td>
<td>Thailand</td>
<td>7</td>
<td>55</td>
<td>Cohort; Follow up after 3 years</td>
<td>AHI based on overnight PSG assessment</td>
<td>18.2% of PS children remained in PS group; 9.1% progressed to OSA</td>
</tr>
<tr>
<td>Marcus et al. (1998)²⁶</td>
<td>USA</td>
<td>1-15</td>
<td>20</td>
<td>Cohort; Follow up after 1-3 years</td>
<td>Apnea index based on overnight PSG assessment</td>
<td>No significant change in group as a whole; 2 out of 20 developed into mild OSAS</td>
</tr>
</tbody>
</table>

AHI: Apnoea-hypopnoea index; OAHİ: Obstructive apnoea-hypopnoea index; OSA: Obstructive sleep apnoea; OSAS: Obstructive sleep apnoea symptoms; PS: Primary snoring; PSG: Polysomnography

Table 2. Summary of literature on blood pressure and primary snoring

<table>
<thead>
<tr>
<th>Author</th>
<th>Region</th>
<th>Age at recruitment</th>
<th>Number of subject of interest</th>
<th>BP measurement methodology</th>
<th>Results</th>
</tr>
</thead>
<tbody>
<tr>
<td>Nisbet et al. (2014)²⁰</td>
<td>Australia</td>
<td>3-5</td>
<td>72 PS 38 Controls</td>
<td>PTT during sleep</td>
<td>Nocturnal dipping is preserved</td>
</tr>
<tr>
<td>Nisbet et al. (2013)²⁰</td>
<td>Australia</td>
<td>3-6</td>
<td>66 PS 36 Controls</td>
<td>PTT during sleep</td>
<td>No significant</td>
</tr>
<tr>
<td>Horne et al. (2013)¹¹</td>
<td>Australia</td>
<td>7-13</td>
<td>61 PS 36 Controls</td>
<td>Continuous BP monitoring during sleep with finger photoplethysmography</td>
<td>Nocturnal dipping is preserved</td>
</tr>
<tr>
<td>Horne et al. (2011)¹²</td>
<td>Australia</td>
<td>7-13</td>
<td>61 PS 36 Controls</td>
<td>Continuous BP monitoring during sleep with finger photoplethysmography</td>
<td>BP is elevated in both wake and sleep in PS</td>
</tr>
<tr>
<td>Li et al. (2009)¹³</td>
<td>Hong Kong</td>
<td>6-13</td>
<td>46 PS 56 Controls</td>
<td>ABP with oscillometric monitor during wake and sleep</td>
<td>BP is elevated in PS</td>
</tr>
<tr>
<td>Kwok et al. (2003)¹⁴</td>
<td>Hong Kong</td>
<td>9.5±2.8</td>
<td>30 PS 30 Controls</td>
<td>Oscillometric device during wake</td>
<td>BP is elevated in PS</td>
</tr>
</tbody>
</table>

ABP: Ambulatory blood pressure monitoring; BP: Blood pressure; PS: Primary snoring; PTT: Pulse transit time (inverse continuous indicator of BP change)
The results yielded in these studies are very interesting as they demonstrate that, firstly, an elevated BP can be found in children with PS in during waking and sleeping hours, and, secondly, nocturnal dipping – the 10-20% BP drop during sleep without which hypertension risk is increased – is preserved in children with PS. The former studies show that, just like OSA, paediatric PS has a direct physiological impact on the cardiovascular system causing higher mean, systolic, and diastolic BP in both daytime and slumber, regardless of age, gender and body size. The latter piece of information shows that, unlike adult SDB patients, paediatric PS children do not have reduced nocturnal BP dipping; this is particularly reassuring as it shows that children with PS have yet to be exposed to a strong hypertensive risk such as that suggested by nocturnal dipping. Such an absence suggests that at childhood, PS should be immediately dealt with while the cardiovascular impairments remain minor and serious consequences have not yet emerged.

Apart from BP, 2 studies on local Hong Kong children, both comparing a PS group with a healthy control group, have shown that PS disrupts normal endothelial function. Kwok et al showed that pulse wave velocity (PWV), and therefore the resting arterial tone, is increased in the PS group. As PWV is strongly associated with the presence and extent of atherosclerosis, PS children, if left unmanaged, are likely to have higher risk of cardiovascular mortality as adults. Li demonstrated that PS is independently associated with a reduced brachial flow mediated vasodilation (FMD). FMD is again strongly associated with higher risk of adverse cardiovascular events. Moreover, another study using conventional Doppler analysis demonstrated that early asymptomatic left ventricular myocardial dysfunction could be observed in PS children when compared to healthy controls. These three publications complement one another in showing that cardiovascular health in children can be severely jeopardised should paediatric PS not be corrected.

**Neurocognitive and behavioural effects**

In a group of 32 paediatric PS patients who underwent overnight PSG with continuous monitoring of cerebral oxygenation, it has been found that the normalised regional cerebral oxygenation (rSO$_2$) did not reach levels measured during wakefulness, when compared with a group of 14 healthy controls; the difference in rSO$_2$ between the two groups is significant in many of the sleep stages. With strong evidence that sustained decrease of rSO$_2$ is related to cognitive decline and poorer neurocognitive performances, we can extrapolate that children with PS may be exposed to a greater range of neurocognitive and behavioural deficits. Table 3 summarises the literature investigating the cognitive facet of the defects.

In all of these studies, children with PS and otherwise healthy controls were selected as subjects for comparison. With the exception of one study, all of these investigations report statistically significant weaker cognitive measurements in children with PS. In short, paediatric PS has a negative impact on the intelligence, academic performance, memory, language ability, learning ability, and reasoning ability of children. These associations can be attributed to the disrupted sleep cycles in PS patients. The increased upper airway resistance and inflammatory effects of the snoring process might have also played a part in the pathogenesis.

The neurocognitive sequelae depicted by the above articles are extremely alarming, as children around this age (<12 years) are going through a period of rapid cerebral and functional development, making them particularly vulnerable to pathological effects; any poor neurological outcome are also likely to be irreversible in nature. The irony of this situation in Hong Kong is marked – as many school children were possibly instructed to sacrifice sleep for academic development, an action that potentiates them to PS and the related detrimental neurocognitive deficits, ultimately impeding academic performance.

Chervin is one of the first to report snoring as a predictor of hyperactivity behaviours. Extensive evidence now demonstrates that children with PS show a significantly greater rate of behavioural problems than healthy controls. Table 4 contains the findings of studies directly comparing paediatric PS patients with control groups. Majority of the studies find a statistically significant relationship between paediatric PS and issues such as poor self-control, poor flexibility, poor execution, inattention, hyperactivity, and so on. The only study that shows no correlation between inattention and PS adopted a test that is not as empirically validated as the ones used in other studies, and therefore the results are not directly comparable.

Readers should note that when comparing scores of PS group with OSA group, Jackman et al discovered that PS children exhibit generally more problematic...
behaviour than children with the more severe forms of SDB such as OSA. While further replication and verification of such data is required, it further shows that PS is a highly harmful entity, and may even surpass OSA in the causation of certain neurobehavioural deficits.

**Quality of life**

Apart from the aforementioned poor specific functional outcomes, the condition of paediatric PS also has a general impact on the quality of life (QOL) of not only the child, but also the parents and the family. Be it self reported by the child or reported by the parents, the impacts and disturbances imposed by PS in children are reported to be very noticeable and affect the family as a whole.

The fact that PS is, at the very least, a condition that warrants as much attention as OSA, is again reiterated by these studies. The degree of impact of PS on QOL (based on QOL questionnaire scores) has been shown to be statistically identical to that of OSA, in other words, SDB of all severities impose at least similar level of QOL deterioration. Jackamn et al once again shows that, after accounting for other factors, it is possible that PS patients experience lower QOL than patients with more severe forms of SDB, an effect which, if true, makes management of PS a greater predictor in QOL improvement than management of other SDB diseases.

### Table 3. Summary of literature on primary snoring, and neurocognitive performance

<table>
<thead>
<tr>
<th>Author</th>
<th>Region</th>
<th>Age at recruitment</th>
<th>Number of subject of interest</th>
<th>Measured parameter and methodology</th>
<th>Results</th>
</tr>
</thead>
</table>
| Jackman et al. (2012)
| Australia           | 3-5              | 60 PS 37 Controls  | - IQ with ABIQ                   | - Minimal differences between cognitive parameters between PS & controls                                                          |
| Brockmann et al. (2012)
| Germany             | 6-12             | 69 PS 410 Controls | - Academic performance based on percentile in maths, science & spelling                            | - Statistically significant higher percentage of poor school performance in the three investigated subjects in PS                   |
| Miano et al. (2011)
| Italy               | 9.1±2.4          | 13 PS 60 Controls  | - IQ with WISC-III               | - Statistically significant lower VIQ, PIQ & FSIQ in PS when compared to controls                                                      |
| Bourke et al. (2010)
| Australia           | 7-12             | 59 PS 35 Controls  | - IQ with WASI; - Academic function with WRAT-3; - Memory with COWAT                             | - Statistically significant lower VIQ & FSIQ was noted in PS - higher rate of academic impairment in PS                                |
| Hamasaki et al. (2007)
| Brazil              | 6-12             | 37 PS 20 Controls  | - Learning ability & memory with RAVLT - IQ with coding & digit subtest from WISC-III         | - Statistically significant lower test performances in PS when compared to controls                                                |
| O’Brien et al. (2004)
| USA                 | 5-7              | 87 PS 31 Controls  | - Reasoning & conceptual ability with DAS - Memory, language, visuospatial processing & learning with NEPSY | - Statistically significant impairment on overall cognitive measures, especially language and visuospatial processing |

Conclusions

Primary snoring is a common condition found in children and our locality of Hong Kong is particularly afflicted by it, possibly as a result of the unique childhood lifestyle. Despite its prevalence, pathogenesis and complications of PS remain under-researched; these correlations should be widely investigated, on the note that children are most vulnerable to the effects of primary snoring due to their developing respiratory and neurological systems, as well as the potential irreversibility of the deficits.

Based on the reviewed articles, it can be safely concluded that PS in children is never the benign entity physicians once thought – as many still do – it was.56 International researches, as well as many local studies, have demonstrated that PS itself is a strong determinant for cardiovascular deficits, neurocognitive and behavioural abnormalities. More children with PS experience higher blood pressure, disrupted endothelial and heart functions, lower intelligence, worse memory, poorer academic performance, lower attention, and more behavioural issues, when compared with healthy children; and all these ultimately extend to affect the entire family unit, causing a general diminution of the household's quality of life. Given the natural history that majority of PS cases do not resolve without management, and that it is even possible for it to progress into OSA, this author believes that reporting

<table>
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<tr>
<th>Author</th>
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</tr>
</thead>
<tbody>
<tr>
<td>Jackman et al. (2013)</td>
<td>Australia</td>
<td>3-5</td>
<td>56 PS 38 Controls</td>
<td>Behavioural problems with BRIEF-P &amp; CBCL/1.5-5</td>
<td>- PS children show occasional scores showing behavioural problems e.g. poorer self control, poorer global execution, after controlling for SRI</td>
</tr>
<tr>
<td>Jackman et al. (2012)</td>
<td>Australia</td>
<td>3-5</td>
<td>60 PS 37 Controls</td>
<td>Attention with NEPSY Behavioural problems with BRIEF-P, CBCL/1.5-5, C-TRF &amp; BAS-II</td>
<td>- Statistically significant more dysexecutive and problematic behaviour than controls</td>
</tr>
<tr>
<td>Brockmann et al. (2012)</td>
<td>Germany</td>
<td>6-12</td>
<td>69 PS 410 Controls</td>
<td>Attention and hyperactive activity with ADHD rating scale</td>
<td>- Statistically significant higher ADHD rating scale mean scores for PS than controls</td>
</tr>
<tr>
<td>Miano et al. (2011)</td>
<td>Italy</td>
<td>9.1±2.4</td>
<td>13 PS 60 Controls</td>
<td>Attention and hyperactive activity with ADHD rating scale</td>
<td>- Statistically significant higher ADHD rating scale mean scores for PS than controls</td>
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<td>Bourke et al. (2011)</td>
<td>Australia</td>
<td>7-12</td>
<td>59 PS 35 Controls</td>
<td>Behavioural problems with BRIEF &amp; CBCL</td>
<td>- Statistically significant higher rate of behavioural problems in PS than controls</td>
</tr>
<tr>
<td>Hamasaki et al. (2007)</td>
<td>Brazil</td>
<td>6-12</td>
<td>37 PS 20 Controls</td>
<td>Attention with Mezulam's symbol cancelling test &amp; letter cancelling test</td>
<td>- No statistical significant difference between PS &amp; OSA control in attention tests</td>
</tr>
</tbody>
</table>

ABAS-II: The Adaptive Behaviour Assessment System - Second Edition; ADHD: Attention Deficit Hyperactivity Disorder; BRIEF: The Behaviour Rating Inventory of Executive Function; BRIEF-P: The Behavior Rating Inventory of Executive Function - Preschool Version; C-TRF: the Caregiver-Teacher Report Form for Ages 1.5-5; CBCL: Child Behaviour Checklist; CBCL/1.5-5: the Child Behavior Checklist 1.5-5; NEPSY: Developmental Neuropsychological Assessment; SRI: Social Risk Index; OSA: Obstructive Sleep Apnoea; PS: Primary Snoring
and management of paediatric PS should be encouraged in parents and physicians.

This review accentuates the urgency for public education on the opaque health detriment primary snoring presents to our children — each and everyone should recognise that loud sleeping sound is not a sign of sound sleeping. Snoring calls for medical attention and management, as it is only without it, can our children truly sleep in a healthy and vitalising slumber.

References


