Early intervention for obstructive sleep apnoea in Down syndrome – making a difference?

Indra Narang[a,b,c,*] *and Chun Ting Au*[a]

[a]Translational Medicine, Research Institute, The Hospital for Sick Children, Toronto, Ontario, Canada
[b]Division of Respiratory Medicine, The Hospital for Sick Children, Toronto, Ontario, Canada
[c]Department of Paediatrics, Temerty Faculty of Medicine, University of Toronto, Ontario, Canada

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Obstructive sleep apnoea is a common condition in children with Down syndrome, with reported prevalence rates ranging from 55 to 98%.[1,2] Untreated obstructive sleep apnoea is associated with a range of adverse outcomes, including pulmonary hypertension, cognitive impairments, behavioural problems[3] and impact on quality of life for children and their families. Screening and early intervention for obstructive sleep apnoea in children with Down syndrome is believed to negate its negative impact, and current guidelines recommend universal polysomnography (PSG) screening by the age of 4 years.[4] However, given that these recommendations are not evidence-based, the optimal timing for screening and intervention for obstructive sleep apnoea in children with Down syndrome remains an important unanswered clinical question. A recent prospective, non-randomised interventional study published in *The Lancet Regional Health—Europe* is one of the first studies to explore the impact of early treatment for obstructive sleep apnoea on neurocognitive development and behaviour in children with Down syndrome.[5]

In this study by Fauroux and colleagues,[5] 40 infants with Down syndrome were enrolled at 6 months of age and underwent regular PSGs every six months until 36 months (Screened Group) with treatment for obstructive sleep apnoea once diagnosed. These children were compared to a group of 40 children with Down syndrome receiving standard care who were enrolled at 36 months of age and underwent a PSG only at 36 months (Standard Care Group). The primary outcome was the Global Quotient of Development (GQD) of the Griffiths Scales of Child Development (Griffiths III) at 36 months, the gold standard for child development testing to 6 years of age although it has not been validated for children with Down syndrome. At 36 months, 14/29 (48.3%) and 1/40 (2.5%) participants had been treated for obstructive sleep apnoea primarily with upper airway surgery in the Screened and Standard Care Groups respectively. Not surprisingly, the frequency of moderate-to-severe obstructive sleep apnoea was lower, 11% vs 46% in the Screened vs Standard Care groups respectively although the frequency of mild persistent obstructive sleep apnoea was very high in the Screened Group (82%). Importantly, the study observed that the Screened Group had higher GQD (median difference = 4.1 [95% CI: 1.3–7.6]), indicating better overall neurodevelopment compared to the Standard Care group. Such results imply that early diagnosis and management of obstructive sleep apnoea may offer long-term benefits such as improvements in social-emotional development, learning, and communication skills. However, while these results are statistically significant, the clinical significance is unclear. A study evaluating the test-retest reliability of GQD over two assessments spaced 2–4 weeks apart reported an intra-class correlation coefficient of 0.991.[6] Given the GQD's standard deviation of 16.8,[6] the minimal detectable difference of GQD was 4.43,[7] which exceeds the point estimate of the between-group difference observed in the trial which may fall within the range of random measurement error. Additional bias was that the neuropsychologists performing the assessments were not only known to the participants as part of their care but were also aware of the participants' group allocation.

Importantly, these results do highlight that there is a high frequency of obstructive sleep apnoea prior to 3 years of age and it would be prudent to screen all children with Down syndrome for obstructive sleep apnoea from 6 months of age. What remains uncertain is whether early and targeted intervention or the intervention per se (regardless of timing) influenced the results observed, as the Screened Group received treatment for obstructive sleep apnoea at various time points within the first 36 months, targeting those with moderate-to-severe disease. Moreover, other unmeasured factors, including variations in socioeconomic status, the likelihood of receiving adjunct therapies and sleep hygiene counselling related to regular monitoring, may have also confounded the study findings. To disentangle the impact of timing and targeted therapeutics, a longitudinal controlled study with interventions at specific time points and long-term neuropsychological follow-up is required to determine whether early diagnosis and treatment of obstructive sleep apnoea during infancy offer additional long-term benefits for children with Down syndrome. This is an important consideration as the risks of airway surgery in

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*Corresponding author. The Hospital for Sick Children, 555 University Avenue, Toronto, Ontario, M5G 1X9, Canada.
E-mail address: indra.narang@sickkids.ca (I. Narang).

early life should be carefully weighed against the potential long-term benefits. Further, surgical risks should be evaluated alongside the benefits of watchful waiting and the natural resolution/improvement in obstructive sleep apnoea in children with Down syndrome especially given that a significant proportion of these children have moderate-to-severe disease before one year of age.[1,8]

This study underscores the importance of diagnosing obstructive sleep apnoea; however, there is a global lack of sleep diagnostic testing compounded by inequities in accessing existing sleep services.[9,10] There is an urgent need for a diagnostic tool that is low-cost, accessible, scalable, easy to use, and accepted by Otolaryngologists to ensure that every child with Down syndrome has the opportunity for targeted management of obstructive sleep apnoea to optimise long-term outcomes.

Contributors

Indra Narang and Chun Ting Au drafted the initial manuscript, revised and edited it, and approved the final version as submitted.

Declaration of interests

All authors have nothing to disclose.

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