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ORIGINAL ARTICLE

Establishing a telehealth model addressing paediatric sleep health in remote and rural Northern Territory Australia: Overcoming the distance barrier

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Aim: This study examined the outcomes of a telehealth model for sleep health assessment among Indigenous and non-Indigenous children residing in remote and regional communities at the Top End Northern Territory (NT) of Australia.

Methods: Video telehealth consultation, that included clinical history and relevant physical findings assessed virtually with an interstate paediatric sleep physician was conducted remotely. Polysomnography (PSG) and therapeutic interventions were carried out locally at Darwin, NT. The study participants were children referred between 2015 and 2020.

Results: Of the total 812 children referred for sleep assessment, 699 underwent a diagnostic PSG. The majority of patients were female (63%), non-Indigenous (81%) and resided in outer regional areas (88%). Indigenous children were significantly older and resided in remote or very remote locations (22% vs. 10%). Referral patterns differed according to locality and Indigenous status – (non-Indigenous via private (53%), Indigenous via public system (35%)). Receipt of referrals to initial consultation was a median of 16 days and 4 weeks from consult to PSG. Remote children had slightly longer time delay between the referral and initial consult (32 vs. 15 days). Fifty one percent were diagnosed to have OSA, 27% underwent adenotonsillectomy and 2% were prescribed with CPAP therapy.

Conclusions: This study has demonstrated that a telehealth model can be an effective way in overcoming logistical barriers and in providing sleep health services to children in remote and regional Australia. Further innovative efforts are needed to improve the service model and expand the reach for vulnerable children in very remote communities.

Key words: aboriginal; adeno-tonsillectomy; children; first nations; polysomnography; sleep apnoea.

What is already known on this topic

1 Sleep disorders are commonly encountered in day-to-day-clinical practice among both Indigenous and non-Indigenous Australian children.
2 Geographical isolation can impose a barrier in accessing specialist care in the diagnosis and management of sleep disorders among children residing in regional and remote communities.
3 There is a sparsity of studies examining the feasibility and outcomes of a paediatric telehealth sleep service model in regional and remote Australian settings.

What this paper adds

1 This study demonstrates the outcomes of a paediatric telehealth sleep service model in a regional and remote setting in the Top End Northern Territory of Australia.
2 Paediatric telehealth sleep service is feasible when approached in a cohesive and collaborative manner by engaging regional specialist services.
3 There are ongoing challenges for access to service, particularly for those residing in very remote areas and for Indigenous children compared to those in urban locality.

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Sleep disorders, including obstructive sleep apnoea (OSA) are common issues in children.[1] Approximately 10% of children habitually snore and 1%–4% have OSA.[1,2] Paediatric sleep disorders may give rise to a myriad of adverse health-related consequences, such as learning and behavioural issues, neurocognitive dysfunction and untoward cardio-metabolic effects.[3] Hence, accurate diagnosis of sleep disorders, including OSA, is crucial for children to lead a better quality of life.[4] However, access to specialised sleep services including physicians and polysomnography (PSG) testing can be a major hurdle for a population residing in regional and remote communities.[5,6]

In the Australian context, the Northern Territory (NT) is the least populated state or territory in Australia, with a population density of just 0.18 people/km^2 and approximately 26%–30% self-identify as Indigenous Australians.[7] The vast majority (80%) of Indigenous people in the NT reside in remote or very remote communities as defined by the Australian statistical geographic standard (ASGS level 4 and level 5).[8] Notably, even Darwin which is the capital city of the NT is classified as Outer Regional (ASGS level 3) (Fig. 1).[8]

In the NT, until 2015, no local in-lab sleep diagnostic services were available for paediatric patients. Children suspected to have sleep issues and requiring polysomnography (PSG) were either not investigated or were referred to another state, the nearest centre being approximately 2600 km away. However, in 2015, a dedicated paediatric sleep service was established alongside the adult sleep diagnostic facility in Darwin, NT. This service includes a remote telehealth model – video paediatric sleep physician consultation, PSG testing locally at Darwin and close collaboration with local Ear Nose Throat (ENT) surgeons and paediatric services.

To date, in the Australian setting, there is a paucity of studies that have comprehensively assessed sleep-related outcomes, inclusive of PSG parameters, continuous positive airway pressure (CPAP) treatment and surgical interventions in a regional or remote residing paediatric population.[9] There is an even greater paucity of data regarding the impact of location (rural vs. urban, Indigenous vs. non-Indigenous). Therefore, in this study, we aimed to assess the impact and related outcomes of establishing a telehealth sleep service model to cater for children residing in regional and remote communities in the Top End Health Service (TEHS) region of the NT of Australia.

Methods

Study setting and ethics approval

This study was conducted at the Darwin Respiratory and Sleep Health (DRSH) centre/Darwin Private Hospital (DPH), the only accredited paediatric sleep diagnostic facility for both public and private patients residing in the TEHS region of the NT of Australia (Fig. 1). This study was approved by the Health Research Governance and Human Research Ethics Committee (HREC), Menzies school of health research of the TEHS, NT (Reference no: HREC-2019-3434).

Study participants

All paediatric patients residing in the TEHS region, including urban, remote and very remote who were referred for

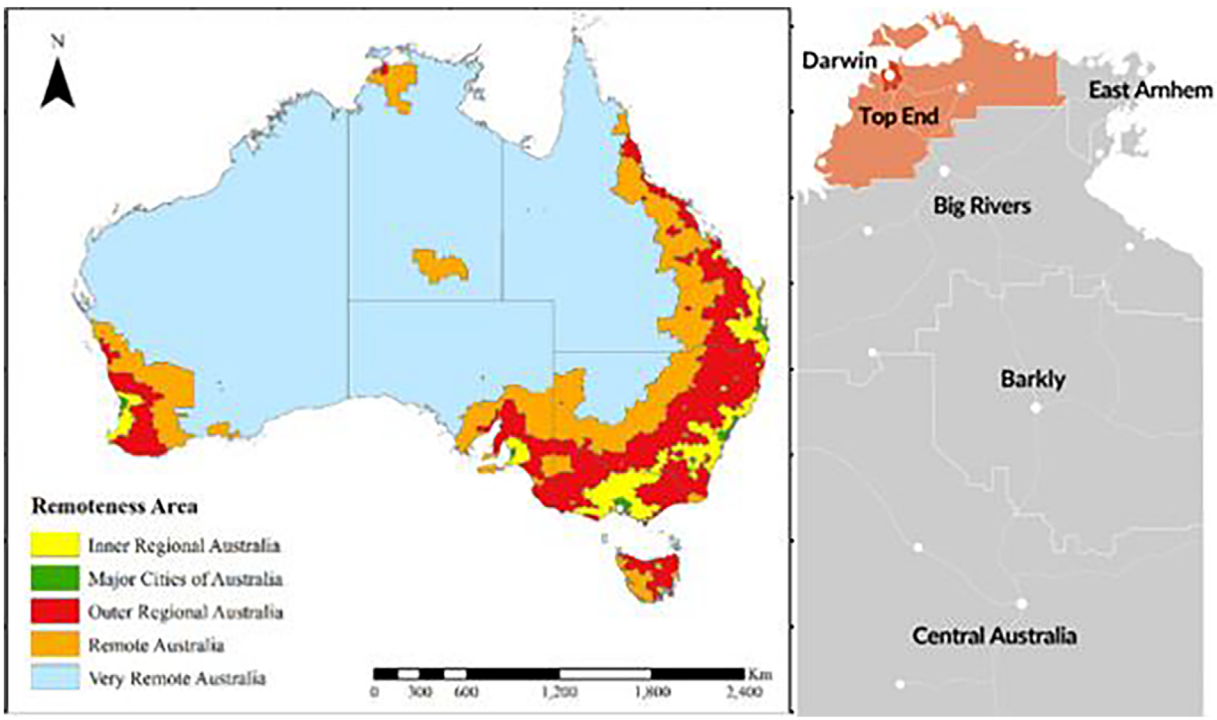

Fig. 1 Map showcasing the Australian remoteness areas and Top End, Northern Territory, Australia. *Source*: Australian Bureau of Statistics (2021), Snapshot of Australia, ABS Website. Top End, Northern Territory Government.

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assessment of clinically suspected sleep disorders to the specialist sleep service at the DRSH centre between 2015 and 2020 were included in this study.

Service model

For both private and public patients, after the receipt of initial referral from general practitioners (GP's), paediatricians, ENT surgeons or remote primary health practitioners, an initial video telehealth consultation was organised with the treating paediatric sleep specialist based in Sydney, Australia. The referrals document the reason for referral, comorbid issues and relevant examination findings. For patients residing in Darwins urban or outer regions, the telehealth consultation took place either at the patient's home or at DRSH facility/DPH. In hospital clinic consultations were usually done for parents or caregivers who were unfamiliar or had difficulty with internet technology. For patients residing in remote communities, the telehealth consultations were predominantly conducted in the home environment. However, for those having difficulty with technology, the telehealth consultation was coordinated at their respective remote community health clinic with the help of local community Indigenous health workers or clinic nurses.

Telehealth consultation

During the initial telehealth consultation, detailed clinical histories and review of manifestations were undertaken. This included symptoms related to sleep issues reported by parents and care givers or by children. Any other relevant medical comorbidities were recorded. Further, children were virtually assessed for dysmorphic features, obvious obesity, jaw/chin malformations, presence of tonsillar enlargement and palate position etc. In certain circumstances during the video telehealth consultation, physicians were able to view the paediatric patient sleeping and parents were able to demonstrate their concerns in real time. The home environment could also be observed. Following the initial telehealth consultation patients underwent a diagnostic PSG if deemed necessary by the paediatric sleep specialist. A follow up telehealth consultation was organised to discuss the sleep study results and therapeutic interventions were facilitated, including pharmacotherapy, behavioural strategies, specialist referral and CPAP therapy. Details of consultation records, including any comorbid medical conditions of concern identified during the consultation were communicated to the referring specialist or GPs to take appropriate action. Finally, the sleep study results and any interventions undertaken were also communicated to the primary referring physician (Fig. 2).

Clinical and polysomnography data

The clinical and PSG protocol at the DRSH/DPH sleep diagnostic facility are described in detail from a previous report from our centre.[10]

Data assessed

Baseline demographic data, residence locality identified through post code (urban/outer regional or remote) and source of primary referral for sleep consultation (GPs, ENT or paediatrician) was recorded. Length of timepoints from referral to initial consult to sleep study and follow up/final consultation were assessed. The number of patients diagnosed to have OSA and who subsequently underwent adenotonsillectomy or CPAP therapy were also recorded.

Statistical analysis

Data are displayed as median (interquartile range (IQR)) for continuous variables or number (%) for categorical variables. Differences in demographic and clinical variables between Indigenous and non-Indigenous patients, urban and remote patients, and between the referral routes were tested via two-tailed chi-squared test utilising Fishers exact test in the case of cells containing <10 for categorical variables, and via Kruskal–Wallis rank sum test for continuous variables. Univariate and multivariate quantile (median) regressions were utilised to test the effect of demographic and clinical variables on the length of time between referral and diagnostic studies. Multivariate models adjusted for age, sex, body mass index (BMI), public/private system, referral route (GP/paediatrician/ENT) and urban/remoteness (ASGS 3 or 4 & 5 respectively). All data were analysed in STATA IC 15 (StataCorp, Texas) and alpha was set to 0.05 throughout.

Results

Referral pathway and patients demographics

In total 886 referrals were received by this service, which included several patients with multiple referrals, resulting in a final 812 unique patients. Several patients failed to attend the initial consult, or the follow up appointments. Hence, in this study we opted to include and assess only the first (earliest) visit of those patients who underwent a diagnostic sleep study (699 patients). Of the 699 patients who underwent a diagnostic sleep study during the study period: the majority were female (63%), non-Indigenous (81%) and resided in Darwin urban areas (88%) of the TEHS region. Indigenous children were significantly older, and more likely to reside in remote or very remote locations than non-Indigenous children (21% vs. 10%). Overall, the most common route of referral was via ENT surgeons (53%) and occurred through the private system. However, for Indigenous patients the most common route was via paediatricians through the public system (35%), while the most common route for non-Indigenous patients was via ENT service through the private system (30%) (Table 1). The total number of patients referred for assessment of sleep health increased progressively since 2015 and 51% were diagnosed to have OSA during this study window (Figs 3, 4).

The clinical characteristics by Indigenous status as per referral pathway by GPs, ENT surgeons or by paediatricians and the number of children referred for assessment are shown in Table 2 and Fig. 5.

Timeline course of referrals to consultation and sleep study

Overall, there was a median of 55 days between receipt of initial referral and patients undertaking the PSG, consisting of a median

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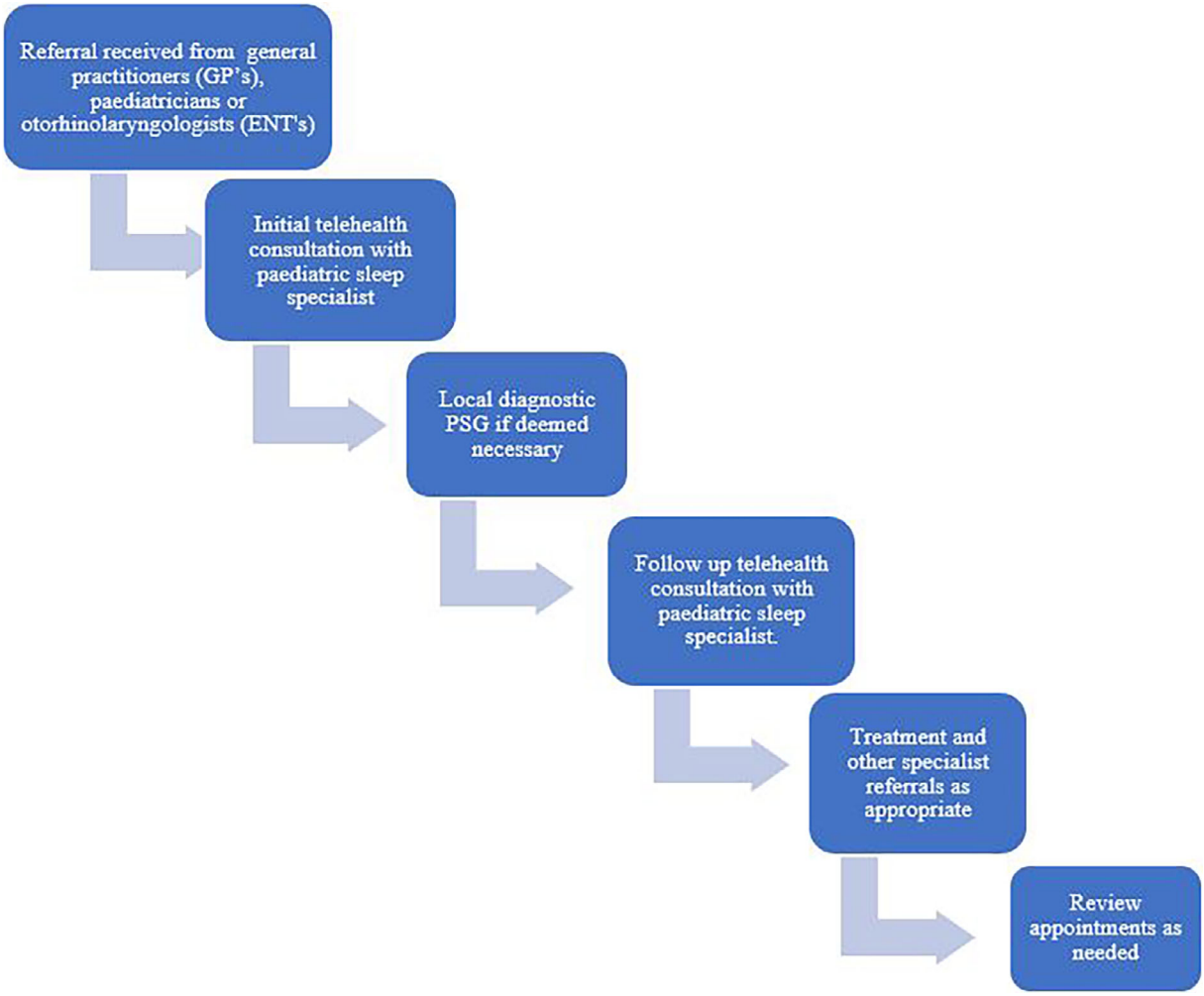

Fig. 2 Service model – showing patient pathway/journey.

Table 1 Patients' demographics and referral pathways for both public and private patients and by Indigenous status

Demographics and referral pathway	Indigenous ($n = 131$)	Non-Indigenous ($n = 568$)	*P*-value
Age	7.15 (3.99, 11.52)	5.15 (3.21, 8.16)	<0.001*
Female	47 (36%)	221 (39%)	0.520
Urban	102 (79%)	495 (90%)	0.001*
Remote/Very remote	27 (21%)	56 (10%)	
Public patient	80 (61%)	240 (42%)	<0.001*
GP referral	3 (2%)	25 (4%)	0.106
Paediatric referral	67 (51%)	234 (41%)	
ENT referral	61 (47%)	309 (54%)	
PDSS	14 (10, 20)	12 (8, 17)	0.001*
OSA	73 (56%)	281 (50%)	0.175

* Statistical significant. ENT, ear nose and throat; GP, general practitioner; OSA, obstructive sleep apnoea; PDSS, paediatric daytime sleepiness scale.

16 days from referral to the initial consult with the paediatric sleep specialist and 4 weeks from the initial consult to PSG. Referrals by ENT surgeons had a significantly reduced time between referral and study by 2–3 weeks compared to GP's and paediatrician referrals, which was mainly due to a shorter timespan between the referral and initial consult (Table 3).

Indigenous children had a significantly shorter time delay between the initial consult and sleep study, however the time from referral to sleep study was the same as for non-Indigenous children (Table 4). Remote children had a significantly longer time delay between the referral and initial consult with a median wait twice as long as for urban patients. However, the time between the initial consult and the study was significantly shortened. Remote children also showed significantly greater variation in the time between each point compared to urban patients (Fig. 6).

Regression analysis

In quantile regression models: age, referral route (via paediatrician or GP's against ENT specialist as reference), healthcare

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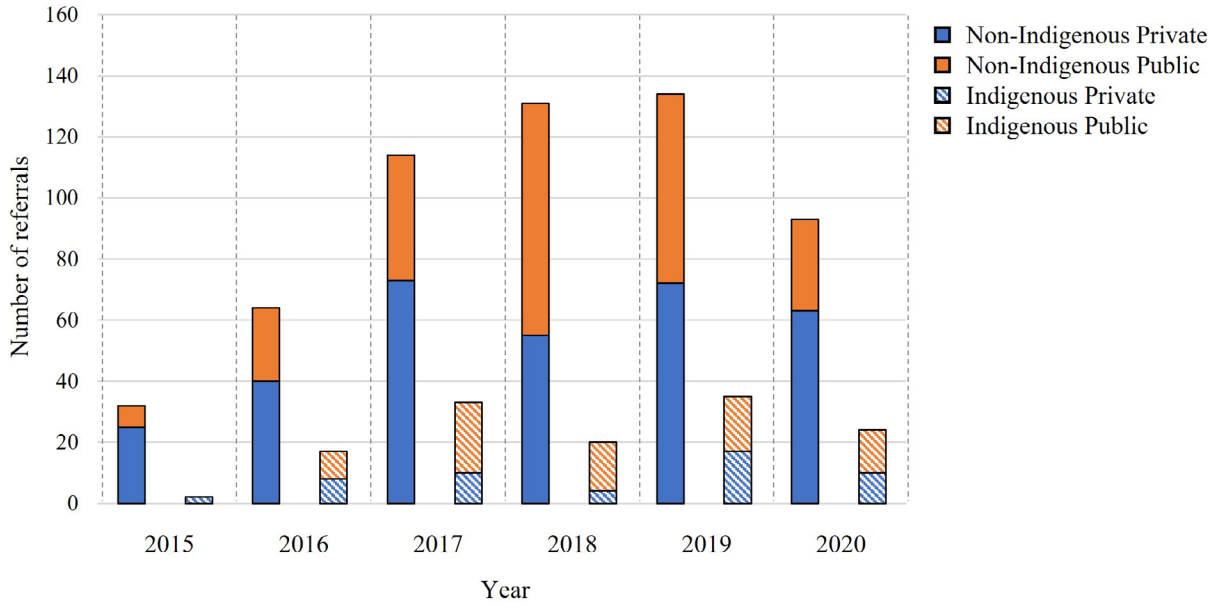

Fig. 3 Number of patients characterised by year, public or private system, and by Indigenous status.

system (public against private as reference) and residence location (remote residence as reference) were significantly associated with the length of time taken between referral and study in both univariate and adjusted models. Older patients who were referred via GPs in the public system and resided remotely showed the longest time delay between referral and sleep study. All other aspects held even; urban residing patients had their sleep study conducted a median 20 days sooner than remote residing patients (Fig. 7).

Treatment outcomes

One quarter of patients ($n = 191$, 27%) were recorded as having had adenotonsillectomy surgery and 14 (2%) as being prescribed CPAP. There were no significant differences in any demographic variables, nor OSA symptoms via PDSS between those who did or did not have surgery (Table 5). In multivariate modelling, the only factor which predicted surgery was presence of OSA (OR 1.92 (95% CI 1.36, 2.7)), with no significant influence of

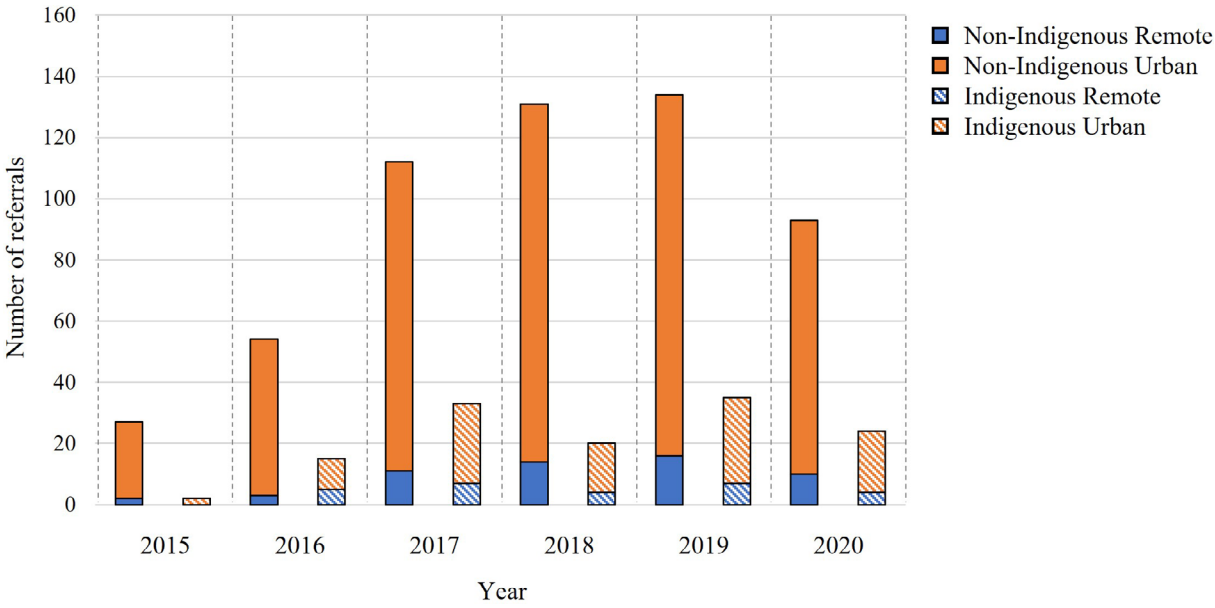

Fig. 4 Number of patients referred per year by location of patient (Urban vs. Remote) and by Indigenous status.

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Table 2 Clinical characteristics as per referral pathway

Demographics	Paediatric referral ($n = 301$)	GP referral ($n = 28$)	ENT referral ($n = 370$)	*P*-value
Age	6.65 (4.29, 10.35)	7.14 (3.53, 10.24)	4.06 (2.54, 6.8)	<0.001*
Female	106 (35%)	10 (36%)	152 (41%)	0.332
Indigenous	67 (22%)	3 (11%)	61 (17%)	0.106
Urban	247 (85%)	23 (92%)	327 (90%)	0.091
Remote/Very remote	44 (15%)	2 (8%)	37 (10%)	
Underweight	39 (13%)	3 (11%)	50 (14%)	<0.001*
Normal weight	142 (47%)	16 (57%)	232 (64%)	
Overweight	57 (19%)	4 (14%)	40 (11%)	
Obese	62 (21%)	5 (18%)	41 (11%)	
Public patient	139 (46%)	7 (25%)	174 (47%)	0.079
PDSS	13 (8, 18)	12 (10, 16)	12 (8, 17)	0.899
OSA	151 (51%)	14 (50%)	189 (52%)	0.963

* Statistical significant. ENT, ear nose and throat; GP, general practitioner; OSA, obstructive sleep apnoea; PDSS, paediatric daytime sleepiness scale.

greater severity of OSA. Figure 8 illustrates the patients journey for initial contact to treatment intervention.

DISCUSSION

This current study demonstrates a service model inclusive of a relatively large proportion of Indigenous children, from both urban/outer regional and remote populations, for which limited data exists in the previous literature.[9] However, although the number of children referred for assessment for sleep issues showed a progressively increasing trend over the years, there was considerable discrepancy between urban and remote residing children. According to Australian Bureau of Statistics (ABS) 2016 Census data – in the TEHS of the NT there were 15 521 Indigenous children (4537 urban residing and 10 989 remote residing) and 27 863 non-Indigenous children (24 720 urban residing and 3140 remote residing). Thus, our centre saw approximately 22.5/1000 Indigenous and 20/1000 non-Indigenous urban children, compared to 2.5/1000 Indigenous & 17.9/1000 non-Indigenous remote residing children.[11] There could be several reasons for why so few remote Indigenous children were referred for sleep assessment: Restricted access to technology and services to make appointments; lack of awareness/literacy on sleep health issues; transient or lack of health practitioners in remote communities; irregular access to specialist care in remote communities and other health priorities are some of the possible reasons.

Nonetheless, presence of sleep disorders can give rise to significant morbidity and mortality in children.[12] Therapeutic interventions may ameliorate long term adverse health consequences. Adenotonsillectomy is the first line treatment modality

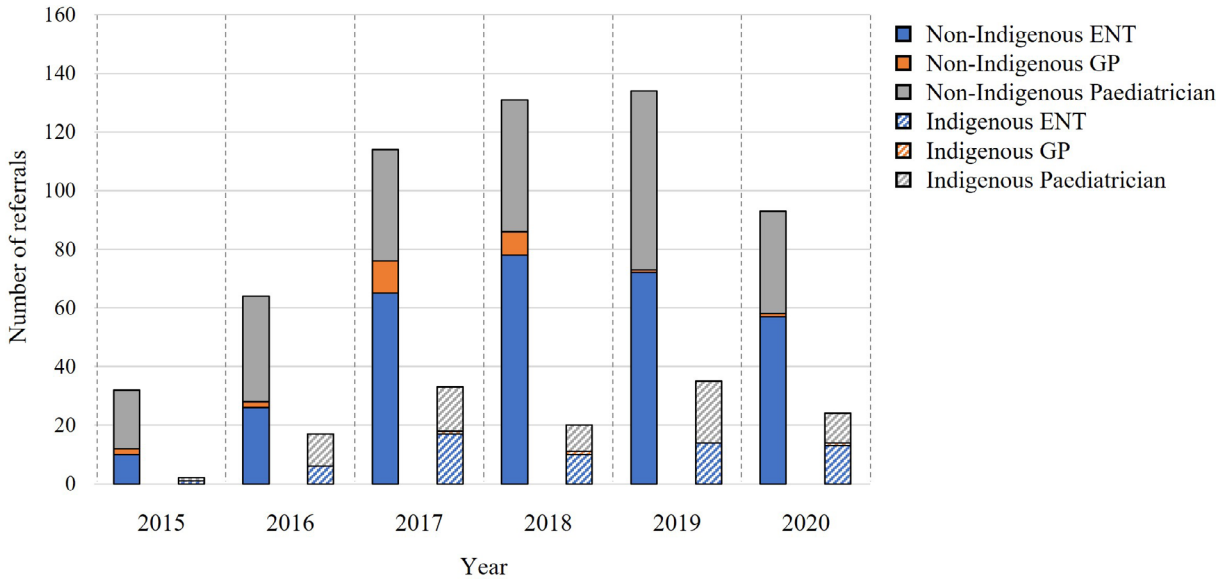

Fig. 5 Number of patients referred per year, referral source and by Indigenous status. ENT, ear nose and throat; GP, general practitioner.

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Table 3 Length of time (days) between timepoints of referral, initial consult, sleep study and follow up by referral source

Referral outcomes	Total	GP	Paediatrician	ENT	*P*-value
Referral to initial consult	16 (6, 40)	12.5 (2, 61.5)	21 (7, 49)	14 (6, 33)	0.004*
Initial consult to study	29 (10, 58)	31.5 (11, 83)	29 (9, 58)	28 (11, 56)	0.769
Referral to sleep study	55 (28, 98)	77.5 (28, 135)	61 (32, 108)	49 (27, 90)	0.023*
Sleep study to follow up consultation	21.5 (14, 39)	26 (14, 54)	24 (15, 43)	21 (14, 35.5)	0.080

* Statistical significant. Data reported as median (IQR). *P*-value obtained via Kruskal–Wallis rank sum test. ENT, ear nose and throat; GP, general practitioner; IQR, interquartile range.

Table 4 Length of time (days) between referral and initial consult to sleep study, and between sleep study and follow up split by Indigenous status and remoteness

Referral outcomes	Indigenous	Non-Indigenous	*P*-value	Urban	Remote	*P*-value
Referral to initial consultation	18 (4, 57)	15 (6, 39)	0.912	15 (6, 38)	32 (5, 92)	0.009*
Initial consult to sleep study	22 (3, 43)	30 (13, 60)	0.001*	30 (13, 58)	10.5 (0, 63.5)	0.003*
Referral to sleep study	50 (27.5, 99.5)	56 (28, 97)	0.565	53 (28, 92)	77 (31, 155)	0.021*
Sleep study to follow up consultation	21.5 (13, 54.5)	21.5 (14.5, 37)	0.864	21 (14.5, 36.5)	21 (8, 49)	0.397

* Statistical significant. Data reported as median (IQR). *P*-value obtained via Kruskal–Wallis rank sum test. IQR, interquartile range.

in children with sleep disordered breathing (OSA), alongside obesity management[13,14] and PAP therapy for selected patients.[15] In this study, we observed 190 patients diagnosed with OSA and who underwent adenotonsillectomy. The prescribed interventions were undertaken within a reasonable time frame and when needed, adenotonsillectomy was performed within 48 hours (data not shown) following a diagnostic sleep study. The number of patients initiated on CPAP therapy was low; but when required, patients were provided with a CPAP device and education soon after the study. Moreover; other appropriate investigations and management of sleep disorders and any comorbid medical conditions identified during the consultation were facilitated as and when appropriate in coordination with the referring physicians and GPs. Hence, this could be considered as a true testament for a positive outcome through this current sleep service model, particularly when engaging and approaching sleep health issues in a multidisciplinary/holistic manner for children residing in remote/regional settings.

Few studies have assessed the feasibility and outcomes of a telehealth model in addressing sleep health issues.[16,17] Telehealth is an excellent modality for people residing in isolated remote communities, but comes with several limitations especially in regard to understanding of or access to technology.[18] In our experience we observed – lack of understanding of how to connect to the internet link provided, not checking or understanding the instructions, not presenting at the appointment time, and internet dropouts during telehealth consultations. Moreover, for

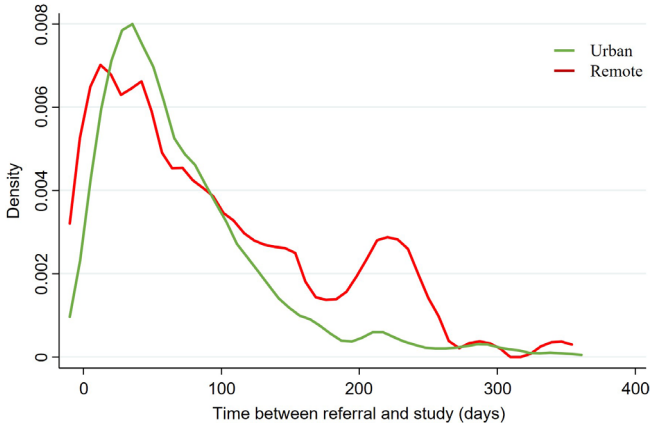

Fig. 6 Kernel density diagram to show the distribution of time between referral and sleep study for urban (green) and remote (red) patients.

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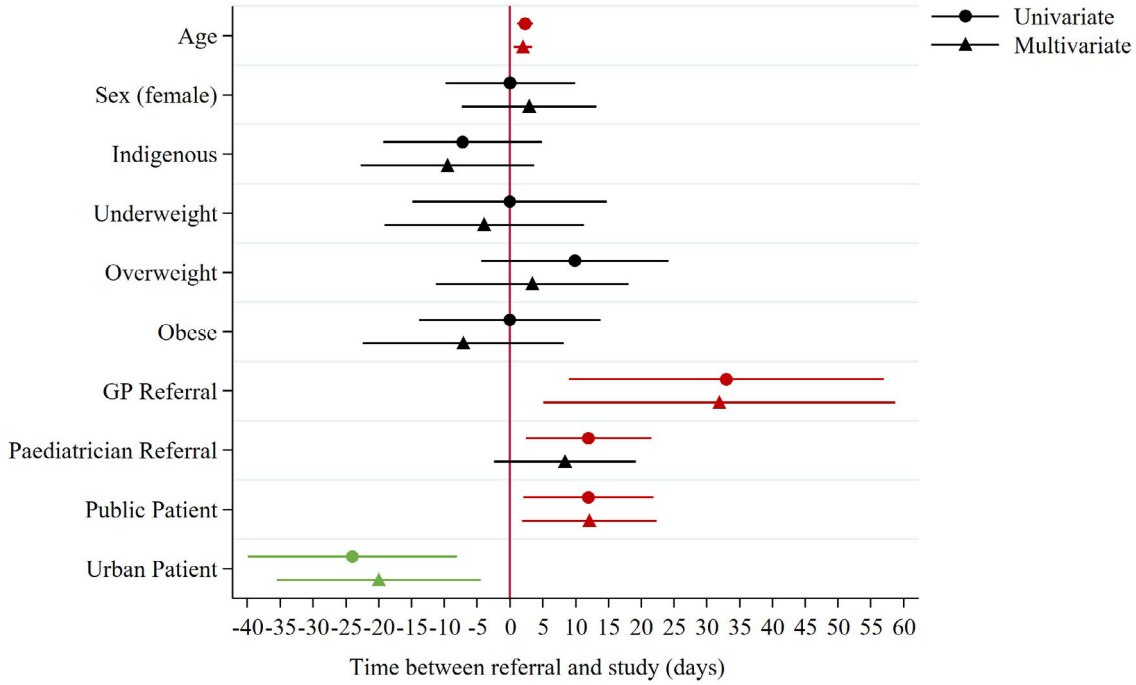

Fig. 7 Coefficients plot for the effect of demographic parameters on time between referral and sleep study.

a few children and families in remote communities, despite telehealth and dedicated nurses to assist with co-ordination of bookings and travel; distance from the sleep centre was a major barrier. Although, vital clinical information and patients' facial/neck morphology could be assessed during the video telehealth consultation, one of the drawbacks of this model is the inability to undertake physical examination in person. Presence of a paediatrician or a GP during the telehealth consultation would have assisted with physical examination, especially for children residing in remote Aboriginal communities. However, realistically, availability of a health professional during the telehealth consultation is not always possible. Moreover, the transient nature of health practitioners in remote and rural Aboriginal health centres could impose a barrier as well. Therefore, plausible and realistic alternate models of care could be explored, such as upskilling and capacity building for community health workers, including, Aboriginal health practitioners and community nurse practitioners to assist with physical examination during telehealth consultations.

Table 5 Patients' demographics with and without surgical intervention

Demographics and referral pathway	No surgery ($n = 508$)	Had surgery ($n = 191$)	*P*-value
Age	5.41 (3.22, 8.69)	4.95 (2.99, 8.11)	0.446
Female	193 (38%)	75 (39%)	0.757
Indigenous	98 (19%)	33 (17%)	0.543
Urban	427 (87%)	170 (91%)	0.126
Remote/Very remote	66 (13%)	17 (9%)	
Underweight	62 (12%)	30 (16%)	0.353
Normal weight	283 (56%)	106 (56%)	
Overweight	80 (16%)	22 (12%)	
Obese	76 (15%)	32 (17%)	
Public patient	229 (45%)	92 (48%)	0.465
GP referral	19 (4%)	9 (5%)	0.761
Paediatric referral	222 (44%)	80 (42%)	
ENT referral	267 (53%)	102 (53%)	
PDSS	12 (8, 17)	12 (8, 18)	0.932
OSA	234 (47%)	119 (63%)	<0.001*
Mild	150 (63%)	71 (59%)	0.393
Moderate	36 (15%)	16 (13%)	
Severe	54 (23%)	33 (28%)	

* Statistical significant. ENT, ear nose and throat; GP, general practitioner; OSA, obstructive sleep apnoea; PDSS, paediatric daytime sleepiness scale.

Another aspect we observed in this study was that the primary direct referral for assessment for sleep issues by GP's were considerably lower in comparison to that of ENT surgeons and paediatricians. It is unclear at this stage if this is related to lack of awareness of the existence of paediatric sleep services in this region or due to under recognition of sleep issues in children or parents not reporting or discussing the child's sleep issues with their GP's. Previously published literature suggests that it is not uncommon for both under recognition as well as under reporting by parents of children's sleep health issues at the primary health care level.[19] Hence, it appears that education and awareness of sleep health issues among children is paramount in this region for both health practitioners and the general population.[20] Studies in the NT have demonstrated that addressing chronic disease

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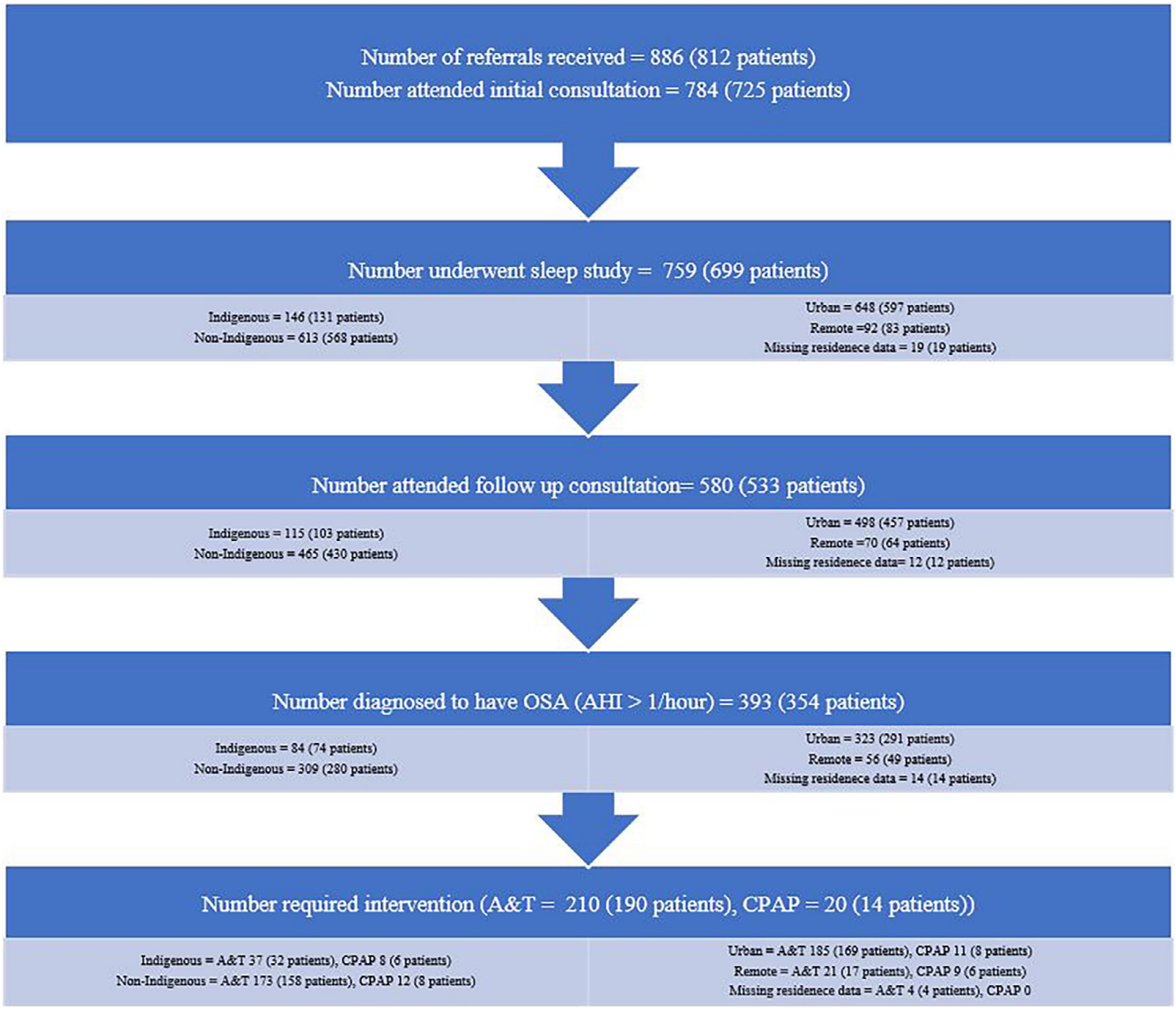

Fig. 8 Showing patients journey through the sleep service and treatment interventions. Numbers are represented in this figure as a total original number for respective service booked and actual numbers attended for respective service indicated in brackets.

at the primary care level in community is likely to result in substantial cost saving.[21] Whether this is also the case for sleep health issues in children, which has several bidirectional impacts on several chronic health issues is uncertain.

Previous studies in both Indigenous and non-Indigenous adults residing in the rural and remote localities of the NT of Australia have demonstrated that a significant proportion have sleep-related disorders.[22] Substantial barriers to access to specialist sleep services are posed by geographical distance.[23] In this regard, our study demonstrates potential avenues in overcoming these logistical barriers, including access to specialist service that would not otherwise be possible due to geographic isolation. Exploring further innovative strategies, including education and awareness in addressing sleep health in a clinically and culturally relevant manner[24] and moreover, investigating opportunities to expand sleep paediatrics telehealth model in a public health paradigm may be key for future management of sleep disorders in this NT Indigenous and non-Indigenous paediatric population.

Limitations

This was a retrospective study in which we reviewed our service, referrals and patient outcomes. The patients referred were a pre-selected group as they had symptoms of OSA. Due to the nature of telehealth and the inability to examine patients, there was a lower threshold for performing PSG's. The time from referral to study and follow up may be skewed due to a few remote patients who had their PSG on the same day as their initial assessment

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due to pre-arranged travel and assessment. In other patients, due to technical issues and rescheduling of appointments, the times were much longer. In a number of cases, post PSG outcomes were not clear and some were lost to follow up. Exploring children who did not undergo surgical intervention or follow up was beyond the scope of this study. In our study, only 19% were Indigenous and were under-represented. Nevertheless, viewed in a different way, a large proportion of Indigenous children were identified and referred for management of their sleep issues which would have been less likely to occur prior to development of our service.

Conclusion

This study reviewed the development of a new service as well as assessing the feasibility of a telehealth model. We have shown a positive impact on all children in the NT, particularly remote and Indigenous. The study has highlighted many of the challenges facing these children- access to specialists, internet, management, and education of physicians and health workers. We also became aware of the cultural issues involved and are looking at ways to make our information and education that are culturally sensitive and relevant.

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