

Virtual consultations for patients with obstructive sleep apnoea: a systematic review and meta-analysis

Sulaiman S. Alsaif^{1,2,3}, Julia L. Kelly ^{1,2}, Stuart Little⁴, Hilary Pinnock ⁵, Mary J. Morrell¹, Michael I. Polkey^{1,2,6} and Phyllis Murphie⁷

¹National Heart and Lung Institute, Imperial College London, London, UK. ²Royal Brompton Hospital, Guy's and St Thomas' NHS Foundation Trust, London, UK. ³Rehabilitation Health Sciences Department, College of Applied Medical Sciences, King Saud University, Riyadh, Saudi Arabia. ⁴Department of Respiratory Medicine, NHS Dumfries and Galloway, Dumfries, UK. ⁵Allergy and Respiratory Research Group, Usher Institute, The University of Edinburgh, Edinburgh, UK. ⁶Sleep and Ventilation Services, Raigmore Hospital, Inverness, UK. ⁷Modernising Patients Pathways Programme, National Centre for Sustainable Delivery, Glasgow, UK.

Corresponding author: Sulaiman S. Alsaif (s.alsaif18@imperial.ac.uk)



Shareable abstract (@ERSpublications) Virtual consultations are as effective as in-person consultations for follow-up management of adult patients with obstructive sleep apnoea treated with continuous positive airway pressure. Moreover, this healthcare strategy appears to be cost-effective. https://bit.ly/3UuyU3M

Cite this article as: Alsaif SS, Kelly JL, Little S, *et al*. Virtual consultations for patients with obstructive sleep apnoea: a systematic review and meta-analysis. *Eur Respir Rev* 2022; 31: 220180 [DOI: 10.1183/ 16000617.0180-2022].

Copyright ©The authors 2022

This version is distributed under the terms of the Creative Commons Attribution Non-Commercial Licence 4.0. For commercial reproduction rights and permissions contact permissions@ersnet.org

Received: 13 Sept 2022 Accepted: 1 Nov 2022

Abstract

Background The coronavirus disease 2019 pandemic has accelerated the adoption of virtual care strategies for the management of patients with obstructive sleep apnoea/hypopnoea syndrome (OSAHS).

Research question What is the effectiveness of virtual consultations compared to in-person consultations for the management of continuous positive airway pressure (CPAP) therapy in adult patients with OSAHS? *Methods* A systematic review and meta-analysis (PROSPERO; CRD42022297532) based on six electronic databases plus manually selected journals was conducted in January 2022. Two researchers independently selected, quality appraised and extracted data. The co-primary outcomes were patient-reported sleepiness, assessed by the Epworth Sleepiness Scale (ESS), and reported cost-effectiveness.

Results 12 studies (n=1823 adults) were included in the review. Seven studies (n=1089) were included in the meta-analysis which showed no difference in the magnitude of improvement in patient-reported sleepiness scores between virtual and in-person consultations (mean difference -0.39, 95% CI -1.38-0.60; p=0.4), although ESS scores improved in both groups. Virtual care strategies modestly increased CPAP therapy adherence and were found to be less costly than in-person care strategies in the three Spanish trials that reported cost-effectiveness.

Conclusion The findings of this review suggest that virtual care delivered by telephone or video consultations is as effective as in-person consultations for improving subjective sleepiness in patients with OSAHS treated with CPAP. This clinical management strategy may also improve CPAP adherence without increasing the costs, supporting its potential as a follow-up management strategy, where patients prefer this approach.

Introduction

Obstructive sleep apnoea/hypopnoea syndrome (OSAHS) is estimated to affect nearly 1 billion adults worldwide, with increasing prevalence [1, 2]. OSAHS is associated with debilitating symptoms, reduced neurocognitive performance and quality of life (QoL), increased risk of cardiovascular and metabolic morbidity and occupational accidents [3, 4], and overall represents a major public health concern [5].



Continuous positive airway pressure (CPAP) improves QoL [6]. However, its effectiveness is contingent on optimal adherence [7]. The recently published National Institute for Health and Care Excellence guidelines [8] recommend a consultation with the patient within 1 month of CPAP initiation and subsequent follow-ups

based on patients' needs until optimal outcomes are achieved. This intensive management strategy adds to the strain on respiratory sleep services already coping with increasing demand.

Accelerated by the coronavirus disease 2019 (COVID-19) pandemic [9], digital health interventions such as telemonitoring and virtual consultations have been introduced into sleep medicine to meet this growing demand [10, 11]. Such transition has the potential to revolutionise the way healthcare is delivered, improving accessibility and affordability [12], though there are concerns about exacerbating inequalities, particularly in disadvantaged communities such as minorities and rural populations [13].

In a previous systematic review, we reported on the applicability and feasibility of telemonitoring and virtual consultations in reviewing patients with OSAHS using CPAP. At the time, evidence on clinical and cost-effectiveness of these strategies was scarce [14]. Subsequent reviews [15–18] have reported promising findings on the potential of digital interventions to improve adherence to CPAP therapy, but have not evaluated the effectiveness of such strategies on patient-reported outcomes and cost-effectiveness. We, therefore, aimed to systematically review the effectiveness of virtual consultations compared to in-person consultations on patient-reported sleepiness and cost-effectiveness for adults with OSAHS treated with CPAP therapy.

Methods

This systematic review and meta-analysis was registered in the International Prospective Register of Systematic Reviews (PROSPERO; CRD42022297532) and reported in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement [19].

Search strategy and data sources

A comprehensive search of six electronic databases (MEDLINE, EMBASE, Scopus, Cochrane Library (Central), CINAHL and medRxiv) was conducted from database inception to 10 January 2022. No limits on publication year or language were imposed. The search strategy, developed in consultation with a medical librarian, used the search terms and keywords: "Sleep Apnoea, Obstructive", "OSA", "OSAHS", "Sleep-disordered breathing" AND "Positive airway pressure", "PAP", "CPAP" AND "Virtual consultation", "Remote consultation", "Telemedicine" (see supplementary material, section 1 for full search strategy).

The search results were de-duplicated in EndNote 20 (Clarivate, Philadelphia, United States). The reference lists of the included studies, other published reviews and relevant sleep medicine and respiratory journals (*e.g. Sleep, Sleep Medicine* and *Thorax*) were searched manually to identify any additional relevant studies.

Eligibility criteria

The population, intervention, control and outcomes (PICO) framework for eligibility criteria is summarised in table 1. Studies were included if they were randomised controlled trials (RCTs), quasi-randomised controlled trials or controlled clinical trials (CCTs) of adult patients with OSAHS using CPAP, comparing virtual consultations to in-person consultations for the follow-up of CPAP therapy.

TABLE I Popu	nation, intervention, control and outcomes framework for inclusion and exclusion criteria
Population	Adults with a clinical diagnosis of OSAHS, either naïve or established users of CPAP; recruited from any healthcare or community setting. Studies were excluded if they investigated patients with other types of sleep disorders such as central sleep apnoea.
Intervention	Any form of a virtual consultation between a patient and a healthcare provider. This included either synchronous or asynchronous communications made <i>via</i> telephone or videoconferencing, with or without real-time telemonitoring of CPAP. No limitations were imposed regarding the number of consultations, methods of CPAP initiation or the duration of consultations. Trials that investigated automated interventions, without direct input from a healthcare professional, were excluded.
Comparator	The comparator group were allocated to a clinical in-person consultation, with or without real-time telemonitoring of CPAP therapy.
Outcomes	 The primary clinical outcome was subjective sleepiness, assessed by the Epworth Sleepiness Scale and the primary organisational outcome was cost-effectiveness of the intervention. Additional outcomes were clinical, patient and/or clinician-reported, and environmental impact outcomes (see supplementary material, section 2, for definitions).
Study design	Studies were included if they were randomised controlled trials, quasi-randomised controlled trials or controlled clinical trials.
CPAP: continue	nus positive ainway pressure: OSAHS: obstructive sleep appoea/bypoppoea syndrome

Study selection and data extraction

We conducted the selection process using Covidence software (Veritas Health Innovation, Melbourne, Australia). Two review authors (S.A. and P.M.) independently screened titles and abstracts using the pre-defined inclusion and exclusion criteria. The full-text reports of potentially eligible studies were then assessed independently by the same authors to determine inclusion eligibility. Any discrepancies between reviewers at either stage were resolved by a third review author (J.K.) or the wider review team (S.L., H.P., M.M. and M.P.).

Data extraction was performed independently by S.A. and P.M. using a customisable form in Covidence, with comparison and discussion of the findings. The following data were extracted:

- Study design, methodology, follow-up duration, and participants' demographic and baseline data.
- Details of intervention, including mode of delivery, intensity and duration.
- Details of comparator.
- · Relevant findings, specifically those related to our primary and additional outcomes.

When needed, corresponding authors were contacted to either seek missing data or clarify unclear methodologies. If the required data were not available in the correct format, the study was excluded from the quantitative meta-analysis.

Risk of bias assessment

The risk of bias (RoB) for each included trial was independently assessed by S.A. and P.M. using the Cochrane Collaboration's tool for assessing risk of bias in randomised trials [20]. In the event of a disagreement between the reviewers, a third review author (J.K.) arbitrated. The assessment of reporting bias through funnel plots was not appropriate in this review due to the small number of studies included in the meta-analysis [21].

Data synthesis

A summary of the included trials is presented, specifically focusing on clinically relevant outcomes including patient-reported sleepiness, cost-effectiveness and adherence to therapy. Where appropriate data (*i.e.* mean±standard deviation) were available for the key outcomes, findings were pooled for a meta-analysis. A random-effects model, with mean differences for continuous data, was performed to pool the results and to calculate 95% confidence intervals and the p values for key outcomes between the virtual consultations group and the control group. The end-point data after exposure to the intervention were used for the analyses. The I^2 statistic was used to assess the statistical heterogeneity of the included studies; a value greater than 50% was considered an indicator of substantial heterogeneity. Subgroup analyses by the mode of delivery of the virtual intervention and by study follow-up duration were performed. All statistical analyses were conducted using the Cochrane Collaboration's Review Manager Software (RevMan, version 5.4.1).

Results

Overview of eligible studies

The literature search identified 875 records. After deduplication, 324 studies were retained for initial title and abstract screening. Of these, 63 studies were retrieved for full-text review and assessment for eligibility. 51 studies did not meet the inclusion criteria and a total of 12 studies [22–33] were included in the review as outlined in the PRISMA flow diagram (figure 1). The commonest reason for exclusion was "wrong intervention" (n=16). Nine of these studies appeared initially to meet the inclusion criteria, but were excluded because they required an in-person consultation for patients randomised to virtual care; see supplementary material, section 3, for further details.

Study characteristics

A summary of the included studies is presented in table 2. 11 of the 12 studies were RCTs and one was a CCT, published between 2006 and 2021. These trials had study sample sizes ranging between 45 to 306 participants and follow-up durations spanning from 30 days to 6 months. In these trials, multimodal digital health interventions were used to deliver virtual consultations to patients with OSAHS using CPAP. In total, the 12 included studies represented 1823 adult participants with the majority being male (averaging 78%) and overweight, diagnosed with moderate to severe OSAHS (table 3). Only four studies reported ethnicity data [22, 23, 25, 33] and two [22, 32] reported only minimal data on socioeconomic status.



FIGURE 1 Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow diagram of the records identified and assessed during the study selection process. CENTRAL: Cochrane Central Register of Controlled Trials; CINAHL: Cumulative Index to Nursing and Allied Health Literature.

RoB and quality of evidence assessment

A summary of the RoB assessment for the included studies is provided in figure 2. Overall, the assessment showed variation in the RoB among the included studies due to the complex nature of the intervention. As would be expected, there was a high RoB in all included studies, because of the inability to blind participants and personnel to allocation. Additionally, the RoB arising from allocation concealment was found to be unclear. Imprecision of the findings is unlikely due to the large sample size in the meta-analysis. However, the certainty of evidence was rated down because of the indirectness in the studies. There was variation in how the intervention of interest, virtual consultations, were delivered. Additionally, all the trials recruited the population of interest (patients with OSAHS using CPAP) and investigated the outcomes of interest. Taken together, with reference to the Grading Recommendations, Assessment, Development and Evaluations (GRADE) framework, the evidence to support the use of virtual consultations for improving the primary outcome, Epworth Sleepiness Scale (ESS) scores, in patients with OSAHS is moderate.

Primary outcomes

Patient-reported sleepiness

The change in subjective sleepiness, assessed by the ESS, was reported by eight studies [23–25, 28–31, 33]. These studies found an improvement in ESS scores from baseline to follow-up in both virtual and in-person consultation groups, with two trials [29, 30] reporting a significantly greater reduction in ESS scores in the virtual consultation group. No difference was observed in the ESS scores for virtual compared to in-person consultations (mean difference (MD) -0.39, 95% CI -1.38-0.60; p=0.4; moderate-certainty evidence) in the seven studies (n=1089) that had the end-point data in the correct format for a meta-analysis (figure 3). However, considerable statistical heterogeneity was observed (I²=72%).

TABLE 2 Summary of the included studies											
Study (year, country)	Study design and overall RoB	Number of participants (n=1823)	Population description	Intervention	Intensity and duration	Mode of delivery	Main findings				
Taylor <i>et al.</i> [22] (2006, USA)	RCT FU: 30 days High RoB	Total: 114 VC: 56 IP: 58	Adult patients with OSA who were initiating CPAP therapy	Telemonitoring <i>via</i> the Health Buddy. OSA patients with "high-risk" responses were contacted within 24 h.	Patients were contacted as needed to resolve issues	Telephone consultation	ESS was not reported for post-intervention follow-up				
Stepnowsky <i>et al.</i> [23] (2007, USA)	RCT FU: 2 months Moderate RoB	Total: 45 VC: 24 IP: 21	Adult patients newly diagnosed with OSA	Telemonitoring via flow generator data. Objective and subjective patient reports triggered patient contact.	Patients were contacted as needed based on a pre-defined clinical pathway	Telephone consultation	No significant differences in ESS scores between the study groups at baseline and post-intervention				
ISETTA <i>et al.</i> [24] (2015, Spain)	RCT FU: 6 months Moderate RoB	Total: 139 VC: 69 IP: 70	Adult OSA patients requiring CPAP treatment	Telemonitoring <i>via</i> a website developed for this study. Input evaluation triggered patient contact.	Virtual consultations via Skype were scheduled at 1 and 3 months. Consultation duration: 38.97±12.04 min.	Video consultation	Improvement in ESS at 6 months, but no significant difference in change from baseline between the study groups. The telemedicine-based strategy had a lower total cost compared to standard care.				
FRASNELLI <i>et al.</i> [26] (2015, Switzerland)	CCT FU: 30 days High RoB	Total: 223 VC: 113 IP: 110	Adult patients with sleep apnoea	Telemonitoring <i>via</i> CPAP. A colour-coded algorithm triggered patient contact.	Patients were contacted as needed for a duration of ~30 min	Telephone consultation	ESS was not reported for post-intervention follow-up				
Fields <i>et al.</i> [25] (2016, USA)	RCT FU: 3 months Moderate to high RoB	Total: 60 VC: 32 IP: 28	Adult patients with OSA from two community-based outpatient centres	Telemonitoring <i>via</i> APAP. Scheduled follow-up contact and if needed.	Initial evaluation visit for 40 min with a 10 min (or less) follow-up call at week 1. Virtual consultations scheduled at 1 and 3 months for 20 min each.	Initial evaluation <i>via</i> real-time CVT. Telephone consultation for follow-up.	No significant difference in the change of ESS scores from baseline to 3 months follow-up between the study groups				
Turino <i>et al.</i> [27] (2016, Spain)	RCT FU: 1 and 3 months Moderate RoB	Total: 100 VC: 52 IP: 48	Adult patients with newly diagnosed OSA requiring treatment with CPAP	Telemonitoring <i>via</i> MyOSA – Oxigen Salud web database. Automatic alarms triggered patient contact.	Patients were contacted as needed to resolve issues	Telephone consultation	ESS was not reported for post-intervention follow-up. The total average cost per randomised patient was 28% lower in the VC group than in the IP standard care group.				

https://doi.org/10.1183/16000617.0180-2022

ы

Continued

Study (year, country)	Study design and overall RoB	Number of participants (n=1823)	Population description	Intervention	Intensity and duration	Mode of delivery	Main findings					
Lugo <i>et al</i> . [28] (2019, Spain)	RCT FU: 3 months Moderate RoB	Total: 186 VC: 94 (32 with CPAP) IP: 92 (40 with CPAP)	Adult patients with suspected OSA who were referred to the sleep unit	Telemonitoring <i>via</i> CPAP. Input in a custom web application triggered patient contact.	Virtual consultations were scheduled at 3, 6 and 12 weeks for no more than 15 min each	Video or telephone consultation	No significant differences in the ESS scores between the study groups. The costs of the VC were cheaper than those for IP standard care and the Bayesian analysis showed that the VC was cost-effective.					
Nilius <i>et al.</i> [29] (2019, Germany)	RCT FU: 6 months Moderate to high RoB	Total: 80 VC: 40 IP: 40	Adult OSA patients who had suffered an ischaemic stroke within the last 3 months	Telemonitoring. A colour-coded algorithm triggered a more detailed evaluation and patient contact if needed.	Patients were contacted as needed for a duration of 5 min	Telephone consultation	VC group had a significantly lower ESS scores at 6 months follow-up					
Pépin <i>et al.</i> [30] (2019, France)	RCT FU: 6 months Moderate RoB	Total: 306 VC: 157 IP: 149	Adult patients with severe OSA and high cardiovascular risk	Telemonitoring <i>via</i> CPAP and the multimodal system. Automatic algorithms triggered patient contact.	Patients were contacted as needed. Regular assessments at day 8 and months 1 and 6.	Telephone or teleconsultation	ESS scores significantly improved in both study groups, but the size of improvement was significantly higher in the VC group					
Tamisier <i>et al.</i> [31] (2020, France)	RCT FU: 6 months Moderate RoB	Total:206 VC: 102 IP: 104	Newly diagnosed adult patients with OSA and low cardiovascular risk who were referred for CPAP therapy	Telemonitoring <i>via</i> CPAP and the multimodal system. Automatic algorithms triggered patient contact.	Patients were contacted as needed based on an automatic algorithm	Telephone or teleconsultation	ESS scores significantly improved in both study groups, with no significant difference between the groups					
FIETZE <i>et al.</i> [33] (2021, Germany)	RCT FU: 6 months Moderate to high RoB	Total: 224 VC: 110 IP: 114	Adult patients with moderate to severe OSA	Telemonitoring <i>via</i> APAP. Pre-defined criteria triggered patient contact.	Patients were contacted as needed based on pre-defined criteria	Telephone consultation	Change from baseline to 6 months in ESS scores was not significantly different between the two groups					
Koou <i>et al.</i> [32] (2021, Netherlands)	RCT FU: 4 weeks, 12 weeks, 24 weeks Moderate RoB	Total: 140 VC: 70 IP: 70	Adult patients diagnosed with moderate or severe OSA who require CPAP treatment	Telemonitoring. Not achieving pre-defined objectives (e.g. adherence and residual AHI) triggered patient contact.	Patients were contacted as needed Scheduled follow-ups at 1 and 4 weeks	Video and telephone consultation	ESS was not reported for post-intervention follow-up					

AHI: apnoea-hypopnoea index; APAP: automatically adjusting positive airway pressure; CCT: controlled clinical trial; CPAP: continuous positive airway pressure; CVT: clinical video tele-health; ESS: Epworth Sleepiness Scale; FU: follow-up duration; IP: in person; OSA: obstructive sleep apnoea; RCT: randomised controlled trial; RoB: risk of bias; VC: virtual consultation.

TABLE 3 Participant baseline characteristics												
Study (year)	Number of participants	Age (years)	Ge (ma	nder Ile %)	BMI (k	g∙m ⁻²)	AHI (events per hour)				
	(n=1823)	VC	IP	VC	IP	VC	IP	VC	IP			
Taylor <i>et al.</i> [22] (2006)	Total: 114 VC: 56 IP: 58	45.8±10	44.6±8.5	66	71	NR	NR	NR	NR			
Stepnowsky <i>et al.</i> [23] (2007)	Total: 45 VC: 24 IP: 21	60±10.8	58±13.7	98	98	33.3±4.9	30.5±5.1	44.8±17.9	37.6±14.3			
Isetta <i>et al.</i> [24] (2015)	Total: 139 VC: 69 IP: 70	51.0±8.9	47.0±10.9 [¶]	85	87	32.8±7.3	33.6±8.3	45 (35–70) [#]	52 (35–62) [#]			
Frasnelli <i>et al.</i> [26] (2015)	Total: 223 VC: 113 IP: 110	55 (47–63)#	55 (45–61)#	76	78	NR	NR	37 (23–62) [#]	40 (20–69)#			
FIELDS <i>et al.</i> [25] (2016)	Total: 60 VC: 32 IP: 28	46.7±13.1	58.2±14.4	93	94	33.2±6.0	32.9±3.8	32.0±33.9	30.1±30.0			
Turino <i>et al.</i> [27] (2016)	Total: 100 VC: 52 IP: 48	56 ±13	54 ±12	77	77	35±7	35±7	52±25	53±26			
Lugo <i>et al.</i> [28] (2019)	Total: 186 VC: 94 (32 with CPAP) IP: 92 (40 with CPAP)	50.39±11.31	50.82±12.15	70	66	29.97±6.19	31.50±10.91	24.68 ±21.01	33.60±28.96			
Nilius <i>et al.</i> [29] (2019)	Total: 80 VC: 40 IP: 40	55.4±10.4	58.6±9.3	73	73	31.7±5.4	30.1±6.6	41.2±19.0	37.6±18.4			
Р _{ÉРIN} <i>et al.</i> [30] (2019)	Total: 306 VC: 157 IP: 149	60.8 (53.8–66) [#]	61.8 (54.7–66.1) [#]	73	74	32.4 (29.6–36.5) [#]	31.4 (28.1–35) [#]	47 (35–60.5) [#]	45 (35.4–61.2) [#]			
Тамізіег <i>et al.</i> [31] (2020)	Total: 206 VC: 102 IP: 104	51.9 (44.8–58.8) [#]	49.4 (40–57.9) [#]	58	68.3	30.5 (27.5–34.9) [#]	31.3 (26.6–35.2) [#]	45 (34–58) [#]	42.3 (33–57.6) [#]			
FIETZE <i>et al.</i> [33] (2021)	Total: 224 VC: 110 IP: 114	53.6±11.8	53.1±10.6	83	79	32.8±6.4	31.7±5.6	35.3±17.6	37.0±20.3			
Коол <i>et al.</i> [32] (2021)	Total: 140 VC: 70 IP: 70	52.3±12.4	54.3±11.9	83	76	BMI >30 n=51 (73%)	BMI >30 n=46 (66%)	31 (22–46) [#]	30.5 (20–42) [#]			

Data are presented as n or mean±sp, unless otherwise stated. AHI: apnoea–hypopnoea index; BMI: body mass index; CPAP: continuous positive airway pressure; IP: in person; NR: not reported; VC: virtual consultation. [#]: Median (interquartile range). [¶]: Significantly different value from the other group.

Subgroup analysis by the mode of delivery of the virtual intervention (telephone alone or video and/or telephone consultations) and by study follow-up duration (\leq 3 months or 6 months) showed no differences between the study groups (see supplementary material, section 4.1A and 4.1B).

Cost-effectiveness

Three trials [24, 27, 28] from Spain investigated the cost-effectiveness of virtual compared to in-person care for patients with OSAHS, from patient and provider perspectives. ISETTA *et al.* [24] reported that a telemedicine-based strategy had a slightly lower total cost, with a 69% estimated probability that it would be cheaper than in-person care. When considering only obstructive sleep apnoea (OSA)-related costs, the probability increased to 98% with estimated costs of ϵ 150.90 and ϵ 114.00, respectively, for the in-person and virtual care strategies. In another RCT, TURINO *et al.* [27] found the total average cost for each randomised participant to be 28% lower in the virtual care group (ϵ 123.60) than in the in-person care group (ϵ 170.90), with a ϵ 47.32 difference between the two study groups. The investigators also reported the incremental cost-effectiveness ratio per quality-adjusted life year (QALY), which was estimated at



FIGURE 2 A summary of the risk of bias assessment for the included studies.

 \notin 17 358.65 (QALY). The third study conducted by Lugo *et al.* [28] showed, using Bayesian analysis, that the telemedicine-based strategy was cost-effective; OSA-related costs for the virtual and in-person care groups were \notin 264.96 and \notin 412.03, respectively.

Secondary outcomes

Adherence to CPAP therapy

CPAP therapy usage, assessed objectively, was reported by all included studies (n=12). The average CPAP usage at follow-up assessment ranged from 3.5 to 5.6 h per night with virtual consultations and 2.1 to

Virtual			ι	li li	1-pers	on		Mean difference	Mean difference				
Study or subgroup	Mean	SD	Total	Mean	SD	Total	Weight (%)	IV, random, 95% CI	Year	IV, random, 95% CI			
STEPNOWSKY et al. [23]	9.2	6.6	20	9.9	5.2	20	5.4	-0.70 (-4.38-2.98)	2007				
ISETTA et al. [24]	6.52	4.14	64	5.89	3.51	64	15.5	0.63 (-0.70-1.96)	2015				
Lugo et al. [28]	8.5	4.4	80	7.05	4.31	74	15.2	1.45 (0.07-2.83)	2019				
NILIUS et al. [29]	3.7	3.2	36	6.1	4.1	37	13.2	-2.40 (-4.080.72)	2019				
PÉPIN <i>et al</i> . [30]	4.58	3.88	157	6.05	4.07	149	18.3	-1.47 (-2.360.58)	2019				
TAMISIER et al. [31]	4.51	4.08	102	5.19	4.32	104	16.7	-0.68 (-1.83-0.47)	2020				
FIETZE et al. [33]	8	4.7	90	7.8	4.2	92	15.7	0.20 (-1.10-1.50)	2021				
Total (95% CI)			549			540	100.0	-0.39 (-1.38-0.60)		•			
Heterogeneity: Tau ² =1.18; Chi ² =21.14; df=6 (p=0.002); l ² =72%													
Test for overall effect: $Z=0.77$ (p=0.44) -4 -2 0 2 4										-4 -2 0 2 4			
N /										Favours (virtual) Favours (in-person)			

FIGURE 3 Forest plot of the mean difference in patient-reported sleepiness scores, assessed by the Epworth Sleepiness Scale, in patients randomised to virtual compared to in-person consultations for the management of obstructive sleep apnoea/hypopnoea syndrome. The diamond represents the 95% confidence interval of the pooled estimate of the mean difference. df: degrees of freedom; IV: inverse variance.

5.6 h per night with in-person consultations. Nine studies [22–25, 27, 28, 31–33] found no statistically significant difference in the mean hours of CPAP usage between the two care strategies, while the other three studies [26, 29, 30] showed a significantly higher adherence among participants randomised to virtual consultations. The pooled analysis of 10 RCTs [22–25, 27–31, 33] comprising 1299 participants demonstrated a significant difference in favour of virtual consultations (MD +0.43, 95% CI 0.06–0.80 h per night; p=0.02; moderate-certainty evidence); see figure 4. This equates to an average of 26 min increase in CPAP usage per night.

Subgroup analysis by the mode of delivery of the virtual intervention (telephone alone or video and/or telephone consultations) and by study follow-up duration (≤ 3 months or 6 months) were performed. The analyses showed attenuation of the effect size in studies with 6 months follow-up compared to studies with ≤ 3 months follow-up durations (see supplementary material, section 4.2A and 4.2B).

Change in QoL

10 trials [22–25, 27–31, 33] reported the change in QoL from baseline using multiple different generic and disease-specific instruments, including the European Quality of Life Five-Dimensions questionnaire (EQ-5D; n=2), the 12-Item Short-Form Health Survey (SF-12; n=3), the Functional Outcomes of Sleep Questionnaire (FOSQ; n=4) and a modified FOSQ (M-FOSQ; n=1). Seven of these trials reported a general improvement in QoL among participants randomised to both virtual and in-person consultations, with no statistically significant differences between the two groups (EQ-5D n=1/2; SF-12 n=2/3; FOSQ n=3/4, M-FOSQ n=1/1). Three trials found a greater improvement of QoL in the virtual consultations group compared to the in-person consultations group (European Quality of Life–visual analogue scale n=1/2; SF-12 physical component n=1/3; FOSQ n=1/4).

Environmental impact outcomes

None of the trials reported environmental impact outcomes (including travel distances from home to clinic, mode of transport and carbon footprint).

Sensitivity analyses

The substantial heterogeneity among the included studies in the meta-analyses prompted *post hoc* sensitivity analyses. NILLUS *et al.* [29] met our inclusion criteria, but was thought on clinical grounds to be an outlier, due to its specific patient population (OSA patients with recent ischaemic stroke), which may have resulted in unexpected between-group differences in the study outcomes, specifically ESS scores and CPAP adherence. However, when this study was removed from the meta-analyses, the results remained unchanged: the pooled weighted mean difference for subjective sleepiness was -0.09 (95% CI -1.07-0.89); p=0.86) and for CPAP usage was +0.30 (95% CI 0.00-0.60 h per night; p=0.05).

		Virtua	l	In-person				Mean difference					
Study or subgroup	Mean	SD	Total	Mean	SD	Total	Weight (%)	IV, random, 95% CI	Year	IV, ra	ndom, 9	5% CI	
TAYLOR et al. [22]	4.29	2.15	56	4.22	2.05	58	9.5	0.07 (-0.70-0.84)	2006				
STEPNOWSKY et al. [23]	4.1	1.8	20	2.8	2.2	20	5.7	1.30 (0.05-2.55)	2007				
ISETTA et al. [24]	4.4	2	69	4.2	2	70	10.6	0.20 (-0.46-0.86)	2015				
FIELDS et al. [25]	3.68	0.62	32	2.92	0.61	28	14.4	0.76 (0.45-1.07)	2016		-		
TURINO <i>et al.</i> [27]	5.1	2.1	52	4.9	2.1	48	8.9	0.20 (-0.62-1.02)	2016	-			
Lugo et al. [28]	5.68	1.38	32	5.63	1.64	40	10.2	0.05 (-0.65-0.75)	2019	_			
NILIUS et al. [29]	4.4	2.5	40	2.1	2.2	40	7.1	2.30 (1.27-3.33)	2019			-	
PÉPIN <i>et al.</i> [30]	5.28	2.23	157	4.75	2.5	149	12.1	0.53 (0.00-1.06)	2019				
TAMISIER et al. [31]	4.73	2.48	102	5.08	2.44	104	10.5	-0.35 (-1.02-0.32)	2020				
FIETZE <i>et al.</i> [33]	4.38	2.04	89	4.32	2.28	93	11.0	0.06 (-0.57-0.69)	2021	-	-	_	
Total (95% CI)			649			650	100.0	0.43 (0.06-0.80)					
Heterogeneity: Tau ² =0.22	2; Chi ² =2	7.98; d	f=9 (p=0	.0010); I	² =68%			_	1	1			1
Test for overall effect: Z=	2.29 (p=0	0.02)							-2	-1	0	1	2
									Favou	irs (in-pers	son) Favo	ours (virt	:ual)

FIGURE 4 Forest plot of the mean difference in continuous positive airway pressure usage (hours per night) in virtual compared to in-person consultations. The diamond represents the 95% confidence interval of the pooled estimate of the mean difference. df: degrees of freedom; IV: inverse variance.

Deviations from protocol

Although the heterogeneity assessment was higher than specified in the protocol ($I^2 > 50\%$), we decided to perform meta-analyses due to the complex nature of the intervention. We have also conducted subgroup and sensitivity analyses.

Discussion

To our knowledge, this is the first systematic review and meta-analysis to evaluate the effectiveness of virtual compared to in-person consultations on both patient-reported sleepiness and also cost-effectiveness. The main findings are that virtual consultations using telephone or videoconferencing are as effective as in-person consultations for improving patient-reported sleepiness (moderate-certainty evidence) in patients with OSAHS treated with CPAP, and appear to be cost-effective. Additionally, virtual consultations modestly increased the average CPAP usage by nearly half an hour per night when compared to in-person consultations. No published studies compared the environmental impact of virtual *versus* in-person follow-up strategies.

Subjective sleepiness, assessed by the ESS, is an important outcome for the management and monitoring of patients with OSAHS [8]. In this meta-analysis, there was no difference in the magnitude of improvement of ESS scores between virtual and in-person consultations, though ESS decreased in both groups. Importantly, this observation persisted with subgroup analyses by the mode of delivery of the virtual consultation (telephone alone or video and/or telephone consultations) and by study follow-up duration (\leq 3 months or 6 months), suggesting that virtual care delivered in a variety of strategies remained as effective as in-person care.

CPAP adherence was higher in the virtual consultations group than the in-person consultations group, although the 26 min increase in CPAP usage is slightly lower than the minimal clinically important improvement of 30 min per night [34]. This increase in CPAP usage is consistent with earlier meta-analyses which focused on the use of virtual care strategies specifically to investigate the effect on CPAP adherence [15–17]. However, recent post-COVID-19 data has shown a reduction in CPAP adherence with remote CPAP set-up, compared to in-person [11, 35]. While CPAP set-up is just one part of the process of initiation onto therapy, these data highlight the need for robust assessment of new healthcare strategies before they become mainstream. In the current review, an assessment of whether specific characteristics of the virtual consultation could have influenced the adherence to CPAP was performed. Subgroup analysis by follow-up duration showed a trend for the effect size to attenuate in studies with 6 months compared to \leq 3 months follow-up durations, which may support the efficacy of long-term follow-up interventions [36].

Virtual care strategies were less costly than in-person strategies in the three studies that reported cost-effectiveness [24, 27, 28]. Whilst these data need to be interpreted with caution, it is reassuring that there is no suggestion that virtual follow-up is more costly. The cost savings were mainly driven by fewer in-person follow-up visits, savings on transport and less loss of productivity. However, the cost-utility analyses were limited to one healthcare context (*i.e.* Spain) and there was little clarity in how all the calculations of the mean cost per patient were performed. Further global data is likely to emerge as such virtual strategies are continued following the COVID-19 pandemic. Well-designed studies are important to establish whether virtual consultations are a clinically and cost-effective strategy for the management of patients with OSAHS using CPAP, particularly from the patient, societal and healthcare provider perspectives.

There has been a growing interest in the importance of reporting ethnicity and socioeconomic status data in clinical trials as well as addressing their potential effect on health inequalities [37]. A concerning finding of the current review is that only one-third of the studies reported data on ethnicity and two studies presented minimal data on socioeconomic status. This reduces interpretability and generalisability of their findings, emphasising the need for explicit reporting of trial participants' demographics. Such data facilitates investigations of OSAHS phenotypes, where heterogeneous clinical manifestations exist. This data will also inform discussions around health inequalities as we move towards personalised medicine approaches for OSAHS care [38].

Virtual care strategies have the potential to improve access to healthcare services and address geographical barriers to delivering high-quality care [39, 40]. However, patient-focused concerns exist, specifically about increasing health inequalities ("the digital divide") [41], emphasising the need to ensure that the care is tailored to patients' context, needs and preferences.

One unanticipated finding in this review was that none of the included studies reported data on any of the environmental impact outcomes, despite increased awareness of the climate emergency [42]. This observation signifies a potential gap in evidence related to virtual care strategies for the management of OSAHS. A critical and interesting direction for future research would be to assess the impact of virtual compared to in-person consultations on the environment, reinforcing the need for sustainable delivery of healthcare.

Strengths and limitations

The review was conducted in accordance with current recommendations and guidelines [19, 21]. However, there are several limitations to consider when interpreting the findings. The studies were relatively small in size, with most containing between 100 and 200 participants. They were also varied in terms of the methodologic quality, identified by the RoB assessment. The studies were heterogeneous in type, intensity and duration of both virtual and in-person care strategies; potentially limiting the interpretation of the results. Care should also be taken when interpreting the subgroup analyses due to the small number of included studies. We suggest future studies should, therefore, be powered to evaluate the different techniques, cost-effectiveness and the environmental impact of delivering virtual consultations for the management of patients with OSAHS. This is to identify the most efficacious components of these virtual care strategies from patient, clinical and economic perspectives. Finally, it is important to stress that our analysis did not address the diagnosis of OSAHS and therefore cannot be used to support a virtual diagnostic pathway.

Conclusion

Our findings suggest that virtual patient care delivered by telephone or video consultations is as effective as in-person consultations for improving subjective sleepiness in patients with OSAHS treated with CPAP. Moreover, virtual consultations modestly increased CPAP adherence, compared to in-person consultations, and were not associated with reduced cost-effectiveness. Virtual follow-up of patients with OSAHS using CPAP should be available as an alternative care strategy to in-person follow-up, where patients prefer it.

Provenance: Submitted article, peer reviewed.

Conflict of interest: The authors have nothing to disclose.

References

- 1 Peppard PE, Young T, Barnet JH, *et al.* Increased prevalence of sleep-disordered breathing in adults. *Am J Epidemiol* 2013; 177: 1006–1014.
- 2 Benjafield AV, Ayas NT, Eastwood PR, *et al.* Estimation of the global prevalence and burden of obstructive sleep apnoea: a literature-based analysis. *Lancet Respir Med* 2019; 7: 687–698.
- **3** Garbarino S, Guglielmi O, Sanna A, *et al.* Risk of occupational accidents in workers with obstructive sleep apnea: systematic review and meta-analysis. *Sleep* 2016; 39: 1211–1218.
- 4 Lévy P, Kohler M, McNicholas WT, *et al.* Obstructive sleep apnoea syndrome. *Nat Rev Dis Primers* 2015; 1: 15015.
- 5 Lyons MM, Bhatt NY, Pack AI, *et al.* Global burden of sleep-disordered breathing and its implications. *Respirology* 2020; 25: 690–702.
- 6 National Institute for Health and Care Excellence, Continuous positive airway pressure for the treatment of obstructive sleep apnoea/hypopnoea syndrome: technology appraisal guidance. www.nice.org.uk/guidance/ ta139 Date last accessed: June 2022. Date last updated: 20 August 2021.
- 7 Rotenberg BW, Murariu D, Pang KP. Trends in CPAP adherence over twenty years of data collection: a flattened curve. *J Otolaryngol Head Neck Surg* 2016; 45: 43.
- 8 National Institute for Health and Care Excellence, Obstructive sleep apnoea/hypopnoea syndrome and obesity hypoventilation syndrome in over 16s. www.nice.org.uk/guidance/ng202 Date last accessed: June 2022. Date last updated: 20 August 2021.
- 9 Pinnock H, Murphie P, Vogiatzis I, *et al.* Telemedicine and virtual respiratory care in the era of COVID-19. *ERJ* Open Res 2022; 8: 00111-2022.
- **10** Johnson KG, Sullivan SS, Nti A, *et al.* The impact of the COVID-19 pandemic on sleep medicine practices. *J Clin Sleep Med* 2021; 17: 79–87.
- 11 Turnbull CD, Allen M, Appleby J, *et al.* COVID-19-related changes in outpatient CPAP setup pathways for OSA are linked with decreased 30-day CPAP usage. *Thorax* 2022; 77: 839–841.
- 12 World Health Organization. Future of digital health systems: report on the WHO symposium on the future of digital health systems in the European region: Copenhagen, Denmark, 6–8 February 2019. https://apps.who. int/iris/handle/10665/329032 Date last accessed: June 2022. Date last updated: 10 October 2019.

- 13 Litchfield I, Shukla D, Greenfield S. Impact of COVID-19 on the digital divide: a rapid review. *BMJ Open* 2021; 11: e053440.
- 14 Murphie P, Little S, McKinstry B, *et al.* Remote consulting with telemonitoring of continuous positive airway pressure usage data for the routine review of people with obstructive sleep apnoea hypopnoea syndrome: a systematic review. *J Telemed Telecare* 2019; 25: 17–25.
- **15** Aardoom JJ, Loheide-Niesmann L, Ossebaard HC, *et al.* Effectiveness of ehealth interventions in improving treatment adherence for adults with obstructive sleep apnea: meta-analytic review. *J Med Internet Res* 2020; 22: e16972.
- 16 Hu Y, Su Y, Hu S, et al. Effects of telemedicine interventions in improving continuous positive airway pressure adherence in patients with obstructive sleep apnoea: a meta-analysis of randomised controlled trials. Sleep Breath 2021; 25: 1761–1771.
- 17 Labarca G, Schmidt A, Dreyse J, *et al.* Telemedicine interventions for CPAP adherence in obstructive sleep apnea patients: systematic review and meta-analysis. *Sleep Med Rev* 2021; 60: 101543.
- 18 Verbraecken J. Telemedicine in sleep-disordered breathing: expanding the horizons. Sleep Med Clin 2021; 16: 417–445.
- **19** Page MJ, McKenzie JE, Bossuyt PM, *et al.* The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. *BMJ* 2021; 372: n71.
- 20 Higgins JPT, Altman DG, Gøtzsche PC, *et al.* The Cochrane Collaboration's tool for assessing risk of bias in randomised trials. *BMJ* 2011; 343: d5928.
- 21 Higgins JPT, Tomas J, Chandler J, *et al.* Cochrane Handbook for Systematic Reviews of Interventions Version 6.3 (updated February 2022). London, Cochrane, 2022. https://training.cochrane.org/handbook
- 22 Taylor Y, Eliasson A, Andrada T, *et al.* The role of telemedicine in CPAP compliance for patients with obstructive sleep apnea syndrome. *Sleep Breath* 2006; 10: 132–138.
- 23 Stepnowsky CJ, Palau JJ, Marler MR, *et al.* Pilot randomized trial of the effect of wireless telemonitoring on compliance and treatment efficacy in obstructive sleep apnea. *J Med Internet Res* 2007; 9: e14.
- 24 Isetta V, Negrín MA, Monasterio C, et al. A Bayesian cost-effectiveness analysis of a telemedicine-based strategy for the management of sleep apnoea: a multicentre randomised controlled trial. Thorax 2015; 70: 1054–1061.
- 25 Fields BG, Behari PP, McCloskey S, *et al.* Remote ambulatory management of veterans with obstructive sleep apnea. *Sleep* 2016; 39: 501–509.
- 26 Frasnelli M, Baty F, Niedermann J, et al. Effect of telemetric monitoring in the first 30 days of continuous positive airway pressure adaptation for obstructive sleep apnoea syndrome a controlled pilot study. J Telemed Telecare 2016; 22: 209–214.
- 27 Turino C, de Batlle J, Woehrle H, *et al.* Management of continuous positive airway pressure treatment compliance using telemonitoring in obstructive sleep apnoea. *Eur Respir J* 2017; 49: 1601128.
- 28 Lugo VM, Garmendia O, Suarez-Girón M, et al. Comprehensive management of obstructive sleep apnea by telemedicine: clinical improvement and cost-effectiveness of a virtual sleep unit. A randomized controlled trial. PLoS One 2019; 14: e0224069.
- **29** Nilius G, Schroeder M, Domanski U, *et al.* Telemedicine improves continuous positive airway pressure adherence in stroke patients with obstructive sleep apnea in a randomized trial. *Respiration* 2019; 98: 410–420.
- **30** Pépin JL, Jullian-Desayes I, Sapène M, *et al.* Multimodal remote monitoring of high cardiovascular risk patients with OSA initiating CPAP: a randomized trial. *Chest* 2019; 155: 730–739.
- **31** Tamisier R, Treptow E, Joyeux-Faure M, *et al.* Impact of a multimodal telemonitoring intervention on CPAP adherence in symptomatic OSA and low cardiovascular risk: a randomized controlled trial. *Chest* 2020; 158: 2136–2145.
- **32** Kooij L, Vos PJE, Dijkstra A, *et al.* Video consultation as an adequate alternative to face-to-face consultation in continuous positive airway pressure use for newly diagnosed patients with obstructive sleep apnea: randomized controlled trial. *JMIR Form Res* 2021; 5: e20779.
- **33** Fietze I, Herberger S, Wewer G, *et al.* Initiation of therapy for obstructive sleep apnea syndrome: a randomized comparison of outcomes of telemetry-supported home-based *vs.* sleep lab-based therapy initiation. *Sleep Breath* 2022; 26: 269–277.
- 34 Patil SP, Ayappa IA, Caples SM, et al. Treatment of adult obstructive sleep apnea with positive airway pressure: an American Academy of Sleep Medicine systematic review, meta-analysis, and GRADE assessment. J Clin Sleep Med 2019; 15: 301–334.
- **35** Stanchina M, Lincoln J, Prenda S, *et al.* The impact of different CPAP delivery approaches on nightly adherence and discontinuation rate in patients with obstructive sleep apnea. *J Clin Sleep Med* 2022; 18: 2023–2027.
- **36** Patel SR, Bakker JP, Stitt CJ, *et al.* Impact of an extended telemonitoring and coaching program on CPAP adherence. *Am J Respir Crit Care Med* 2022; 205: A2571.

- **37** Buttery S, Philip K, Alghamdi S, *et al.* Reporting of data on participant ethnicity and socioeconomic status in high-impact medical journals: a targeted literature review. *BMJ Open* 2022; 12: e064276.
- **38** Edwards BA, Redline S, Sands SA, *et al.* More than the sum of the respiratory events: personalized medicine approaches for obstructive sleep apnea. *Am J Respir Crit Care Med* 2019; 200: 691–703.
- **39** Barbosa W, Zhou K, Waddell E, *et al.* Improving access to care: telemedicine across medical domains. *Annu Rev Public Health* 2021; 42: 463–481.
- **40** Greenhalgh T, Procter R, Wherton J, *et al.* The organising vision for telehealth and telecare: discourse analysis. *BMJ open* 2012; 2: e001574.
- **41** Greenhalgh T, Rosen R, Shaw SE, *et al.* Planning and evaluating remote consultation services: a new conceptual framework incorporating complexity and practical ethics. *Front Digit Health* 2021; 3: 726095.
- **42** Purohit A, Smith J, Hibble A. Does telemedicine reduce the carbon footprint of healthcare? A systematic review. *Future Healthc J* 2021; 8: e85–e91.