THE UNIVERSITY
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‘Reviewing people with Obstructive Sleep Apnoea Hypopnoea Syndrome: Telehealth and Templates’

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Declaration

I declare that this thesis presented for the degree of Doctor of Philosophy of Population Health Sciences has

i) Been composed entirely by myself
ii) Been solely the result of my work
iii) Not been submitted for any other degree or professional qualification
iv) Some results of studies contained within this thesis have previously been presented in publication and abstract format as listed

Phyllis Murphie

Abstract

Introduction

Obstructive Sleep Apnoea Hypopnoea Syndrome (OSAHS) is estimated to affect one billion people globally, carrying a substantial risk of road traffic accidents due to daytime sleepiness with additional other adverse health impacts. Continuous positive airway pressure (CPAP) is an effective treatment but requires long-term adherence. Regular clinical review of CPAP therapy users is important to address any problems, assess symptom relief, and specifically to review implications for driving. Reviews can be face-to-face, by telephone, or via teleconsultation with remote telemonitoring of CPAP usage. My PhD addresses the mode of delivery and content of CPAP reviews for people with OSAHS in three distinct but interlinked projects.

Aims and objectives:

1) *Systematic review:*

To review the evidence for the effectiveness of teleconsultation combined with telemonitoring in the review of people with OSAHS receiving CPAP therapy compared to face-to-face care.

2) *e-Delphi:*

i) To reach consensus amongst a panel of international sleep clinicians, academics, and CPAP users on the most important components to include in a CPAP review, and how often a review should take place.

ii) To develop a structured sleep medicine review template that records the important components identified by the e-Delphi consensus panel.

3) *Implementation study*

To introduce the template in three diverse sleep medicine Centres for use in face-to-face or remote consultations and observe implementation using mixed methods.

Systematic review

I followed Cochrane methodology and I searched ten electronic databases, trial registries, and reference lists for studies that included interventions that combined remote consultations with telemonitoring of CPAP usage. Outcome measures were: the proportion of CPAP users who had a review, adherence to CPAP, symptom control, satisfaction / acceptability, and cost-effectiveness. From 362 potentially relevant papers, I identified four randomised controlled trials and one controlled clinical trial (including 269 patients). The risk of bias was moderate in one, and moderate to high in four trials. Two trials reported the number and duration of reviews with inconsistent results. The teleconsultation / telemonitoring improved CPAP adherence in two trials; two reported no between-groups differences. Two trials, both at moderate to high risk of bias, showed no between-group differences in the Epworth Sleepiness Scale. Satisfaction was generally positive in all five trials; one trial reported that the teleconsultation / telemonitoring patients were 'more likely to continue' with CPAP therapy and one trial reported that CPAP teleconsultation / telemonitoring was cost-effective.

e - Delphi and template development

I recruited an international expert panel to identify the core components of a CPAP review derived from thirteen OSAHS guidelines and asked participants to score components (scale of one to five) over three rounds. Consensus was defined as ≥75% agreement for scores of ≥4. Free-text comments were analysed thematically.

40 participants completed all three scoring rounds. Of 36 potential components, 17 achieved consensus: treatment acceptability, sleep quality, symptom resolution (including reduction in apnoea-hypopnoea index), assessment of sleepiness (including when driving), technical issues (mask fit / humidification / cleaning / filters), CPAP adherence, and quality of life. Participants suggested reviews should be 12 to 18 monthly (more frequent when in early treatment) or "on demand / request." Free text comments

emphasised that reviews should be multidisciplinary, flexible (including telehealth), and focus on symptom control.

Based on the consensus findings I devised a CPAP review template that included the core components suggested by the participants.

Implementation study

I recruited three Centres: City, Urban / Rural, and Rural to a mixed-methods implementation study. There were four stages:

I. Development of a tailored implementation strategy
II. Implementation
III. Evaluation
IV. Refining

Effectiveness of the implementation strategy was assessed by template uptake, feasibility of use, and fields completed. Analysis of the quantitative data was descriptive. Nine clinicians participated in final semi-structured interviews which were analysed thematically.

219 anonymised templates were returned for analysis; all were completed voluntarily by nursing staff. The average completion time was 14 minutes; with six of the eight fields completed in over 90% of reviews. Six themes emerged from nine interviews. The template:

1) Facilitated a structured, standardised review, reducing variation in practice.
2) Was not perceived to affect clinical autonomy or person-centred care.
3) The decision to participate was made at organisational level and individuals in the departments were free to decide whether, or not, to use the template in any / all of their consultations.
4) Use of the template did not impact on how the review was conducted though template completion extended review time in one Centre where double data entry was required.

5) From a service perspective, the template was perceived as improving efficiency and promoting better patient outcomes whether in face-to-face reviews or teleconsultations.
6) The template was seen as potentially contributing to multidisciplinary team working.
With the rapid deployment of Microsoft Teams and NHS Near Me during the COVID -19 pandemic, the template supported remote consultations and might be useful for education and training purposes. Adaptations (structure, content, and layout) were highlighted. An electronic template was suggested, integrated with the electronic health record, obviating the requirement to dictate letters and reducing administrative processes.

Conclusions and context

The three studies add to the evidence base on the regular review of CPAP users. Telemedicine is an option for CPAP reviews for people with OSAHS. The international consensus group identified important components that should be recorded in a CPAP review informing development of a structured review template. Implementation of the template was achieved in three Centres and was considered to provide structure to both face-to-face reviews and teleconsultations with a potentially important role in training staff and standardising care.

Teleconsultation has come of age in the midst of a global pandemic for routine CPAP reviews, and electronic templates adapted to the local setting could support safe and effective care and promote efficient communication.

Lay summary

Background

Obstructive Sleep Apnoea Hypopnoea Syndrome (OSAHS) is a condition where a person may stop breathing numerous times throughout the night. This can result in excessive daytime sleepiness or tiredness and reduced quality of life, and an increased risk of road traffic accidents. OSAHS can also have other important health consequences such as an increased risk of heart-related and circulation-related problems. The treatment for this condition is to wear a mask over the nose or mouth that provides positive pressure to keep the airway open during sleep, and this treatment is called CPAP therapy. People who use this treatment require regular review, and this can be either face-to-face or using video-assisted calls or telephone review.

What did I want to find out and what did I do?:

1. *What was the evidence for using video-assisted or telephone review combined with electronic monitoring of CPAP use overnight?*

I reviewed research papers that looked at combining real-time video or telephone reviews combined with remote monitoring compared to face-to-face traditional care of people using CPAP therapy. I found five papers that concluded that there was no difference in the number of hours that people use CPAP each night when they were reviewed remotely compared to a face-to-face review. Patient satisfaction was generally reported positively in all five papers; one paper reported that a video review combined with overnight monitoring of patients were ‘more likely to continue’ with CPAP therapy and one paper reported that CPAP teleconsultation / telemonitoring was cost-effective. There were no safety concerns identified in any of the papers.

2. *What were the most important components to include in a CPAP review, and how often this should review take place*?

I carried out an international study by email to gain an understanding of participants collective views on the most important components to include in a review. I was then able to establish what were the most important components to include in a CPAP clinic review. I then developed a review template that could be used to support a CPAP review.

3. *What were the opinions of the sleep clinic staff on the use of the template in clinical reviews? and how many templates were completed during the study period?.*

I then implemented this template in three sleep clinics in the UK. I asked clinical teams to use the template for two months and provide me with copies of their completed templates to see how they had used them, and what they thought of them by asking them to take part in recorded interviews. The nurses that participated in the study found the review template to be useful and suggested some alterations and are keen to see a revised version of the template.

Conclusion

The three studies I have carried out support the use of telemedicine as an option for review in people using CPAP therapy, and the importance of having a regular review and suggested core components to include in a review. Using a structured template that includes these core components can guide a review and is a tool to assist the healthcare professional with ensuring the most important components to include in a review are covered. However, the review should be about what matters to the patient and what they want to discuss in a review and this should not be overlooked. Using telemedicine as an option for review in the midst of a global pandemic for routine CPAP reviews and using templates adapted to the local setting could support safe and effective care and promote patient centred care.

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Contents

Contributions to science

Publications and Presentations
Peer-reviewed publications

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Abbreviations

AASM - American Academy of Sleep Medicine
AHI - Apnoea Hypopnoea Index
AMED - Allied and Complementary Medicine
APAP - Automatic Positive Airway Pressure
BMcK - Brian McKinstry
BMI - Body Mass Index
BMJ - British Medical Journal
BNI - British Nursing Index
BQ - Berlin Questionnaire
BTS - British Thoracic Society
CCT - Controlled Clinical Trial
CSA - Central Sleep Apnoea
CFIR - Consolidate Framework for Implementation Research
COVID 19 - Coronavirus Disease
CINAHL - Cumulative Index of Nursing and Allied Health Literature
CPAP - Continuous Positive Airway Pressure
DARE - Database of Abstracts of Reviews of Effects
DVLA - Driver, and Vehicle Licensing Agency
ECG - Electrocardiogram
e- DELPHI - Electronic Delphi Study
EEG - Electro Encephalogram
EMBASE - Excerpta Medica dataBASE
EMG - Electromyogram
EPOC - Effective Practice and Organisation of Care
EOG - Electrooculogram
ERIC - Education Resources Information Centre
ESS - Epworth Sleepiness Scale
ERS - European Respiratory Society
European Union
FOSQ - Functional Outcomes of Sleep Questionnaire

HP - Hilary Pinnock

HSAT - Home Sleep Apnoea Test

LILACS - Latin American and Caribbean Health Service Literature

MAD - Mandibular Advancement Device

MDT - Multi-Disciplinary Team

MEDLINE - Medical Literature Analysis and Retrieval System Online

NHS - National Health Service

NICE - National Institute for Health and Care Excellence

NIV - Non-Invasive Ventilation

NPT - Normalisation Process Theory

OSAHS - Obstructive Sleep Apnoea Hypopnoea Syndrome

PM - Phyllis Murphie

POS - Pilot Observational Study

PRISMA - Preferred Reporting Items for Systematic Review and Meta-analysis

PSG - Polysomnography

QOL - Quality of Life

RCT - Randomised Controlled Trial

ROB - Risk of Bias

RTA's Road Traffic Accidents

SAVE - Sleep Apnoea Cardiovascular Endpoints Study

SBQ - StopBang Questionnaire

SL - Stuart Little

SQ - Stop Questionnaire

StaRI - Standards for Reporting Implementation Studies

TESLA - Transcutaneous Electrical Stimulation on OSA

TLC - Telephone Linked Communication

TRIP -Turning Research into Practice

UK - United Kingdom

USA - United States of America

ZETOC - Z Electronic Table of Contents

Chapter 1 General overview of thesis

1.1 Clinical context of thesis

Obstructive Sleep Apnoea Hypopnoea Syndrome (OSAHS) is a common condition that occurs in all age groups where a person may have repeated complete or partial collapse of the of the upper airway resulting in reduced respiratory effort, intermittent dips in the oxygen saturation level leading to hypoxaemia and disrupted sleep resulting in excessive daytime sleepiness and reduced quality of life (1-3). The gold standard treatment for this condition is Continuous Positive Airway Pressure (CPAP) therapy, although there are other treatment options for those that may not tolerate CPAP (4, 5). Current guidelines recommend regular review of CPAP users however there is no collective agreement regarding the content or components of the review and how often a review should take place (4-9). The mode of consultation may be adapted to the clinical context setting (face-to-face or teleconsultation).

In Chapter 2, I summarise the clinical features of OSAHS, the options for management, and the different modes of consultation used.

1.2 Program of Research and presentation of the thesis

In this thesis, I will explore both the content of a clinical review and mode of delivery and how often this should take place.

- In Chapter 3, I describe my aims and methods.
- In Chapter 4, I will describe the evidence base for use of teleconsultation combined with telemonitoring in the review of CPAP users compared to traditional face-to-face care.
- In Chapter 5, I will describe the findings of an international e-Delphi consensus study and how this has facilitated the formation of a novel structured sleep medicine review template that contains the most important core components suggested by the e - Delphi participants (10).

- In Chapter 6, I will describe the findings of the Implementation study of the use of this template in the clinical setting in three diverse sleep medicine Centres in the UK.

1.3 Relevance of this research

In Chapter 7, I discuss the findings of my thesis in terms of the clinical, healthcare, and policy context.

This work will be of interest and have potential relevance for sleep medicine service providers / policymakers both nationally and internationally. Particularly within the current Coronavirus global pandemic where telemedicine solutions for delivering healthcare services are critically important to remobilisation and the continuation of services and for the longer term. This work may also inform future healthcare guidelines regarding the most important components to consider / include in a clinical review in CPAP therapy users, how often a review should be undertaken and what a review template may look like.

Chapter 2 Obstructive Sleep Apnoea Hypopnoea Syndrome

2.1 History of OSAHS

The evolution of the history of sleep medicine from ancient to modern times makes for captivating reading for 21st-century practicing sleep medicine clinicians, particularly in this era of the SARS-CoV-2 global pandemic where the modern-day history of the delivery of sleep medicine services globally is being redefined.

2.1.1 Ancient history of OSAHS

Some surviving ancient Egyptian medical papyri dating back to 4000 BC include a reference to sleep disorders that include descriptions of sleep problems like insomnia, and treatments for sleep disorders such as narcolepsy, insomnia, and snoring (11).

Hippocrates "The Father of Medicine" may have identified the origins of OSAHS. Hippocrates lived between 460-377 BC and is possibly the first to describe the existence of OSAHS in the literature where he writes about abnormal sleep patterns: *'Others, when their diet bares too great a proportion to their exercise, not only sleep well at night, but are likewise drowsy in the day; the repletion still increases, and their nights begin to grow restless; their sleep afterwards becomes disturbed with frightful dreams of battles'* (12).

A condition that could be described as possible OSAHS is reported in the literature by Aelianus et al in 1666 when Dionysius 'a Greek ruler' in 360 AD is described as having several features that suggest that he may have suffered from sleep apnoea, notably obesity, breathing problems, sleepiness, and that he was difficult to waken (13). Another reference by Athenaeus in 1863 in the same publication by Krieger et al reports that Magas, King of Cyrene (deceased in 258 BC) *'was weighted down with monstrous masses of flesh in his last days; in fact, he choked himself to death'*.

2.1.2 Modern history of OSAHS

The Surgeon Extraordinary to the King was William Wadd, and in 1816 is reported to have written a monograph regarding his observations on obesity and how this could impede exercise and breathing and increase sleepiness. He writes about a case where the individual regularly fell asleep while eating in the company of others (14). The first contemporary descriptions of OSAHS emerge from the 1960s with reports from Lugaresi et al and Gastaut et al both describing cases where they saw an association between obesity and hypersomnolence (15, 16), however, the discovery of OSAHS has been attributed to Jung in 1965. Lavie et al's publication in 2008 does describe reported cases of suspected OSAHS some 50 years earlier with the publication of Burwell et al's paper in 1956 – titled *'Extreme Obesity Associated with Alveolar Ventilation - A Pickwickian Syndrome'* (17). Although the first description of OSAHS is generally attributed to Charles Dickens' *The Pickwick Papers.* In the last 40 years and more recent times, the ground breaking work of Sullivan et al published in 1981 has positioned OSAHS as an important respiratory condition that deserves the full attention of respiratory clinicians globally (18).

2.2 Obstructive Sleep Apnoea Hypopnoea Syndrome - definition

The National Institute for Health and Excellence (NICE) defines OSAHS as the coexistence of excessive daytime sleepiness with irregular breathing at night (9). This quotes the British Thoracic Society (BTS) who use the term OSAHS for people with repetitive apnoeas and symptoms of sleep fragmentation with excessive daytime sleepiness (9). The American Academy of Sleep Medicine (AASM) has defined OSAHS as a sleep-related breathing disorder where there is upper airway narrowing that can impair normal sleep-related ventilation (19). The European Respiratory Society (ERS) similarly defines this condition as intermittent obstruction of the airway during sleep (6).

An obstructive apnoea is the cessation of airflow for at least ten seconds with persistent respiratory effort while the term hypopnoea refers to a reduction in inspiratory airflow (by at least 30%) lasting greater than ten seconds, with associated oxygen desaturation or arousal from sleep (20). The pathophysiology of OSAHS is described in more detail in section 2.2.7. It is a common, treatable condition and a major public health problem for healthcare providers globally with a reported prevalence of 3 – 12.5 % of middle-aged men and 2 – 5.9 % of women (4, 6, 21-24). The prevalence of OSAHS is discussed in more detail in section 2.2.2. It has been recognised as an important cause of morbidity and mortality for almost four decades. People present to health care services with a range of complaints; loud disruptive snoring, witnessed cyclical breathing cessation during sleep, a sensation of nocturnal choking in some cases, and consequent excessive daytime tiredness and often falling asleep in inappropriate situations (23, 25, 26).

2.2.1 Contributing factors to OSAHS

OSAHS is present in all age groups and there are a number of risk factors that increase the likelihood of this condition developing: being overweight or obese (2, 24, 26), having a large collar size (>17 inches), (27), increasing age and male gender (male to female ratio estimated to be 2:1 in the general population) (25, 28), post-menopausal women, existing hypertension (29, 30), a family history of OSAHS (31), a small lower jaw and certain other facial configurations or abnormalities (32, 33), enlarged tonsillar tissue (34, 35), increased alcohol consumption, sedative medications and smoking (29, 36), and hypothyroidism and acromegaly (37).

2.2.2 Prevalence of OSAHS

One reason for the wide divergence in the reported prevalence of OSAHS over time is that there have been substantial variations in methodological reporting of diagnostic tests and thresholds for reporting sleep studies (38). The current accepted metric for diagnosing OSAHS is the Apnoea Hypopnoea Index (AHI), (the number of apnoeic and hyponoeic events recorded per hour of sleep). Section 2.2.6 (Table 1) defines diagnostic thresholds for the AHI (27).

Recent epidemiological data from the United States and Europe have suggested that between 14% and 49% of middle-aged men have clinically significant OSAHS (30). Senaratna conducted a systematic review in 2017 and reports that the prevalence of OSAHS ranged from 9% to 38% in the adult population, from 13% to 33% in men, 6% to 19% in women, with much higher prevalence in the elderly (38). A further recent publication from a European epidemiological study reports the overall prevalence of OSAHS as 44 % in the general population and 22.7% in moderate to severe OSAHS cases (39). The reported rising prevalence of OSAHS is in line with the 'epidemic' of obesity and is a clear indication of the significant disease burden this condition places on health care systems globally.

2.2.3 Economic impact of OSAHS

OSAHS also represents a significant economic burden on healthcare systems and society globally. The overall healthcare costs related to OSAHS include the direct costs of diagnostic testing and the ongoing costs of treating the condition and the indirect costs associated with other co-morbid conditions (obesity, diabetes, cardiovascular and cerebrovascular disease), (30). People with poorly controlled or untreated OSAHS may be less productive in the work environment, with a higher level of absenteeism due to daytime fatigue / somnolence, impaired workplace vigilance / performance, mood disturbances, neurobehavioral impairments, and general malaise (40). OSAHS also impacts occupational safety at work; people with OSAHS have a twofold increase in

work accidents and more commonly suffer work-related injuries (41). OSAHS is a well-documented cause of increased road traffic incidents resulting in serious injury and fatalities which has financial implications for society as a whole and already overstretched and underfunded healthcare systems (41-44). It has been estimated that up to 7% of road traffic accidents for a population of male drivers can be attributed to OSAHS (45). With proper management of OSAHS, significant improvements to an individual's health and public safety will benefit healthcare systems and society as a whole.

2.2.4 Medico-legal implications of OSAHS

With the expanding evidence base of the impact of increased work-related incidents and road traffic accidents in people with OSAHS, there are medico-legal implications for the individual as well as healthcare providers of sleep medicine services globally (40). This has important driving and vehicle licensing implications for people with this condition as well as a major public safety concern as accidents involving people with OSAHS can be accompanied by serious injuries (46). Driving issues and OSAHS are discussed in more detail in section 3.2.

2.2.5 Diagnosis of OSAHS

The diagnosis of OSAHS can only be confirmed by objective tests that are carried out during sleep and the gold standard diagnostic test is considered to be laboratory-based polysomnography (PSG). Home Sleep Apnoea Testing (HSAT) is now considered an alternative option due to the rising prevalence of OSAHS. This has particular relevance due to the increased demand for limited laboratory-based PSG (4, 19, 47).

- A full PSG measures multiple sleep parameters including brain activity (EEG), eye movements (EOG), muscle activity or skeletal muscle activation (EMG), and heart rhythm (ECG), respiratory airflow, and respiratory effort indicators with pulse oximetry to measure oxygen saturation (19).

- HSAT can be conducted in the place of residence as well as any health care setting and is an accepted test for suspected OSAHS. It is a more convenient diagnostic option, it is less expensive than formal PSG, however, there is the disadvantage that HSAT measures fewer parameters and has the potential for misdiagnosis (47). HSAT is poised to become the most prevalent diagnostic test in the current COVID 19 pandemic, and for the longer term with full-in lab PSG testing being paused in many services to reduce the need for face-to-face inpatient diagnostic testing and reduce the risk of virus transmission.

2.2.6 Measurement of severity of OSAHS

The measurement of the severity of OSAHS has traditionally been defined by the number of breathing pauses that occur during sleep and the AHI has been the most commonly used means of defining the severity of this condition (19). The AHI is derived from the total number of apnoeas and hypopnoeas divided by the total sleep time (Table 1).

Severity	Average number of obstructive breathing events per hour of sleep
Normal	<5
Mild	5-15
Moderate	15-29
Moderate/severe	31-50
Very severe	>50

Table 1 Apnoea Hypopnoea Index (AHI) and Severity of OSAHS

However, controversy exists about the AHI score and its link to the severity of OSAHS and this is related to differences in the way that PSG and HSAT scoring criteria have changed and evolved over time (23). A study that explored three scoring criteria methods in a population of 2,162 people with OSAHS was able to demonstrate wide variation in the AHI and this resulted in almost one-third of individuals being misdiagnosed (48). This may be resolved in the future by sleep medicine services adopting a more standardised or uniform scoring method (49).

2.2.7 Pathophysiology of OSAHS

The pathophysiology of OSAHS has been widely studied and numerous anatomical and pathophysiological abnormalities within the upper airway give rise to the occurrence of OSAHS (Figure 1). Other contributory factors are enlarged tongue / tonsillar tissue, excess pharyngeal soft tissue, and retro - or micrognathia (50). Quality of life (QoL) is adversely affected by OSAHS sufferers (6, 50). The underlying mechanism of OSAHS is the repeated intermittent collapse of the pharyngeal airway dilator muscles due to the loss of the sleep-induced compensatory muscle tone resulting in intermittent nocturnal hypoxaemia and sleep fragmentation (51-53). The maintenance of a patent upper airway during sleep is essential for effective nocturnal alveolar ventilation.

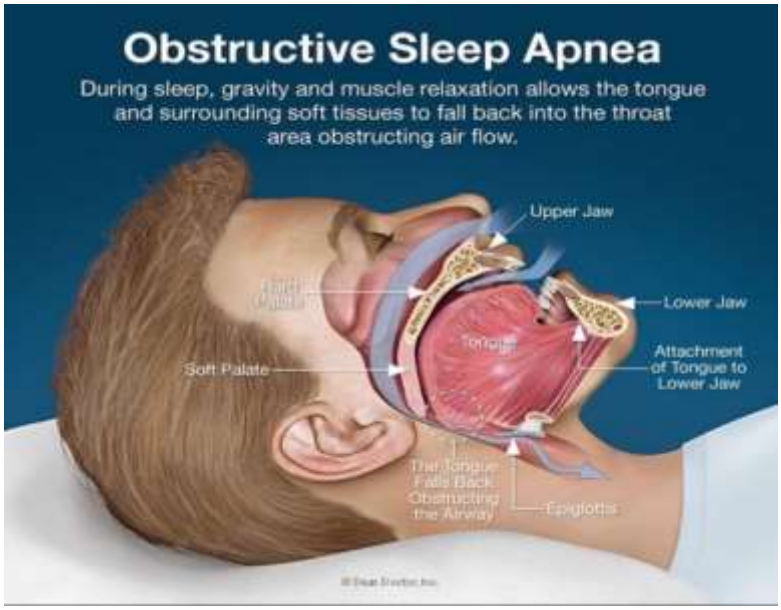


Figure 1 Mechanism of airway obstruction OSAHS

An apnoeic event is the episodic absence or cessation of breathing and OSAHS occurs where an individual experiences repeated episodes of breathing cessation as a result of partial narrowing (hypopnoea) or collapse of the pharyngeal airway during sleep (2, 54). The decrease in the airway muscle tone supporting the airway during sleep is the cause of these apnoeic/hyponoeic events. Complete airway closure (obstruction) stops

airflow and is defined as ‘apnoea’ whereas partial obstruction decreases airflow and is defined as ‘hypopnoea’. OSAHS results in periodic episodes of brief arousal from sleep to restore normal breathing and airway patency (9, 55).

Intermittent hypoxaemic events, numerous sleep arousal events, changes in intrathoracic pressure, and elevated nocturnal blood pressure are considered to be the most likely causal pathway through which OSAHS leads to cardiovascular disease (56). Increased deposition of fat in the tissues surrounding the upper airway and central adiposity can lead to increased upper airway collapse, by increased mechanical loading around the upper airway, reduced tracheal traction on the pharynx, and reduced neuromuscular activity, particularly during sleep (57). Craniofacial abnormalities such as a small maxilla and mandible are common features found in OSAHS (33). The anatomical structures of the upper airway also impact significantly on an individual’s predisposition to have an increased risk of OSAHS (58-60), and more recent research has focused on craniofacial and upper airway morphology as contributing mechanisms in OSAHS in adults (60). Neelapu et al in 2017 conducted a systematic review and meta-analysis of cephalometric studies in people with OSAHS and concluded that there is good evidence to support a reduction in pharyngeal airway space as a causative factor, the inferior position of the hyphoid bone and an increase in the anterior facial height is also implicated (32).

2.3 Treatment options in OSAHS

2.3.1 Weight loss

Obesity is a problem of global epidemic proportions as evidenced by a recent report by the World Health Organisation suggesting that over 600 million people have a body mass index (BMI) greater than 30 kg/m2 (26). The rising prevalence of obesity is directly associated with the increasing incidence of OSAHS and is recognised as the key reversible risk factor in this condition (21,

61-64). Significant and sustained weight loss is arguably the gold standard of treatment for OSAHS where this can be achieved (65, 66), although minorities of individuals are non-obese (6, 21, 67). The AASM makes recommendations for weight reduction by modifying lifestyle factors such as increasing exercise and dietary modifications that promote weight loss (71). Advising on lifestyle changes and weight reduction strategies should be addressed in every OSAHS clinical discussion (22, 63-66). A randomised controlled trial by Lopes-Padros et al in 2020 has shown that an intensive weight loss program in obese patients with severe OSAHS is effective in both reducing weight and the severity of OSAHS and also improved lipid profiles, glycaemic control, and inflammatory markers (68).

2.3.2 Treatment of OSAHS with CPAP

In 1981 Sullivan et al published their seminal paper in the Lancet describing their experience of using CPAP therapy in five patients with severe OSAHS in an inpatient setting. They described the effective reversal of sleep-induced upper airway collapse with the application of CPAP. They concluded that the safety and simplicity of their CPAP apparatus suggested that this could be replicated in the home environment (18). CPAP therapy is now accepted globally as the treatment of choice in the management of OSAHS, however until the later 1990s the effectiveness of CPAP therapy had yet to be firmly established (69). Since then a plethora of research, national and international guidelines, and consensus documents have confirmed the benefits of CPAP therapy as the gold standard in the treatment and long term management of the symptoms associated with OSAHS (3-5, 8, 9, 70-72). CPAP machines have evolved to become very small portable, quiet devices with the capacity to remotely monitor users' adherence to therapy, mask fit, mask leak, efficacy of therapy in treating sleep-disordered breathing, and record oxygen saturation monitoring via mobile networks. Web-based technology may offer the options for reduced clinical review and remote review via teleconsultation(73-75). CPAP therapy is the most common treatment that is now offered on a global

scale and is recommended by all national and international guidelines (6, 8, 9, 67, 76). Figure 2 is a device that is used in the local sleep medicine service and is typical of devices that are available in 2020.

Figure 2 An example of a CPAP device (Airsense Elite 10 CPAP unit)

Following diagnosis, the majority of individuals are established on fixed CPAP therapy, usually following auto-titration to establish therapeutic pressure settings. CPAP therapy works by generating a positive continuous flow of air from an electronic flow generator that is delivered to the user via a hose and nasal or full face mask attached to the face (62, 69). The positive airway pressure generated by the CPAP unit acts to keep the upper airway open during sleep and thus abolish the episodic breathing pauses that occur during sleep (Figure 3), (77). Traditionally the pressure required to prevent upper airway collapse is assessed by manually titrating increased CPAP pressure over time (9, 24, 67). In contrast, Automatic positive airway pressure (APAP) therapy automatically calculates the pressure required to overcome airway obstruction during an apnoeic or hyponoeic episode. The method used to determine the required pressure has been shown to make no difference to longer-term clinical outcomes of therapy (78).

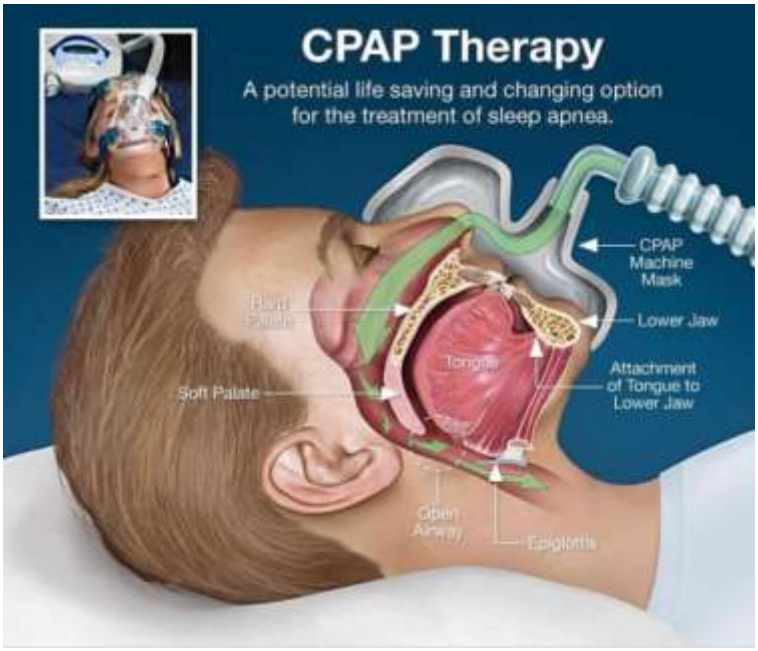


Figure 3 Mechanism of action of CPAP

Since the design of the first CPAP therapy device by Sullivan et at in 1981 (18), CPAP machines have evolved and become smaller and quieter to increase treatment acceptability and adherence rates (62). Newer CPAP devices provide some expiratory relief by reducing the pressure on the expiratory phase of breathing to make exhalation against the positive pressure more comfortable (3). APAP devices can be used where there are difficulties encountered in adhering to a fixed CPAP pressure. These devices work out the 95th percentile pressure required to abolish the apnoeic events and a fixed CPAP pressure can then be prescribed for individual patients or they can continue with the APAP device.

The CPAP pressure required to treat nocturnal events can change over time with weight loss and weight gain and patients need to be advised that they should contact their CPAP service provider if presenting symptoms reoccur such as snoring, nocturnal breathing pauses, or increasing daytime somnolence (79-81). Some sleep medicine Centres provide all patients with an APAP device for long-term treatment, the rationale being that this can

reduce the number of clinical reviews needed over time and are equally effective as CPAP devices. This is an important consideration during COVID 19 for sleep services as APAP devices can reduce the number of face-to-face visits (8, 82, 83).

More sophisticated breathing support device options are also available as Non-Invasive Ventilation (NIV) devices for those with complex sleep-disordered breathing, and some people tolerate NIV therapy better than fixed CPAP if poor adherence is an ongoing problem (83). NIV devices can deliver more modes of ventilation and are often used where people have OSAHS and obesity hypoventilation syndrome (84).

The latest development in recent years in CPAP and other breathing support devices is the ability to remotely monitor therapy adherence and other parameters of interest to the clinical teams such as efficacy of treatment and mask fit information, and the ability to adjust CPAP settings remotely (85-88). The latest CPAP units are so small that they can be fit into the palm of a hand, are much lighter and more portable for those with OSAHS who have to travel for their business or employment purposes.

CPAP therapy can have some side effects and oro-nasal dryness and nasal congestion can be relieved by the addition of humidification (89). Other common side effects reported by users are claustrophobia and mask discomfort which may result in poor adherence. This can be alleviated in some cases by good and responsive sleep clinician support and ensuring the appropriate mask is fitted (79, 89, 90). These factors are discussed in more detail in 2.3.3.

Current guidelines recommend that patients using CPAP should be reviewed regularly to assess adherence, replace disposables such as masks, filters, and hoses, manage side effects, and maintain CPAP devices (3, 8, 9, 23, 67). Although routine review and maintenance are clinically important (87, 90, 91),

this is an on-going resource-intense and time-consuming factor globally and service provision varies in different Centres and healthcare systems (8, 23).

2.3.3 CPAP adherence and challenges

CPAP adherence is problematic for several reasons such as mask fit, comfort, prescribed CPAP pressure intolerance, oral-nasal dryness, claustrophobia, abdominal bloating, psychological and social factors. A NICE technology review reported CPAP adherence as 71% (range 64–83%), in the first year of use and 79% (range 68–90%), in those that used it for > than 12 months (9). However, a systematic review by Rotenberg et al in 2016 identified that over a 20-year time frame CPAP use of greater than 7 hours per night was 34% (92). An explanation for this wide variation in adherence rates may lie in the fact that the definition of CPAP adherence is inconsistent leading to reports of CPAP adherence ranging from as low as 28% to more than 83% in more recent studies (72, 93). Both of these studies defined adherence as 4 hours per night for 70% of nights used.

Modern CPAP devices can measure CPAP wear time and efficacy at effective pressure and provide remote monitoring (72). Low adherence to CPAP limits its therapeutic benefit (72, 74), and encouraging optimal CPAP use is an ongoing challenge for users and service providers. A review by Bakker et al 2019 sought to understand two important unanswered questions regarding CPAP adherence: 1) How many hours of CPAP use are necessary to improve daytime symptoms and reduce cardiovascular risk and 2) What strategies could be implemented to optimise adherence in clinical settings. They found a dose-response relationship between CPAP use and a range of outcomes, including daytime sleepiness, functional status, and blood pressure. They also noted that current data suggests that optimal adherence rates differ depending on the outcome measures of interest. Following this review, the authors were unable to come to a definitive answer regarding their first question and recommended further RCTs are needed to define this. Regarding their second question, they suggested that combining theory-driven behavioural interventions with telemedicine could be the answer to improving CPAP adherence rates in the real-life clinical setting, and they recommend that further randomised studies are required to explore this theory (94).

The results of the Sleep Apnea Cardiovascular Endpoints (SAVE) study by Qui et at 2019 confirmed that CPAP treatment is beneficial in improving daytime somnolence and quality of life and it provided evidence that CPAP therapy in OSAHS can reduce depression and improve work place productivity even when adherence was < 4 hours per night (95). The MERGE study published in 2020 by Kelly et al also showed that there was a beneficial effect of CPAP in quality of life in people with mild OSAHS when used for up to 4 hours per night (96).

2.3.4 Mandibular Advancement Devices (MADs)

Other treatment modalities beyond CPAP therapy are available such as custom-made MADs, (Figure 4) which can be successful in managing the symptoms of mild, moderate, and severe OSAHS (36) (97-99). The evidence base for the use of MADs to treat OSAHS is well established (98, 99), however, these devices can result in side effects such as temporomandibular joint stress and pain and changes in dental morphology over time and it is recommended that users should be monitored routinely (100, 101).

Figure 4 Mandibular Advancement Device

A MADs mechanism of action is to advance or protrude the length of the mandible while simultaneously increasing the space in the upper airway (35,

99, 101). CPAP therapy can be difficult to tolerate in some individuals for several reasons as listed previously and may result in sub-optimal adherence to treatment so referral to an orthodontic service for consideration of a MAD may be a better or more aesthetic option for some individuals.

2.3.5 Bariatric surgery

Bariatric surgery is recognised nationally and internationally as a therapeutic treatment option for morbid obesity and can offer suitable individuals a more permanent weight reduction solution that can also convey beneficial effects on diabetes, hyperlipidaemia, hypertension, and the severity of OSAHS although critical complications of bariatric surgery for OSAHS have been reported (66, 102-104). Wong et al in 2018 conducted a systematic review and meta-analysis of the effect of bariatric surgery in people with OSAHS and concluded that surgical weight loss reduces the severity of OSAHS and improves symptoms (26). The successful outcomes of permanent weight loss post-bariatric surgery is an attractive long-term treatment option however it should be acknowledged that people with OSAHS are at high risk from serious post-surgical complications including death (104).

2.3.6 Palatal surgery and other surgical options

A systematic review by Rotenberg et al 2016 suggests that some individuals with OSAHS may be managed more effectively with palatal surgery than CPAP, avoiding the need for long term treatment adherence (92), and Yu Li 2019 advocates hybrid palatial surgery techniques for treatment of OSAHS in individuals who cannot tolerate CPAP therapy (105). Nasal surgery interventions such as septoplasty may have some beneficial effects in selected cases in terms of reducing the severity of the OSAHS, respiratory disturbance, reducing snoring, improving subjective daytime somnolence, quality of life, and improving nasal airway patency to improve CPAP adherence although in isolation does not have a consistent effect in people with OSAHS (106, 107). Other anatomical abnormalities can be surgically corrected with good outcomes and there is a growing evidence base for some surgical options. Facial examination can identify retrognathia of maxillary retrusion which may

be amenable to corrective maxillary / facial procedures. Obvious external nasal septal deviation may also reveal other proximal nasal defects of the nasal septum contributing to symptoms that may improve symptoms of OSAHS with corrective surgery (108).

2.3.7 Positional therapy

Positional therapy has emerged in the last decade as a simple and easy-to-use therapy to manage positional related OSAHS although its use is not widely used by the sleep medicine community due to a paucity of evidence regarding its effectiveness (109). It works by providing a vibrating feedback mechanism in a position sensor worn around the chest that alerts a person when they are lying supine to change position. A recent systematic review by Ravesloot 2017 provides good quality evidence that newer generation positional therapy devices can be an effective means in reducing the severity of OSAHS (110). A Cochrane review in 2019 concluded that CPAP is the most effective treatment for OSAHS in terms of reducing the AHI, however long-term adherence is problematic. Positional therapy user's self-reported adherence was better, but with less effect compared to CPAP on measures like the AHI. Future studies should record objective adherence to positional therapy and also establish its effect on other important outcomes like quality of life and clinical outcomes like cardiovascular events (111). The Cochrane review 2019 findings are further supported by Mok et al's publication in 2020 where they conducted the first RCT crossover study of its kind comparing positional therapy versus CPAP therapy in 40 somnolent patients who were suspected of having OSAHS. The non-inferiority primary endpoint for positional therapy was not met and their results were inconclusive (112).

2.3.8 Hypoglossal nerve stimulation

There is emerging evidence beyond clinical trials that hypoglossal nerve stimulation (HNS) implant surgery may be a successful alternative option for treating OSAHS (113-115), and has been approved by several health care systems. Weeks et al in 2018 reported that upper airway stimulation by hypoglossal nerve implant can be successfully implemented in non - academic

hospital and clinic settings (116). A systematic review by Kompelli et al in 2018 concluded that HNS is a safe and effective treatment for OSAHS in those who find CPAP therapy difficult to tolerate. It was associated with good adherence and significantly improved subjective and objective outcomes of sleep with minimal adverse effects (117), however, further trials are needed to compare invasive and non-invasive methods with other surgical interventions.

2.4 Consequences and Complications of OSAHS

2.4.1 Cardiovascular consequences of OSAHS

The underlying mechanisms of cardiovascular injury are multifactorial with frequent intermittent hypoxaemia, surges in elevated blood pressure following apnoeic spells, and oxidative stress, triggering an inflammatory cascade of events that results in repetitive injury to the vascular endothelial system with subsequent inflammation, accelerated atherosclerosis, hypercoagulation, and activation of the sympathetic nervous system (118, 119). The cardiovascular consequences of OSAHS have been extensively studied over the last three decades. OSAHS is considered to be an independent risk factor for many forms of cardiovascular disease. It is associated with increases in the incidence and progression of coronary heart disease, heart failure, stroke, hypertension, and atrial fibrillation and the potential for sudden cardiac death (5, 20, 61, 118-121). Eighteen years ago Stradling et al established that OSAHS is an independent risk factor for diurnal hypertension (122), and in subsequent years numerous studies have firmly established an association between OSAHS and hypertension, independently of age, weight, and other confounding factors (20, 118, 123-125).

2.4.2 Cardiovascular effects of adherence to CPAP therapy

Despite a clear association between cardiovascular disease and OSAHS,

prospective observational studies and RCTs have disappointingly not been able to show that cardiovascular outcomes are improved by adhering to CPAP therapy. This is a controversial subject that remains to be clarified in future research (126-129). A recent meta-analysis by Abuzaid et al in 2017 has been more promising in that they found a cardiovascular protective effect when CPAP use was greater than four hours per night (129). Quan et al 2018 reanalysed data for the SAVE study and their results have suggested that CPAP therapy may offer protective cardiovascular effects in people with OSAHS and diabetes, and improved adherence should be encouraged in these individuals (130). There is a push to study physiological and clinical phenotypes in people with OSAHS and precision medicine is entering the arena in recent publications (131-133). There are still unanswered questions regarding the true effects of CPAP therapy on cardiovascular outcomes and the sleep medicine world is waiting in anticipation of definitive answers to these questions.

2.4.3 Other Specific complications of OSAHS

2.4.3.1 Heart failure in OSAHS

Another manifestation of cardiovascular disease in OSAHS is the development of heart failure and these conditions commonly co-exist with the prevalence of heart failure in the general population greater in people with OSAHS, estimated to be as high as 50-70% (21, 134, 135).

2.4.3.2 Resistant Hypertension and OSAHS

The relationship between hypertension and OSAHS has been known since 2001 and since then numerous observational studies have confirmed this association (20, 127). OSAHS prevalence has been reported to be as much as 100% in patients with refractory hypertension (uncontrolled hypertension despite treatment with at least five antihypertensive drugs), (136, 137). The Wisconsin Sleep Cohort study followed 709 patients with OSAHS and found a linear, dose-dependent relationship between the severity of OSAHS at

baseline and the risk of developing hypertension during follow-up (137).

2.4.3.3 Atrial fibrillation and OSAHS

The prevalence of atrial fibrillation in people with OSAHS is reported as 40 to 50% and significantly impacts increased morbidity and mortality and is an independent predictor of stroke (137-143). Evidence points to OSAHS also being an independent risk factor for atrial fibrillation however its role in new-onset atrial fibrillation is unclear (143-145).

2.4.3.4 Cerebrovascular consequences of OSAHS

Multiple studies have shown that OSAHS is an independent risk factor for stroke disease and ischaemic stroke, and the severity of OSAHS also increases stroke incidence (146, 147). People presenting with a stroke or a transient ischaemic attack are three to four times more likely to have OSAHS than the general population and OSAHS is prevalent in up to 50% of people affected by stroke (148). Undiagnosed and untreated OSAHS also exacerbates the damage produced by a stroke once it has occurred and increases the risk for a subsequent stroke.

2.4.3.5 Metabolic consequences of OSAHS

Metabolic Syndrome is a collection of metabolic and cardiovascular abnormities such as impaired fasting glucose, type-II diabetes, hypertension, dyslipidaemia, and central obesity (147, 149, 150). The reported global prevalence of metabolic syndrome is between 20 - 40% and a systematic review by Xu et al in 2015 has confirmed a positive association between OSAHS and metabolic syndrome (150, 151). Although causality is not proven, there is evidence that in people with moderate to severe OSAHS, three months of CPAP treatment lowers blood pressure and partially reverses metabolic abnormalities (152).

2.4.4 Assessing excessive daytime somnolence in OSAHS

Untreated OSAHS fragments the sleep pattern and is known to have a negative impact on the physical and psycho-social aspects of life (29, 40, 42, 43, 151). Excessive daytime somnolence or sleepiness is commonly reported by people presenting to their health care provider with a suspected diagnosis of OSAHS. Simple screening tools that can assist in early detection of OSAHS are currently in use in clinical services that may support a decision for referral for diagnostic testing. There are several scales or scoring tools that can be used to measure the level of daytime sleepiness however the evidence base regarding their use as a diagnostic tool are controversial (153).

2.4.4.1 The Epworth Sleepiness Scale (ESS)

The ESS developed by Johns in 1991 is popular and used in many clinical settings (154). Despite the limitation of inaccurate reporting of daytime sleepiness, a score greater than ten is generally considered to be abnormally sleepy. The ESS sleepiness scale is intended to be a self-administered questionnaire regarding the likelihood of falling asleep in certain situations and provides a numerical measurement of an individual's general level of daytime somnolence or sleepiness. It was validated in 30 adult control subjects and 150 adults with sleep disorders, and the total ESS score significantly distinguished normal people from those with OSAHS, narcolepsy, and idiopathic hypersomnia. A more recent pictorial ESS was developed in 2011 to address any misunderstandings on how to use the scale, and the validation study found that a pictorial version of the ESS was comparable with the traditional ESS and may be a suitable alternative in cases with limited literacy (155).

2.4.4.2 STOP-Bang Questionnaire (SBQ)

In 2008 Chung et al developed the snoring, tiredness, observed apnoea, high BP, BMI, age, neck circumference, and male gender (STOP-Bang) questionnaire as a tool to screen individuals for possible OSAHS (156). The tool contains eight dichotomous (yes / no) items related to the clinical features of OSAHS and the total score range is zero to eight with the individual risk of OSAHS being greater with higher scores (157). A recent meta-analysis by Chiu et al in 2017 concluded that compared with the Berlin questionnaire, STOP questionnaire, and Epworth sleepiness scale, the STOP-Bang questionnaire is a more accurate assessment tool with both a higher sensitivity and diagnostic odds ratio for detecting OSAHS (156).

2.4.4.3 STOP questionnaire (SQ)

The snoring, tiredness during the daytime, observed apnoea and high blood pressure (STOP) questionnaire includes four questions and was developed and validated by Chung et al in 2008 as a tool to screen for possible OSAHS in preoperative surgical patients (157). It is a shortened version of the STOP-Bang questionnaire. It was found to be quick, concise, and easy to use and shows a moderately high level of sensitivity and specificity in detecting OSAHS in surgical patients (156).

2.4.4.4 Berlin Questionnaire (BQ)

The Berlin questionnaire (BQ) is a useful, brief, and clinically validated screening tool that identifies persons who are at high risk for OSAHS. It is used in some sleep medicine Centres and includes questions about snoring, daytime sleepiness, body mass index (BMI), and hypertension (158). Ramakant et al 2018 recently compared the ESS and the BQ and concluded that the ESS is a less sensitive tool in predicting OSAHS compared to the BQ (159).

2.4.5 Impact of sedative medication and alcohol on OSAHS

The role of sedative medications as a predisposing factor in OSAHS has been raised and there is concern that the administration of these drugs to people with co-existing OSAHS may worsen the condition. However, a Cochrane review in 2015 reviewed 14 studies that investigated ten sedative drugs and did not find convincing evidence that sedatives increased the severity of OSAHS, though they concluded that more studies in a larger cohort for a longer duration were needed (160). A metanalysis by Nigam et al in 2019 did not find that the use of non-benzodiazepine sedating drugs worsened the severity of existing OSAHS in the majority of participants (161).

In contrast to these findings, there is evidence that sedative and opioid-

containing drugs worsen central sleep apnoea which has a prevalence as high as 24% in chronic opioid use (162), and is reversible on discontinuation of opioid medication (163). Central sleep apnoea co-exists with OSAHS in methadone replacement programmes (164, 165).

There is sparse literature on alcohol consumption and its effects on OSAHS however a systematic review by Simou et at in 2018 concluded that alcohol consumption is associated with an increased risk of OSAHS and this could be explained as alcohol consumption reduces airway muscle tone, predisposing to upper airway collapse and generally increasing upper airway resistance (36).

2.5 Driving and OSAHS

The recognition of OSAHS as a causative factor in Road Traffic Accidents (RTA's) has been known for 40 years and a meta-analysis by Bioulac in 2017 estimated a twofold increased risk of RTA's due to sleepiness whilst driving (166). The costs to society and healthcare systems of RTA's are significant (40), and it has been estimated that 7% of RTA's for a population of male drivers can be attributable to OSAHS (44).

2.5.1 European Union Regulations

In 2016 the European Union directive 2014/85/EU - New rules on driver licensing for patients with obstructive sleep apnoea were published (167). The directive states the following:

- Applicants or drivers where moderate or severe obstructive sleep apnoea syndrome is suspected shall be referred for further authorised medical advice before a driving licence is issued or renewed. They may be advised not to drive until confirmation of the diagnosis.
- A driving licence may be issued to applicants or drivers with moderate to severe obstructive sleep apnoea syndrome who show adequate control of their condition and compliance with appropriate treatment and improvement of sleepiness, if any, confirmed by authorised medical opinion.
- Applicants or drivers with moderate to severe OSAHS under treatment shall be subject to a periodic medical review, at intervals not exceeding three years for drivers of group 1 (non-commercial drivers) and one year for drivers of group 2 (commercial drivers), to establish the level of compliance with the treatment, the need for continuing the treatment and continued good vigilance.

A European Respiratory Society task force Statement published in 2020 on Sleep Apnoea, Sleepiness, and Driving Risk provides a collective statement for clinicians advising on fitness to drive in people with OSAHS (45). All clinicians undertaking CPAP reviews regardless of the mode of consultation need to take all of these four factors into account when assessing people's fitness to drive safely. The key messages of this statement are:

- Sleep apnoea severity as assessed by the AHI alone does not predict fitness to drive in people with obstructive sleep apnoea.
- Excessive sleepiness is a major factor in determining accident risk in Obstructive sleep apnoea but does not relate to the AHI and may be partly due to other non-related sleep apnoea factors.
- Where doubt exists regarding the validity of self-reported sleepiness, further investigation such as maintenance of wakefulness test is warranted, especially in professional drivers.
- Effective and compliant treatment of obstructive sleep apnoea with CPAP largely reverses the increased accident risk and driving can resume once demonstrated.

2.5.2 United Kingdom Driving regulations

In 2018 the British Thoracic Society published a position statement advising medical practitioners and specialist sleep medicine teams of their roles and responsibilities regarding advice to be given to individuals who are suspected of having OSAHS and have excessive daytime sleepiness that may affect their ability to drive safely (90). The roles and responsibilities of the clinicians involved in the care of people with OSAHS are outlined below:

A general practitioner or other referring healthcare practitioner should:

- Recommend that a patient does not drive if excessive sleepiness is having or likely to have an adverse effect on driving, whatever the cause.
- Arrange onwards referral for specialist review if indicated and ensure prioritisation of cases as urgent where patients report an impact on

vigilant critical activities such as driving, flying, or operating dangerous machinery.

The sleep specialist team member (clinician, sleep nurse, sleep physiologist) should:

- Advise the patient that if they hold a driving licence, they must follow the Driver and Vehicle Licensing Authority (DVLA) guidance.
- Help the patient to assess the likely impact of their symptoms on their safety to drive with a detailed driving history including distances driven, episodes of drowsiness whilst driving, and information from family members, identifying any crashes or near misses which could be attributed to sleepiness or inattention.
- Advise the patient whether they have just OSA (no need to contact DVLA), or OSA plus sleepiness (need to contact the DVLA). The DVLA's specific forms give options for "Which condition have you been diagnosed with?" including "Sleep Apnoea (with excessive sleepiness during normal waking hours), or OSAS or other sleep condition". The clinician should record the discussion and recommendations about driving, and whether the DVLA should be notified, in the patient's notes.

The Patient:

- Report their symptoms honestly to their clinicians.
- Notify the DVLA regarding a diagnosed medical condition when required to do so, including OSA which affects their ability to drive safely.
- Comply with the instructions of the DVLA

The DVLA website A-Z page provides a link to the forms that need to be completed. On receipt of the appropriate form, the DVLA opens enquiries for car / motorcycle license holders and for lorry and bus licence holders, which are then sent to the patient's GP / hospital consultant to be completed, as necessary.

2.6 Organisation and delivery of care in OSAHS

2.6.1 Clinical review

National and international guidelines recommend regular review of people with OSAHS who are using CPAP therapy however there is no consensus on the frequency and modality of the clinical review (3, 24, 168). Long-term follow-up may vary in frequency, or potentially be “on-demand / or request,” and can be conducted by a range of healthcare clinicians with the option of using telemonitoring and teleconsultation (168, 169). A practice guideline by the American Academy of Sleep Medicine recommends review should include monitoring and troubleshooting of objective effectiveness of CPAP therapy including usage data to ensure adequate treatment and adherence is being achieved. This review should be delivered by a qualified clinician (e.g., physician or advanced practice provider, trained healthcare provider), following CPAP initiation and during treatment of OSAHS (67).

2.6.2 Who conducts the clinical review

A variety of healthcare clinicians can undertake a review of people who are using CPAP therapy. Physicians, respiratory scientists and therapists, sleep technologists, specialist nurses, and other sleep specialists may all be involved in the early clinical review and longer-term review of CPAP users (3, 169, 170). Given the increased prevalence of OSAHS and the number of people requiring CPAP therapy, overstretched sleep medicine services need to rethink their service delivery and design pathways and by whom, and where will future services be delivered to meet the rising demand for the service with no increased capacity.

There is evidence that a nurse-led service for home diagnosis, CPAP treatment initiation, and ongoing management of selected patients with OSAHS is feasible and cost-effective, with outcomes comparable to conventional inpatient services (169, 171-174). Comer 2017 concluded that a nurse-led respiratory sleep medicine service for diagnosis and initiation of

CPAP therapy was fit for purpose and was well received by patients (169). A study in 2015 also concluded that a simplified nurse-led model of care has demonstrated non-inferior results to physician-directed care in the management of symptomatic moderate-severe OSAHS while being less costly (61). An internet search reveals that many Centres across the UK offer specialist nurse and specialist respiratory physiologist-led CPAP review with respiratory consultant oversight, thus freeing up the physicians to see more complex cases.

In the USA, there is a major concern regarding the rapid decline in board-certified sleep medicine specialists and some solutions that have been suggested are increased training for generalist clinicians, including non-physicians, in the diagnosis and management of symptomatic, uncomplicated OSAHS (175-178). An RCT conducted in Europe in 2015 provided evidence that follow-up in primary care following suitable training showed no worsening CPAP compliance compared with a specialist model and was a cost-effective alternative (174).

Chakrabarti et al 2020 concluded that an intelligent computer-guided consultation programme creates opportunities in sleep medicine management pathways to utilise non-specialists working effectively and safely under specialist supervision to assist with diagnosis of OSAHS and undertake clinical reviews (179).

2.6.3 Method of Clinical review

2.6.3.1 Traditional face-to-face review

The review of people using CPAP therapy has traditionally been face-to-face in a secondary care clinic setting. There are advantages to this model of care as the clinician and patient can interact directly and any physical or clinical examination or required tests can be conducted at this review (182). The clinician can stay in the same place and avoid travel, and this may be time-efficient for clinical service provision. However, there are also some disadvantages of face-to-face review for CPAP users, such as time taken off work and the distance to travel to tertiary Centres, increased fossil fuel consumption, and dealing with traffic congestion which has a negative environmental impact in terms of increased carbon emissions. There is also the potential for increased road traffic incidents (168, 180, 181).

2.6.3.2 Teleconsultation clinical review

The introduction of telemedicine to the stage in the last two decades has driven a revolution in OSAHS management and changed the landscape for the treatment and long-term management of this condition (181-185). There are several potential ways in which technology can support CPAP reviews: (168, 185-188).

- Facilitation of remote reviews
  Telemedicine can support remote reviews and this can have benefits for both clinicians and patients and may be a more convenient mode of service delivery.

- Telemonitoring of CPAP adherence
  Adherence to CPAP therapy can be overseen remotely and important parameters such as CPAP hours of use, reduction in the AHI, and mask leak can also be monitored, reducing the need for face-to-face clinic

reviews. CPAP settings can also be changed remotely reducing the need for face-to-face reviews.

- Reducing Carbon footprint

  By obviating the need to travel to satellite clinics for clinicians and reducing face-to-face visits for CPAP users, the miles travelled for review can be significantly reduced with the beneficial effects of reduced fossil fuel consumption and reduced carbon emissions.

- Improved access to care

  Telemedicine can improve access to care, particularly where there are large geographical distances to travel to main sleep medicine Centres. It can help to reduce waiting time for investigations and treatment and improve patient-clinician safety and deliver more person-centred care.

- Reduced traffic congestion and work productivity

  Traffic congestion can also be avoided, of particular importance in densely populated cities. It can also reduce the loss of work productivity in terms of time taken off work to travel for face-to-face reviews.

2.7 Summary of chapter 2

OSAHS is a common and increasingly prevalent condition that is being driven by the global obesity epidemic. Weight reduction is the first treatment recommendation and may reverse this condition, however, CPAP therapy remains the gold standard therapy of choice for most people. Other treatment options are discussed, and research is beginning to identify phenotypes of OSAHS that may lead to a precision-medicine approach to the diagnosis and treatment of this OSAHS in the future. This introductory chapter outlines the increased risk of accidents and cardiovascular, cerebrovascular, and metabolic complications of having a diagnosis of untreated OSAHS and the impact that some sedative medication and alcohol can contribute to the

severity of OSAHS. The various assessment tools that are available in clinical practice to assess the level of daytime somnolence are described, as this is directly related to assessing an individual's fitness to drive until their symptoms are controlled on CPAP therapy. The vehicle and licensing agencies rules and reporting regulations for clinicians and people with OSAHS to maintain individual and societal safety by avoiding RTA's in sleepy drivers are presented.

The importance of comprehensive reviews of people with OSAHS is discussed and the current lack of consensus about what this should include. With the rapid deployment of telemonitoring and teleconsultation options for CPAP therapy users, there are many opportunities to streamline and provide more efficient, person-centred, safe, and cost-effective delivery of sleep clinic review services.

Chapter 3: Aims and objectives of PhD

In the last chapter, I outlined the history of OSAHS and its contributing factors, its prevalence, economic impact, medicolegal implications, diagnostic options, measuring its severity, the pathophysiology of this condition, and its treatment options. I outlined the consequences and complications of OSAHS, tools to assist with assessing the level of daytime somnolence, UK and European driving regulations, the organisation and delivery of care, and the options available for current reviews including telehealth. Current guidelines recommend regular review of CPAP users however there is no collective agreement regarding the content or components of the review and how often a review should take place. In this thesis, I will explore both the content of a clinical review and mode of delivery and how often this should take place.

Aims and objectives:

I aimed to inform the mode of delivery and content of CPAP reviews for people with OSAHS in three distinct but interlinked projects:

I. *Systematic review:*

   To systematically review the literature on the effectiveness, acceptability, and health service resource implications of using the combination of real-time telemonitoring of CPAP usage data plus remote consultations to undertake routine reviews of patients using CPAP compared to face-to-face care.

II. *e-Delphi:*

   a) To reach consensus amongst a panel of international sleep clinicians, academics, and CPAP users on the most important components to include in a CPAP review and how often a review should take place.

   b) To develop a structured sleep medicine review template that records the important components identified by the e-Delphi consensus panel.

III. *Implementation study*

To introduce the template in three diverse sleep apnoea Centres for use in face-to-face or remote consultations and observe implementation using mixed methods.

Chapter 4: Systematic review

There is increasing interest in telehealth as an option for the clinical review and longer-term management of CPAP users in sleep medicine services. In this chapter, I will explore the evidence base for the role of telehealth (teleconsultation and telemonitoring), in CPAP therapy users.

The systematic review is published:

- Phyllis Murphie (PM), Stuart Little (SL), Brian McKinstry (BMcK), Hilary Pinnock (HP). 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

4.1 Rationale for the systematic review

Technology-enabled care is rapidly evolving and transforming healthcare on a global scale and telemedicine and telemonitoring in sleep medicine have emerged as additional options in diverse health care settings in this specialty over the last few decades. Telehealth methods to support the clinical review of CPAP users have the potential to deliver cost-effective and convenient care (87-89, 185, 186, 189). Potential benefits include increased therapy adherence, user satisfaction (184), and reduced loss of workforce by not attending for a face-to-face review, avoidance of traffic congestion in urban Centres, and consequent positive effects on the environment with reduced fossil fuel consumption and carbon emissions (87, 181, 187, 189). Recent evidence has suggested that the use of CPAP telemonitoring may improve 90-day treatment adherence, however, the literature regarding combining both telemonitoring with teleconsultation is limited (187), and longer-term studies are needed to assess the duration of the effect on adherence (75). There is also evidence to support teleconsultation as an acceptable alternative to face-to-face review (74, 85, 189).

The AASM in 2015 published a position statement on the use of telemedicine for the diagnosis and treatment of OSAHS (4). They convened a task force that recommended clinical standards of care for sleep telemedicine services should be comparable to face-to-face review, including all aspects of diagnostic and therapeutic treatment plans compared to usual care. My specific interest was in routine reviews as the increasing prevalence of OSAHS is straining limited healthcare resources and viable alternatives are needed that can deliver safe, effective, and person-centred care closer to their place of residence.

Aims and objectives of the systematic review

I aimed to systematically review the literature on the effectiveness, acceptability, and health service resource implications of using the combination of real-time telemonitoring of CPAP usage data plus remote consultations to undertake routine reviews of patients using CPAP compared to face-to-face care in any healthcare setting.

4.2 Methods

The systematic review protocol is registered with the international prospective register of systematic reviews (Appendix 2 – PROSPERO registration: CRD42015019455). I did not make any changes to the protocol.

An ethics self-audit checklist was submitted to the University of Edinburgh, Centre for Population Health Sciences Research Ethics Group for this systematic review (Appendix 1). The systematic review is reported according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement for reporting on systematic reviews (190). PRISMA is an evidence-based minimum set of items for reporting in systematic reviews and meta-analyses that focuses on the reporting of reviews evaluating randomised trials. The PRISMA checklist is in Appendix 3. I followed the methodology described in the Cochrane Handbook (190-192).

4.2.1 Inclusion and exclusion criteria

		Definitions
Population	Adults with a diagnosis of OSAHS on CPAP therapy	Sleep apnoea is defined as a condition in which a person experiences repeated episode of apnoea because of a narrowing or closure of the pharyngeal airway during sleep
Intervention	The application of telehealth to review people with a diagnosis of OSAHS and using CPAP	This was defined as the use of telehealth to: review patients remotely (e.g. telephone or video consultation) and telemonitoring of CPAP usage I excluded telephone calls without telemonitoring and also telemonitoring without remote consultation
Comparator	Usual clinical care without telehealth	Normally delivered face-to-face but may include some telephone calls (but without telemonitoring of CPAP data)
Outcomes	*Primary outcome*: Primary outcomes of interest were: • the proportion of patients who had received a telehealth review • adherence with prescribed CPAP therapy. *Secondary outcomes*: Secondary outcomes of interest included current control of symptoms of OSAHS, patient and clinician acceptability and satisfaction with the telehealth review, and health service resource use/cost implications compared to face-to-face care	CPAP adherence which is typically defined as recorded use of CPAP therapy for > 4 hours per night however details vary between studies Clinical symptoms include validated measures of sleepiness, quality of sleep, and quality of life
Setting	Any setting	Typically, the patient will be in the community, but the healthcare practitioner may be based in primary or secondary care
Study design	Randomised controlled trials (RCTs), quasi-RCTs, and controlled clinical trials (CCT).	

Table 2 PICOS Search strategy

4.2.1.1 Reason for my decisions on inclusion and exclusion criteria

- Population

My interest was in adults with a diagnosis of OSAHS who were using CPAP therapy.

- Intervention

I chose to only include interventions that had both a consultation remote to the main health-care facility (video or telephone consultation) and the facility for reviewing real-time telemonitoring of data from the CPAP device (defined as data transmitted and monitored by, or available to the reviewing clinical team via a web-based platform at any time). I excluded telephone calls without telemonitoring and also telemonitoring without remote consultation. This decision was based on my interest in published research that provided access to almost identical CPAP review care with the same real-time face-to-face teleconsultation and access to real-time adherence data.

- Comparator

The comparator was the usual face-to-face review of CPAP therapy users who did not use telehealth.

- Outcomes

My primary outcome of interest was the proportion of patients who had received a telehealth review and adherence with prescribed CPAP therapy. My Secondary outcomes of interest included current control of symptoms of OSAHS, patient and clinician acceptability and satisfaction with the telehealth review, and health service resource use / cost implications compared to face-to-face care.

- Setting

Any healthcare setting was included with typically, the patient being in the community, but the healthcare practitioner may be based in primary or secondary care.

- Study design

I searched for randomised controlled trials (RCTs), quasi-RCTs and controlled clinical trials (CCTs) in CPAP therapy users that compared teleconsultation combined with daily telemonitoring of CPAP data with face-to-face care. I chose to only include these types of studies as I considered these would provide the most reliable evidence on the effectiveness of any telehealth intervention compared to usual care and minimise the risk of bias. My interest was in routine reviews of people established on CPAP, as opposed to the coaching and additional support associated with the initiation of treatment.

4.2.2 Search strategy

I searched ten electronic databases; Allied and Complementary Medicine (AMED); British Nursing Index (BNI); Cumulative Index of Nursing and Allied Health Literature (CINAHL); Cochrane Library; Database of Abstracts of Reviews of Effects (DARE); Excerpta Medica dataBASE (EMBASE); Latin American and Caribbean Health Sciences Literature (LILACS); Medical Literature Analysis and Retrieval System Online (MEDLINE); Web of Science and Z Electronic Table of Contents (ZETOC) (Table 3). The bibliographies of included studies were scrutinised to identify possible additional studies and I also hand searched relevant sleep medicine and respiratory journals (Sleep Medicine, Journal of Clinical Sleep Medicine, Sleep, Sleep Medicine Reviews, Thorax, European Respiratory Journal, Breathe, British Medical Journal Open Respiratory Research, Respiratory care, American Journal of Respiratory and Critical Care Medicine, Chest, European Respiratory Review, Respiratory Medicine, Nature Partner Journals Primary Care Respiratory Medicine). Search dates were not limited as the literature review identified possible research of the use of telemedicine in sleep medicine over the last 20 years. I only included studies published in English as I did not have access to the resources to permit translation. The search was carried out in November 2015.

Search strategies for MEDLINE, EMBASE, AMED
1. Sleep apnea syndromes/ or sleep apnoea, obstructive/ 2. Sleep-disordered breathing. mp. or Sleep apnea Syndromes/ 3. Continuous positive airway pressure/ 4. CPAP therapy. mp. 5. Nasal CPAP.mp. 6. Sleep apnoea syndrome. mp. 7. 1 or 2 or 3 or 4 or 5 or 6 8. Obstructive sleep apnoea hypopnoea syndrome. mp. 9. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 10. telemedicine/ or teleconsultation/ 11. (Telehealth* or ehealth or telemonitoring*).mp. 12. 10 or 11 13. Clinical trial/ or controlled clinical trial/ or randomised controlled trial/ 14. quasi-randomised. mp. 15. 13 or 14 16. 9 and 12 and 15 17. adherence.mp. 18. compliance.mp. 19. exp "patient acceptance of health care"/ 20. Satisf*.mp. 21. 17 or 18 or 19 or 20 22. 9 and 12 and 21
Search strategy for The Cochrane Library, ISI Web of Science, CINAHL, LILACS, ZETOC, DARE and British Nursing Index
1. Sleep apnea syndromes/ or sleep apnoea, obstructive/ 2. Sleep-disordered breathing. mp. or Sleep Apnea Syndromes/ 3. Continuous positive airway pressure/ 4. CPAP therapy 5. Nasal CPAP 6. Sleep apnoea syndrome 7. S1 OR S2 OR S3 OR S4 OR S5 OR S6 8. Obstructive sleep apnoea hypopnoea syndrome 9. telemedicine/ or teleconsultation/ 10. telehealth or e-health or telemonitoring 11. S9 OR S10 12. Clinical trial/ or controlled clinical trial/ or randomised controlled trial/ 13. quasi randomised 14. S12 OR S13 15. S7 AND S11 AND S14 16. adherence or compliance 17. exp "patient acceptance of healthcare" 18. patient acceptance of healthcare 19. satisfaction 20. S16 OR S17 OR S18 OR S19 21. S7 AND S11 AND S20

Table 3 Search strategies

4.2.3 Selection of studies

Selection was carried out in three stages:

- I carried out an initial sift of the papers that had been retrieved and rejected any obviously unrelated abstracts. The reason for this preliminary step was the time and resource restraints of undertaking this part-time PhD whilst working in my NHS role.
- I adhered to the principles of this methodology and included the study setting and quasi-RCTs and controlled clinical trials as some non RCTs warranted inclusion in the review.
- I then screened the titles and abstracts with 25% checked by HP or BMcK (agreement 100%). This was also a practical decision based on time constraints however, to ensure that I did not overlook potentially eligible trials, I included all abstracts in which any form of telehealth and OSAHS and / or CPAP were mentioned. I included interventions that had both a teleconsultation remote to the main health-care facility (video or telephone consultation), and the facility for reviewing real-time telemonitoring of data from the CPAP device (defined as data transmitted and monitored by, or available to the reviewing clinical team via a web-based platform at any time).
- The full text of all potentially eligible studies was retrieved and independently assessed against the inclusion criteria by two reviewers (PM, and SL or HP); any disagreements were resolved by team discussion. The selection process was summarised using a PRISMA flow diagram (Figure 5).

4.2.4 Quality assessment

The methodological quality of included studies was assessed (by PM and HP) using the Cochrane Risk of Bias (ROB) tool (192) and the Cochrane Effectiveness and Practice Organisation of Care (EPOC) guidelines (190). The Cochrane risk of bias tool covers six domains of bias: selection bias, performance bias, attrition bias, detection bias, reporting bias, and other

biases. The following seven domain-based parameters were used to assess bias in the systematic review: adequate sequence generation, allocation concealment, blinding of participants and personnel, blinding of outcomes, how incomplete outcome data were addressed, completeness of reporting, and freedom from other sources of bias. Included studies were given an overall risk of bias by PM and HP and any differences of opinion were resolved by discussion with BMcK and SL.

4.2.5 Dealing with lack of information

If, after full-text assessment, it was still unclear whether a study fulfilled the inclusion criteria, or if I required clarification of any details relating to the intervention, study reporting, or outcome data, I contacted authors by email for further relevant information.

4.2.6 Data extraction

I used a piloted data extraction sheet to extract characteristics of included studies under the headings; author and study year, country, risk of bias, type of trial, number of participants, follow up, setting, intervention, and comparator, patient demographics, personnel, arrangements for remote consultation, telemonitoring of CPAP adherence, aspects of care addressed, duration and intensity of intervention, and care provided to the comparator groups.

4.2.7 Outcomes of Interest

The primary outcomes of interest were:

- The proportion of patients that received both a teleconsultation and telemonitoring review.
- Adherence with prescribed CPAP therapy that was recorded by telemetric data transmission.

The Secondary outcomes of interest were:

- Current symptom control using a validated score (e.g. the Epworth sleepiness scale or the Functional Outcomes of Sleep Questionnaire).
- Patient and clinician acceptability and satisfaction with the teleconsultation / telemonitoring model of clinical review.
- The health service resource use / cost compared to face-to-face care.

4.2.8 Data synthesis

I expected that I would identify a limited number of eligible studies (based on my preliminary scoping work). I, therefore, planned to undertake a narrative synthesis of the data. The heterogeneity or variation in the data reporting in the included studies in this systematic review would be assessed using Cochrane methodology (190, 192).

4.3 Results

362 potentially relevant publications were identified from the literature search, from which 17 full-text articles were reviewed with 12 being excluded (71, 86, 87, 184, 186, 193-199), as they did not match my inclusion criteria. Table 4 – describes the reasons for excluding these studies.

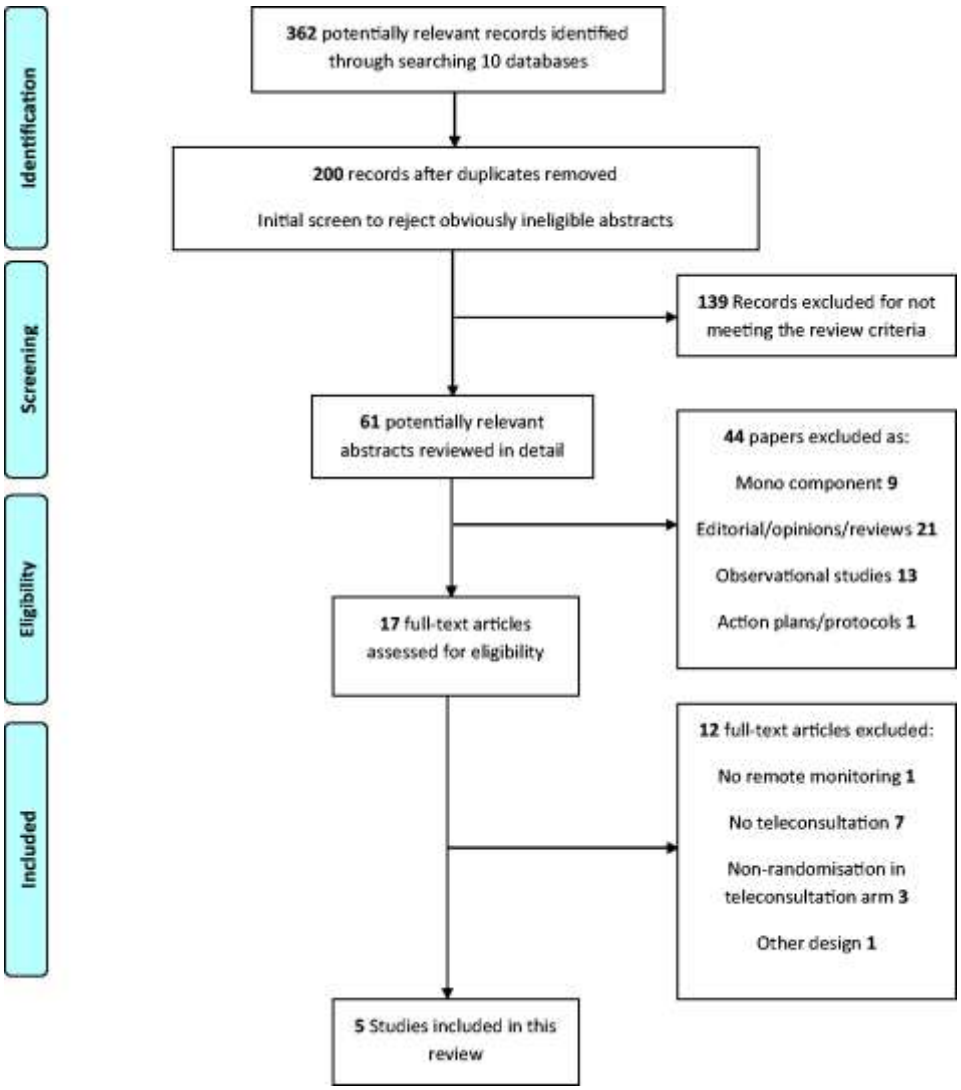


Figure 5 PRISMA diagram

Reference Country	Study design, number of participants Follow-up	Intervention and comparator	Target group Participants	Reason for exclusion
Abreu et al 2013 (193) Portugal	RCT n=51 FU: 1m	Telemonitoring of CPAP compliance and efficacy data compared with active phone call care or usual care	Adults with OSAHS Mean age 54 % male 82	Abstract only published No teleconsultation used in study
Dellaca et al 2011 (194) Spain	Pilot observational study n= 20 FU: 1 week	Remote CPAP pressure titration with TM system in the patient's home by a sleep lab technician No comparator	Adults with OSAHS Mean age 56 yrs % male not reported	No control group and no teleconsultation
DeMolles *et al* 2004 (195) USA	Pilot RCT n=30 FU: 2m	Use of TLC technology system in addition to UC Comparator - UC	Adults with OSAHS Mean age 50 yrs % male not reported	No daily telemonitoring or teleconsultation
Farre 2009 (196) Spain	Editorial			Journal editorial
Fraysse *et al* 2012 (197) France	Observational multicentred study n=90 FU: not reported	Feasibility of CPAP telemonitoring No comparator	Adults initiating CPAP for OSAHS	No results published to date
Isetta *et al* 2014 (86) Spain	Study 1 - prospective observational study n= 50 FU: same day	Study 1- Patient satisfaction with teleconsultation No comparator	People with OSAHS Mean age 62 % male 64	Study 1- no comparator
Isetta et *al* 2014 (86) Spain	Study 2- RCT: n=40 FU: same day	Study 2 CPAP training application via teleconference Comparator- usual care	People with OSAHS being commenced on CPAP therapy Mean age 59 % male 80	Study 2- no daily telemonitoring
Isetta *et al* 2015 (87) Spain	Multicentre RCT: n=139 FU: 1,3 6 m	Bayesian cost-effectiveness analysis of a Telemedicine- based CPAP strategy for follow-up Comparator face-to-face follow up	People with OSAHS requiring CPAP therapy Mean age 49 % male 86	No telemonitoring of CPAP data Data were downloaded via the data card at the end of the study (not for review purposes)
Kwiatkowska 2009 (199) Canada	Proposed framework for development of a telehealth support system	Proposed Telehealth - Framework for Supporting the Treatment of OSA	People with OSAHS living remotely from specialist sleep clinics	No study Description of a framework
Kwiatkowska 2010 (198) Canada	Editorial			Journal editorial

Nilius *et al* 2012 (71) Germany	RCT: n=84 FU:3m	Weekly telephone support re CPAP use for 12 weeks Comparator- usual care	Adults with OSAH new to CPAP therapy Mean age 50 % male 62	No telemonitoring of CPAP data usage
Parikh et al 2011 (184) USA	Prospective survey of patient satisfaction n=90 FU: 2 weeks	Teleconsultation review remotely Comparator- usual care	Adults with OSAHS prescribed CPAP therapy Mean age 51 % male 43	No telemonitoring of CPAP data usage
Sparrow et al 2010 (186) USA	RCT n=250 FU: 12 m	Theory-driven interactive voice response system designed to improve CPAP adherence using a telephone-linked communications system Comparator- educational control TLC system Provided general health education via a TLC system	Adults with OSAHS prescribed CPAP therapy Mean age 55 % male 82	No telemonitoring of CPAP data usage

Table 4 Reasons for excluding studies

4.3.1 Five papers fulfilled our inclusion criteria

Table 5 describes the characteristics of the studies included and Table 6 describes the aspects of care addressed in these studies.

Four studies were from North America: A Large private hospital in San Diego (200), The University of Kansas / St Francis Hospital (201), The University of British Columbia, Vancouver, Canada (202), The Walter Reed Army Medical Centre in Washington (203) and one from Europe, Burgos in Spain (204). All the telehealth-based interventions were delivered in the community setting. The five studies included a total of 269 participants and all included real-time telemonitoring of CPAP adherence; three studies utilised telephone consultations as a means of communication with participants (202, 204), and the other two utilised teleconsultation for remote consultations (201, 203).

The heterogeneity or variation in the data reporting in the included studies in this systematic review was assessed using Cochrane methodology (192). The only homogenous data reported in all five included studies were that all participants were newly diagnosed with OSAHS before commencing CPAP therapy and all the studies reported that adherence data was transmitted telemetrically. There was significant heterogeneity in other outcomes of interest: one of five included studies was a CCT (204), two had very small sample sizes (201, 204), and there was wide variation in the length of follow up between one to six months in all studies. The proportion of people reviewed was only reported in two studies, only two included the Epworth sleepiness scale (200, 202), with only two reporting on the cost of the interventions (201, 204), and patient / clinician acceptability was not reported in four studies (200, 201, 203, 204). This level of heterogeneity in the included studies allowed me to conclude that a meta-analysis of the data was not appropriate (Table 5).

Reference Country ROB	Study design No of participants FU	Intervention and comparator	Target group participants	Proportion reviewed	Adherence to CPAP	Symptom control	Satisfaction acceptability	Costs	Comments
Coma del Corral et al 2013 (204) Spain Mod/high-risk RoB	CCT n = 16 FU: 6 m	TH (video consultation with telemonitoring of CPAP usage) v UC (with CPAP telemonitoring)	Adults with OSAHS Mean age 53 years 63% male	Not measured	There was no between-group difference in adherence to CPAP: Telehealth 75% v UC 85% (significance not reported)	Not measured	The visual analogue scale used (1–10) reported as 9.5 for both groups Clinician satisfaction not reported	Staff and polysomnography and disposables were reported Telemedicine / teleconsultation costs not reported for CPAP users	TH to remote sites feasible; may provide specialised sleep medicine services to geographically distant populations
Fox et al. 2012 (202) Canada Mod/high risk RoB	RCT n = 75 FU: 3 m	TH (telemonitoring with telephone support if needed) v UC (with telephone support where needed)	Adults with OSAHS Mean age 53 years 80% male	Compared with UC, more time was spent with the TH patients: TH: 210 min (SD 42) - vs UC: 143 min (SD 48) p< 0.0001	Adherence greater in TH group: mean hours /night: TH 3.2 v UC: 1.8 (p = 0.006)	Epworth sleepiness score - no significant between-group differences (TH 5.1 v UC 5.2)	Not measured	Not measured	TH may improve adherence to CPAP at initiation of therapy
Smith et al. 2006 (201) USA Mod/high-risk RoB	RCT n = 19 FU: 3 m	TH support for CPAP adherence via telehealth equipment with inbuilt camera enabling clinician consultation with patient's v TH (W-B = well-being advice)	Adults initiating CPAP for OSAHS Mean age 63 years % male not reported	Not measured	TH support for CPAP adherence improved proportion achieving 80% use rate. TH (CPAP support): 90% vs TH (W-B): 44% (p = 0.033)	Not measured	Proportion rating service 'positively' TH (CPAP support) 100% v TH (W-B) 75% (significance not reported)	TH (CPAP support) costs were less than TH (W-B) costs: TH (CPAP support): US $420 vs TH (W-B) US $1500 (significance not reported)	Structured video-consultations may facilitate problem-solving strategies via visual camera observation and overcome barriers to adherence
Stepnowsky et al. 2007 (200) USA Mod/high-risk RoB	RCT n = 45 FU: 2 m	TH (regular telemonitoring and telephone calls) v UC supported by telephone calls	Adults initiating CPAP for OSAHS	Not measured	Non-significant trend to greater adherence in TH group. Hours/ night:4.1	Epworth sleepiness score. No significant between-group	Likelihood of continuing CPAP higher in TH than UC group on a score of 1 to 5 (4.8 vs 4.3: p = 0.05)	Not measured	TH monitoring of CPAP adherence with rapid clinician support to guide

			Mean age 59 years 98% male		(SD 1.8) vs UC: 2.8 (SD 2.2) (p = 0.07)	difference TH 9.2 (SD 6.6) v UC 9.9 (SD 5.2) p = 0.72			management may improve adherence and commitment to using CPAP
Taylor et al. 2006 (203) USA Mod risk RoB	RCT n = 114 FU: 1 m	TH via Health Buddy with daily automated feedback v UC Both groups had telephone support if needed	Adults initiating CPAP for OSAHS Mean age 45 years 71% male	TH group had fewer 'walk-in visits' than the UC group (1-week vs 3-week)	No between-group differences in adherence. Hours/night TH: 4.29 (SD 2.15) v UC: 4.22 (2.05) p = 0.87	Modified Functional Outcome of Sleep Quality (reported - no differences between TM and UC group	No between group differences in client Satisfaction Questionnaire. TH: 28.5 (SD 3.1) v UC 28.0 (SD 3.5) p=0.43	Not measured	Very short follow-up (28 days)
ROB- Risk of bias, FU- follow up, CCT- controlled clinical trial, n-number, m-month, TH-telehealth, v-verses, UC- Usual care, RCT-randomised controlled trial, SD- standard deviation, WB-wellbeing, $-dollar									

Table 5 Characteristics of included studies

Study year	Country, Setting	Patient demographics	Description of delivery of Intervention	Aspects of care addressed	Duration and intensity	Control
Coma del Corral *et al* 2013 (204)	Burgos, Spain. Hospital-based study led from main site to nurse supported remote site	New diagnosis of OSAHS commenced on CPAP therapy Demographics of those randomised not reported. 16 randomised and 7 received intervention, 9 controls	Central-based sleep research unit and distant centre 80Km away. Team members included physicians at central unit and nurses at remote site Face-to-face consultation for commencement of CPAP at central unit and via nurse assisted videoconference remotely	3 of 5 outcomes of interest addressed Compliance/Adherence - reported as CPAP therapy for > 4 hours per night for 5 nights per week reported Control of symptoms- not reported Patient/clinician acceptability - not reported Patient satisfaction - reported in both groups Clinician satisfaction- not reported Cost of telemedicine - reported versus usual face-to-face care	Interventions conducted by a physician based centrally and nurse assisted remote teleconsultation arm Duration of intervention 6 months. All received at least 2 consultations during the study	Conventional face-to-face follow up
Fox *et al* 2012 (202)	Sleep disorders program, University of British Columbia, Vancouver, Canada	Patients with new diagnosis of OSAHS commenced on CPAP therapy. Demographics 80% male, mean age 53.5 39 received intervention, 36 controls	Introductory CPAP session conducted for all patients by trained respiratory therapists, other team members were physician-researchers	3 out of 5 outcomes of interest reported. Adherence-reported as number of minutes of use per day Control of symptoms – Epworth sleepiness score used Patient/Clinician acceptability - not reported Patient satisfaction - measured using a visual analogue scale Clinician Satisfaction - not reported Cost of Telemedicine - not reported	Interventions delivered by trained respiratory therapists, face-to-face and telemedicine review conducted by a physician and respiratory therapist Standard care group contacted at day 2 to assess progress /adherence and discuss any problems Further face-to-face review at 4-6 weeks with physician / CPAP coordinator. CPAP adherence/efficacy information downloaded at this visit, week 8-and 3-month clinic visits Telemedicine group data sent daily via modem directly to web-based	Usual standard face-to-face care

					database monitored by research co-ordinator. Contacted by a research co-ordinator at day 2 and at other times if needed to discuss compliance and any problems. Review by physician 4-6 weeks and at 3 months with adherence data	
Smith *et al* 2006 (201)	University of Kansas/ St Francis Hospital USA. Home-based nurse-led study intervention via telehealth link	Patients with new diagnosis of OSAHS (had been non-adherent across 3 months) recommenced on CPAP therapy Mean age 63 No difference in severity of OSAHS. % male - not reported. 10 received intervention, 9 controls	Nurse-led study in patient's homes using telehealth equipment with built-in two-way cameras 2-way telehealth sessions over the 12-week period regarding adherence to CPAP therapy and how to manage any problems encountered in the intervention group and in the control group the 12-week health care sessions provided information on importance of daily vitamin intake	4 of 5 outcomes of interest addressed. Compliance/Adherence- reported adherence of > 4 hours/night for 9 of 14 nights Control of symptoms- not reported Patient acceptability - reported by both groups Clinician acceptability - not reported Patient satisfaction - reported using evaluation survey Cost of telemedicine- reported	Study interventions delivered in patient's homes by nurses familiar with CPAP and clinical trial research. Duration of intervention was 12 weeks (3 x 30 min telehealth visits in first week and subsequent 1 x 30 min visits/week for next 11 weeks).	Both groups commenced on CPAP therapy but only one group received coaching in importance of CPAP adherence and monitoring of problems encountered on treatment. Placebo-controlled trial model to control for Hawthorne effect.

Stepnowsky *et al* 2007 (200)	Veterans Affairs San Diego Healthcare System (VASDHS), San Diego, CA, USA. CPAP clinical staff and research team at a large private hospital	Patients with new diagnosis of OSA commenced on CPAP. Demographics 98% male, mean age 59, 20 received the intervention, 20 controls	CPAP therapists (non-blinded) delivered all study interventions, Initial face-to-face start-up visit for all participants Other team members were research physicians and statisticians	3 out of 5 outcomes of interest were addressed Compliance/Adherence – reported as percentage of use Control of Symptoms – Epworth sleepiness score used Patient /Clinician acceptability - not reported Patient satisfaction -not reported Clinician Satisfaction - not reported Cost of telemedicine - not reported	Study interventions conducted by CPAP therapists /research staff. Usual care group -1-week telephone follow-up then 1-month face-to-face review. Helpline number in the event of problems. CPAP compliance data downloaded manually at 1 month Telemonitored group had CPAP compliance/ efficacy data nightly monitored for 2 months via secure website. Participant interactions were guided by defined clinical pathways	Usual face-to-face care
Taylor *et al 2006* (203)	Walter Reed Army Medical Centre, Washington DC, USA. Sleep medicine practitioners based at the Sleep disorders centre	Patients diagnosed with OSAHS prescribed CPAP therapy Demographics 71% male, mean age 45, 56 received intervention, 58 controls	Sleep medicine practitioners delivered initial face-to-face CPAP introductory session before randomisation	2 out of 5 outcomes of interest were addressed Adherence – measured in average hours of CPAP use Control of Symptoms - Not reported Patient/Clinician acceptability - not reported Patient Satisfaction -reported Clinician satisfaction - not reported Cost of Telemedicine- not reported	Sleep medicine practitioners delivered face-to-face care and reviewed the data daily Usual care group, one-month review and subsequent review as needed. Telephone support/walk-in clinic visits provided for both groups Intervention group received prescheduled CPAP-related questions they entered daily in a home computer-based telehealth system for 30 days. Guidelines used to advise on appropriate action where necessary	Control was traditional face-to-face care

Table 6 Aspects of care addressed

4.3.2 Description of the study characteristics

Four of the included studies were RCTs; (201-203), the other was a CCT (204). The follow-up period in the studies ranged from one to six months. Two of the studies reported the proportion of reviews achieved. The outcome measures included CPAP adherence in all five studies, (200-202, 204), and ESS in two studies (200, 202). Patient satisfaction and acceptability were reported in all five studies (200-204), but clinician satisfaction and acceptability were not reported in any of the studies. The costs of providing the telemonitoring / teleconsultation intervention were reported in two studies (201, 204). The setting for the studies varied with the main sleep Centres either being hospital or university-based and the telehealth interventions being delivered remotely in people's homes (200-202), and in one study via teleconsultation at a district general hospital, 80 km's from the main research Centre (204). The characteristics of included studies are described in Table 5 and the aspects of care addressed are described in Table 6.

4.3.3 Methodological quality

The results of the methodological quality assessment are detailed in Table 7. One study was assessed as a moderate risk of bias (203), and four were at a moderate-to-high risk of bias (200-202, 204). The commonest sources of bias were lack of information regarding sequence generation, allocation concealment, completeness of reporting, no published study protocol, and the inability to blind the study personnel to the study intervention. The risk of bias in the included studies is represented in Table 8.

Study year ROB	Design	Adequate sequence generation	Allocation concealment	Blinding of participants / clinicians	Blinding of outcome assessment	Incomplete outcome data addressed	Free of selective reporting	Free of other bias	Notes /limitations
Comma-Del - Corral *et al* 2013 {Coma-Del-Corral MJ, 2013 #290} High ROB	CCT	Not reported	Not reported	No (not possible)	Yes - analysis performed blinded and non-consecutively by same observer	Yes - loss of data transmission in 1 patient technical problems in transmission of data in 1 patient problems in 1 of 20 teleconsultations	Unclear (no published protocol, or statement about deviations from protocol)	Yes	Small sample size - underpowered Included participants with Apnoea hypopnoea index > 10 but no further details given on severity of OSAHS
Fox *et al* 2012 {Fox N, 2012 #617} High ROB	RCT	Yes - patients randomised to standard care or telemedicine (1:1 ratio) using sequentially numbered envelopes	Unclear, states envelopes were sequentially numbered, but not that they were identical/ or opaque	No (not possible)	No (described as a 'non-blinded RCT)	Not reported	Unclear (no published protocol, or statement about deviations from protocol)	Yes	Single centre study in only patients with moderate to severe OSAHS included so results relate to this group only Low adherence rate in usual care group No sham telemonitoring device used as control
Smith *et al* 2006 (201) High ROB	RCT	Not reported	Not reported	No	Not reported	Yes	Yes	No	Small sample size - underpowered Participants had been non-adherent to CPAP across 3 months following diagnosis before enrolment Severity of OSAHS not reported
Stepnowsky *et al* 2007 (200) High ROB	RCT	Yes - the randomisation scheme was generated by the project statistician and carried out by research staff	Yes- randomisation scheme was concealed until the time in which the interventions assigned.	No (not possible)	Not reported	Yes (attrition from CONSORT diagram was 1/21 in control v 4/24 in intervention group	Unclear (no published protocol, or statement about deviations from protocol)	Yes	No published protocol Included only those with moderate to severe OSAHS 100% accuracy of wireless data transmission reported in comparison to manual download

Taylor *et al* 2006 (203) Mod ROB	RCT	Yes- randomisation to either telemedicine or traditional group was balanced within each stratum	Yes- randomisation to either telehealth or traditional group was balanced within each stratum through the use of a software-generated blocked randomisation schedule	Yes. this (complicated) description of blinding appears to be adequately concealed with randomised selection of blocks	Not reported	Yes – attrition was 9 in the control group and 9 in the intervention group	Unclear (no published protocol, or statement about deviations from protocol)	No	Telephonic communication delays reported, and delay in delivery of equipment may have resulted in failure to demonstrate benefit of telemedicine ? 30 days follow up not adequate time to measure adherence to treatment 7 in intervention group withdrawn because they did not activate the Health Buddy, those left will have been more compliant which may have introduced bias Severity of OSAHS reported
CCT- controlled clinical trial, RCT- randomised controlled trial.									

Table 7 Methodological quality of included Studies

	Adequate sequence generation (selection bias)	Allocation concealment (selection bias)	Blinding of participants/clinicians (performance bias)	Blinding of outcome assessment (performance bias)	Incomplete outcome data addressed (attrition bias)	Free of selective reporting (reporting bias)	Free of other bias
Comma de Corral 2013							
Fox et al 2012							
Smith et al 2006							
Stepnowsky et al 2007							
Taylor 2006							

Table 8 Risk of bias

Key: High ROB  Moderate ROB  Low ROB 

The main areas of bias were lack of information regarding sequence generation, allocation concealment, and completeness of reporting, no published study protocol, and the inability to blind study personnel to the study intervention.

4.3.4 Description of the interventions

The characteristics of the included studies are described in detail in Table 5. All of the interventions were conducted by physicians, trained nurses, respiratory clinicians, researchers, or CPAP therapists. All of the participants in the five included studies had received a face-to-face consultation by a respiratory clinician to initiate the introduction of CPAP therapy before entry to the study.

4.3.5 Telemonitoring of CPAP usage data

Real-time telemonitoring (via a modem) of CPAP adherence and efficacy was included in three studies (200, 202, 204). Fox et al also transmitted the CPAP adherence data via an attached modem (EncoreAnywhere®, Philips Respironics Inc.), (202). Stepnowsky et al used CPAP flow generators that were fitted with a ResTraxx wireless transmitter (ResMed Corp, Poway, CA), (200). Two studies used a home web-connected telemonitoring system (participant activated), with telephone support (201, 203). In all the included studies, clinical personnel could access and monitor participants and intervene were indicated using predefined pathways that could assist with problems where encountered. Stepnowsky et al used a clinician intervention management matrix in the telemonitored group that could assist with identified problems associated with the mask fit, leak, CPAP pressure issues, and other encountered side effects (200). Fox et al used daily telemetric monitoring of the participants and data were transmitted daily via modem for review by the research co-coordinator regarding adherence, pressure delivery, mask leak, and residual respiratory events. Participants were contacted after two days by the research coordinator to enquire regarding progress and adherence, and to troubleshoot problems with CPAP therapy. The research coordinator reviewed the data every working day and made contact with the participant if problems were identified: a detected mask leak > 40 Litre/min for > than 30% of the night, < 4 hr of CPAP use for two consecutive nights, observed AHI > 10 events/hr, and the 90th percentile of pressure > 16 cm water pressure (202).

4.3.6 Teleconsultation for remote review

Additional telephone support by the clinical research staff was used to recommend appropriate action where necessary in four studies (200, 201, 203). Coma-del- Corral et al used a video conference system at a remote site hospital, 80 km distant to the central base of the sleep research unit to provide the teleconsultation intervention (204). Smith et al used a home-based telemedicine system with a built-in modem and two-way camera to allow the study nurses to deliver a structured 12 week CPAP education / support program (201). Both groups were commenced on CPAP therapy but only one group received coaching in importance of CPAP adherence and monitoring of problems encountered on treatment.

4.3.7 Effectiveness of interventions

The main findings and interpretation of the studies are summarised in Table 9. Below is a description of the findings of each included study, and section 4.3.8 - and 4.3.12 presents the findings according to the primary and secondary outcomes of interest.

Comma-del-Corral found some evidence that teleconsultation combined with telemonitoring from the main sleep medicine Centres to a distant site was feasible and could potentially provide specialised sleep medicine services to a geographically distant population. There was 85% adherence to CPAP therapy for greater than four hours per night in the usual care group compared to 75% in the telemonitored group. Participants in both groups were satisfied with the intervention they received; however, clinician satisfaction and costs of the telemedicine intervention were not reported. This model of sleep medicine review may reduce the service user's travel / expenses and improve efficacy and optimisation of specialist services with better use of community-based hospital beds and be more environmentally friendly (204).

Fox et al found some evidence that adherence to CPAP is improved with the use of telemonitoring / teleconsultation at the time of the initiation of CPAP

therapy. The adherence / compliance time was defined as minutes per day used in this study however there was significantly greater adherence in the telemedicine arm (3.2 hours / day), versus the usual care group (1.8 hours per day), hours over 3 months (p = 0.006). There were no significant between-group differences in the ESS or participant satisfaction and cost implications were not reported (202).

Smith found some evidence that a structured telehealth / teleconsultation intervention delivered over 12 weeks had the potential to improve CPAP adherence, facilitate problem-solving strategies via direct visual camera observation, and overcome barriers to adherence. There was a higher percentage adhering to CPAP therapy in the telemedicine group compared to the usual care group: 90% (n = 9 out of 10), compared to 44% (n = 4 out of 9), (p = 0.03). Participant satisfaction in the telemedicine group was 100% and 75% in the usual care group. The total costs of the telemedicine intervention per patient were reported as $420 versus $1,500 in the usual care group providing some evidence that a telehealth / teleconsultation model may be more cost-effective than face-to-face care (201).

Stepnowsky et al found some evidence that telemonitoring of CPAP adherence and efficacy with rapid clinician support to guide collaborative management was as effective as usual care in improving adherence and patient satisfaction. The telemedicine group adhered to CPAP on 78% of possible nights compared to 60% in the usual care group (p = 0.07), however, adherence was not defined by hours of night-time use in this study. No between-group differences were reported in the ESS and participant satisfaction was rated highly in the telemonitored group and the likelihood of continuing CPAP treatment was higher than the usual care group (4.8 vs 4.3: p = 0.05). The telemedicine costs were not reported (200).

Taylor et al found no differences in adherence to CPAP therapy, functional status, and client satisfaction in the telemedicine versus usual care group. Adherence was reported telemetrically via the Health Buddy computer system

and via an automated data recorder at the end of the study. Adherence was defined as CPAP use ≥ 4 hours over 30 days with the telemedicine group reporting 4.29 hours versus 4.22 hours in the usual care group ($p = 0.87$). There were no between-group differences in participant satisfaction with the telemedicine intervention (69%) versus usual care (71%). The costs of the telemedicine intervention were not reported (203).

	Adherence/Compliance	Symptom control of OSAHS	Satisfaction/acceptability	Costs implications	Interpretation
Coma-del-Corral 2013 (204)	Monitored telematically in both groups Reported as use of CPAP therapy > 4 hours per night For 5 days per week. 85% adherence in UC compared to 75% in TM arm	Not reported	Measured using visual analogue scale (1-10), reported as 9.5 for both groups Clinician satisfaction not reported	Telemedicine costs not reported	Some evidence that teleconsultation and telemonitoring from main sleep medicine centres to remote sites feasible and can potentially provide specialised sleep medicine services to a geographically distant population May also reduce service user's travel / expenses and improve efficacy and optimisation of specialist services with better use of community-based hospital beds
Fox et al 2012 (202)	Adherence reported telemetrically via attached modem in the telemonitoring group Not defined as ≥ 4 hours use Significantly greater in telemedicine arm (321mins per day) v standard arm (207 mins per day) UC arm 1.8 hrs. v 3. 2 hours over 3 months- (p= 0.006)	Epworth sleep score measured in all participants at baseline and post-intervention- No statistically significant differences reported in Epworth sleep score. (TH 5.1 vs UC 5.2)	No significant differences between intervention and UC measured using a visual analogue scale (0 - 10) larger number representing more favorable response regarding subjective sleep quality, 5.13 in intervention and 6.4 in controls	Not reported	Some evidence that adherence to CPAP is improved with use of telemonitoring / teleconsultation at initiation or therapy
Smith et al 2006 (201)	Measured by CPAP unit timer recorder. Reported as ≥ 4 hour's use of nightly CPAP / 9 out of 14 days Higher percentage adhering to CPAP therapy in TM compared to the UC group. 90% (n =9 out of 10) compared to 44%. (n= 4 out of 9) (p = 0.03)	Not reported.	Patient satisfaction measured by evaluation survey 100% of surveys in TM and 75% in UC group rated the telehealth sessions positively (n =17)	Total costs of the TM intervention per patient reported as $420 v $1,500 usual care	Some evidence that structured telehealth / teleconsultation over 12 weeks has the potential to improve CPAP adherence, facilitate problem-solving strategies via visual camera observation and overcome barriers to adherence Some evidence that telehealth/teleconsultation model is more cost-effective than face-to-face care

Stepnowsky et al 2007 (200)	Adherence reported via telemetry to a secure website – but not defined by ≥ 4 hours of use per night TM arm used CPAP on 78% of possible nights compared to 60% in UC arm (p = 0.07)	Epworth sleep scale measured in all participants at baseline and post-intervention Epworth sleepiness scale - no significant between-group differences TH 9.2 (SD 6.6) vs UC 9.9 (SD 5.2) p=0.72 Other measures were CPAP therapy efficacy, health-related quality of life, and psychological factors at baseline and post-intervention	Patient and clinician satisfaction assessed using questionnaires that included both Likert – type items and open-ended questions Participants asked to rate satisfaction with care using scale (1 = poor; 5 = excellent) TM arm rated satisfaction highly and likelihood of continuing CPAP treatment higher than UC group (4.8 vs 4.3: p = 0.05) Both groups reported not being concerned with being telemonitored	Not reported	Some evidence that telemonitoring of CPAP adherence and efficacy with rapid clinician support to guide collaborative management as effective as usual care in improving adherence and patient satisfaction
Taylor et al 2006 (203)	Adherence reported telemetrically via Health Buddy system and via automated data recorder Defined as CPAP use ≥ 4 hours over 30 days UC arm 4.22 hours v 4.29 in TM arm (p=0.87)	Modified Functional Outcome of Sleep Quality (reported - no differences between TM and UC group	Client satisfaction questionnaires were utilised. No difference between UC and TM groups 69% in intervention and 71% in control	Not reported	Evidence that no differences in adherence to CPAP therapy, functional status, and client satisfaction observed in TM v UC group
TM-telemedicine, UC- usual care, v-versus, $-dollar, n-number, SD- standard deviation.					

Table 9 Main findings and interpretation of the studies

4.3.8 Proportion reviewed

Two studies reported on the number and / or duration of participants reviewed. Fox et al found that on average an additional hour over the three-month trial was spent with the telehealth patients compared with the usual care group (202). In contrast, the teleconsultation / telemonitoring patients in Taylor et al's study in new users of CPAP had one 'walk-in visit' per week compared to the usual care group who had three per week (203).

4.3.9 Adherence to CPAP

The Taylor et al study (at moderate risk of bias) recruited patients with a range of OSAHS severity. Adherence to CPAP with teleconsultation / telemonitoring was similar to usual care TH 4.29 hours per night v UC 4.22 hours per night (p=0.89).The proportion of nights used in the TH intervention was 47% v 50% in UC group (p=0.61) (203). Three studies of a lower quality reported improved CPAP adherence after the teleconsultation / telemonitoring intervention, although two were relatively small pilot studies. Fox et al reported adherence to CPAP were greater in the telehealth group: mean hours per night: with telehealth 3.2 versus usual care: 1.8 (p=0.006), (202). Smith reported telehealth (CPAP support) improved the proportion achieving 80% use. Telehealth CPAP support: 90% vs telehealth wellbeing : 44% (p=0.033), (201). Stepnowsky et al reported a non-significant trend to greater adherence in the telehealth group. Hours of CPAP use per night : 4.1hrs (SD 1.8) versus usual care: 2.8hrs (SD 2.2) (p=0.07), (204). Coma del Corral reported adherence as 75% in the teleconsultation / telemonitoring intervention versus 85% in those who received usual care (200)(207). Table 9 reports the adherence data.

4.3.10 Control of symptoms

Two studies (both at moderate to high risk of bias) used the ESS to record symptom control (200, 202). Fox et al reported the ESS - no significant between-group differences with telehealth (5.1hrs) versus usual care (5.2hrs). Stepnowsky et al reported the ESS as no significant between-group differences with telehealth 9.2hrs (SD 6.6), versus usual care 9.9hrs (SD 5.2), (p=0.72). One study also used the Functional Outcome of Sleep Questionnaire (FOSQ) and found no statistically significant differences between the intervention and usual care group for any of the component measures of this tool (203). Two of the included studies did not measure symptom control (201, 204). Table 9 - page 63.

4.3.11 Patient / clinician acceptability / satisfaction

All five studies reported on participant satisfaction with teleconsultation / telemonitoring versus face-to-face care. Four studies using visual analogue scales (200, 202-204), or questionnaire ratings (201, 203), found no difference in satisfaction between teleconsultation / telemonitoring and usual care. One study (at moderate / high risk of bias) reported on patient acceptability and satisfaction of telemonitoring compared to the control group (201). The authors undertook a thematic analysis of the free-text comments on their satisfaction survey; 100% of the surveys in teleconsultation / telemonitoring and 75% in usual care group rated the telehealth sessions positively (no statistical analysis reported); a key theme was that teleconsultation / telemonitoring helped reinforce the importance of CPAP adherence and participants felt more supported with this intervention. None of the studies reported on acceptability to clinicians. Table 9.

4.3.12 Costs of telehealth intervention

Smith et al reported that the cost of delivering 14 teleconsultations / telemonitoring sessions for a single participant was United states $420 compared to $1500 for face-to-face visits (201). Coma del Corral et al reported

on the overall teleconsultation / telemonitoring costs (which included diagnostic testing) but did not report the direct costs associated with delivering the CPAP telehealth review component of the study (204). Table 9.

4.4 Discussion

4.4.1 Summary of findings

The evidence base for the clinical effectiveness of the combined interventions of remote consultations plus telemonitoring in the clinical review of adults who are using CPAP therapy was limited to five studies. The findings of two small studies (201, 204), and three larger studies (200, 202, 203), four of which were assessed as being at moderate to high risk of bias (200-202, 204), and one at moderate risk of bias (203), are reported. Collectively these studies do not provide definitive evidence of the effectiveness (in terms of adherence and symptom control), of teleconsultation combined with telemonitoring in CPAP users; however, there is no suggestion of any adverse harm to people who are using CPAP therapy from the telemedicine / teleconsultation interventions. The free-text comments from a single study suggest that teleconsultation combined with telemonitoring was well received and perceived as being supportive (201).

4.4.2 Strengths and limitations

In conducting this systematic review, I searched a wide range of databases and kept the search strategies broad, but there is the risk that I may have missed some potentially relevant studies. I had access to limited resources in the conduct of the systematic review and this meant I was unable to arrange a duplicate independent screening at all stages of the selection process of excluded studies, but I undertook duplicate screening of 25% of the excluded abstracts with my co-authors and after discussion achieved 100% agreement.

A key challenge for the systematic review was my definition of telehealth. Published studies on the use of telehealth in the clinical review of people with OSAHS who are using CPAP therapy utilise a wide variety of terms to describe telehealth interventions. I defined the use of telehealth for the purposes of the

systematic review as an intervention that included both remote consultation either by telephone or videoconferencing, and also the capability for real-time telemonitoring of CPAP adherence using a web-based platform to transmit CPAP adherence data to the main sleep medicine Centre clinical research teams. This meant that I rejected interventions (for example) that used remote data on CPAP usage but expected the patient to travel to follow-up face-to-face consultations. However, I recognise that by including the dual requirements of the provision for real-time telemonitoring and remote consultations in combination in my review that I have excluded a number of important studies that have utilised other aspects of telehealth in their study. I regarded both telemonitoring and teleconsultation as important components in maximising the benefits of telehealth solutions in the delivery of future OSAHS healthcare services. Nevertheless, I recognise that the studies that I rejected may have some significant lessons of relevance to this review. I have described the characteristics of the twelve excluded studies in Table 5.

The main reasons for excluding these studies were: Abreu et al only published an abstract of their telemonitoring study that did not include a teleconsultation intervention (193). Dellaca et al did not include teleconsultation or a control group (194). De-Molles et al's study did not use daily telemonitoring or teleconsultation (195). Farre was a journal editorial (205). Fraysse et al conducted a feasibility study that included telemonitoring but there was no comparator and there are no published results to date (197). Isetta et al reported two studies and the first one measured satisfaction with teleconsultation but there was no comparator (86), their second study used teleconsultation for CPAP initiation but not telemonitoring (86). The same authors published a study in 2015 which did not utilise daily telemonitoring of CPAP adherence (87).

In summary, six of the excluded studies did not have the capacity to enable real-time telemonitoring of CPAP adherence (71, 86, 87, 184, 186, 197), although two retrieved these data at the end of the study (86, 87, 197), using

a data card to download this information to include in healthcare records. Not having access to real-time telemonitoring CPAP adherence and efficacy data may have reduced the potential for the clinicians to detect real-time problems in remotely monitored participants such as poor adherence, poor mask fit / leak, and elevated AHI that could have facilitated early clinician intervention to resolve any detected problems with the continuous remote monitoring. The remote consultations in the excluded studies were generally well-received, (86, 87, 184) and one trial found the teleconsultations to be cost-effective (travel costs and lost work time were the most important sources of savings), (87).

Adherence remains a major and important clinical issue for users of CPAP therapy. Echoing the findings of this systematic review, three of the excluded RCTs that used a teleconsultation approach showed no benefit in terms of adherence, (71, 87, 184), daytime sleepiness, or quality of life (87). In contrast, the excluded study by Sparrow et al tested a telephone-delivered, theoretically based, motivational intervention and showed a significant improvement in adherence, symptoms, and functional status (186).

The reasons for non-adherence to CPAP therapy are extremely complex and in an overview of the many factors that influence adherence to CPAP therapy, Shapiro et al cite features of the actual CPAP device; treatment side effects; individual patient factors (such as clinical condition / co-existing comorbidity, family / home support context, socio-economic status, personality and cognitive functioning), the attitude and communication skills of healthcare professionals; availability and efficiency of healthcare services; and the national policy and the funding of sleep medicine services (93).

An important factor in the long-term adherence of CPAP therapy users is that they need to demonstrate to driver and vehicle licensing agencies that they meet the driving standards of these agencies and this may have a significant and important impact on adherence (52). Sleep medicine clinicians need to regularly monitor adherence, provide the facility to educate and coach their

patients on the benefits of long-term CPAP adherence, and reinforce this education at every clinical review. Access to real-time telemetric CPAP adherence usage data is a tool to inform and facilitate clinical review discussions regarding long-term CPAP treatment benefits as well as meeting the requirements of local and national driving and vehicle licensing agency's regulations.

An overview of the collective findings of these five studies suggests that a combined remote monitoring and teleconsultation model of clinical review for people with OSAHS who are using CPAP therapy is worth pursuing. Higher quality, larger trials with a low risk of bias in their design are needed. They need to explore the effect of this intervention on adherence, symptom control, service user and clinician satisfaction, socio-economic and environmental cost / benefits on an individual and population basis in terms of reduced travel, reduced loss of work-based productivity, reduced fossil fuel consumption, and reduced carbon emissions for service users and clinicians.

4.5 Interpretation in the light of published literature

Telehealthcare-enabled solutions in the delivery of clinical services are a rapidly developing field in sleep medicine and the AASM have recently published their systematic review, meta-analysis, and grade assessment of treatment of Adult Obstructive Sleep Apnoea with Positive Airway Pressure 2019. They reported a clinically significant improvement in CPAP adherence in adults where telemonitoring was utilised compared to usual care with a high quality of evidence for adherence (3). Since I undertook the initial searches at the outset of this systematic review, several further relevant studies have been published that would have met the inclusion criteria. These studies are discussed below.

The first is a three month prospective parallel group pilot RCT by Fields et al (85), which randomised 60 participants to telehealth (real-time telemonitoring and teleconsultation with telephone review at one week, one month, and three months after CPAP initiation) or usual (face-to-face) care. They found no statistical between-group differences in CPAP adherence, patient satisfaction, functional outcomes, and dropout rates, although those randomised to telemedicine showed greater improvements in mental health scores. The participants formal and verbal feedback with all parts of the study was overwhelmingly positive. Fields et al's results reinforce the findings of this systematic review, with their conclusions suggesting that larger-scale trials are warranted to establish the benefits (or not) of a combined telemonitoring and teleconsultation approach for the review for CPAP therapy users.

The second study published in 2017 that would have met the systematic review inclusion criteria is a one month non-blinded prospective RCT by Munafo et al that included 122 CPAP therapy users that compared a telehealth web-based coaching program that also included real-time telemonitoring and telephone support from clinicians triggered by responses to automated texts / e-mails compared to usual care in people newly diagnosed with OSAHS (88). Again, reflecting the findings of my systematic review, there were no statistical between-group differences in adherence to CPAP therapy or change in ESS with the combined telehealth intervention versus usual care. However, the telehealth intervention did significantly reduce the time spent providing education and coaching making it a more time-efficient clinical service delivery option which is noteworthy and of added value to services.

The third study published in 2018, that would have met the systematic review inclusion criteria is a three month RCT by Hwang et al 2018 (187). They studied the effect of telemedicine education and telemonitoring on CPAP adherence. This four-arm study recruited 556 participants who were prescribed CPAP to treat OSAHS. Two telemedicine interventions were implemented: 1) a web-based OSAHS education intervention, 2) CPAP

telemonitoring initiated with an automated patient feedback intervention. Patients were randomised to receive 1) usual care, 2) usual care plus tele-education 3) usual care plus telemonitoring and 4) usual care plus tele-education and telemonitoring. The primary endpoint in this study was 90 day CPAP adherence rates with secondary endpoints being CPAP clinic attendance rates and change in ESS. This study concluded that the combination of the use of CPAP telemonitoring with automated feedback messaging improved the 90 day adherence rates in CPAP therapy users. The telemedicine-based education intervention did not significantly improve CPAP adherence but did increase the clinic attendance rates for service users.

The fourth study published by Schoch et al in 2019 was a six month large randomised, controlled phase III trial that collected data on the use of telemedicine in the CPAP treatment adherence of 240 patients with OSAHS over a six-month follow-up period. Participants were randomised to a telemedicine or control arm with the telemedicine arm also receiving teleconsultation via phone calls during the first month of treatment where poor adherence or excessive mask leakage was identified via the remote CPAP telemonitoring system. The primary outcome of interest was adherence to CPAP therapy at six months with a secondary outcome being the measurement of sleep-related quality of life. The study established that the telemonitoring triggered intervention in the first month of CPAP use did not increase adherence in the study population, however, it had a beneficial effect in those with milder OSAS and improved in sleep-related quality of life (206).

The fifth study by Lugo et al 2019 was an open three-month follow-up RCT in 186 patients that compared routine review with a virtual sleep unit review in review of CPAP users. They aimed to compare any clinical improvement and cost-effectiveness in a non-inferiority analysis. The virtual sleep unit review included teleconsultation and telemonitoring. The virtual sleep unit offered a more cost-effective option of improving quality-adjusted life years (QALY's), than a hospital review and also in terms of overall cost-effectiveness indicate

that a virtual sleep unit could assist with the management of CPAP therapy users and adherence was similar in both groups at > five hours per night (207).

Taking into consideration the above studies published since I undertook the systematic review that collectively included 1164 participants (85, 88, 187, 206, 207), all of these studies would have met the inclusion criteria for the systematic review. Four of these studies showed no differences in the recorded CPAP adherence rates between those randomised to telemedicine / teleconsultation versus usual care (85, 88, 206, 207), which mirrors the findings of my review where the included studies showed no benefit of teleconsultation combined with telemonitoring versus usual care but with no observed safety concerns. Only one of these studies showed an increased 90 day CPAP adherence rate in the participants and this study included CPAP telemonitoring initiated with an automated patient feedback messaging intervention which may have added additional support and encouragement to new CPAP therapy users (187). Regarding other secondary outcomes of interest, only one study reported a statistically significant increase in reported quality of life at one month in the telemonitoring / teleconsultation group (206), however, this was not found to be significant at six months follow up. One study reported that virtual sleep unit review compared to hospital review was more cost-effective in both in terms of QALY's and overall service delivery (207).

The follow up period in all the studies included in the systematic review ranged from one to six months and the five subsequent studies that have been published since my systematic review was conducted also had a follow up period of one to six months. This may not be a long enough period of time in order to address the outcomes of interest such as longer term adherence rates to CPAP therapy and the effects of telemonitoring and teleconsultation and patient and clinician satisfaction with this model of CPAP review. A longer follow up period such as 12 - 24 months in future studies may allow more confidence in the findings of the outcomes of interest in my systematic review. However, expecting patients to participate in an RCT for this length of time

may be problematic due to drop out rates with patients being unwilling to participate in extended RCT's rates. There is also the issue of obtaining funding for conducting lengthy RCT's to consider. Perhaps prospective observation studies over a longer period of time may be an option to conduct future research on these outcomes and add to the evidence base and the option of accessing longer term telemonitoring data may assist in this process.

When combining the results of these recently published studies with my systematic review findings published in 2017 it would appear that there is a consistent message emerging. Combined telemonitoring and teleconsultation results in similar adherence to CPAP and clinical outcomes to traditionally delivered face-to-face care but is generally acceptable to patients and may offer an organisational advantage to the health service.

4.6 Implications for research and future practice

The technology to implement this combined model of sleep medicine service delivery has been available for more than a decade and is evolving at pace globally (particularly in relation to the sleep medicine specialties response to the global pandemic). It is becoming widely available and being utilised in many healthcare settings and driving more research in this area. Sarmiento et al 2019 are leading the field in the national expansion of sleep telemedicine for veterans in the USA (208), and many more services are now adopting this technology.

Based on the results of the five trials included in the systematic review the evidence for remote teleconsultation with daily telemonitoring of CPAP usage in OSAHS was far from clear at the time of conducting the systematic review and I had originally maintained that well-designed, adequately powered studies were required to clarify the role of teleconsultation combined with real-time telemonitoring in the routine clinical review of people using CPAP therapy.

When considering the combined findings of the five studies that have been published since my systematic review was conducted (85, 88, 187, 206, 207), and reflecting on the free-text comments in one of the studies, telemonitoring was perceived as reinforcing the importance of adherence, whilst remote consulting has the potential to facilitate convenient reviews especially for those living at a greater distance from sleep Centres. With the emergence of new technologies that enable remote teleconsultation, and real-time telemonitoring capability in-built into newer CPAP units, there is an opportunity for trials to build on the current evidence base and inform future practice (189). A recent meta-analysis that included 18 studies and investigated the effectiveness of a range of telehealth interventions in improving CPAP adherence concluded that a broad range of electronic telehealth interventions in adults with OSAHS can improve CPAP adherence early in treatment initiation, increasing the night time adherence by 30 minutes however, there is still a gap in the literature regarding

the duration, timing, intensity, and methods of telehealth delivery that are most effective using technology-enabled healthcare (74).

The impact of the arrival of the SARS-CoV-2 pandemic in early 2020 has had a huge impact on the delivery of sleep medicine services globally, with many services being completely suspended as respiratory clinicians were redeployed to the front line to assist acute services during this pandemic. Services were just beginning to explore ways to remobilise as the second wave of the pandemic appeared, and the use of innovative teleconsultation and telemonitoring solutions in the diagnosis and the review of CPAP therapy users is at the forefront of all current services.

These technologies are not new and despite this, there has been a slow uptake in some services before the pandemic. It is now absolutely necessary to continue to develop and utilise these technologies moving forward beyond the pandemic as services have adapted rapidly to deliver as many clinical review options using non face-to-face telehealth solutions. Incorporating telehealth into the diagnosis, treatment, and review pathways for CPAP therapy users need to be delivered at the same standard level for face-to-face reviews using guideline-based care. The current COVID 19 pandemic although a global disaster with catastrophic loss of lives continues, however does force sleep medicine services to redesign service pathways using already proven telehealth solutions that can help transform health care and people's lives on a global scale.

The positive environmental impacts of delivering more remote consultations using telehealth solutions are already an important factor in healthcare delivery. It reduces the time taken away from the place of employment, also reducing the travel time from the place of work to attend a centrally based sleep medicine service, and the associated subsequent reduced fossil fuel consumption, and is more environmentally friendly than traditional face-to-face reviews. It reduces the environmental impact of atmospheric pollutants emitted

by vehicles by reducing the number of journeys made for face-to-face visits and thus contributes to environmental sustainability. Globally there is an ever-increasing drive to reduce greenhouse gas emissions with Scotland having an ambitious target of an 80% reduction in emissions by 2050. If these targets are to be realised, then sleep medicine providers nationally and internationally need to develop their telemedicine / teleconsultation services at pace to keep up with the growing body of research in this technology-enabled care driven environment. A widespread paradigm shift in the sleep medicine community is needed where telehealth solutions are viewed as an essential component of health care delivery and not just considered for those who lack access to care due to geography, or other constraints. The pandemic may be the catalyst for this paradigm shift.

4.7 Conclusions

The combination of a remote consulting service and continuous real-time telemonitoring of CPAP therapy use has the potential to offer efficient, acceptable, and convenient care that has similar outcomes in terms of promoting adherence to CPAP and symptom control. Recent trials corroborate these conclusions. Future research needs to include other secondary outcome measures beyond adherence, treatment efficacy, symptom control, and patient and clinician satisfaction. They need to include the impact of teleconsultation and telemonitoring on travel reduction, reduction in lost time in terms of work productivity, reduced fuel consumption, and carbon emissions. In the light of the increasing prevalence of OSAHS globally, specialist sleep medicine service clinicians and providers should consider investing in telehealth services as a strategy to manage the rising demand within the limited capacity of this specialist area of respiratory medicine. The global pandemic has added additional urgency to this agenda.

4.8 Summary of Chapter 4

This chapter reports the findings of a systematic review of the literature published in 2015 and also summarises a number of trials published shortly after the completion of my review. It is clear that, although some unanswered questions remain, teleconsultations / telemonitoring now has an established place in the routine management of people using CPAP for OSAHS.

My initial plan was to conduct a pilot RCT comparing telemonitoring and teleconsultation with usual face-to-face care however in combining the findings of my review and the subsequent publication of several large trials I concluded that this was no longer required. During my VIVA I reflected on the five studies included in my Systematic review and their collective relatively small sample size and their moderate to high risk of bias. My current thinking around this has now changed and I would no longer make this assertion. I will consider updating my systematic review and include the five studies that have been published since then and any more recent studies published since submission of this thesis. I would define usual care as a routine face-to-face consultation usually in a hospital clinic or outreach clinic that requires the clinician and the individual to attend in person for a review and include both teleconsultation and real time telemonitoring as the comparator in an updated systematic review.

I turned my attention from the mode of delivery to the content of a clinical review of CPAP therapy users and how often such a review should take place. National and international guidelines recommend regular reviews but do not provide clarity on what components should be included in a clinical review and the frequency.

In the next chapter, I will describe an e-Delphi study that established an international consensus on the most important components and frequency of a clinical review in CPAP therapy users (10).

Chapter 5: International e - Delphi study and design of Sleep Medicine review template

In the previous chapter, I conducted a systematic review and established that telehealth is an effective and alternative mode of conducting a clinical review with some advantages for CPAP therapy users, reducing the need for travel for face-to-face reviews, with benefits in terms of reduced time taken off work and thus being more convenient and environmentally friendly.

In this chapter, I describe an international e-Delphi in which I reached a consensus on the core components of a CPAP review which would be applicable in all routine reviews of people with OSAHS.

The e-Delphi is published:
Murphie P (PM), Little S (SL), Paton R (RP), McKinstry B (BMcK), Pinnock H (HP). Defining the Core Components of a Clinical Review of People Using Continuous Positive Airway Pressure Therapy to Treat Obstructive Sleep Apnea: An International e-Delphi Study
J Clin Sleep Med 2018;14:1679–1687

5.1 Introduction and rationale for the e-Delphi

5.1.1 OSAHS as a lifelong condition

OSAHS is usually a lifelong condition that requires long-term treatment such as CPAP therapy to relieve symptoms, and the potentially serious impact of disturbed sleep and daytime sleepiness and reduced quality of life (9, 67, 209).

5.1.2 Arrangements for CPAP review in the UK

In the UK CPAP review and frequency varies amongst Centres with some seeing people on an annual basis and some providing open access or on request review. A recent publication in the BMJ best practice recommends a time interval of 6 - 12 monthly review, however, this has not been firmly established. Vocational drivers do require an annual review to meet the UK

DVLA requirements (209)(212). Stradling in 2016 reported there were approximately 230 000 patients on CPAP in the UK (210). A recent editorial in the Lancet estimates that around 8 million people aged 30–69 years might be affected by OSAHS in the UK alone, which represents a huge burden to the NHS, and already overwhelmed sleep medicine services across the country (211). There has been a shift to nurse-led, physiologist and clinical scientist review services across the UK to free up respiratory physicians to see more complex cases, and there is evidence that this is an effective way of delivering services with similar outcomes to nurse versus consultant reviews (171, 212). An internet search has identified many services across the UK that work in an organised and multidisciplinary team with the oversight of a consultant.

5.1.3 Frequency and components of a CPAP review

At the time of developing the International e-Delphi study, the published guidelines and best practice statements recommended regular clinical review for people who are using CPAP therapy. They suggested a range of components that could be included such as subjective assessment of sleepiness and other symptoms, adherence to CPAP therapy, practical issues with mask fit, equipment issues, pressure settings, guidance on driving license advice, and weight and blood pressure monitoring. However, the guidelines gave conflicting advice, and some did not offer any advice regarding the core content of a clinical review (Table 10), and how often this should take place. I felt that this represented a gap in the evidence base regarding the core content of a clinical review and there was, therefore, a need to reach a consensus. I decided to conduct an international e-Delphi study that included the views of respiratory clinicians, specialist nurses, physiologists, physician academics, and CPAP therapy users, with the objective being that this method would allow me to achieve an international consensus.

5.2 e-Delphi methodology

Stemming from the RAND Corporation in the 1950s (213), a Delphi study delivers a series of questionnaires over (typically) three rounds in which expert panellists contribute their ideas independently, and in subsequent rounds individual responses may be influenced by feedback of the collective participant responses from previous rounds, facilitating arrival at a consensus. The e-Delphi is an electronic structured research method that can be used for reaching a common viewpoint or consensus among a panel of experts where there is limited evidence on the priority attached to a range of items (214, 215). It has been used extensively within a health and social care research setting to aid the decision-making process regarding health care practices and to assist with developing guidelines based on the consensus outcomes. Communication can take place by email, enabling participation both nationally and internationally over a short timeframe. With the use of the e-Delphi technique, experts participate in different periods, at their own pace within the set timescales of the research project (213, 215).

5.2.1 Other Consensus methodology options

5.2.1.1 Nominal group technique

The nominal group technique (also known as the expert panel method), is a moderated and structured group activity that encompasses identification of the problem, developing solutions to facilitate decision making. Groups of different numbers can participate, with the aim being to arrive at a consensus decision in a timely manner, by a group vote with all attendees opinions being heard. It requires face-to-face meetings and the method aims to prevent the domination of the discussion by a single person, encouraging all members to contribute, aiming to arrive at an agreed set of solutions or recommendations that represent the group's views or preferences (216). This method is popular and has been used to scrutinise the suitability of healthcare interventions that may influence the decision-making process regarding treating decisions for patients

(217). It has advantages in that groups can meet face-to-face and disadvantages with the main disadvantage being the travel time and economic cost involved with a face-to-face meeting.

5.2.1.2 RAND-UCLA appropriateness method (RAM)

The RAM - also developed by the RAND Corporation and the University of California - Los Angeles was developed in the mid-1980s, primarily as a method to quantify the overuse and underuse of healthcare procedures. It is an internationally recognised technique that applies a combination of expert opinion and current evidence to arrive at a consensus. The consensus between participants is then applied to inform the content of guidelines or policies under development. This method is used when the evidence base underpinning guidelines or practices may need to be refined for a particular setting or group, before being introduced or when there is incomplete evidence to support decision making. It is a modified Delphi technique that aims to precisely find out participants own experiences and views to assist with informing the relevance of different healthcare practices. Again its main disadvantages are that it requires a face-to-face meeting with the added travel and economic costs incurred and it can take a long time to reach consensus (218).

5.2.1.3 Consensus development conference method

This method is intended to assist in the development of a guideline that reflects the views of the expert panel that has examined and discussed the scientific data, with the aim being to create a multidisciplinary approach to the subject of interest. A consensus development conference can be national or international with the aim being to gather a large group of healthcare professionals and others to meet, and jointly interact to evaluate an existing technology or evidence base regarding healthcare policies or guidelines. These panels do require in-person communication to allow discussion and

deliberation and consensus of priority issues, and they can be helpful where there are conflicting opinions among participants. The method has some advantages in facilitating both the creation of the best available information and supports the reaching of consensus and validation between participants, for whom this work will have the most relevance for this consensus process (219, 220). However, the disadvantages are that the process of interactions with participants is less structured and there is also the requirement for travel and time take from work activities.

5.2.2 Why did I choose an e-Delphi

I chose to use an e-Delphi research technique for this study to be able to include an international audience of participants from different healthcare settings and disciplines who were living in different parts of the world. I considered this was the best and most practical method in which to obtain or achieve consensus, as it would have been difficult to arrange to conduct three rounds either face-to-face or via telemedicine due to the differing time zones of participants globally from different continents.

5.2.3 Aims and objectives of the e-Delphi study

a) To reach consensus amongst a panel of international sleep clinicians, academics, and CPAP users on the most important components to include in a CPAP review and how often a review should take place.

b) To develop a structured sleep medicine review template that records the important components identified by the e-Delphi consensus panel.

5.3 Methods

5.3.1 Ethics

I obtained ethical approval for the International e-Delphi study from the Usher Institute of Population Health Sciences and Informatics, University of Edinburgh (application number 1700), (Appendix 3). All participants provided electronic written consent to participate in the study. The e-Delphi study protocol is described in Appendix 6.

5.3.3 Developing the initial list of components

5.3.3.1 Guideline review

I identified current national and international guidelines for the management of people with OSAHS therapy, searching Medline, Turning Research Into Practice (TRIP), databases and Google Scholar using the following search terms '*sleep apnoea/apnoea syndrome', 'CPAP therapy', 'national / international guidelines' 'clinical review'* and *'follow up'.* I scrutinised all of these guidelines for their recommendations about the suggested content of a regular review of CPAP therapy users and then extracted all the suggested components from the guidelines to populate an initial list.

5.3.3.2 Pilot work to refine the list

I then piloted the e-Delphi process in our local sleep medicine service with ten of our clinicians who were invited to add any additional components they considered to be important.

5.3.4 Recruitment of an expert panel

Delphi panels generally have fewer than 50 participants, and the majority of Delphi studies have included between 20-30 respondents (214). I invited by e-mail, 80 international experts (including some CPAP therapy users), intending to recruit up to 30 participants to the study. I aimed to include

participants with clinical practice experience, physicians with academic expertise as well as CPAP therapy users. My recruitment strategy involved inviting healthcare professionals who were actively involved in the clinical review of people with OSAHS who were using CPAP therapy (e.g., clinical academics, respiratory physicians, general practitioners, clinical nurse specialists / nurse practitioners, respiratory therapists, and physiologists). I also invited up to five individuals with OSAHS who were using CPAP therapy from a local sleep medicine service. All the suggestions, comments, and data that were collected during the study were anonymised, however, all the participants were offered the option of being acknowledged as participants in subsequent publications.

5.4 Analysis

I calculated the collective median scores for each component of the clinical review and the proportion of respondents scoring each item as 4 or 5. The median is the middle score for a set of data that has been arranged in order of magnitude. I used the median because it is less affected by outliers and skewed data. Consensus was defined as ≥75% agreement for the priority scores of 4 or 5.

Prioritised components were grouped in the following way: treatment acceptability; technical / CPAP issues; sleepiness assessment; treatment adherence; symptom resolution; assessing sleep quality; driving issues; quality of life; lifestyle issues / sleep hygiene. These proritised components were then mapped to an initial template which could be used to facilitate a standardised clinical review.

5.4.1 Free text comments from participants

Participants were invited to contribute additional free-text comments in all rounds. I analysed the free text comments thematically to identify the key issues from the perspective of the participants (Table 13).

5.4.2 The three rounds of the e-Delphi

I followed the recommended consensus methodology (213, 215), and anticipated that it would require up to three rounds to reach consensus with a fourth-round planned if this was required. The data collection sheets for the three rounds are in Table 10.

Table 10 Round One - Three data collection sheets

	An International e-Delphi exercise to define the components of a clinical review of people using CPAP therapy	Participant Study ID 3
There are three parts to the study: (1) specification of necessary components of a CPAP review, (2) specification of timing/frequency of CPAP review, and (3) opportunity to provide free text comments. We request that you complete sections 1 and 2 as a minimum.		
Thank you for agreeing to help with this e-Delphi exercise. Listed below are the possible components of a clinical review of a patient using CPAP therapy. We invite you to add any additional components and/or comments that you think are important.		
Clinical CPAP review	**We estimate it will take 15 mins to reflect on CPAP therapy review, and complete the questionnaire.**	
Important!	**Save this spreadsheet to your desktop, or a folder on your computer. If you don't do this you will lose all your responses when you close the spreadsheet**	
Section 1. Specification of necessary components of a CPAP review. Below are some suggestions from the literature. Please add any addition components that you think are important in the blank rows below our list. We will use this to generate the list for scoring in the next round.		
Recording Epworth Sleep Score		
Verbally asking about CPAP adherence		
Recording of CPAP adherence by data download via memory card		
Recording adherence by remote monitoring		
Checking mask fit		
Chest auscultation		
Checking for treatment side effects		
Checking electrical safety of CPAP unit		
Changing CPAP filters		
Checking body weight		
Checking blood pressure		
Asking about current driving status - Car/Heavy goods Vehicle license		
Checking that any relevant Vehicle Licensing agencies are aware of the condition		
Section 2 - Specification of timing/frequency of CPAP review. This section is intended to seek opinions on how often a clinical review should take place. Please add any alternative timing/frequency of review you feel should also be considered by the panel in the process of iterating to consensus.		
Annual review		
Six monthly review		
Section 3 - Opportunity to provide free text comments that are relevant to a CPAP clinical review.		
Professional role or interest in sleep apnoea: - Please enter yes to all that apply.		
	Academic	
	Clinician	
	Guideline development	
	Role in professional society	
	Journal editor	
	Healthcare management	
	Policy maker	
	Role in sleep apnoea patient group / charity	
	Person with sleep apnoea	
On completion of your response please save to your desktop and return to me by email attachment to: s1470044@ed.ac.uk		

Table 10 – Round One - Three data collection sheets

	**Round 2 - An International e-Delphi exercise to define the components of a clinical review of people using CPAP therapy**
There are three parts to this round of the study: (1) Scoring the importance of the components of a CPAP review, (2) specification of timing/frequency of CPAP review, and (3) opportunity to provide free text comments. We request that you complete sections 1 and 2 as a minimum.	
Thank you for agreeing to help with this e-Delphi exercise. This is round 2. **Listed below are the possible components of a clinical review of a patient using CPAP therapy.** **We now invite you to score the importance of the componets listed below with 0 being unimportant and 5 being very important**	
Clinical CPAP review	**We estimate it will take up to 15 mins to complete the round 2 questionnaire.**
Important!	**Save this spreadsheet to your desktop, or a folder on your computer. If you don't do this you will lose all your responses when you close the spreadsheet**

Section 1. Consists of components of a CPAP clinic review generated from round 1 of the e-Delphi study. Please specify the importance of these with 0 = unimportant and 5 = very important.

Component of the CPAP review	Score
Recording Epworth Sleep Score	
Verbally asking about CPAP adherence/Sleep time	
Recording of CPAP adherence/efficacy by data download via memory card/remote monitoring	
Checking mask fit issues	
Chest auscultation	
Checking for treatment side effects	
Checking electrical safety of CPAP unit	
Checking body weight	
Checking blood pressure	
Asking about current driving status - Car/Heavy goods Vehicle license	
Checking that any relevant Vehicle Licensing agencies are aware of the condition	
Review of medical history, medication, any new co morbidities in realtion to symptoms/need for hospitalisation	
Checking partner feedback / quality of life	
Checking patient quality of life	
Checking patient physical activity/exercise	
Checking patient quality of sleep /feeling refreshed on waking	
Asking about quality/quantity of sleep/sleep routine times	
Asking about work schedule/Shift Pattern	
Ask if any problems with sleepiness while driving driving	
Checking if initial symptoms for referral have improved e.g. tiredness/sleepiness/hypersomnalence/ concentration/memory	
Patients preparedness to continue with treatment	
Check for control of witnessed residual snoring or apnoeas, chocking spells	
Recording the altitude where the patient lives and the altitude where the sleep study was carried out	
Cognitive/developmental issues	
Support system at home (Help with mask on/off fitting and filling of humidifier if used) /partner engagement	
Examination of the nasal passage and throat	
Ensuring CPAP is resolving Apnoea Hypopnoea index	
Advice re air travel	
Ask about factors that help/hinder CPAP use	
CPAP unit noise level	
Nocturia/Frequency of getting up to pass urine	
Requirement to repeat diagnostic study with significant weight loss/weight gain	
Enquiring re possibility of other sleep disorders/i.e. restless leg syndrome	
Fatigue and depression scale /as interfering with epworth sleep score	
Requirement for humidification	
Frequency of cleaning mask interface and circuit/changing filters	
Section 2 - Timing/frequency of CPAP review. This section is intended to gain consensus on how often a clinical review should take place. Please indicate by putting a cross in the final column your preferred timing for routine checks of a patient established on CPAP	
Within 3-30 days after initial CPAP introduction	
Telephone reviews at 1,4,8 weeks following starting of trial	
3 monthly	
4 monthly	
6 monthly	
12-18 month review	
Every 2 years	
Every 5 years	
If poor compliance/usage seen every 6 months	
Follow up via -Telemonitoring where used	
On demand/open access review/based on patient need	
Forced review - ie request from traffic agency/pre planned surgery or similar	

Section 3 -	**Additional free text comments**

On completion of your response please save to your desktop and return to me by email attachment to:
s1470044@sms.ed.ac.uk

Table 10 – Round One - Three data collection sheets

	Round 3 - An International e-Delphi exercise to define the components of a clinical review of people using CPAP therapy	**Participant ID**
There are three parts to this round of the study: (1) Scoring the importance of the components of a CPAP review, (2) specification of timing/frequency of CPAP review, and (3) opportunity to provide free text comments. We request that you complete sections 1 and 2 as a minimum.		
Thank you for agreeing to help with this e-Delphi exercise. This is round 2. **Listed below are the possible components of a clinical review of a patient using CPAP therapy.** **We now invite you to score the importance of the componets listed below with 0 being unimportant and 5 being very important**		
Clinical CPAP review	**We estimate it will take up to 15 mins to complete the round 2 questionnaire.**	
Important!	**Save this spreadsheet to your desktop, or a folder on your computer. If you don't do this you will lose all your responses when you close the spreadsheet**	
Section 1. Consists of components of a CPAP clinic review generated from round 1 of the e-Delphi study. Please specify the importance of these with 0 = unimportant and 5 = very important.		

Component of the CPAP review	Score	Median score from round 2
Recording Epworth Sleep Score		
Verbally asking about CPAP adherence/Sleep time		
Recording of CPAP adherence/efficacy by data download via memory card/remote monitoring		
Checking mask fit issues		
Chest auscultation		
Checking for treatment side effects		
Checking electrical safety of CPAP unit		
Checking body weight		
Checking blood pressure		
Asking about current driving status - Car/Heavy goods Vehicle license		
Checking that any relevant Vehicle Licensing agencies are aware of the condition		
Review of medical history, medication, any new co morbidities in realtion to symptoms/need for hospitalisation		
Checking partner feedback / quality of life		
Checking patient quality of life		
Checking patient physical activity/exercise		
Checking patient quality of sleep /feeling refreshed on waking		
Asking about quality/quantity of sleep/sleep routine times		
Asking about work schedule/Shift Pattern		
Ask if any problems with sleepiness while driving driving		
Checking if initial symptoms for referral have improved e.g. tiredness/sleepiness/hypersomnalence/ concentration/memory		
Patients preparedness to continue with treatment		
Check for control of witnessed residual snoring or apnoeas, chocking spells		
Recording the altitude where the patient lives and the altitude where the sleep study was carried out		
Cognitive/developmental issues		
Support system at home (Help with mask on/off fitting and filling of humidifier if used) /partner engagement		
Examination of the nasal passage and throat		
Ensuring CPAP is resolving Apnoea Hypopnoea index		
Advice re air travel		
Ask about factors that help/hinder CPAP use		
CPAP unit noise level		
Nocturia/Frequency of getting up to pass urine		
Requirement to repeat diagnostic study with significant weight loss/weight gain		
Enquiring re possibility of other sleep disorders/i.e. restless leg syndrome		
Fatigue and depression scale /as interfering with epworth sleep score		
Requirement for humidification		
Frequency of cleaning mask interface and circuit/changing filters		
Section 2 - Timing/frequency of CPAP review. This section is intended to gain consensus on how often a clinical review should take place. Please indicate by putting a cross in the final column your preferred timing for routine checks of a patient established on CPAP		
Within 3-30 days after initial CPAP introduction		
Telephone reviews at 1,4,8 weeks following starting of trial		
3 monthly		
4 monthly		
6 monthly		
12-18 month review		
Every 2 years		
Every 5 years		
If poor compliance/usage seen every 6 months		
Follow up via -Telemonitoring where used		
On demand/open access review/based on patient need		
Forced review - ie request from traffic agency/pre planned surgery or similar		
Section 3 -	**Additional free text comments**	
On completion of your response please save to your desktop and return to me by email attachment to: s1470044@sms.ed.ac.uk		

Round 1: Open round

Round 1 was an open round to compile the list for prioritisation of components of a clinical review in CPAP users (Table 10). I developed a data collection sheet (using an excel spreadsheet), of potential clinical review components that were identified from my literature review of published guidelines and combined this with additional suggestions from our local pilot work. I emailed the Round 1 data collection sheet with the initial list of possible components to the expert panel, requesting that they add any additional components that they felt were important to include (Table 10). They were also asked to add any additional free-text comments on the initial components. I asked for their opinions regarding the suggested frequency of a clinical review. I collated all the additional suggested components at the end of Round 1 completion to create the final list for the scoring prioritisation consensus in round 2.

Round 2: First scoring round

Round 2 was the first scoring round in the e-Delphi study and the panel of participants were asked to review the list of components that were generated from the free-text in round 1 and then prioritise these components using a scale of 1-5 (1 = unimportant and 5 = very important). The results were collated, and a median score was calculated for each component in preparation for round 3.

Round 3: Second scoring round

The Round 3 data collection sheet included the median scores for all the components from round 2 as well as the individual participants own round 1 score (Table 10). In round 3 the participants were given the opportunity to revise their scores (or not), on the priority of the clinical review components in the light of the median score findings from all participants of the previous round by again ranking each component on a score of 1 to 5 (where 1 = unimportant and 5 = very important). I predicted an acceptable level of agreement on priority components with 3 rounds however a final 4th round (following the method of round 3) could be conducted if required.

5.5 Results

Guideline review and preparation of the initial list of possible components.

The literature review identified thirteen national / international guidelines and best practice statements published between 2003-2016 that made recommendations on the long-term management and follow-up of CPAP therapy users (Table 11). From the guideline recommendations, an initial list of twelve components was compiled. No additional components were suggested by the ten local sleep medicine clinicians who participated in the pilot process.

- Weight monitoring
- BP monitoring
- Subjective measure of sleepiness
- Reported symptoms
- CPAP adherence
- Mask interphase issues
- Pressure setting / comfort
- Reported side effects
- Chest auscultation
- Technical and safety check
- Driving / DVLA guidance
- Follow up

Guideline body, date [ref]:	Weight monitoring	BP monitoring	Subjective measure of sleepiness	Reported symptoms	CPAP adherence	Mask interphase issues	Pressure setting/ comfort	Reported side effects	Chest auscultation	Technical and safety check	Driving/ DVLA guidance	Follow up
Scottish Intercollegiate Guideline Network, 2003 (70)												
National Institute for Health and Clinical Excellence 2008 updated 2015 (9)	√	√			√							√
BTS Impress Improving and Integrating Respiratory Services (8)				√	√			√		√	√	√
American Academy of Sleep Medicine 2009 (67)	√		√	√	√	√	√	√		√	√	√
Australasian Sleep Association 2009 (221)				√	√	√		√		√		√
Hellenic Society of Sleep Disorders 2009(222)			√			√	√			√		√
Spanish Pulmonology Society 2011 (7)	√			√	√							√
Canadian Thoracic Society 2011 (76)												
BMJ Best practice 2016 (223)				√	√							
American College of Physicians 2014 (24)	√		√		√							
European Respiratory Society White book 2014 (6)												√
India Institute of Medical Sciences 2015 (224)												
International Geriatric Sleep Medicine task force 2016 (78)												

Table 11 National and international guidelines

5.5.1 International expert panel

Of the 80 potential participants approached, 47 consented to participate from 21 countries (Europe n=37, Australasia n=5, Asia n=3, North America n=2). Professionals (some represented more than one group), encompassed respiratory physicians (n=29), academics (n=25), journal editors (n=9), CPAP therapy users (n=6), specialist respiratory nurses (n=5), respiratory physiologists (n=3), and respiratory therapists (n=1), (Figure 6). 44 completed round one; 41 and 40 participants completed rounds two and three, respectively.

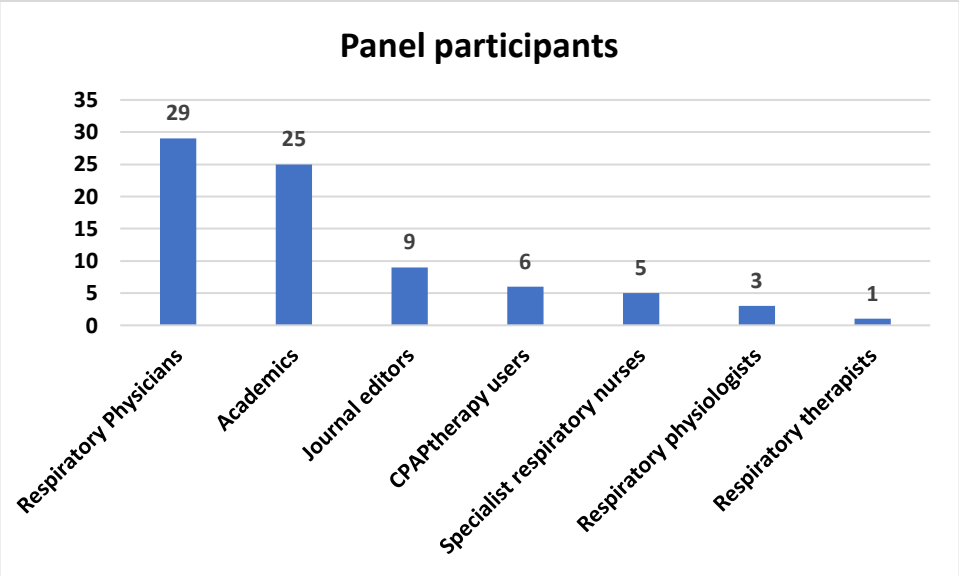


Figure 6 Panel participants

5.5.2 Final list of components for scoring

An additional 24 components to include in the CPAP review were suggested in the free text in round one and included in the list for the scoring rounds making a total of 36 components (Table 10):

- Recording Epworth sleepiness scale
- Verbally asking about CPAP adherence / sleep time
- Recording of CPAP adherence / efficacy by data download via memory card / remote monitoring

- Checking mask fit issues
- Chest auscultation
- Checking for treatment side effects
- Checking electrical safety of CPAP unit
- Checking body weight
- Checking blood pressure
- Asking about current driving status - Car / Heavy goods vehicle license
- Checking that any vehicle licensing agencies are aware of the condition
- Review of medical history, medication, any new co-morbidities in relation to symptoms / need for hospitalisation
- Checking partner feedback / quality of life
- Checking the patient quality of life
- Checking patient physical activity / exercise
- Checking patient quality of sleep / feeling refreshed on waking
- Asking about quantity / quality of sleep / sleep routine times
- Asking about sleep schedule / shift pattern
- Ask if any problems with sleepiness while driving
- Checking if initial symptoms for referral have improved e.g. tiredness / sleepiness / hypersomnolence / concentration / memory
- Patients preparedness to continue with treatment
- Check for control of witnessed residual snoring / apnoeas / choking spells
- Recording the altitude where the patient lives and the altitude where the sleep study was carried out
- Cognitive / developmental issues
- Support system at home (help with mask on / off / fitting and filling of humidifier if used) / partner engagement
- Examination of the nasal passage and throat
- Ensuring CPAP is resolving apnoea hyponoea index
- Advice re air travel
- Ask about factors that help / hinder CPAP use

- CPAP unit noise level
- Nocturia / frequency of getting up to pass urine
- Requirement to repeat a diagnostic study with significant weight loss / weight gain
- Enquiring re possibility of other sleep disorders / i.e. restless leg syndrome
- Fatigue and depression scale / as interfering with the Epworth sleep scale
- Requirement for humidification
- Frequency of cleaning mask interphase and circuit / changing filters

5.5.3 Components reaching consensus threshold

17 components achieved a priority consensus of ≥75%, indicating agreement that these components were important to include in a clinical review. Eight components reached a consensus of ≥90% suggesting these were core components. The percentage consensus for all 36 components at the end of round three, are listed in order of percentage agreement with the priority scores in (Table 12).

Components of a clinical review, listed in order of proportion of respondents who gave priority score of 4 or 5	% agreement with priority scores	Grouped into key categories
Components achieving the priority threshold for consensus (75% agreement with the priority score)		
Checking for treatment side effects	98	Acceptability
Checking mask fit issues	98	Technical
Recording subjective assessment of sleepiness / somnolence e.g. Epworth Sleepiness Scale	95	Sleepiness assessment
Recording of CPAP adherence / efficacy by data download via memory card / remote monitoring	95	Adherence check
Ask if any problems with sleepiness while driving	95	Sleepiness assessment
Checking if initial symptoms for referral have improved e.g. tiredness / sleepiness / hypersomnolence / concentration / memory	93	Symptom resolution
Checking patient quality of sleep / feeling refreshed on waking	90	Assess sleep quality
Ensuring CPAP is resolving the Apnoea Hypopnoea Index (AHI)	90	Symptom resolution
Asking about quality / quantity of sleep / sleep routine times	88	Assess sleep quality
Patients preparedness to continue with treatment	88	Acceptability
Asking about current driving status - Car / Heavy goods Vehicle license	85	Driving
Checking patient quality of life	83	Quality of life
Verbally asking about CPAP adherence / Sleep time	80	Adherence check
Requirement for humidification	78	Technical
Frequency of cleaning mask interface and circuit / changing filters	78	Technical
Check for control of witnessed residual snoring or apnoeas, choking spells	76	Symptom resolution
Asking about work schedule / Shift Pattern	76	Lifestyle issues / seep hygiene

Table 12 Components achieving the priority threshold for consensus (75% agreement with the priority score)

Components of a clinical review, listed in order of proportion of respondents who gave priority score of 4 or 5	% agreement with priority scores	Grouped into key categories
Components not achieving the priority threshold for consensus but with >50% agreement with priority scores		
Review of medical history, medication, any new comorbidities in relation to symptoms / need for hospitalisation	73	General medical assessment
Ask about factors that help / hinder CPAP use	73	Technical
Checking body weight	71	General medical assessment
Support system at home (Help with mask on / off fitting and filling of humidifier if used) / partner engagement	65	Technical
Recording the altitude where the patient lives and the altitude where the sleep study was carried out	61	Technical
Checking that any relevant Vehicle Licensing agencies are aware of the condition	60	Driving
Requirement to repeat the diagnostic study with significant weight loss / weight gain	60	General medical assessment

Table 12 Components not achieving the priority threshold for consensus, but with >50% agreement with priority scores

Components of a clinical review, listed in order of proportion of respondents who gave priority score of 4 or 5	% agreement with priority scores	Grouped into key categories
Components with <50% agreement with priority scores		
Checking partner feedback / quality of life	48	Quality of Life
Fatigue and depression scale / as interfering with objective assessment of sleepiness e.g. Epworth sleepiness scale	43	Sleepiness assessment
Checking blood pressure	40	General medical assessment
Checking electrical safety of CPAP unit	33	Technical
Nocturia / Frequency of getting up to pass urine	33	General medical assessment
Examination of the nasal passage and throat	28	General medical assessment
Advice re air travel	25	Lifestyle issues/seep hygiene
CPAP unit noise level	25	Technical
Cognitive / developmental issues	23	General medical assessment
Chest Auscultation	0	General medical assessment

Table 12 Components with <50% agreement with priority scores

5.5.4 Themes emerging from the free-text comments

Seven themes emerged from the free text comments:

- The first two were regarding checking for symptom resolution and the fifth one was about the partner's perspective on symptom resolution.
- The third theme was regarding the clinician's perspective on the importance of monitoring adherence to CPAP therapy.
- The fourth theme was regarding managing the technical aspects related to CPAP treatment.
- The sixth theme was regarding the requirement for further general medical assessment and input where needed.
- The seventh theme related to the clinician's perspective on the frequency and mode or clinical review. (See table 13)

Table 13 Themes from free-text comments

Assessment of symptom resolution
"Assessment should be focused on whether CPAP has improved the symptoms responsible for initial presentation as CPAP is mainly a treatment for symptoms" (Respiratory Physician and Academic). *"Should also have some documentation of residual AHI on treatment although I am cautious of putting this in as it is often a software derived number and in a significant number of cases never fully settles to <5."* (Respiratory Physician).
Assessment of sleepiness and its impact on activities including driving
"Questioning whether CPAP is used for all sleep episodes, daytime napping, etc., and documentation of whether excessive sleepiness has improved" (Respiratory Physician). *"I would take a detailed sleep history for a patient with residual sleepiness despite apparently effective CPAP".* (Respiratory Physiologist). *"Professional drivers should have annual nurse review at least".* (Respiratory Physician). *"We tend to offer 12 monthly reviews for HGV drivers".* (Specialist Nurse). *"Professional drivers will continue to be seen yearly".* (Respiratory Nurse Specialist).
The importance of monitoring adherence to CPAP therapy
"Telemedicine for CPAP adherence tracking should be encouraged and reimbursed" (Respiratory Physician). *"Download of the CPAP device should be performed or supplied ahead by the supplier"* (Respiratory Physician).
Technical issues regarding CPAP therapy
"Checking the operational status and cleanliness of the CPAP humidifier and mask. Need for replacement of same". (Respiratory Physician). *"Some of the content of CPAP reviews, such as electrical safety testing will also depend on whether the machine is provided by the CPAP clinic, rented or purchased by the patient"* (Respiratory Physician).
Checking partner feedback and quality of life
"Clinical review that incorporates the patient's bed-partner and / or other close family may be useful to help identify and manage any potential problems". (Specialist Nurse).
The requirement for general medical assessment
"Requirement to repeat CPAP titration study or conduct a review if the download information or patient report is inconsistent helps with mask issues CPAP pressure etc.". (Respiratory Physician).
Frequency and mode of review
"An early review after the first visit to initiate CPAP is in my view crucial for increasing the chance of long-term CPAP compliance." (Respiratory Physician). *"The frequency of a clinical review varies globally depending on local health care systems and providers, and professional drivers should have an annual review."* (Respiratory Physician) *"Clinical review provides the opportunity for education / support and reinforcement of treatment ".* (Respiratory Nurse Specialist).

5.5.5 Frequency of review

There was general agreement that the timing of a review should be flexible to meet the clinical needs of the patient as well as being compatible with the healthcare delivery context. Frequent review (face-to-face or telephone) was considered important to support initiation of CPAP therapy. A consensus on the frequency of review once CPAP therapy was established was more varied among participants, with twelve to eighteen-month follow up recommended by the majority, although review could more frequent in those with poor adherence to CPAP therapy. A flexible approach to review that offered 'open access' or follow up 'on-demand / or request' review was prioritised by 80% of the participants. Similarly, nearly half the participants prioritised the need to review CPAP users when there was a specific request from a traffic agency, or before elective surgery. Review via a telemonitoring option, where available, was also an acceptable option (Figure 7).

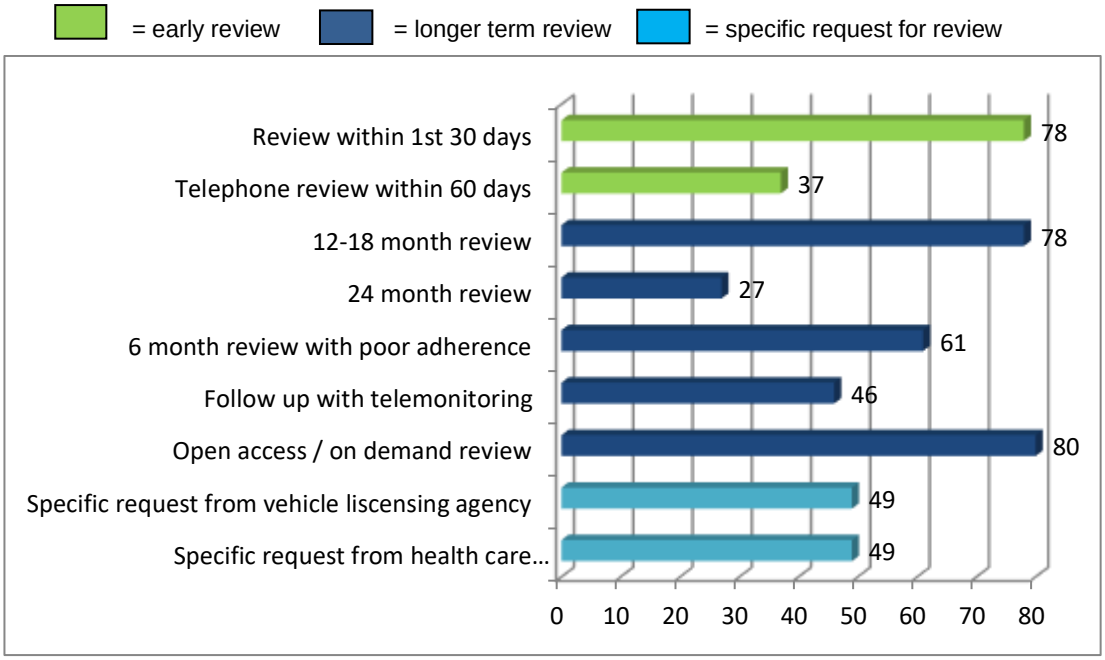


Figure 7 Frequency of review expressed as a percentage of respondents

5.6 Discussion

5.6.1 Main findings

This is the first study to provide an international consensus on the most important components that should be considered when reviewing people using CPAP therapy. From a list of 36 components, 17 reached a consensus (≥75%), and were considered the most important to include during a CPAP therapy review. The components identified have been grouped into key categories: Technical aspects (n=8 components); General medical assessment (n=7); Sleepiness assessment (n=3); Symptom resolution (n=3); Acceptability of treatment (n=2); Adherence check (n=2); Assessing sleep quality (n=2); Driving issues (n=2); Quality of life (n=2); and Lifestyle issues / sleep hygiene (n=2). The need for flexible follow-up arrangements was highlighted by the free-text comments indicating that clinical review arrangements should focus on individual patient needs. I mapped these prioritised components to a suggested prototype template that may support clinical reviews (Table 14). I concluded that the clinical review of CPAP therapy users should be flexible, frequent in the early stages of commencing CPAP, moving to 'on-demand' and / or remote follow-up for maintenance. This can be either face-to-face, via telephone, or teleconsultation with the oversight of remote telemonitoring of CPAP adherence and efficacy data. The findings of this study may inform future guideline recommendations for reviewing CPAP users.

5.6.2 Strengths and limitations

I generated an extensive list of potential components of a CPAP therapy review by amalgamating recommendations from current guidelines with suggestions from an international and multidisciplinary participant panel of clinicians and academics involved in sleep medicine reviews, and also from CPAP therapy users. Forty participants completed all three rounds of the study; only one person withdrew between the two scoring rounds enabling the consensus process, and this is important and may represent minimal risk of bias.

There are some strengths and weakness in using the e-Delphi method in conducting consensus research. This methods strengths are that communication can take place among experts in the field that enables linking of existing knowledge and can enable agreement/disagreement to be mediated thus avoiding confrontation. Experts can contribute to the understanding and resolution of important unknown problems where there are perceived gaps in evidence based guidelines. Another strength is that the e-Delphi does not require face-face meeting and can be conducted electronically on a large global scale in different time zones. Some weaknesses also exist with this method such as a lack of agreed standards on selection of participants and a lack of a universally agreed definition of consensus and interpreting and analysing the findings. There is also the limitation of generalising the findings to a wider population due to sample sizes and the differing expertise within the included panel.

An important strength of the study is that the participants are from a range of healthcare backgrounds involved in the delivery of sleep medicine services, representing 21 countries with a broad range of economic backgrounds and healthcare systems. Although the number of participants recruited to the expert panel is larger than other similar e-Delphi studies, (229), they may not represent the full range of perspectives from sleep medicine clinicians / providers as the delivery of sleep medicine services varies widely in healthcare systems globally. For example in the USA, sleep medicine physicians need to be board certified to deliver this specialist service. In less well-developed countries with less mature healthcare services access to this specialist service may be more limited and there is a risk that the participants in the e-Delphi may represent much more well-developed services.

There is also the consideration that the composition of the panel may have influenced the findings of the e-Delphi study. The e-Delphi panel comprised of 29 Respiratory physicians, 25 academics, 9 journal editors, 6 CPAP therapy users, 5 specialist nurses, 3 Respiratory physiologists and 1 respiratory

therapists. The Physicians/Academic/Journal editors may have been more likely to participate in the panel as many were active researchers in this field already and therefore their views may not represent those of the non-medically qualified participants which were of a much lower number therefore their views may be under represented. The inclusion of patients in the e-Delphi also has some advantages and disadvantages to consider. The advantages could be that the patients get to have their views represented about what they think is important to them however as there were only 6 in the panel their views may also have been under represented. Perhaps conducting separate e-Delphi's in these groups is a future research option, which may allow clinicians to compare and contrast what matters to patients regarding a clinical review.

I also focused initially on predetermined clinical review components, derived from current guidelines which may have resulted in the participants being less open to making their own suggestions regarding their usual clinical practice. However, I invited free text comments throughout all three rounds of the study, to allow opportunities for participants to comment, and this resulted in more than doubling the number of components included in the scoring rounds. Another strength I believe is that I used the free text comments, to assist with interpretation and development of the initial outline review template following on from a structured mapping of components (Table 13).

5.6.3 Interpretation in the light of other literature

5.6.3.1 Comparison with guidelines

Current clinical guidelines regarding the review of CPAP users collectively suggest ten components that should be included in a CPAP review, with the guideline from the AASM being the most comprehensive and identifying eight of twelve prioritised components (67). There is however wide variation in the guideline recommendations published between 2003 (70), and 2016 (78), highlighting the need for an international consensus on what is important to include in a clinical review, and how often this should occur. Some components

(e.g. asking if any problems with sleepiness while driving, objective assessment of sleepiness e.g. ESS, checking for any mask interphase issues), were strongly prioritised (>90%) in the e-Delphi study but were not always recommended by the guidelines. Specifically, current guidelines do not mention checking for resolution of the AHI despite it being given a priority consensus of 90%.

The Veteran Affairs / Department of Defence in the USA published a Clinical Practice Guideline for the Management of Chronic Insomnia Disorder and Obstructive Sleep Apnoea in 2019, and although the guidelines recommend support for adherence to CPAP and other therapies that may be beneficial and weight management recommendations, they do not specifically cover the clinical review process and components and how often this should occur (225).

The National Institute for Health and Care Excellence is currently updating their guidelines regarding OSAHS, and are due to be published in early 2021, and the draft scoping document that was published in 2018 does mention that monitoring of obstructive sleep apnoea / hypopnoea syndrome, determining the efficacy of treatment, how to monitor and how to improve adherence is within the scope of this guideline (226).

In 2019 the AASM published Treatment of Adult Obstructive Sleep Apnoea with Positive Airway Pressure: An American Academy of Sleep Medicine Clinical Practice Guideline. They recommend adequate follow-up, including troubleshooting and monitoring of objective efficacy, and usage data to ensure adequate treatment and adherence. This should occur following CPAP therapy initiation and during the treatment of OSAHS. The guidelines specifically mention periodic review by a qualified clinician is necessary to confirm adequate treatment, assess symptom resolution, and promote continued adherence to treatment, particularly in the early phase of CPAP initiation. They also recommend the scheduling of the review after CPAP initiation will be

person-dependent, with review in the few initial weeks to months after CPAP acclimatisation to support adherence and assess treatment effectiveness. Yearly review by a trained clinician is recommended, although extended follow-up may be appropriate for some patients. They also recommend that in patients with persistent or recurrent sleep-related complaints of persistent difficulties with CPAP use, more frequent review may be necessary. Finally, they recommend objective monitoring of CPAP therapy adherence to complement patient reporting of difficulties with CPAP use (3). These recent AASM guidelines mention 9 of the 12 possible review components identified in previously published guidelines (Table 11). Chest auscultation and BP monitoring were not mentioned in the guidelines; however, these components could be considered to be part of clinical routine review based on the individual clinician's decision as to whether this was necessary.

5.6.3.2 Driving regulations

In the UK, this prioritised component of a clinical review may reflect the recently updated guidance from the DVLA which states that in moderate to severe OSAHS subsequent licensing will require; control of the condition; improved sleepiness, and treatment adherence (90). A recent EU taskforce publication by Bonsignore et al 2020 has developed a statement to support clinicians who advise patients with OSAHS on fitness to drive and this may influence clinicians to make this component part of their routine review (45). This is an important finding of the e-Delphi study and driving-related issues need to be given a higher priority in future clinical review and guideline development. An Official American Thoracic Society Clinical Practice Guideline: Sleep Apnoea, Sleepiness, and Driving Risk in Non-commercial drivers was published in 2013, and recommendation nine states clinicians should familiarise themselves with the presentations and complications of excessive sleepiness as well as local and state statutes or regulations regarding the compulsory reporting of high-risk drivers with OSAHS (227). Based on current driving guidelines it would appear that asking about driving issues should be a core

component of a CPAP review which also applies to commercial drivers.

5.6.3.3 Tailoring the review to the individual context

The study identified a number of components that were considered as being less important (≤75%), suggesting these may be optional and included according to specific clinician judgment (Table 12), suggesting that there is a core of components that may always be included in a CPAP review; however, other components may sometimes be applicable. For example, a nurse-led or non-medical review may include the more highly proritised components, and not focus on a general medical assessment as this will have been part of the initial review at the point of diagnosis. In contrast in more complex CPAP review cases, a medical-led review may focus on other more detailed general medical assessment issues.

5.6.3.4 Supported self-management in CPAP users

Ongoing / long-term review also provides the opportunity for education, support, and reinforcement of the treatment rationale in CPAP users. Asking about quality and quantity of sleep, sleep routine times and work schedules / shift patterns, assessing quality of life, and reviewing patients preparedness to continue with treatment, all reached a priority consensus of >75%, although few current guidelines specifically recommend these components. This may reflect the clinical focus of guidelines which may overlook the nuances of how OSAHS affects people's lives. The consensus gained from the e-Delphi study has highlighted the importance of considering including these components in a review. Supported self-management is not a new concept and is used in many other respiratory conditions. A good example is in asthma care where supported self-management should not be considered as optional but an essential component of asthma long-term care, that is personalised to meet the needs of an individual and assist them in optimising their condition (227, 228). People need to be coached or taught how to recognise deterioration in

their condition to better self-manage, and the same principles can be applied to recognising deterioration in the symptomatic management of OSAHS so that patients take appropriate self-management action and contact their healthcare provider where review and intervention are required. A recent study by Daines et al, again related to asthma self-management concluded that patients reported learning intuitively how to self-manage, and clinicians should support and educate patients proactively (228). The same principles could be applied in CPAP therapy users, where attention to education and self-management at the outset is crucial to obtaining longer-term adherence and management of symptoms (229).

5.6.3.5 Practical Checks of CPAP devices

The priority attached to checking practical maintenance of CPAP equipment depended on the organisation of sleep medicine services; in some systems, this was not part of a clinical review. Practical checks such as electrical safety checking of CPAP devices are conducted in some services by the actual service provider or may be a contracted out service. The manufacturers of modern CPAP units do not recommend that this is necessary on an annual basis but some organisations do an annual safety check as this is part of their equipment service governance process. This can often be done at the time of a clinical review, however, with more remote monitoring of adherence data, it is easy to view where there are issues that need corrected like mask fit issues or adjusting pressure and humidification remotely.

5.7 Implications

5.7.1 Templates and their pros and cons

One option for implementing these clinical review components is to provide a clinical review template. While opinions vary on the use of templates in clinical practice, they can facilitate a structured review process and improve consistency of care and also reduce practice variation (230). They can support a guideline-based review that ensures evidence-based care is delivered.

However, there are limitations to the use of templates; for example, they may not address all issues with CPAP therapy usage from a patient's perspective so some flexibility in their use is important in the review. The use of templates to guide a CPAP review may be an attractive option for services, particularly for service evaluation, quality assurance, and audit purposes. However, the clinician needs to be cognisant of the fact that the review should remain patient centred, and about what matters to the patient and not simply become a 'tick box' exercise that could affect the patient / professional relationship. Building on the e-Delphi findings I have developed a suggested prototype template based on the components prioritised in the e-Delphi study that could assist sleep medicine clinicians to provide a structured review (Table 14).

5.7.1.2 The core components that will form the basis of the template

Acceptability of treatment
Checking for treatment side effects Preparedness to continue with treatment
Technical aspects of therapy
Checking mask fit issues Requirement for humidification Frequency of cleaning mask interface and circuit/changing filters
Subjective assessment of sleepiness
Recording subjective assessment of sleepiness / somnolence e.g., Epworth Sleepiness Scale Ask if any problems with sleepiness while driving
Measurement of adherence to CPAP therapy
Recording of CPAP adherence / efficacy by data download via memory card or remote monitoring Verbally asking about CPAP adherence/Sleep time
Resolution of symptoms
Checking if initial symptoms for referral have improved e.g., tiredness, sleepiness, hypersomnolence, concentration, memory Ensuring CPAP is resolving the Apnoea Hypopnoea Index Check for control of witnessed residual snoring or apnoeas, choking spells
Assessing sleep quality
Checking patient quality of sleep / feeling refreshed on waking Asking about quality / quantity of sleep/sleep routine time
Driving / Vehicle licensing agency issues
Asking about current driving status - Car / Heavy goods vehicle license
Quality of Life
Checking patient quality of life
Lifestyle issues/Sleep hygiene
Asking about work schedule / Shift pattern

Table 14 First prototype of the CPAP review template

Note: This table lists the prioritised components of a review. The vision is that this might be used as the basis of a (potentially computerised), template which would:

- Start with an open question setting the agenda for the review (What does the patient wish to discuss?).
- Act as a 'checklist' to prompt delivery of important components of a review.
- Include space for free-text entries.
- Be followed by a second page with the components that did not reach consensus, but which will be important in some contexts.

5.7.2 Implications for service models

A key finding from the e-Delphi study is that there needs to be flexibility in the delivery of services, both in frequency and the mode of clinical review. Early and frequent review is recommended as a priority for new CPAP users and those having difficulty with adherence, or practical problems such as treatment side effects, and reducing to annual / biannual review when more stable and acclimatised to CPAP therapy. The option of offering an 'open access' review service in which the patient could determine their need for review appealed to 80% of the respondents in the study. With the ever-increasing demand for sleep medicine services globally particularly in the midst of this global pandemic, this may be seen as an attractive option for healthcare providers. However, there is currently no published literature to inform this practice. Furthermore, with the rise in the implementation of sleep telemedicine services CPAP review can be facilitated with telemonitoring and overseen remotely.

5.8 Conclusion

The international expert panel agreed that the most important components of a clinical review of people using CPAP therapy to treat OSAHS were assessing: treatment acceptability; technical aspects of therapy; subjective sleepiness assessment; recording adherence / treatment efficacy verbally or by data download via memory card / remote monitoring; symptom resolution; driving issues; sleep quality; quality of life and lifestyle issues / sleep hygiene. There were diverse opinions on the optimal frequency of review but general agreement that relatively frequent review should be undertaken in the newly diagnosed patient. Long-term review will be less frequent, or potentially 'on-demand', and can be provided by a range of professionals with the option of using telemonitoring where available. The findings of the e-Delphi may inform future guideline recommendations in the delivery of care for people with OSAHS using CPAP therapy.

5.9 Summary of Chapter 5

This chapter outlines the findings of the international consensus study that has defined the most important core components of a clinical review. I have mapped these components into a prototype sleep medicine review template that aims to assist clinicians to conduct a patient-centred, structured, and evidence-based clinical review. In the next chapter, I will describe a mixed-methods evaluation of the implementation of the prototype template in three sleep Centres in Scotland.

Chapter 6: A Mixed Methods Implementation Study of a Structured Sleep Medicine Review Template in the Clinical Setting

In the last chapter, I described an International consensus study that defined the most important core components of a clinical review which I used to develop a prototype sleep medicine review template to guide clinicians conducting CPAP review for people with OSAHS. In this chapter, I will describe how I recruited three diverse sleep Centres in Scotland to participate in an implementation study of a prototype of the structured review template and to develop a strategy for using the template in their clinical practice (Figure 8). I then evaluated the use of the template in the three real-life clinical settings using a mixed methods research design. To inform more generally the impact of the implementation of the template in consultations, I explored perceptions of the impact on the dynamics of the consultation and specifically any potential disadvantages to patient-centeredness, and any potential benefits to the completeness of the review.

Name/Address label	Date	Age	Gender

The patient agenda for the review. What does the patient want to discuss? Any concerns?	Wt.: Ht: Adherence: Hours per night: BP: SPO2: TCC02: ABGs:

Acceptability of treatment	Are there any side effects from the treatment? Is the patient happy to continue treatment? Free text
Technical aspects of CPAP	Does the mask fit correctly? Is humidification required / satisfactory? Is cleaning / filters changing routine satisfactory?
	Free text
Subjective assessment of sleepiness / driving issues **<u>Epworth</u>** **1.Sitting** **2.TV** **3.Lying** **4. Reading** **5.Talking** **6.Public** **7.Passenger** **8.Driving**	Subjective assessment of sleepiness (Epworth Sleepiness Scale) : Are there any problems with sleepiness while driving? What are they driving: Car / Heavy Goods Vehicle?
	Free text
	Have witnessed snoring, apnoeas, choking spells been controlled? Have initial symptoms improved / resolved?
	Has CPAP resolved the Apnoea Hypopnoea Index? AHI: Mask Leak:
	Free text
Assessing sleep quality / quality of life	What is the quality of sleep? Do they feel refreshed on waking? What are their sleep routines? How much sleep do they get? Quality of life Free text
Lifestyle	Ask about work schedule / Shift pattern Free text

Additional optional review components that may be useful in some reviews	
Technical aspects	Factors that help / hinder CPAP use. Support system at home / partner engagement Altitude where lives / Altitude where sleep study was done Electrical safety of CPAP unit/ CPAP unit noise level
General medical assessment	Review of medical history, medication review, any new co morbidities in relation to symptoms / need for hospitalisation Examination of the nasal passage and throat Chest Auscultation findings: Need to repeat diagnostic study with significant weight loss / weight gain Nocturia / Frequency of getting up to pass urine Cognitive / developmental issues
Sleepiness assessment	Fatigue and depression scale / as interfering with subjective assessment of sleepiness e.g. Epworth Sleepiness scale Checking partner feedback / quality of life Are Vehicle Licensing agencies aware of the condition? Advice re air travel
Lifestyle	
Quality of life	
Driving issues	

We are assessing the use of this template and would value any feedback you can give us.

Did you find the template useful in this review? Yes ☐ No ☐ Not sure ☐

Did use of the template increase the length of the consultation Yes ☐ No ☐

Time taken to complete during consultation …….

Time taken to complete after consultation ……

Review method: Face-to-face ☐ telephone ☐ videoconsultation ☐

Do you have any feedback about how the template worked (or didn't) in this review?

Please tick box that represents your position

Respiratory Physician/doctor ☐ Physiologist ☐ Specialist Nurse ☐

Thank you for the feedback Name: Date:

Figure 8 Prototype template

6.1 Templates and their role in clinical review

The use of templates as tools to assist with the delivery of healthcare reviews can facilitate a structured and more guided review process and also support consistency of care and, have been used in clinical practice for more than two decades (230, 231). The use of electronic templates is widespread in general practice and was introduced to implement improvements in the quality of guideline-based care delivered for people with long-term conditions, as well as to standardise data collection linked to the Quality and Outcome Framework (232, 233). However, by directing the content of the consultation, templates risk overriding the patient's agenda and have the potential for preventing them from expressing their concerns or may constrain the clinical review process (233-235).

A recent study by Stanhope et al 2019 has shed further light on the impact of using a template-based electronic health record on person-centred care. They highlighted that there is a requirement to take a balanced approach to this form of consultation to provide some assurance that the use of electronic templates during a clinical review is both patient-centred and facilitates high-quality evidence-based care. Including an initial question in a standardised review template about the patient's main concerns has been suggested as a way to avoid potential disadvantages and facilitate a more patient-focused review (236). In the secondary care setting, there is evidence that the use of a structured template can facilitate a safe handover of patients between clinical teams, and also improve patient safety measures in ward rounds by reducing variation and standardising practice (237, 238). There is also evidence that the use of a standardised surgical template improves the quality and quantity of documentation regarding patient information compared to freehand text and is preferred by health professionals and can support the reliability of the audit quality and control process (239, 240).

The NHS Patient Safety Strategy was published in 2019 and central to this publication is a safer culture, safer working systems, and safer patient care.

Within this document, there is reference to the patient safety incident response framework and introducing standard reporting templates as a means of reducing variation nationally (241).

There has been no evidence on the use of a specific paper-based or electronic CPAP clinical review template until very recently. In 2020 Chakrabarti et al describe the use of an intelligent computerised guided decision support systems as a novel way to safely assess and review people with OSAHS who are using CPAP therapy. This system is based on the use of computerised templates with decision-support creating the possibility for non-sleep medicine specialists to conduct comprehensive guided consultation. This could support the entire diagnostic and management pathway and create capacity within healthcare systems – which the authors suggest could be valuable as the world recovers from the impact of the global pandemic (179). This system is not widely available within the UK to date. Their system offers a much more guided decision support pathway with safety net features within the computer program that can provide alerts and advice to non-specialist clinicians to support them to conduct an evidence-based consultation. It differs from my prototype template. My template is a means of recording the content of a CPAP review which can be either paper-based or computer-based.

6.2 Implementation Research

More than a decade ago Goldsmith et al 2007 et al 2009 described implementation research as a method of merging both quantitative and qualitative data sources that if successfully brought together and considered in one review could promote the uptake of research findings into routine practice (242). The field of implementation research has evolved over recent years as a methodology to understand and evaluate strategies for implementing evidence-based interventions in the real-life clinical setting (243-245)(248- 250). A core aim is to shorten the time it takes to translate evidence-based practice into routine clinical practice and hasten the integration of research policy and practice to improve health outcomes.

A core concept promulgated by the StaRI reporting standards and reflected in the terminology used in this implementation study is the distinction between the evidence-based intervention (e.g. core components of an OSAHS/CPAP review), and the implementation strategy which is used to promote the use of the intervention (e.g. the use of a template integrated within the existing routines of the clinical setting (246, 247). In an implementation study, it is the implementation strategy that is the primary focus of the evaluation (e.g. uptake and the use of the template in the clinical review).

Preparatory phases of implementation studies involve understanding existing routines and adapting the intervention and the strategies to improve local conditions in a specific health care delivery setting. Implementation research (in distinction to local quality improvement studies), seeks to create generalisable findings by adopting a theoretical approach and / or implementing in a theoretically determined range of settings (e.g. computerised / non-computerised clinics; mostly face-to-face / remote reviews; rural / urban setting). To inform more generally the impact of the implementation of the template in CPAP reviews, I aimed to explore the participating clinicians perceptions of its impact on the dynamics of the review, clinical autonomy, any potential benefits to the completeness of review, and also any potential disadvantages to patient-centeredness.

6.3 Need for research in this area

A search of the literature (AMED, (Allied and Complementary Medicine) 1985 to May 2020, Embase 1974 to 2020 May 06, ERIC 1965 to January 2020, Ovid MEDLINE(R) ALL 1946 to May 07, 2020, Books@Ovid May 04, 2020, Journals@Ovid Full Text May 07, 2020, NHS Scotland Journals@Ovid) did not identify any publications regarding the use of a sleep medicine review template in routine clinical practice.

6.4 Mixed methods research

The fundamental purpose of using mixed methods studies is that merging quantitative and qualitative data facilitates a better understanding of the findings than either approach alone. The overarching purpose of combining these methods is to expand and strengthen a study's conclusions and its contribution to the evidence base (248). I chose to use mixed methods in this study as the use of the differing approaches had the potential to provide a greater depth and breadth of study data information which I thought was not possible using either approach in isolation.

6.5 Aims and objectives of the implementation study

This multicentre study used both qualitative and quantitative mixed methods to develop and evaluate implementation strategies and explore the use / applicability of the sleep medicine review template in established CPAP therapy users. Quantitative data aimed to assess the effectiveness of the implementation strategy (e.g. uptake of the template by Centre and by the clinician), the feasibility of using the template (e.g. time taken to complete), and its ease of use as an intervention (fields completed, additional fields required). The qualitative data aimed to explore the perceptions of sleep medicine clinicians on the ease of use or otherwise, and any advantages or disadvantages of the use of the template in patient reviews. See Appendix 10 Implementation study protocol.

Aim

To introduce the template in three diverse sleep medicine Centres for use in face-to-face or remote consultations and observe implementation using mixed methods.

Objectives

I had four objectives which aligned with four phases of the study:

1. **Objective I**: To develop an implementation strategy adapted to the routines of the different Centres.
2. **Objective II**: To implement the use of templates in routine practice in each of the Centres.
3. **Objective III**: To undertake a mixed-methods evaluation, assessing uptake and use of the template and exploring perceptions of template use.
4. **Objective IV**: To refine the intervention and implementation strategy.

6.6 Methods

6.6.1 Study design

The CPAP review template was implemented over two months in the routine clinical review of people with OSAHS using CPAP therapy in three diverse settings. Ethical approval was obtained by the NHS Health Research Authority in July 2019 (Appendix 11). ACCORD was the study sponsor. I used the Standards for Reporting Implementation Studies (StaRI) checklist to assist in accurately reporting the findings of this implementation study (246, 247), (Appendix 12).

6.6.2 Recruitment of Centres

Three sleep medicine Centres in the UK consented to participate in the study. These Centres were selected to represent different contexts: City, Urban / rural, and Rural. Participation was discussed with senior clinical and / or management staff to gain management approval for the Centre to participate.

The participating Centres clinicians were invited to use the template when they were conducting a review of CPAP therapy users over the study period. Reviews could be undertaken face-to-face, by telephone, or teleconsultation review according to usual clinical practice. All the clinicians from the participating Centres were invited to use the sleep medicine review template in all reviews, and Centres were asked to ensure that a blank template was attached to each set of records, or the templates being readily available to the staff undertaking the review. Clinical staff had the choice of whether or not to use the template in their reviews. In addition, whether the template was used for any individual review or whether specific components were addressed was entirely at the discretion of the clinician.

6.6.3 Recruiting participants for the qualitative research

I aimed to recruit between five and seven clinicians (respiratory physicians, physiologists, specialist nurses), from each Centre (in total up to 20), who were actively involved in reviewing people using CPAP therapy to treat OSAHS to participate in a final qualitative interview. Interviewees were selected to offer a maximum variation sample of professional backgrounds, frequent / occasional / non-users of the template. All participants provided written informed consent before the interview.

6.6.4 Study procedures

In line with the four objectives (section 6.5), the study procedure followed four phases. The study objectives and procedures are summarised in Table 15 and mapped to the objectives.

Objective		Procedures
I	To develop an implementation strategy adapted to the routines of the different Centres	The implementation strategy for each site was developed during a series of site visits in discussion with the participating clinicians and the principal investigator. Each site had different routines and developed different strategies for ensuring a template was available for every consultation (e.g. attached to the clinic documentation or in a room where the review was being conducted. A Specific consideration was the choice of a paper or electronic template. The preference of the participating site teams was identified and materials prepared accordingly.
II	To implement the use of templates in routine practice in each of the Centres	At the baseline visit to set up the study, each site was provided with anonymised templates to suit their service (all sites indicated their preference for a paper version) – training, explanation on completion of the template was provided. A mid-study visit to check for any problems (and potentially make any minor amendments if feasible) was offered if required. Potentially this 'visit' could be done by telephone or teleconference.
III	To undertake a mixed-methods evaluation, assessing uptake and use of the template and exploring perceptions of template use	**Quantitative data collection** Anonymised copies of the template were collected for analysis of uptake The fields completed on the template The overall proportion of CPAP reviews **Qualitative exploration of clinical perception of template use (n=20 clinicians):** The discussion covered the following questions What were the advantages / disadvantages of using the template in clinical practice? How was the template used (during the consultation as a prompt, after the consultation as a means of recording the review content) What is the impact of the template on the conduct of the review, clinical autonomy, patient centred care, and patient / professional relationship?. Are there any components that should be added or removed from the template
IV	To refine the template and implementation strategy	Further refinement of the structured sleep medicine review template will be informed by the evaluation of the feedback from the semi-structured interviews and free text comments entered on the returned templates.

Table 15 Study objectives and procedures

6.6.5 Development of an implementation strategy adapted to the routines of the different Centres (Objective I)

I planned to visit each of the Centres at least once at the onset of the study. At one of these initial visits, I planned to convene a group discussion of participating clinicians during which I would explore perceptions of current arrangements, the components that they usually include in the clinical review of CPAP therapy users, and any benefits or concerns they may have about the use of a template

- The implementation strategy for each site would be developed during these Centre visits in discussion with the participating clinicians. Each site may have different routines and develop different strategies for ensuring a template be available for every consultation (e.g. attached to the clinic documentation or readily accessible within the clinic rooms).
- A specific consideration for the Centres would be the choice of using a paper or electronic template. The preference of the participating site teams would be identified and materials prepared accordingly.

Data collection: Field notes would be kept of all visits (including minutes of any formal discussions / meetings), telephone calls, e-mails, etc. All decisions about implementation strategies would be documented.

6.6.6 Implementation of the use of templates in routine practice in each of the Centres (Objective II)

Implementation would be the responsibility of the Centres, but I would support the implementation strategy by:

- Providing templates to suit the routines in the different services
- Providing training and an explanation on completion of the template.
- Offering a mid-study visit to check for any problems (and potentially support any minor adaptations to the template or implementation strategy, if required / feasible). Potentially this 'visit' could be done by telephone or teleconference.

Data collection: Field notes would be kept of all visits (including minutes of any formal discussions/meetings), telephone calls, e-mails, etc. Any adaptations suggested by Centres to improve implementation would be noted.

6.6.7 Mixed-methods evaluation, assessing uptake and use of the template and exploring perceptions of template use (Objective III)

6.6.7.1 Quantitative data collection

I planned to undertake a quantitative assessment of the effectiveness of the implementation strategy at promoting the uptake of the intervention (template use). A process of secure collection of anonymised copies of completed templates for analysis would be agreed with each Centre. Anonymised copies of completed templates would be collected at each site visit. Paper copies would be anonymised by removing any patient-specific identifiable information from the completed templates before collection at subsequent site visits or scanned and returned by NHS email. The templates would indicate the Centre, the clinician's profession, and mode of review (face-to-face, teleconsultation, telephone), and an estimate of the time taken to complete.

Data collected were:

- The total number of templates completed at each Centre
- The mode of clinical reviews undertaken by clinicians in the participating Centres as reported by the Centres
- The clinician's specialty that completed the template
- The age and gender of the patients reviewed
- The number of components completed in the templates
- The total proportion of CPAP reviews conducted in each Centre.
- Estimated time to complete the template during or after the review. For the purposes of the research, the template included a box for the estimated duration of the review
- Any free text comments added by the clinician to the template

6.6.8 Qualitative exploration of the clinical perception of template use

At the end of the study, I planned to recruit between three and seven clinicians from each site (in total up to 20), to participate in semi-structured interviews. These interviews would explore how the templates were used in the clinical setting. I planned to audio record all the interviews and transcribe them into the NVIVO software program for thematic analysis.

The discussion would cover the following questions (Table 16) detailed topic guide).

Main question	Additional questions	Clarifying questions
1. I am interested in your views on whether you think there is any value or not in using a structured template for a clinical review – for example, is it worthwhile. Do you already use one in your service?	If you do can you clarify what is different about your usual way of working or if you do not how did using the template impact your usual way of working?	• Can you please clarify what you mean by ....? • Can you please expand a little on ...? • Can you please give some examples of ...? • In particular, what do you think of...?
2. I am interested in your views of any evidence to support this way of conducting a clinical review.		
3. Are you aware of any evidence to support the use of guided templates in a clinical review?	Can you clarify why you think it is or it is not worthwhile using a template in a clinical review?	
4. Was the decision to participate in the completion of the implementation of the template team led or organisationally led? And did you perceive there was any pressure to participate?	How well do you think the template was implemented over the study period in your service?	
5. Do you think you as an individual or your service were given adequate support/instruction on how to implement the template?	Can you clarify what additional support/instruction may have helped with the implementation of the template?	
6. Do you perceive any differences (even if you did not use the template) in the way that a consultation would be carried out as a result of using the template in a clinical review?	Can you clarify what perceived differences would using the template have made to the consultation process/if any?	

7. What is the impact of the template on the way the clinical review is conducted regarding: • your clinical autonomy? • your patient/professional relationships? • patient centred care. 8. Are there any additional components that could / should be added or removed from the template, or how it could be improved 9. Do you think you or your service will continue to use the template or adapt it to suit your service after study completion? 10. What factors do you think may have improved the uptake of the template in your department/service? 11. Do you think the use of a structured template will modify or change the way that you deliver your clinical review or the service in the future? 12. I am interested in your views about the team individual's knowledge and skills and whether you feel the template is useful or not in your personal opinion and your style of practice / consultation 13. I am interested in the structure of your service and how individual members of the team conduct clinical reviews of CPAP therapy users I am interested in how the MDT network and communicate within your service and how each MDT member contributes to developing and delivering the service I am interested in your views on service evaluation and improvement and readiness for implementation of a structured review template in your service	Can you clarify any additional impacts, either positive or negative of using the template on the clinical review regarding: • autonomy • patient/profession relationship • patient centred care Can I ask you to clarify why any additional components you would like to see are added/removed from the template Please can you clarify why you may or may not wish to use the template or adapt or modify it for your service Can you clarify what factors may have helped more clinicians to participate in the study? If so, can you clarify why this may change your future practice or service delivery	

Table 16 Semi-structured interview topic guide

- What were the advantages / disadvantages of using the template in clinical practice?
    - How was the template used (during the consultation as a prompt, after the consultation as a means of recording)?
- What is the impact of the template on:
    - The way the clinical review is conducted.
    - Clinical autonomy.

- Patient / professional relationships.
- Patient-centred care
- Are there any components that should be added or removed from the template?

6.6.9 To refine the template and implementation strategy (Objective IV)

This would be informed by the evaluation of the feedback from the semi-structured interviews and free text comments entered on the returned templates. Further adaptation of the structured sleep medicine review template could then be provided to the Centres for future implementation.

6.7 Data analysis

6.7.1 Quantitative data analysis

The analysis of the quantitative data would be limited to descriptive analysis and summary statistics and the key outcomes would be the effectiveness of the implementation strategy as assessed by:

- The uptake of the template by the participating clinicians and sites (the number of templates completed at each site by the clinicians and the proportion of all clinical reviews).
- The feasibility of using the template in terms of perceived time taken to complete the template; participants would be asked to record this timing at the end of the template.
- The usefulness of the template as an intervention in terms of the number of fields completed and any additional fields suggested by participants.
- Subgroup analysis - the use of the template in relation to the mode of clinical review and the background of the clinician who conducted the review

6.7.2 Qualitative data analysis

The following qualitative data would be available for analysis:

- Initial group discussion with clinicians at participating sites (from field notes at initial visit) about components of the review they normally include in their clinical practice, and practicalities of implementing the use of the template.
- Free text comments added on the anonymised templates by clinicians during reviews which could provide immediate insights into positive / negative perceptions of review
- Clinician / semi-structured interviews would be audio-recorded at the final visit end of the study.

- The intention would be that these interviews would be conducted as soon as possible after the end of the two month study period, but the COVD-19 pandemic meant that data collection had to stop after three interviews. As soon as I was allowed to recommence research activities, I completed the interviews (by Microsoft teams video-conferencing) but this was 4-5 months after the end of the study. I planned to conduct a thematic framework analysis of the qualitative data.
- I aimed to use constant comparison, seeking a wide range of views, particularly seeking out contradictory views, and aimed to recruit participants until no new views related to the research objectives were expressed i.e. data saturation.
- It was agreed that more than one researcher would extract 5% of the data to ensure that there was attention to the potential for and control for interviewer bias.
- I aimed to use NVIVO software to thematically analyse the qualitative data.

6.8 Framework analysis

The Framework Method for the analysis of qualitative data has been used since the 1980s and is increasingly popular in medical and health research (249). It is a method that exists within a wide range of thematic methods (see section 6.9) and is described as an adaptable, organised, and precise method of analysing qualitative data that can facilitate transparent and clear results particularly with regards to theme-based and case-based analysis (250). It can assist the researcher to structure and assist logical thinking in an organised way and is a model that can direct and enable sense-making and understanding of research outputs.

I chose to use a framework analysis to interpret the qualitative study data because it is particularly suited to multidisciplinary health care research and it is not aligned to any particular philosophical, epistemological, or theoretical method, and can be used with a variety of qualitative methods (259). It facilitates a coordinated approach to qualitative data interpretation. In addition, the systematic process of framework analysis, and the ability for analysis to be completed with the help of computer software provides useful structure and guidance for novice researchers (249).

Framework analysis uses a step-by-step process for thematically analysing research data and these steps are outlined in Table 17. The large volume of research data generated by qualitative interviews means it is vital for researchers to categorise the data and be able to manage the data in an organised way to ensure that it is available for analysis and to provide evidence of the research's accuracy and reliability. It lends itself well to the evaluation of implementing healthcare interventions, and it can be used with a mixture of narrative methods of collecting data, such as interviews and focus groups (250, 251).

There are some challenges with framework analysis. As with all thematic analysis, it is a lengthy process that consumes a lot of researcher resources and time and generates a vast amount of data (249, 252). I decided that framework analysis had more strengths that outweigh the challenges of this method and considered that it was well suited to analysing the data from the implementation study. Table 17 describes the steps in the framework analysis.

Stages of framework analysis (250)	
Transcription	A good quality audio recording and, ideally, a *verbatim* (word for word) transcription of the interviews are required. The process of transcription is a good opportunity to become immersed in the data and is strongly encouraged for new researchers. The transcription of the audio recordings can be uploaded to NVIVO software to assist with the data analysis and coding process.
Familiarisation with the interviews	Becoming familiar with the whole interview using the audio recording and any contextual or reflective notes that were recorded by the interviewer is a vital stage in interpretation. It can also be helpful to re-listen to all or parts of the audio recording before coding the data.
Coding	Coding aims to classify all of the data so that it can be compared systematically with other parts of the data set. After familiarisation, the researcher carefully reads the transcript line by line, applying a paraphrase or label (a 'code') that describes what they have interpreted in the passage as important. Again this can be done manually with a software program such as NVIVO and can be used to conduct the coding process. A deductive approach to thematic coding of the interviews is best suited where a theory is being tested for example the participants responses on the usefulness and uptake of the template in this implementation study.
Developing an analytical framework	After coding the first few transcripts, the researchers involved should meet to compare the codes they have applied and agree on a set of codes to apply to all subsequent transcripts. Codes can be grouped into categories (using a tree diagram if helpful), which are then clearly defined. For this study NVIVO was again used to generate the codes. This process results in generating a working analytical framework.
Applying the analytical framework	The working analytical framework is then applied by indexing subsequent transcripts using the existing categories and codes. Computer-Assisted Qualitative Data Analysis Software (CAQDAS) is particularly useful at this stage because it can speed up the process and ensures that, at later stages, data is easily retrievable. In this study, NVIVO software was used to analyse the framework.
Charting data in the framework matrix	Qualitative data are voluminous and a 30-minute interview can generate many pages of text. Being able to manage and summarize (reduce) data is a vital aspect of the analysis process. The text transcripts can be tagged automatically if you are using CAQDAS to manage your data. (N-Vivo version 9 onwards can generate framework matrices). I used NVIVO 12 which was able to automatically generate a coding matrix.
Interpreting the data	It is useful throughout the research to have a separate notebook to record field notes or a computer file to note down impressions, ideas, and early interpretations of the data. If the data are rich enough, the findings generated through this process can go beyond the description of particular findings to explanation of, for example, reasons for the emergence of a phenomenon, predicting how an organisation or in this research study multidisciplinary teams may be likely to instigate or continue to use the intervention in their future service.

Table 17 Stages in the framework analysis

6.9 Alternatives to Framework analysis: Grounded theory

There are other methods of analysing qualitative data such as grounded theory. Grounded theory is flexible, but also a structured methodology. It can be applied when there is not a lot known regarding a phenomenon, with the objective being to generate an explanatory concept that reveals or exposes characteristics of the subject of the qualitative research (253). It refers to a set of systematic inductive methods for conducting qualitative research aimed towards theory development and can follow three main types of approach: Classical grounded theory, Modified grounded theory, and Constructive grounded theory.

A classical grounded theory approach is based on the Glaser and Strauss 1967 book “The Discovery of Grounded Theory”, an approach viewed as theory generation rather than just an analytical method. The aim is to scrutinise the data and look for new theoretical insights. All data are considered in the process including fieldwork notes and other data that are generated. With this approach, it is recommended that the data generated are not examined first which avoids creating an early bias with the focus being to create a conceptual theory that may explain a relevant pattern of behaviour relevant to the research participants.

Modified grounded theory is based on representational interactionism from a sociological perspective that relies on the representational sense that people attribute to the social interaction processes. It addresses the subjective meaning people place on behaviours or objects or even events based on what they believe is their truth. Depending on the different participant perspectives, a modified grounded theory might be more action-focused and may permit more theory to come from both the researcher as well as the data itself (254).

Constructionist grounded theory fully involves the role of the participant and researcher in the process of generating knowledge and interrogating the nature of the knowledge generated and theory generated. It aims to seek numerous perspectives or points of view and also explores and discovers patterns and connections among categories in the data. It encourages researchers to engage with various interpretations of the phenomenon of interest and accepts that the analysis is generated from collective experiences and interactions between the researchers and the participants (254, 255).

I chose not to use a grounded theory approach for the qualitative data collection and analysis of the implementation study as it uses an inductive approach that is concerned with the generation of new theories or themes from the data. The more deductive approach of framework analysis is aimed at testing existing theory which in the implementation study was the content and structure and the use of and uptake of the structured sleep medicine review template in participating Centres, with the contents of the template already informed by the findings of the international e-Delphi consensus study. I aimed to understand the perceptions and practicalities of implementing templates in routine clinical care, not new theory generation (10).

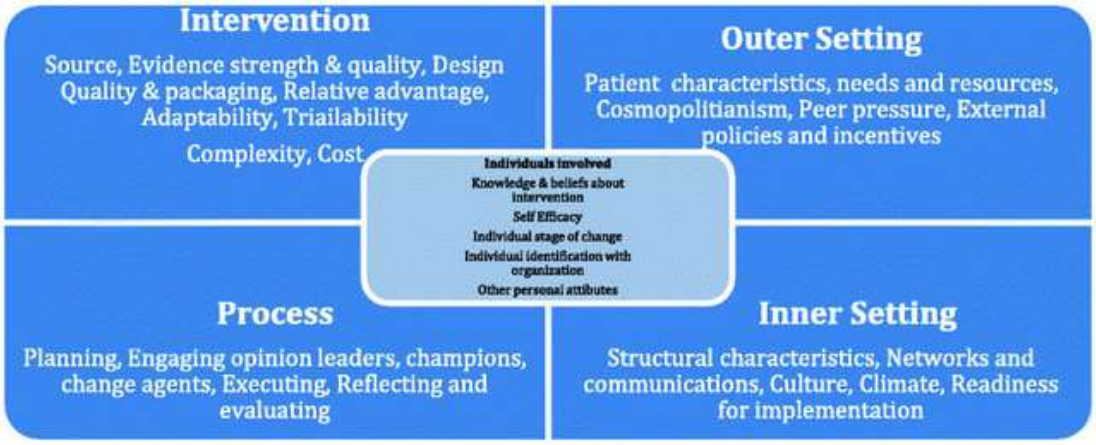
6.10 Frameworks that could inform my framework analysis

6.10.1 The Consolidated Framework for Implementation Research

The Consolidated Framework for Implementation Research (CIFR) is a theoretical framework designed to support the systematic assessment of multilevel implementation settings to characterise components that might impact implementation and its effectiveness in healthcare settings (256). A number of evidence-based effective interventions in healthcare services research are often unsuccessful in their translation into clinical practice, with missed opportunities for contributing to improving patient care. The CIFR can be used to characterise and comprehend contextual factors that may impact the implementation of complex multicomponent health care delivery

interventions. It consists of five domains that can all impact a healthcare intervention and is particularly suitable for rapid cycle appraisal of complex healthcare intervention delivery. The domains are described in Figure 7. There are also a number of constructs within each domain that the researcher needs to consider however, it is designed to be a flexible tool that the researchers can adapt to fit the intervention, design, factors, and research situation.

Figure 9 Consolidated Framework for Implementation Research (CFIR) domains



I considered these domains when I was developing my topic guide (to ensure I had addressed key factors that might have determined implementation); in developing a coding framework; and finally in the discussion of the qualitative data findings of the implementation study.

6.10.2 The Normalization Process Theory (NPT)

The NPT is a sociological toolkit that can be utilised to assist researchers to understand the processes of developing, implementing, embedding, and integrating some new technology or complex intervention in healthcare (257, 258). The NPT proposes four conceptual elements or constructs that represent different methods of working patterns around implementing a new practice: Coherence, Cognitive Participation, Collective Action, and Reflexive Monitoring (258, 259). In parallel with CFIR, I considered using the NPT as a framework to inform coding, analysis, and interpretation of the data from the qualitative interviews.

6.11 Steps in the framework analysis

I followed the seven steps in the Framework analysis to assist me in analysing the qualitative data results (Table 17). After completion and thematic coding of the qualitative interviews using NVIVO 12 software, I chose to incorporate the CFIR (260, 261), in the discussion section of this chapter. I chose to do this at this point as its domains of outer setting, inner setting, intervention, process, and participants felt like a good fit with the themes that emerged from coding the semi-structured interviews (Table 20). I did initially also plan to use the NPT framework in addition to the CFIR, however, after completing and coding the thematic analysis of the interview data I felt that the NPT framework was a much more subjective than objective way to analyse the data compared to the CIFR framework, therefore I chose not to use the NPT framework.

6.12 Synthesis of quantitative and qualitative research data

The limitations of qualitative and quantitative research individually in making sense of all forms of evidence-based healthcare have been recognised for almost two decades (262, 263). In only concentrating on only one of these methods, there is the risk that important evidence-based findings may be overlooked or disregarded (264). Combining the results of both quantitative and qualitative data can be defined as a mixed-methods approach with the aim being to strengthen and compensate for respective limitations of the different methods (131, 262).

More recently there has been increasing acknowledgment that amalgamating both quantitative and qualitative evidence in a mixed-method combination can be beneficial and support the understanding of how complex healthcare interventions may impact in certain settings or environments. This approach can potentially provide a better understanding of how multifaceted interventions may work and for whom, in which healthcare systems and how systems respond or adjust to the implementation, (in this case the

implementation of a structured sleep medicine review template). I used a parallel-results convergent synthesis design where qualitative and quantitative data are analysed and presented separately with integration occurring in the interpretation of results in the discussion section of this chapter. This mixed-methods approach for the synthesis of the quantitative and qualitative data can also use triangulation to analyse the quantitative and qualitative data separately and then compare the research findings to develop a complete understanding and oversight of the clinical effectiveness or otherwise of the implementation strategy and the clinicians perceptions on how the use of the template affected (positively or negatively) the efficiency, completeness and / or patient-centeredness of the review. Triangulation provides opportunities for convergence and corroboration of results that are derived from different research methods (265, 266). Each component of the study data is assessed for agreement (convergence), complementarities, or contradictions (dissonance). The interpretation of the qualitative data from the thematic framework analysis would be compared with the quantitative data to corroborate or contextualise themes that emerged from the qualitative findings that may inform further refinement of the review template for future implementation.

6.13 Reflexivity

As a novice with no previous experience in qualitative research methods the concept of ‘reflexivity’ was completely new to me. Reflexivity is described as a means of helping qualitative researchers to achieve consistency, accuracy, and quality in their research, and is important for defining the reliability and fidelity of qualitative research. All qualitative research takes place within a specific place and time and may involve two or more people as well as the person who is conducting the research. It is essential to show transparency in all the components of the research so that the reader can comprehend the research setting or situation and its validity (266). It includes the requirement to challenge and articulate any social and cultural factors that may affect the

context of the research setting (267). Reflexivity can improve the trustworthiness and reliability of the research and contribute to one of five quality features for publishing (the others are credibility, dependability, transferability, and confirmability of the research findings).

Critical distance and impartiality in the data analysis was a vital component to consider in conducting the implementation study as I was the principal investigator and also a practicing sleep medicine clinician with 23 years of experience in this speciality with my own beliefs and experience. Therefore, as an 'inside researcher,' it was important to consider the potential impact of my preconceived personal biases while conducting this study. I was very keen to hear all the views of the participants regarding the template and how it may (or may not), be helpful in other sleep medicine services but recognise that my personal bias was a factor I needed to keep in mind throughout this implementation study. I actively encouraged and sought out any criticisms of the template from the participants during the interviews. In other respects, the 'inside researcher' position was a strength, as I had a deep understanding of the subject area and the context in which it was delivered and I felt this was an advantage in linking the theoretical and the empirical parts of the study.

I was also interested in exploring where service improvements may be possible by reducing variation, standardising processes, and sharing best practices, but I tried to be open to all critical comments and suggestions. Reflecting on the national context there is a national sleep medicine service improvement and remobilisation workplan ongoing in which I am involved and this will have influenced my understanding and questioning. It may also have affected responses I received as many of the interviewees would know about the national improvement Workplan and my role in it. In addition, I was keen to share any learning from this implementation study with the national team.

I had developed the structured sleep medicine template based on the international e-Delphi study, and this was another factor on which I had to be

reflexive because of the risk of preconceived bias, however, I tried to be open to suggestions about the structure and content of the template and its helpfulness or not in a clinical review and its usability. Objective IV in the study was to refine the template based on feedback from the participants so I felt that this was a really important factor in controlling my personal bias regarding suggestions about how the template could be improved for future use. Some helpful feedback and suggestions were provided that have assisted me in refining the template which may be used as a paper or electronic version. I kept a research diary, where I would record reflexive notes, impressions of the data, and thoughts about analysis throughout the process.

I knew four of the nine clinicians who participated in the semi-structured interviews very well through a national sleep medicine network and this will have affected the interviews in that I already had a personal relationship with these individuals, and also had some prior knowledge of how their services were delivered. This may also have introduced some 'scope drift' in the way in which the topic guide was used and how the interviews were conducted. One positive reflective consideration is that this also made the interviews feel very comfortable and it did allow a relaxed and open-ended style of asking questions. Following on from the first three interviews I was able to have a discussion with my co-researchers about the content of the first three interviews and how I needed to be more reflexive in my future interviews and this helped me to develop the topic guide further to address the research objectives in which I was interested. This assisted me in the final six interviews which generated more useful and richer qualitative data content.

I also needed to recognise and take into account the ongoing potential for bias as the study progressed and continue to apply critical reflection to ensure the relevance of the data collected and the appropriateness of the analysis methods I used to develop the themes that emerged from the study. Good record-keeping at all stages of the study was conducted by keeping field notes and a written journal recording all communication with my co-researchers as

well as any emails with the study participants to maintain a transparent and consistent view of the data collection and interpretation. I also compared and contrasted similarities and differences from the interviews which were assisted by the use of the NVIVO software in the analysis of the responses from interviewees to ensure differing opinions / views were captured as reliably as possible in the thematic analysis. I also continuously engaged with my co-investigators throughout the study to reduce the potential for personal research bias as much as possible.

The initial two semi-structured interviews were the first time that I had conducted qualitative interviews. Although I used a topic guide to assist with the interviews, after transcribing these first interviews I could see that I tended to speak too much about my clinical service and experience and did not ask the participants to expand on some of the responses they gave me. This will have influenced how they responded to the questions and may have reduced divergent opinions as interviewees aligned with my views. A factor that may have improved my qualitative interview techniques prior to commencing the study would have been to ask one or two of my colleagues to participate in a recorded interview to allow me to develop a more reflexive approach to the semi-structured interviews. A total of nine semi-structured interviews, one start-up group discussion, and the free-text comments on the templates were transcribed and analysed by following the framework analysis process described in Table 17. It was evident that in the later six interviews that more themes were emerging that I think reflects these final semi-structured interviews were of a higher quality as I developed more reflexivity in my interview style.

Another reflection is that I conducted the first three interviews before the SaRs-Cov-2 pandemic stopped research activities. This meant I had very limited data which threatened the credibility, dependability, transferability, and

confirmability of the research findings to the point that conducting a thematic analysis would not have been possible. However, due to a respite in the pandemic in July 2020, I was able to conduct the final six interviews using the secure NHS Microsoft teams teleconsultation facility. These final interviews enabled the implementation study to meet the study objectives. Reflecting on the use of Microsoft teams to conduct these interviews, it was a very positive experience for me as the participants all consented to have their interview audio recorded via Microsoft teams and this made the interview transcription process easier, clearer, and smoother to complete.

A final reflective observation is that of maintaining the confidentiality of the sites and the participants identity as the country in which this study was conducted is small, however, there are several similar City sites, Urban / rural and Rural sleep medicine services throughout the country that reduced the risk of the Centres being identified.

6.14 Impact of COVID 19 on the study

Despite a carefully planned implementation study protocol, due to the arrival of the Coronavirus global pandemic in the UK on the 28th February 2020, this resulted in a delay in qualitative data collection however I was able to meet the aims of the study protocol (Appendix 10). I was able to complete the final six interviews in July 2020 due to some respite in the pandemic, and I believe that despite this the qualitative and quantitative data will be able to meet the four study objectives. These final later interviews were conducted almost five months after completion of the study which may have impacted on the recall of the experience of those who used the template, although it had the advantage that I was able to determine that one site continued to use the template beyond the study perhaps indicating that there may be some sustainability of the intervention either in paper or electronic form in future service delivery.

6.15 Results

I recruited three Centres to participate in the study: Centre one – City, which was a large tertiary sleep medicine service, Centre two, Urban / Rural also a city-based sleep medicine service but also serving a large rural catchment area and Centre three Rural, covering a mostly remote and rural wide catchment area. The Centres will be referred to as City, Urban / Rural, and Rural in the results section.

The City Centre was a large mostly urban-based sleep medicine Centre that serves a population of around 800,000. This was a large and well-established city-based sleep medicine service within a dedicated unit where diagnosis and treatment of a range of sleep disorders are conducted. The multidisciplinary team consisted of respiratory physicians, speciality doctors, physiologists, and nurses.

The Urban/Rural Centre was a sleep medicine service serving a population of around 400,000 in a large city but with a very wide rural catchment area and geography again being a significant challenge in delivering services. The multidisciplinary team at this site also consisted of respiratory physicians, speciality doctors, physiologists, and specialist nurses.

The Rural Centre was a sleep medicine service serving a population of around 310,000 in a remote and rural region with significant geographical challenges for clinicians and patients to travel to deliver and receive services in the remote clinic sites as well as in the main Centre. Again the multidisciplinary team at this site also consisted of respiratory physicians, speciality doctors, physiologists, and specialist nurses.

6.15.1 Objective I To develop an implementation strategy adapted to the routines of the different Centres

Visits were conducted face-to-face or virtually at all three participating Centres to establish the local routines of these services to develop tailored implementation strategies. Specific practical considerations were how to ensure a template was available for use during reviews (e.g. attached to the clinic documentation or readily available within the clinical environment to complete during the review). The City Centre and the Urban / Rural Centre were conducted by face-to-face visits and the Rural Centre was conducted using video consultation. All sites chose to use a paper version of the template. These could be anonymised (patient name and CHI number blanked out) before copies of the templates were made available for the evaluations.

City Centre: Developing the Implementation strategy

I was invited by the Lead respiratory consultant to present my research protocol at a multidisciplinary team (MDT) meeting with approximately 20 clinicians, and eight clinicians indicated they were interested in using the templates (three consultants and five registered nurses). I was given the e-mail addresses of the interested participants and I arranged to provide the study resources electronically.

Following the MDT meeting, I met with four of the nurses from the sleep medicine department for an initial group discussion. They were based within the sleep medicine unit and conducted sleep medicine clinical reviews regularly both in the ward and the outpatient clinic setting. These four nurses provided written consent to allow me to audio record the group discussion which involved asking the participants what they normally did within a sleep medicine review using a topic guide. I also made field notes. I wanted to learn about the routines within the department and how they normally structured a clinical review. I explored perceptions of current arrangements, the components that they usually included in the clinical review of CPAP therapy

users, and any benefits or concerns they may have about using the template. We discussed the best way to facilitate the completion of the templates at the time of the clinical review and how to collect these anonymised templates after study completion. The nurses agreed that they would attach the paper templates provided to the clinical notes, and they would keep the anonymised templates securely within the department until the next site visit when I would collect them.

The completion of the template was in addition to the paperwork they already had to complete so they were concerned that this may extend the time of their consultations. They normally wrote their clinical reviews in long-hand text and then also had to enter this in the electronic care record so I understood how implementation at this site might be impacted by the additional work of completing the template. I felt that this was a good start to the implementation study however I had very little time with the consultant medical staff due to clinic workload and I noted that I would need a further site visit to facilitate their engagement.

I was also able to meet with the Service lead for the department who agreed that she would speak to the team and encourage them to use the template. I was informed that the department was already undergoing a service redesign and improvement plan as there were very long waits for diagnosis and treatment of OSAHS and commencing CPAP therapy. I was able to conduct an initial discussion with one of the Consultants early in the study by telephone and he also agreed to use the template however he was then redeployed to another service and did not participate further.

Urban/Rural Centre: Developing the Implementation strategy

A start-up visit group discussion was undertaken with a specialist nurse and one respiratory consultant. The specialist nurse completed all the clinical reviews for CPAP and NIV patients and the consultants mostly saw the new patients and did not conduct any CPAP reviews. The specialist nurse already used a template in her clinics however was happy to use my template for the duration of the study. She commented that the content of my template was very similar to hers except hers was structured differently and she used separate templates for CPAP and NIV reviews. Severe train delays impacted our scheduled appointment time; therefore, the initial group discussion was a bit too short to have a full discussion about what the specialist nurse included in her clinical review and we agreed that we could continue the conversation at a later date. This was clarified at the final interview. It was agreed that completed templates would be anonymised and scanned and emailed to me by secure NHS mail. It was clear that this was very much a nurse-led service at this site and the consultants were unlikely to use the template.

Rural Centre: Developing the Implementation Strategy

The first visit was conducted by teleconsultation using NHS Near Me with two respiratory physicians and two specialist nurses who all agreed on the use of templates. A member of the administrative team was tasked with providing a template for all the consultations and collating the anonymised templates for return by collection in person or by registered post at the end of the study. It was agreed by the Lead respiratory consultant that they would commence the study the first week in January for eight weeks. A template would be made available for all the clinical reviews and the decision was team led by the Lead respiratory physician. It was also highlighted that this service was also looking at service redesign and they were keen to participate in this study as the template was ready for use and could support their local initiative. There was clear enthusiasm for participation in the study by this whole team.

6.15.2 Objective II To implement the use of templates in routine practice in each of the Centres

The study was implemented in the three Centres between October 2019 and closed in March 2020. (Appendix 13 outlines the site visit schedules and field notes from these visits).

City – Progress with implementation

The City Centre was the first site to participate and two mid-study site visits were conducted during the implementation phase.

I attended a site visit where I sat in on a morning clinic with one of the consultants to understand the medical clinic routine and spoke to the clinic outpatient staff to see how they might assist with attaching the templates to the case notes for completion. This was agreed however, no templates were completed and there was no medical participation in the study at this point. Another mid-study visit was conducted where I attended the ward team meeting again and several medical staff members and nurses expressed interest.

Urban/Rural

No mid-site visit or telephone support was required at the Urban / Rural Centre.

Rural

No mid-site visit or telephone support was required for the Rural Centre.

6.15.3 Objective III To undertake a mixed-methods evaluation, assessing uptake and use of the template and exploring perceptions of template use

6.15.3.1 Final data collection Centre visits

City fourth visit 8-2-20

A further visit was conducted again meeting with the department manager and several other nursing team members did agree to participate at this fourth visit however, the Coronavirus made an appearance in March 2020 and the priorities for all respiratory teams was to focus on patient care of those affected by COVID 19. This would also have required a protocol amendment to extend the study time and all studies were halted by ACCORD at this time.

Urban/Rural Final Telephone review 12-2-20

A final structured recorded interview with one respiratory nurse specialist who completed the study was conducted by telephone following informed consent. The quality of the interview was much reduced by not being able to pursue a video consultation as the technology was not working properly that day. I did take extensive field notes as well as recording the interview which helped to capture most of the content of the semi-structured interview and I was invited to telephone this participant if I needed to clarify anything regarded the recorded interview content.

Rural final visit

Email correspondence from the Lead respiratory physician from the 13th March 2020 confirmed that they had completed the study as planned however due to the arrival of the COVID 19 pandemic they were asked by their NHS board to cancel non-urgent visitors to the hospital so a final face-to-face visit to undertake the semi-structured interviews was not possible. I sent the final semi-structured interviews topic guide to the participants on the 4th April 2020 and I suggested in this email correspondence that the final interviews could be conducted by video consultation and this was achieved with two specialist nurses and one respiratory physician in July 2020 using MICROSOFT teams

which they all consented to be audio recorded. The completed templates were returned to me by registered post for the quantitative data analysis.

6.15.3.2 Quantitative assessment of the effectiveness of the implementation strategy

Assessment of the effectiveness of the implementation strategy (e.g., uptake of the template by site and by clinician). There were a total of 520 clinical reviews in which the template could have been used across the three Centres, and 219 completed templates were returned for data analysis suggesting that the template was used in 42% of the reviews.

This proportion varied between Centres:
The City Centre returned 58 templates completed by three nurse specialists; 26% of 224 CPAP reviews. The Urban / Rural Centre returned 12 templates completed by one specialist nurse; 13% of the 96 CPAP reviews. The Rural Centre returned 137 templates completed by specialist nurses plus 12 competed jointly with a respiratory physician; 75% of 200 CPAP reviews.

Where recorded there were 169 face-to-face reviews, 30 were video consultation reviews and 20 were telephone reviews (Figure 10). One Urban / Rural and one Rural Centre were already using teleconsultation CPAP review before participating in the study using NHS Near Me.

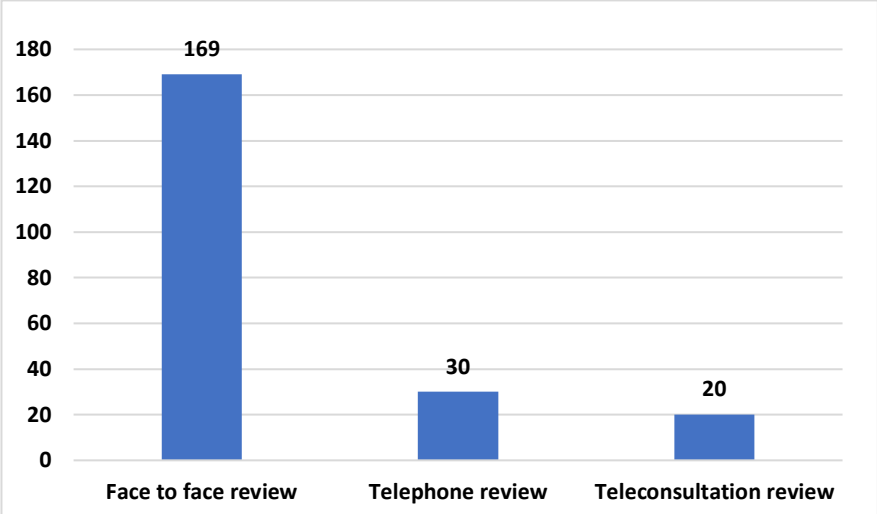


Figure 10 Mode of review

6.15.3.3 The feasibility of using the template (e.g. time taken to complete)

The average time taken to complete the template when this was done during the review with the patient present was fourteen minutes (132 respondents) and the average time to complete the template after the patient left the review was four minutes (123 respondents), (Figure 11). Forty out of 219 (18%) participants responded that it increased the overall review time. Where recorded the average time to complete the template was 14 minutes during the review and an average time of 4 minutes to complete after the review.

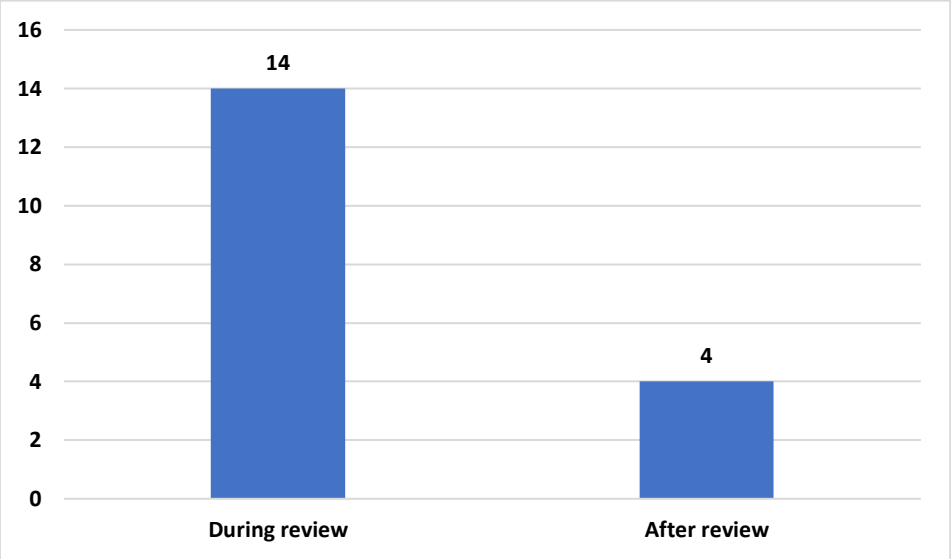


Figure 11 Average time in minutes taken to complete the template

6.15.3.4 The use of the template (fields completed; additional fields required)

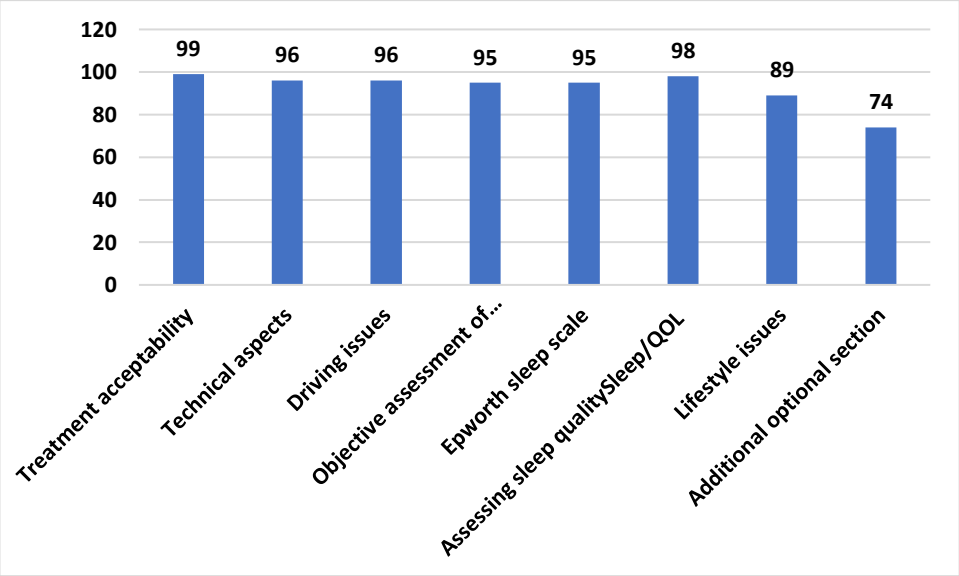


Figure 12 Percentage of fields completed in the template

Of the 219 templates completed 154 (70%) were male, the average age of patients was 60 years (SD 12.6). Adherence was recorded in 204 / 219 (93%) participants,14 of which had no recorded adherence, 28 had < four hours per night adherence and 168 had > four hours per night adherence. Nine patients had OSAHS but were not using CPAP therapy.

The fields completed on the template were; treatment acceptability 216 / 219 (99%), technical aspects 210 / 219 (96%), subjective assessment of sleepiness 211 / 219 (96%), ESS 209 / 219 (95%), symptom resolution 170 / 219 (78%), AHI 70 / 219 (32%), sleep quality and quality of life in 214 / 219 (98%). Of the 210 using CPAP, mask leak issues were recorded in 69 / 210 templates (33%), The lifestyle section of the template was completed in 194 / 219 (89%) with the additional/optional fields completed in 162 (74%). Driving issues were completed in 213 / 219 (97%). Weight, height, BP, and oxygen saturation were recorded in 138 (63%), 96 (44%) ,123 (63%) and 35 (18%) respectively.

Several of the specialist nurses reported that it was not possible to get adherence and efficacy data from older CPAP machines via telephone / teleconsultation review. The section asking the clinician to indicate whether or not the template was useful was completed for 181 (83%) consultations. On 148 (68%) occasions the clinician regarded the template as ‘useful’ and seven responded ‘not sure’ where this was completed (with two responding that it was their first time using it and they were unfamiliar with it). Only 12 (0.05%) indicated that the template was not useful and eight of these were in NIV reviews. Figure 12 represents the percentage of fields completed in the template. There were comments about adaptation to the template in the free text comments which will also be addressed in the qualitative data analysis (Table 18). The additional optional fields were considered less useful (Figure 8).

Free text comments on the templates
A separate NIV template would be preferred
We would adapt the template to record diagnosis, current settings, device, interface
The plan would probably be to have a separate template for the NIV clinic. Not everything is applicable for an NIV clinic
Finding navigating document is getting easier throughout the clinic
First attempt at using the template for review
Took longer than normal at the beginning but this is to be expected
In the clinic, we retained the data card for download and then posted it back to the patient
Finding the template really useful in sleep clinics
Finding that getting easier throughout clinic
First attempt at using template review took longer than normal, but this will be expected.
We are unable to obtain apnoea-hypopnea index and mask fit from some of our older device in the clinic so have retained data card for download and then to be posted back to the patient afterwards
Unfortunately, the patient had not been using correctly throughout and there was no data, and she has not been receiving any therapy
Box for patient further appointment or discharged for open access review would be helpful
Possible outcome should also be included as a box
First time use so unfamiliar with the template.
Reminded me to start patient on humidification.
Layout of questions would be helpful as my personal preference only.
Straightforward appointment with patient who has previously struggled, but is now managing well and moved open discharge
More used to using template now but completing this did take extra time as I have to put information now on the track system as well may
Be useful for patients in outpatient department & this can be guided by the template rather than text
You do not need to put on track in the outpatient department
Layout of questions could be different from my personal preference
Diagnosis box on the sheet would be helpful
We are used to completing the template now and only completing the relevant
Questions patient on long-standing CPAP with previous good compliance
Free text boxes could be bigger
First time I have used template – took longer
For an NIV review – I do not feel that this template is suitable
I do not feel that this template works for an NIV review
Need to add free text into form from NIV machine tidal volume, minute ventilation, R/R, AHI
Sort of worked – Longer clinic time needed
Getting used to template, only using questions relevant to patient and self
Only completed what needed for individual
Older CPAP machines do not provide data on AHI

Table 18 Free text comments on the template

6.15.4 Qualitative exploration of clinical perception of template use

Nine participants (five specialist nurses, one service lead and three respiratory physicians), provided semi-structured interviews. Two interviews were conducted face-to-face, one via telephone, and the other six used Microsoft TEAMS with their consent gained before recording the interviews. Table 19 outlines the characteristics of the participants.

	Profession	Male / Female	Duration in post
Centre 1	Specialist Nurse	Female	>10 years
	Specialist Nurse	Female	>10 years
	Physiologist	Female	>10 years
Centre 2	Physician	Male	>10 years
	Physician	Male	>10 years
	Specialist Nurse	Female	>10 years
Centre 3	Physician	Female	>10 years
	Specialist Nurse	Female	>2 years
	Specialist Nurse	Female	>2 years

Table 19 Characteristics of participants in recorded interviews

A Summary of emerging themes from the thematic analysis of qualitative data is outlined in Table 20.

Themes		Sub Themes
1	Use of a structured template in the clinical review of CPAP users	Structure / content of the template and adaptation evidence for use of a structured sleep medicine template template value, worthwhile, helpful, useful
2	Impact of the template on clinical autonomy, patient-professional relationships, patient centred care	Impact on clinical autonomy impact on patient-professional relationship impact on person-centered care
3	Decision to participate	Organisational or team led / pressure to participate uptake of the template support and instruction continue to use the template in the future
4	Impact on any differences in way the consultation was conducted as a result of using the template /impact in efficacy of review	Impact on different ways of working, standardisation / structured review / equity / reducing variation/consistency impact of template on the time of consultation
5	Impact of the template on service evaluation / service improvement / service structure	
6	Impact of the template on the service - multidisciplinary team working / team knowledge and skills / education and training	

Table 20 Themes and sub-themes form the qualitative data analysis

6.15.4.1 Theme 1 - Use of a structured template in the clinical review of CPAP users

Six specialist nurses used the template in their clinical reviews and one site was already using their clinic review template. As the CPAP reviews were all nurse-led services in the Centres for the majority of cases, the respiratory physicians that were interviewed did not use the template in their reviews which were generally more complex problems as opposed to routine reviews. The physicians were aware of template use as they were available to the specialist nurses for additional support and more complex case discussion if required. There was a comment from one Respiratory physician that using a template guided approach where there were several nursing staff conducting reviews could facilitate a more structured way of doing things and may be a more reliable way of capturing the necessary information. There was variation in the way that CPAP reviews were conducted in the three Centres however, there was general agreement that structured templates were a tool that can assist clinical review.

RNS 3 - *"We have our own template which is pretty similar in the content to yours".*

RNS 5 - *" We used to have our own template that we followed but it wasn't nearly as in-depth as your one. For instance, we didn't ask questions about sleep hygiene. We always ask them do they sleep well but not specifically 'What time do you go to bed?' and 'What time do you get up?'. ……. we found we out more about the patient and more out of the clinic review".*

RP 2 - *"I think the template is useful on the basis that certainly in our service, most of our patient interactions and clinical service, is actually delivered by nursing staff and a variety of them so having a more structured way of doing things that means that it will be a more reliable way of capturing the necessary information".*

RP 3 - *"So, I didn't directly use it with the patients. I used it for gathering information to then give the consultant input. So, the people who were the*

direct users in our service were our nursing staff".

Structure / content of the template and adaptation

The feedback in the interviews and the free text comments on the returned templates offered some useful comments about the structure and content of the template and how it could be refined. A common comment was that separate CPAP and NIV templates were needed as the review of CPAP and NIV users contains different components. One Centre said they did not find the template useful for NIV reviews and they did not use it for this purpose, which supports the case for having two separate templates. The comment about having a component on the NIV template regarding discussing anticipatory care planning is a very important point as often these patients have considerable comorbidities and are much more at risk of admission to hospital when a clear summary of this information would be useful.

Additional space for free text comments and including the diagnosis, outcomes from the review and a follow-up plan, and adding partners' comments were also raised by the specialist nurses as a means of improving the template and making it more user-friendly. There were some comments about some of the content (e.g. chest auscultation was not carried out by nurses or ear nose and throat examination), being less relevant to a nurse-led CPAP review and that these could be removed and also a comment about including a section to record the device used, settings and mask interphase.

There was feedback that some of the older devices do not prove information on the AHI and mask leak as they do not have the inbuilt capacity for remote monitoring that newer devices can do in real-time, via a web-based platform. An important point made by two of the respiratory physicians was that an electronic template embedded in the patient's electronic health record could replace a dictated letter, thus improving efficiency and reducing administrative processes.

RNS 1 - *"We would adapt the template to record diagnosis, current settings, device, and mask interface and include a place to record the outcome and I have written comments on the completed template too".*

SL 1 - *"And in terms of adaptations, I know that staff we're talking about having somewhere to put partners, comments as well……."*

RNS – *3 "We didn't find it so useful for NIV patients, basically because we asked different questions to them".*

RNS 4 -*"The plan would probably be to have a separate template for NIV clinic. Not everything is applicable on the template for an NIV clinic".*

RNS 5 -*"Data capture. So, it asked for the AHI and some of our machines do not record this,…So, we just wanted to have space to see what kind of machine they had, and what type of mask they use and the pressure settings too".*

RP 3 - *"I think we would have two separate ones because the nurses found it very useful as it existed for their CPAP reviews and didn't describe that they changed their structure of doing their clinic".*

RP 3 - *" Oh, I'm going to just go back to the additional component question. Now, this is less relevant to CPAP review but the one thing we feel that we should be doing in an NIV clinic with the more complex patients is making sure we do anticipatory care planning, so, I think from what I remember, that there wasn't a box for that……..".*

Evidence for use of a structured sleep medicine template

None of the clinicians reported any awareness or knowledge of any published evidence on the use of a structured clinical template in a CPAP review, apart from the fact that one of the interviewees was aware of my previous work in preparation for the study. Three of the participants were members of the e-Delphi panel.

SL 1 - *"I don't think I would be able to specifically name any published templates that are used within Sleep services. I do know that there's a lot going on at the moment about minimising variation and improving patient outcomes*

and particularly with the getting it right first-time proposal that's being initiated.."
RP 1- *"No, I've not seen any evidence and I've not really looked for any".*
RP 2 -*" Well, only really the stuff that you have sent me for the purpose of your research……".*

Template value, worthwhile, helpful, useful

Regarding the theme of whether the template was of value, useful, helpful, worthwhile, there was general agreement among the specialist nurses, service lead, and the respiratory physicians that the template was beneficial in conducting a CPAP review as a way of structuring and also standardising the consultation, enabling a consistent and reliable approach to capture the necessary information. Highlighting diversity in practice and styles of clinical review, one respiratory physician thought that more senior, experienced staff didn't need a template to guide their consultation, whilst another considered that structured templates were very useful. It was considered that junior clinicians and new staff might learn from the process of using a structured template for the review. Also, it was felt that it could be used as a prompt to ensure that key elements relevant to a CPAP review were not missed, such as driving status and type of driving licence held which is of particular importance for vocational drivers.

RNS 2 -*" Finding the template really useful in sleep clinics".*
RNS 5 -*" As well as asking about you've got your witnessed snorting, apnoeas and choking spells being controlled, I think just being able to jog your memory to ask all these questions that are specific to the review was really good as well. So, yeah, I found it brilliant".*
SL 1 -*" I do think that it does hold a lot of value within any service and not just within sleep medicine…… It's something that I would see being of a lot of value within the service here and some of the reasons that I've taken a note of is that everything is centralised within one document".*
RP 1 - *"I would say some kind of proforma for a large team who [are] all trying to do the same thing I think is helpful and indeed to try and bring some*

consistency to the approach in safety and all the rest of it……”.

RP 1-*“ I think obviously the more senior people probably don't need a template. But as I said earlier, it's helpful just to probably make it more efficient for them actually. But for junior folk or new people coming into the service, I think it's useful to have these structured templates so you can learn more quickly what's important and what's not. At the same time, still, deliver the same care for the patients that you're seeing”.*

6.15.4.2 Theme 2 - Impact of the template on clinical autonomy, patient-professional relationships, patient centred care

Impact on clinical autonomy

All the interviewees responded that the template did or would not impact negatively on their clinical autonomy or their consultations, and there was an appreciation that it promoted a more structured and thorough consultation. It was described as a tool to assist with the consultation process by one respiratory physician and that *'it does not do the job for you'*. He also commented that it was important to focus on the patient and not the template in a clinical review to maintain that patient-clinician relationship and focus on delivering person-centred care. One respiratory physician considered that it probably impacted positively in terms of guiding the review and there was also feedback from two of the RNS's that they felt it improved their clinical reviews. However, one respiratory physician was concerned about a potential negative impact of the template on clinician autonomy though he did not think that it would personally affect him. Only one of the respiratory physicians used the template in 12 joint consultations therefore it's not possible to explore in any detail whether clinical autonomy of the medical staff would have been affected by the use of a template.

RNS 2 -*" It did not change the way I did the review but the good thing about it was that going through it and having a chat with the patient it highlighted what we had covered in the consultation …….. so it made sure it was a bit more of a thorough consultation …."*.

RNS 3 -*"No I didn't think it affected my autonomy or the consultation by using the template and I use a very similar template with similar content. So no, I don't think it changed the way I conducted my consultation"*.

RNS 4 -*"I think it improved the review and we loved this template and I think it's made our consultations better. We thought we were doing pretty good anyway, but it definitely was more thorough and I think it improved our relationship with the patient and the way they spoke to us as well"*.

RNS 5 - *“I think I would say that it probably improved as well. And I don't think it affected my autonomy. I think it just probably made things a bit more thorough, actually”.*

RP 1 -*“As a clinician, it shouldn't really impact too much. I mean, you're just using it as a tool to help you get through your work. It's not it's not doing the job for you. And you still have to understand why you're asking the questions…… So, I don't think it really impacts clinical autonomy negatively anyway”.*

RP 3 -*“So, my clinical autonomy, I don’t feel that it takes away from that because you can still work around a template. It doesn't alter your patient-professional relationship…… And we've discussed already about the patient centred care is maybe just to bring in that question about what matters to them in the consultation”.*

Impact on patient /professional relationship

Regarding the theme of the impact of the template on the patient-professional relationship, there was general agreement that the use of the template did not impact negatively on the patient-professional relationship, and that it may provide some reassurance for the patient that the review was more inclusive and thorough. However, there was a perception by one respiratory physician that the use of the template could have a negative impact on the relationship if it was used as ‘a tick box’ exercise and prevented a focus on the patient sitting in front of them. Again as only one respiratory physician used the template in 12 joint consultations it’s not possible to explore in any detail whether the patient / professional relationship in the medical staff would have been affected by the use of the template.

RNS 5 -*“A positive impact, definitely a positive. ……...and I think they probably felt a bit more included”.*

SL 1 -*“I would like to think that the patients would see that everything is being covered and see that as a positive. And again, it comes back to making sure that things are structured, not being missed, and making sure that the*

consultation is as full as it could be… … And I think that would probably reassure the patients more as well that everything was being conducted as fully as possible".

RP 1 - *"I suppose it depends on how you use it. I mean, if you just have it in front of you and just look at the form and tick a box, then the relationship might break down. But you still get the same information from people having a long conversation, which I think it depends on the individual for that one".*

RP 3 -*"It doesn't alter your patient-professional relationship. And we've discussed already about the patient centred care is maybe just to bring in that question about what matters to them in the consultation"*

Impact on person-centered care

Most participants described a positive impact of the template on patient-centered care, particularly the specialist nurses with one commenting that it supported a holistic review. One of the physicians highlighted the importance of adapting the consultation to what matters to the patient and not focusing entirely on the template. The opening question of the template about the patient's agenda and what they wanted to discuss was seen as a positive way to begin the review, and some of the RNS's felt that it improved the review for both parties. As with the previous two subthemes as only one respiratory physician used the template in 12 joint consultations, it's not possible to explore in any detail how template use by medical staff would have impacted person-centered care.

RNS 3 -*" addressing what is important to the patient in practice is central to my consultations, I feel the template supports a holistic review ".*

RNS 4 -*" certainly the very first question, actually, and when you know what the patient wants to discuss and how do they have any concerns. I thought that was a great way to start in consultation…… We're starting off our consultations with is there anything you want to discuss first or do you have any concerns with your treatment which we wouldn't have done previously".*

RNS 5 -*" I think I say would that the review probably improved as well. And I don't think it affected person-centered care. I think it just probably made things a bit more thorough, actually.*

LS 1 -*" Absolutely a positive. And as discussed earlier on, I'm quite in favour of minimising variation in patient outcomes and offering equity in all standards of patient care. So it's definitely a positive for me".*

RP 1 -*" I suppose it's as the template is asking the right questions and puts the patient at the centre of the consultation ……",*

RP 2 -*" I mean, I think it's important to adapt a consultation to suit the patient in front of you, everyone's different and that's a skill that not just doctors but nurses do to run their clinics".*

6.15.4.3 Theme 3 - Decision to participate in the implementation study

Organisation or team led/pressure to participate

The decision to participate was made at organisational level and individuals in the departments were free to decide whether, or not, to use the template in any or all of their consultations. This played out differently in the three Centres. One of the Centres (Rural) had a much greater uptake of the template and this may reflect leadership and championing of the implementation study. All the interviewees responded that they did not feel there was any pressure to participate in the study despite the fact that all the participants were known to me except two of the specialist nurses.

Uptake of the template

There was a comment from the service lead at the City Centre that if they had decided to use this as an alternative to their current documentation process and to use the template as a whole department trial for a period of time rather than having the staff complete the template in addition to their usual continuation sheets that would probably have improved uptake. One of the respiratory physicians at the Remote / Rural Centre commented that, in retrospect, he and his colleague could have been more involved in the study, but that this option had not been clear to them at the initial start-up visit. The Centre with the largest uptake was in the process of beginning to undertake service redesign at the time of the first start-up meeting and they were engaged in wanting to try out new ways of working. This Centre however felt the template was only suitable for CPAP reviews and a separate one for NIV reviews would be preferred. There was a whole team buy-in to participating in the study in this Centre and so there was a readiness from the whole team to explore the potential of the template in their service improvement work.

One Centre used its own template already and this was likely a factor in the lower uptake in this Centre. The low uptake of the template in the respiratory physicians at all three Centres is likely due to multifactorial issues such as,

they do not conduct many CPAP reviews as this was a nurse-led service in all three Centres and the respiratory physicians focused on more complex cases therefore as experienced physicians may not feel that they need to use a structured template to guide their review. If I were undertaking a future study with a refined template, I would take more time to explain the importance of getting multi-disciplinary feedback from the whole team and would place more emphasis on the importance of medical staff participation.

The RNS's who used the template commented that they did so because they were keen to try new ways of working. Other comments from the RNS's were it was not applicable to use the template at the short three-month review however it was used for every other CPAP clinic review.

RNS 4 -*" I think because x is new in the past two years and I've only been in the service maybe three and a half years, so we're still quite new. So, we're quite happy to take part and try new things. But I think when you have been in the job a long time, you're more resistant to change. But we certainly didn't feel pressurised ……."*.

LS 1 - *"So I don't think that there was any pressure for anybody to be involved in the study and those that did wanted to participate did. Those that didn't did not. And that was purely an individual choice. So nothing was ever imposed or forced upon any of the staff members. And I would say that there was probably a little bit of both in terms of it being organizationally led. So encouragement for myself but also a decision within the individuals themselves to want to try this."*

RP 3 -*"So, it was team led and no pressure. You didn't put any pressure on us? It was very much we were keen to embrace the use of templates, particularly because you've done all the work".*

RNS 4 -*" And what x said earlier about the 3-month reviews we would not use it then as it just a quick 10-minute consultation but the patients that are new going on treatment.".*

RNS 4 -*"We just said at the beginning we're going to use it for all of our*

patients, all of our clinics. And then once we had sampled it with the NIV patients we did not use it for that but we used it for all our sleep clinics".

LS 1 -*" I think that the only thing that comes to mind is if we had decided to use this as an alternative to the documentation that we would usually scan into the patient notes. So if we decided to do this as a trial for a period of time, perhaps, and, you know, rather than having staff do this as well as usual continuation sheets and things that they would send for scanning, and I think that that may be one of the things that probably would have improved uptake if we had decided to ask everybody to do this as potentially a pilot".*

RP 3 -*"So, I think the nurses embraced it as soon as we said we would do it, which I think was the beginning of January. I'm fairly sure it was well implemented. I didn't hear any mutterings or anything. I think it was embraced".*

Support and instruction

Regarding the sub-theme of support and instruction, there was general agreement that all the Centres were happy with the level of support and instruction on the use and completion of the template that was provided at the start-up visits and any subsequent visits. Two of the Centres had a start-up visit face-to-face with one Centre using NHS Near Me teleconsultation

RNS 4 -*"We are really lucky in that if there are any problems, we've got each other and the senior sleep Nurse and the consultants and we've got was a huge support……...".*

RNS 5 -"No, we didn't get stressed about it. And the fact that we just kind of thought, well, as you read through all of this, it's quite self-explanatory. And once we just got used to doing it and just kind of flowed……".

S L 1-*" And I don't think that that support was just upfront either, I feel that that was something that was ongoing. And I felt that you regularly touched base to find out how things were going……. And certainly from, you know, support from your end, I would say that that was more than adequate and excellent all the way through".*

RP 3 -*" No, we definitely felt that we knew what we were doing after that first*

meeting, once we decided on the date, which was the crucial thing to embrace it, that we didn't and I certainly didn't need to provide any further support…….".

Continue to use the template in the future

One Centre continued to use the template beyond the study period, and all the Centres have requested to see a refined version of the template with the intention to potentially incorporate these refined templates into service improvement projects and also explore electronic template options in some Centres as well.

RP 3 - *"So, I've waited really for your feedback, before we've done anything with it. But I think I'd be quite interested to know what you find from the study before we implement it. Yes, very much. We'd like to implement it".*

RNS 4 - *"But it's definitely something we would want to continue to use and are still using for our Sleep patients".*

SL1 - *"I think that we would probably like to introduce a template like this within the service, particularly now that we have been forced to conduct remote management of our patients and remote set up of our patients. And I think that it just gives us that good guide to follow during a consultation, particularly over the phone by video link and adopting it as a different way of working essentially".*

6.15.4.4 Theme 4 - Impact on any differences in way the review was conducted as a result of using the template /Impact in efficacy of review

Impact on different ways of working

Comments regarding the impact of the template on ways of working varied but were generally positive. The specialist nurses did not think that it impacted on the way the actual review was conducted, and two from the same Centre felt that it was really helpful in guiding the review. In contrast, one Centre used the template in addition to their usual documentation and all the nurses reported that it extended the consultation time initially, although this was less of an issue once they became familiar with the template. One respiratory physician commented that the use of a template may facilitate a more economical way of working with the large volume of clinical work in sleep medicine review services. Some commented that the use of the template could facilitate a patient-focused review and that it did not negatively impact on usual ways of working.

One respiratory physician highlighted that differences in medical and nursing training might affect how the approach to a review, commenting that nurses may be more guideline and process-oriented as the nursing profession is more process-driven. There was also a comment from the same respiratory physician that in sleep medicine the review process is more formulaic compared to a lot of other conditions and is much easier to define and quantify, and so it probably lends itself much more to the use of a template than other conditions. Additionally, the same individual commented that having a more protocolised or focussed way of conducting clinical reviews with less experienced staff was an important factor. Another respiratory physician commented that particularly with the arrival of the Coronavirus pandemic and the significant impact of COVID 19 on clinical review services there would have to be more remote reviews implemented, and the template may help clinicians to focus on telephone and video clinics.

RNS 1 -*"It did not change the way I did the consultation; I did not do it when I saw the patient at first and I waited and did it afterward. Some of the content was irrelevant and I did not use it as it was not appropriate."*

RNS 2 - *"Once I got a bit more familiar with it I took it in with me and although I would usually ask the patient how are you getting on and then make the time to lead to the chat ……...and I found it quite useful to actually take in with me as I got more familiar with it".*

RNS 3 -*"No I didn't think it affected my autonomy or the consultation by using the template and I use a very similar template with similar content, So no, I don't think it changed the way I conducted my consultation".*

RP 1 -*"I suspect COVID will be changing the way we deliver our service much more, that's going to do a lot more remote stuff. So actually, it may actually help focus the mind if you're doing telephone or video clinics. You don't necessarily get the same, you certainly don't get the same signals when you're on the phone to somebody for a clinic, as you do either on video or face to face. So, it actually might provide a bit more focus than just a rambling conversation, actually".*

RP 2 -*"I am not sure what the answers to that is, I mean, I think there is a difference between doctors and nurses generally in how we approach things. Training is a bit different. Nurses are much more taught to follow guidelines and rules and processes, whereas while we are increasingly having to do that too, but there is a need for doctors to, for the benefit of patients, to have the courage and confidence, sometimes to go outside the box. I think sleep is a bit different because it is so formulaic compared to a lot of other conditions. It is much easier to define, quantify and so it probably lends itself much more to the use of a template approach like this than other conditions does".*

RP 2 - *"It is high volume clinical work and so a template like this might facilitate a more economical way of delivering a service because of the numbers involved and the volume of referrals that we get".*

RP 3 -*"You do miss out questions, because quite often when you're trying to lead the patient, so it is a patient-focused interview. Then you can get side-tracked from following your template……."*

Standardisation / structured review / equity / reducing variation / consistency

Themes regarding the impact of the template on the clinical review and patient care were positive in terms of describing reduced variation in practice, standardising the clinical review process, and providing structure and consistency to the process. Also reducing paperwork and having all the information in one document was seen as beneficial. Having free text options to input information and incorporation of the Epworth sleepiness scale was also helpful with comments about having everything centralised in one document. There were also comments about supporting new staff and inexperienced members of the team to keep them on track and ask the right questions, using a consistent way of recording CPAP treatment outcomes. The respiratory physicians had differing opinions on template use and one commented that it may be more helpful in larger services to support consistency in a review. One physician commented that the free text box at the beginning of the template helped focus on what matters to the patient and it could assist with consistency in the review.

RNS 2 -*"It would be a good idea for us all to be following the same protocol it would be easier……... The structure might be different for different services it might suit our service different to your service".*

RNS 5 -*"I find that with me being quite new to the service then it was just a really nice sort of structure to conduct your review of the person. And how x was saying about finding out more about the quality of peoples sleep and things that we don't necessarily ask so much in-depth normally clinic about it".*

LS 1 -*"And certainly I think that it holds a lot of value in the form of reducing variation. So you're not missing anything out and you know, you are standardising, patient care essentially, and that everybody has the same equity and will help to minimize variation and the outcomes within that".*

LS 1 -*"What I like about the template is just that you know we use an awful lot of paperwork and it's all sent for scanning into the patient notes. And it's nice to have everything condensed there within one standardised template. So*

that's one of the things I like about it. I quite like as well is that there are free text section options and I do like the part with the Epworth score being included within the template as well because usually we do that as a separate piece of paperwork and I like that it's all incorporated within the one document ".

RP 2 -*"If we're using more inexperienced people and a wider group of people will help to have something like this, to keep them on track, to asking the right questions and consistent way of recording outcomes from treatment".*

RP 3 -*"So the free text, gave you the opportunity to feedback in real-time rather than trying to remember at the end when you do a semi-structured review. So, I think that free text box was clear".*

RP 3 - *"And I get to the end of the review and go I didn't ask that because I've been trying to listen and let the patients direct the review. So, I guess that's where the advantage might be that if you can still do or bring in the template at the top what matters to the patient about this consultation ……..".*

Impact of template on the duration of consultation

Regarding the impact of using the template on the consultation duration, there was negative feedback because it extended the duration of the review due to a number of factors. The first was that it was a new way of working for the RNS's and they were unfamiliar with the layout of the template; once they became familiar with the use of the template most of them said it did not increase the time of the review. The second factor that increased the time of the consultation in two participants was that they had to complete their usual paperwork as well as the template, so this did extend the time of the consultation. However, the lead for that service observed that if the template was the only source of documentation then this could save time. The service lead also said that if the template was implemented in the future it would be the sole documentation that the staff members would be using and it wouldn't be seen as something additional. There were also some negative comments regarding the use of the template in NIV users in that one RNS felt it was not suitable for an NIV review suggesting the need to have a separate template for CPAP and NIV reviews. One respiratory physician agreed that the template was not suitable for NIV reviews.

RNS 1 -"*More used to using template now but completing this did take extra time as I have to put information now on the TrakCare system as well".*
RNS 2 -"*We all have different ways of working when there are umpteen of us but the fact that we have to write enough for the secretaries to type a letter and they are not at the moment au fait with working with this, but if in the future if they were happy to come up with a letter from this then you could just sit and do this at the clinic but I found it easier just to do this when we're up here in the ward because you have a longer consultation time to type up your notes so I did add a bit of time but if it was the only documentation that you had it would save you time".*
RNS 3 -*" It took a bit longer initially to complete because of the different way of working in my service…….and we have our own template which is*

structured differently but has similar content".

RNS 3 -*"We do have different templates for if it's a CPAP/NIV and an Airview review. I don't think it changed the way I conducted the consultation and as I got used to it, I was quicker and I don't think it changed my routine practice".*

RNS 4 -*" So the first week it took about fifteen minutes roughly and then as the weeks went on, we were noticing we were writing on the back of the form, the box at the bottom. It was taking us less and less and less time as we got used to it".*

RNS 5 -*"We really got into the format of it so we didn't need to constantly look at the template for your next question. But after a few patients, it was pretty much just taking the same amount of time as before. Yes. It didn't take us any longer than previously. But for NIV it did take a bit longer initially".*

LS 1 -*" I think that the template itself was favoured and that the feedback from the team members was they felt that it was time consuming only because they had to fill out the standard documentation that they usually would in addition to the template as well. ……".*

RP 3 -*"When we used your template, the nurses bring it through and we were filling it in, yours did not extend the review using the template. It certainly did not extend the CPAP reviews and I don't think they felt it did for the NIV. It was just they couldn't use the template in its completeness for an NIV review".*

6.15.4.5 Theme 5 - Impact of the template on service evaluation/ service improvement/service structure

There were some comments that the template could impact positively on service delivery by creating more efficiency and better patient outcomes. The Service lead thought this was of particular importance if the template were the sole documentation to be completed in the review, whether this is face-to-face or by teleconsultation. She also highlighted due to the impact of COVID 19 the requirement to rapidly deploy teleconsultation and remote monitoring solutions to minimise face-to-face contact that the template would be beneficial in this process. It was suggested by two of the respiratory physicians that an

electronic template could be developed that could be integrated directly into the health record, reducing the requirement to dictate formal letters and reduce administrative processes. All the services were severely disrupted by the pandemic and they were all looking at ways to remobilise their services and the template impact use and uptake was a very small factor in service redesign.

RNS 2 -*" I think it would be useful to use the template at the clinic downstairs if you just had the one template. Because we don't dictate the letters, we have to type it all out and that is the only documentation and then it all has to go into a letter which is your only record".*

RNS 2 -*" The first thing I thought was as are moving to more telephone-based consultations that the plan for the future if that patients are going to be initially reviewed over the phone I think this will be invaluable then. It is a bit more difficult to have that rapport over the phone that you have face to face with somebody. You can give them a call and go through the template then it means we will definitely cover everything".*

LS 1 -*" I think it holds a lot of value and I think that at the moment, while services are starting to remobilise post-CoVID pandemic there's a greater requirement to look at the way that we offer services to patients and how these things can be integrated into different ways of working, essentially which will now essentially be remote".*

LS 1 -*" I think that there is a place for a nurse or physiology-led clinics and it's something that we hope to implement at some point within the future. And we've started a framework for that with physiologist clinics of patient results just now. So yes, we will continue to use the template, and that's something that we'll look at implementing here in the future".*

RP 1 -*" I think the key thing for this is to try and get the template integrated into some electronic format, our previous patient templates are all paper, we need to not have as much paper but certainly for these sorts of interactions just to be able to do it electronically".*

RP 3 -*"What we also did with your template was we're hoping that it's going to*

replace potentially doing a clinic letter because we uploaded your clinic template as our record to sky store. So, at the moment, we didn't make that final change to not doing the letter. We still did the letter on top where if you got that right, that we'd hope that we would actually reduce our administrative processes".

6.15.4.6 Theme 6 - Impact of the template on the service - Multidisciplinary team working / Team knowledge and skills / education and training

Based on the responses from the participants there is clear evidence on the value of MDT working for all members of the team whether this is formal or informal meetings. Two of the sites found that formal MDT weekly meetings were a good way to support the whole team providing the opportunity for shared learning, consistent and collaborative working, whole team support, and this governance approach was appreciated by the service lead and the RNS's. With the rapid role out of Microsoft teams and NHS Near Me during the COVID 19 pandemic, this has supported remote consultation for the teams in more remote and rural services, and there were also comments that the template may be useful for education and training purposes of newer members of staff, however more senior specialist nurses may find the template less useful. Regarding this wider theme of service impact and MDT working the refined template may be of use to some services which relate to achieving Objective IV of the study and refining the template.

RNS 3 -*" If I am struggling with a difficult patient the Respiratory physicians will obviously see them on request. The consultants will dictate their letters to go to GP and they do not use the local template at all".*

RNS 4 -*" So, we have a consultant who comes up regularly from another hospital and we do a weekly Teams MDT with him, with all our new patients. And he comes up every six weeks to do clinics and to see people with more*

complex disorders……...”

RNS 5 -*“ And we're really lucky in that if there are any problems, we've got each other and the senior sleep Nurse and the consultant and we've got a huge support……..”.*

SL 1 -*“ Although we take the patient results to the MDT for discussion with the doctor, it's not just for discussion with the doctor, but very much from input from everybody as well. And we regard all staff members to be extremely knowledgeable and value everybody's input. So it is very much a team network ….”.*

SL 1 -*“ The template is something that could certainly be incorporated into training and as part of evidential reviews for training as well, and to make sure that staff are as knowledgeable as you can be using it”.*

RP 2 -*“We did initially have an MDT, and then it is kind of fell by the wayside. So, we do it very informally. So basically, we work in close proximity to where the sleep nurses work and the physiologists work and if there are issues, they come and tap on our door. ……..”.*

RP 3 -*“And also, the other thing that's lovely about NHS Near me is that you can get trainees doing it now. And they can send a link to me so that I come into the review remotely. So, you can have your three-way remote review. And that is working well. ……. It would have been interesting if we had been doing this during CoVID”.*

RP 3 -*“ With your remote monitoring capability sleep medicine could not be better set up to do this. This is just the thing where it's going to make a difference. So, one of my team has even kept going with NIV reviews even though I had to come out of doing clinics for NIV, she's kept going on her own because she's felt able to and just asked me for advice about the difficult ones”.*

6.16 Discussion

6.16.1 Statement of principal findings

All participating sites engaged with developing an implementation strategy and the template was used in 26%, 13%, and 75% of consultations in City, Urban/Rural, and Rural Centres respectively with varying uptake of the templates in two Centres.

The quantitative data fields completed in the returned templates in this real-life implementation study align closely with the consensus results achieved in the international e-Delphi study (10), apart from recording the AHI and mask leak which was considered of lesser importance by the implementation study participants. Free text feedback on the completed templates suggested that clinicians perceived that templates aided a thorough and holistic consultation and that it was helpful / useful and easy to use with familiarisation. It was not possible in some cases to get data from older CPAP machines via telephone / teleconsultation and users suggested that additional free text space for the model of CPAP / NIV device used, tidal volume, minute ventilation, respiratory rate, and AHI should be added to the refined templates.

There were six main themes from the qualitative data (Table 20). The template was regarded as being a useful tool to assist a clinical review, though clinicians suggested that the structure and content of the template could be revised with the addition of fields to include device type and mask interface, diagnosis, follow-up arrangements, and additional noteworthy outcomes of the consultation. The template was not reported to adversely affect the way the clinical review was conducted or perceived clinical autonomy, patient / professional relationships, and patient-centred care.

The decision to participate in the study was organisational and team lead with individuals able to decide for themselves whether to use the template. There were some comments regarding how the uptake of the template could have

been improved with wider departmental and clinician participation, and if there was no requirement (in one Centre) for additional data entry. The uptake of the template was particularly high in one Centre where the timing of the study coincided with local service improvement work, All Centres felt that they were given sufficient support and instruction regarding requirements for the study and one site was continuing to use the template. All the Centres have requested to see the refined template.

Participants were clear that there was no negative impact of the template on clinical autonomy, patient-professional relationships, or patient-centred care. In general, comments suggested that the template standardised ways of working and reduced practice variation. The use of the template initially extended the consultation time, however with familiarisation in its use this was less problematic. In the City Centre where the template was an additional task, this extended the duration of the consultation, though it was noted that this could be resolved by adopting a refined template as the sole means of capturing the content of a CPAP review. As a means of increasing and optimising the time allocated for conducting a review the option to ask the patient to complete some aspects of the template prior to the consultation could be a consideration in any future research. For example completion of the epworth sleep score and the section at the top of the template on what the patient wants to discuss may help with a more patient focussed review making best use to the time allocated for this.

There were comments regarding the positive impact of the template on CPAP reviews by creating more efficiency, particularly if the template was the sole means for completion in the review. An electronic template that could be integrated directly into the clinical portal was suggested and this is already achievable within current electronic recording systems.

The template was considered to support valued MDT working, team knowledge and skills / education and training, and shared learning, whole team support, and governance being appreciated by the service lead and the RNS's. Other comments were with the rapid role out of Microsoft Teams and NHS Near Me during the pandemic, the template supported remote consultation for the teams in more remote and rural services. There were comments that the template may be useful for education and training purposes.

6.16.2 Strengths and limitations

I Followed implementation research framework

I followed an implementation research framework and used the StaRI checklist to ensure that I was following agreed standards for reporting the implementation study findings. I distinguished but also focused on both the evidence-based intervention and the implementation strategy. In combining the quantitative and qualitative data using a framework analysis approach to develop the thematic analysis, and subsequent development of the six main themes and then using the CIFR domains to discuss the findings of the study has allowed an in-depth analysis of the study results.

Robust development of the intervention

I developed the review template based on a search of global guidelines, recommendations, and best practice statements on what should be included in a CPAP review plus the findings from the international e-Delphi study. One of the strengths of this research is that no other publications address the most important components to review in CPAP therapy users in the real-life setting. In combining the findings of the international e-Delphi study with the quantitative and qualitative results from this implementation study, both contribute to an evidence-based approach to the use of a template in supporting a structured clinical review in the CPAP users.

Codesign of the implementation strategy with the Centres

The aim of the initial visit to each Centre was to ‘co-design’ the implementation strategy to explore basic things such as did the Centres want the template in a paper or electronic format; how were they going to ensure that templates were readily available within the clinic setting and if there had been any other strategies that might have helped support the implementation at each Centre.

Mixed methods enabled triangulation

Using mixed methods enabled triangulation whereby I was able to analyse the results of the data using multiple approaches to enhance the credibility of the implementation study findings and compensate for any weaknesses in the study methodology. The quantitative data on the template fields completed resonate with the e-Delphi consensus results regarding what are the most important components to include in a clinical review. By combining the quantitative and qualitative data using a framework analysis approach to develop the thematic analysis and subsequent development of the six main themes, and then using the CIFR domains to discuss the findings of the study has allowed an in-depth analysis of the study results.

Uptake and potential use of a refined template

The uptake of the template in the rural Centre was much more successful than the other sites and this may because this Centre was already planning service redesign and there was a strong leadership focus with the whole MDT being keen to try new ways of working. The lesser uptake in the other two Centres may be due to the fact one Centre already used their own template and in the City Centre, there was a requirement for double data entry with no additional clinical time allocated to facilitate this. The reasons for no medical staff participation in template use may be multifactorial, firstly all the CPAP reviews were nurse-led services, and secondly, I may not have been explicit about the medical team using the template at the developing the implementation strategy stage. One of the physicians interviewed said that experienced senior physicians did not need to use a template in a review and this may be a factor in no medical staff using the template. The fact that the study was conducted over the winter period when pressures are greatest for respiratory teams may also have been a factor in the Centres with lesser uptake. Trying to engage more senior physicians in participation may require a different approach or methods to encourage the use of a refined template.

There is the potential for a refined template or templates to be utilised in increased telephone and teleconsultation reviews in the coming years as telemedicine use is adopted more widely in services during and beyond the pandemic. The ability for services to have oversight of telemonitoring data in CPAP and NIV users is already embedded in routine clinical practice nationally and internationally and combined with the use of a structured clinical review template may support the delivery of safe, person-centered care that is effective, economical and environmentally friendly for future sleep medicine services to adopt.

Data saturation

I aimed to recruit up to 20 clinicians from three Centres, conducting up to 15 semi-structured interviews at the end of the study which would have allowed me to conduct a more robust qualitative thematic framework analysis of the recorded and transcribed interviews. Regarding data saturation, I believe this was achieved in the specialist nurse interviews as no new themes were presented during the final interviews. Typically data saturation often requires a much larger sample size in qualitative studies however this was a group of specialist nurses who were very familiar with this speciality and conducting nurse led reviews and may therefore have represented a much more homogenous group allowing data saturation in a much smaller group. However, I am unable to say that I reached data saturation in the physician interviews as none of them used the template and the interviews were already delayed due to COVID 19 and there was no time to do this in my already delayed PhD.

Limited diversity amongst interviewees

Having a larger group of medical interviewees or other clinicians could have resulted in the planned qualitative analysis methods providing much richer data and more confidence in the outcomes of interest in the qualitative study data.

COVID-19 and delayed interviews

The final six interviews were completed in July 2020 - five months after study completion in February 2020. There is the potential for those that used the template to perhaps not recall their thoughts on the use or the content of the template, however, there was the facility within the template for immediate feedback via the free text comments that I was able to analyse.

Use of a theoretical framework to aid interpretation

By using the CFIR and its five main domains of Outer setting, Inner setting, Intervention, Process, and Individuals involved to explore the six main themes from the interviews this has enabled me to be more organised, consistent, and inclusive in systematically reporting the qualitative findings of the implementation study.

Reflexivity

Section 6.14 covers reflexivity in much more detail however some key points on reflexivity are noteworthy. I knew many of the interviewees as long-standing professional colleagues over a number of years which both opened doors and made discussion easier and can be considered a strength, however, this may also have prevented me from asking some questions that may have identified basic issues with template use in CPAP reviews. A limitation is that I developed the template and had been using it in my clinical practice for some time therefore thus believed in it and this may have resulted in my personal bias being a factor in the interviews.

Developing experience

I was inexperienced in qualitative research at the beginning of the study. However, I have undertaken a three-month qualitative research module with the University of Aberdeen before undertaking this study and have read extensively on qualitative research methodologies as well as seeking and following guidance from my PhD supervisors. Nevertheless being a novice

qualitative researcher may mean I have missed some important themes, particularly in the early interviews when I was still learning the skills of undertaking qualitative interview techniques, however, the later interviews were substantially improved.

6.16.3 Discussion of the CFIR domains in relation to implementation study results

I chose to also use the CIFR domains to assist with the discussion of the results of the implementation study (Figure 9) as the five domains of Outer setting, Inner setting, Individuals involved, Intervention, and Process can all be used to interpret the implementation study findings (261, 268). I considered the CIFR framework and its domains to be a good fit to discuss the qualitative data results and themes and to inform understanding of the various factors that influenced the success (or otherwise), of the implementation strategy.

6.16.3.1 Outer setting

Definition (261, 268): Outer setting includes the features of the external context or environment that might influence implementation. Four constructs are included in the outer setting (e.g., external policy or influences and incentives).

There is a national remobilisation of sleep medicine services improvement work steam underway to assist services in a recovery plan in the midst and beyond this pandemic. As this work-plan progresses any new national service incentives, developments, or initiatives such as the wider use of teleconsultation and telemonitoring services and the development and implementation of this template (electronic or paper-based), may influence and support new ways of working and delivering services.

6.16.3.2. Inner setting

Definition (261, 268): Inner setting, which includes features of the implementing organization that might influence implementation. Twelve constructs are included in inner setting (e.g., implementation climate, leadership engagement).

I recruited three Centres that represented three distinct sleep medicine services, City, Urban /Rural, and Rural to gain an insight into how these different settings may impact on and be able to inform and meet the study objectives. All of the Centres had very well-established sleep medicine services with the lead collaborators in each site all being consultant respiratory physicians. Two of the Centres, City, and Rural were already planning future service redesign when I initially made contact with them pre-study to seek their interest in participation because of their very long waiting times to have a diagnostic test for OSAHS, and then to commence CPAP therapy. This may be a factor in that they were perhaps more receptive to participating in the study. The Urban/Rural Centre was imminently facing recruitment issues due to senior specialist nurse staffing changes and this site had already used their own templates (one for CPAP and one for NIV reviews), which may reflect the lower uptake of the template in this Centre.

All the participating Centres had very experienced staff within their teams, however, all the CPAP reviews were all nurse-led services where physicians did not see these patients unless there was a degree of complexity outside the scope of practice of the specialist nurses requiring physician oversight. Furthermore, a long-established national network of sleep clinicians met on a biannual basis for clinical education and peer support pre-COVID 19 and this was a real advantage in recruiting the sites to participate again allowing all the study objectives to be met. The 18-week target of diagnosis of OSAHS to treatment set by the government was not achievable in any of the Centres with long waiting times pre CoVID 19.

The delay in being able to undertake the qualitative interviews provided an opportunity to explore sustainability. In the remobilisation of all sleep medicine services in the midst of the first and second wave of the pandemic in the participating Centres, there was a clear interest in having sight of the refined template in both a paper and electronic format indicating an interest or readiness for further implementation. The deployment of video consultations (rapid deployment of NHS Near Me), and more use of the remote monitoring capabilities in newer CPAP and NIV devices have been accelerated both locally and supported from the Government healthcare perspective as a response to the pandemic. It is clear from the thematic analysis of the qualitative data that working in a standardised and evidence-based way that reduces variation and supports best evidence-based practice and person-centred care is valued by clinicians and patients alike.

6.16.3.3 Characteristics of Individuals Involved

Definition (261, 268): Characteristics of individuals involved in implementation that might influence implementation. Five constructs are related to characteristics of individuals (e.g., knowledge and beliefs about the intervention).

The three participating Centres all has specialist nurse-led CPAP review services, and the respiratory physicians did not routinely conduct CPAP reviews but focused on reviewing the more complex patient who were using NIV. In the Rural Centre with the highest uptake of templates expectation from the lead clinician may have been a factor in achieving this greater uptake, however, participation in the study was seen as a very positive experience by all the participants and reflected in the themes, sub-themes and the free text comments in Table 18. In the Urban Centre, there were a larger number of staff who initially indicated their interest in using the template however, the fact that completing the template was in addition to their usual work may have been

a negative factor in the lower participation at this site where extra work was being asked of them, and this may explain the lower uptake at this site.

The individuals involved and their personal attributes contributed to the implementation strategy achieving all of its objectives. There was clear and transparent evidence of joined-up MDT working in all the participating Centres. This is an important factor as MDT working is widespread across the NHS and a team atmosphere that facilitates whole team involvement, with task allocation can support improvements in healthcare delivery and is a key factor to ensuring team decisions are implemented (283). This was evident in all the Centre visits I conducted (there were good team interactions with myself and all the team members expressing their views regarding participation in the study), during the implementation phase of the study (Objective II), and also in the participant interviews about the impact of the template on the service. Multidisciplinary team working / team knowledge and skills / education and training and their interest in seeing and potentially implementing a refined template post-study (Objective II and IV).

Leadership is an important factor in the adoption and uptake of evidence-based healthcare improvement initiatives, and the NHS invests significant time and resources in leadership development for health professionals. Good leadership is central to team effectiveness, from a clinical and financial performance perspective, and for implementing and improving innovative healthcare solutions (276, 277). Service redesign initiatives were already underway in all the Centres and the teams were all in a state of readiness and open to new ways of working. The negative impact of the template in terms of increasing the consultation time initially in all the Centres was more problematic at the City Centre because of a requirement to maintain the ‘old’ way of working. This could be mitigated by adopting the refined template in an electronic format as the sole means of recording the CPAP review content, thus reducing paperwork and supporting the Scottish Governments TEC agenda of moving to paperless systems. It was reassuring that the participants

who used the template did not feel that it impacted their clinical autonomy or on the patient / professional relationship in any significant way and used it as a tool to guide a structured and thorough consultation. The Centre that had the highest uptake of the template reported that the template had a positive impact on their review process, and they felt that their reviews were of a better standard for both themselves and the patients as a result of using the template.

6.16.3.4 Intervention

Definition (261, 268): Intervention characteristics, which are the features of an intervention that might influence implementation. Eight constructs are included in intervention characteristics (e.g., stakeholders' perceptions about the relative advantage of implementing the intervention, complexity).

Although widely used in other contexts, the literature review did not reveal any evidence of a specific structured sleep medicine review template, and the participants were not aware of any evidence to support this format of conducting a CPAP review. However, two Centres used their own template or had done so previously and many other services likely use their own template to guide such a review, although this has not been translated into any formally published evidence. The comments regarding the structure and content of the template will facilitate the adaptation of the template for further evaluation or trialability. Also supported by the participants responses regarding whether the template was of value, useful, helpful, worthwhile, there was general agreement among the specialist nurses, service lead, and the respiratory physicians that the template did not add any additional complexity to a review. It was viewed as beneficial in conducting a CPAP review as a way of structuring and also standardising the review using a more consistent and reliable approach to capture the necessary information. This is also the case for other conditions such as asthma where templates can facilitate a patient-centred but also structured review that focuses on self-management strategies

and also addresses the patient agenda (228, 229).

There was a comment that more senior staff may not need to use a template in their CPAP consultation which may be reasonable in relation to the clinical autonomy and variation in clinicians' consultation styles. However more junior staff and new staff may learn from the process of using a structured template for the review, and larger teams may benefit from this approach in terms of consistency, reducing variation, and supporting safer care (228, 237). Also, it was felt that the template could be used as a prompt to ensure that key elements relevant to a CPAP review were not missed such as driving status and the type of driving licence held which is of particular importance for vocational drivers.

Regarding the impact on clinical autonomy, all of the interviewees were clear that the template did not impact negatively on their clinical autonomy or their consultations and there was positive feedback that it assisted a more structured and thorough consultation and it was described as a tool to assist with the consultation process. For many of the interviewees, it impacted positively in terms of guiding the consultation, and there was also feedback from two of the nurse specialists that they felt it improved their clinical reviews.

The only costs related to the intervention was in terms of time to conduct the study with no further developmental costs to the NHS as an electronic version of the refined template could easily be integrated into the electronic healthcare records, and the technology is already in place at NHS boards to support this process should any teams wish to adopt the template in the future.

6.16.3.5 Implementation process

Implementation process, which includes strategies or tactics that might influence implementation. Eight constructs are related to the implementation process (e.g., engaging appropriate individuals in the implementation and use of the intervention, reflecting, and evaluating).

Objective I - I aimed to involve the Centres in developing the implementation strategy. I was able to work with each Centre at the initial visit and any subsequent visits to decide on the best strategy to support the implementation of the template within their teams.

Regarding Objective II - Implementation, I conducted two semi-structured interviews; face-to-face with two specialist nurses pre COVID 19, one telephone interview with a specialist nurse, and six video teleconsultations using Microsoft TEAMS with one service lead, two specialist nurses, and three respiratory physicians. Additionally, one initial recorded group discussion with two specialist nurses, and the free-text comments entered on the completed templates were also included in the coding of the data to develop the themes outlined in section 6.15.4. I had originally aimed to conduct between 10-20 semi-structured interviews across all the sites however this was not possible despite extending the recruitment time for the study to the end of February 2020 to engage more participants. The main factor in not achieving more interviews was that in the three Centres all CPAP reviews were nurse-led services and then the additional impact of the appearance of SARS-CoV-2 virus in the UK in early 2020 meant I could not extend the study period further.

Following discussion with my co-authors, we agreed to attempt to analyse the very limited qualitative data I had; however, it was apparent that the qualitative data did not meet the aims and objectives of the study. Following a further discussion, I contacted the participating Centres to ask if any of the clinicians would agree to an audio-recorded interview in July 2020 as the COVID 19

hospitalised affected individual cases were low at this point. Using secure Microsoft teams I was able to conduct a further six semi-structured interviews that generated greater qualitative data for the thematic analysis. I used NVIVO software to undertake the thematic analysis and generated six main themes described in the results section 6.15.4 and Table 20. I used the StaRi checklist recommendations for implementation studies to measure what standards I have met with the results achieved (Appendix 12). The earlier section 6.13, regarding the need for reflexivity, outlines how I tried to be as reflexive as possible at all stages in the study and the evaluation of the study results.

The highest uptake of the templates was in the Rural Centre with 149 / 200 (75%) completed templates returned for analysis. Several factors are likely to have facilitated this higher uptake at this site. The first being the fact that the principal collaborator at this site was the Lead respiratory physician in the service, and the second being they also have a research interest. Thirdly, this service was currently in the process of looking at service redesign and they indicated at the first virtual site visit that they were open to trying out new ways of working. Another factor in this higher uptake of the template is that the two specialist nurses at this site were relatively new to the speciality and were keen to try out new ways of working. The City was the largest Centre and returned 58 / 224 (26%) completed templates for analysis by three specialist nurses. They had to complete the templates in addition to their usual documentation practice and this was a negative factor that impacted on increasing the time of their consultations. None of the respiratory physicians in the City Centre participated in the completion of the template, however, there was interest in participation. Several factors may have impacted this. CPAP reviews were nurse-led and the respiratory physicians reviewed only the more complex patients. The second factor was that the study was conducted over the winter and respiratory services face many conflicting winter pressures that may have impacted their adopting new practices.

This highlights the importance of engagement with a lead collaborator at each Centre and the need for a local champion to be closely engaged in the study which was the case in the Rural Centre. Recent publications have consistently found that the role of champions is an important influence in how effective implementation can be achieved and their role in supporting, promoting, influencing, and motivating teams through implementation by overcoming possible resistance that the intervention may provoke in a healthcare setting (276, 277). The Urban / City Centre returned 12 completed templates for analysis. They had their own template in place already and this may have impacted the number of templates returned from this Centre.

6.17 Interpretation of findings in relation to previously published work

Contents of a CPAP review

When compared to national and international guidelines published between 2003 - 2016 (6-9, 24, 67, 70, 76, 78, 209, 221, 224), the e-Delphi did identify additional components that could be included in a CPAP review that were not always included in these guidelines. For example, specifically asking if any problems with sleepiness while driving, measurement of the ESS, weight, and blood pressure monitoring, checking for any mask interphase issues which were strongly prioritised (>90%), in the e-Delphi study but were not always recommended by the guidelines. Asking about quality of life, and quantity and quality of sleep, asking about work schedule / shift pattern working, preparedness to continue with CPAP therapy, and the requirement for humidification was all prioritised as >75%, but were not specifically included in these guidelines. Also, these guidelines do not mention checking for resolution of the Apnoea / hypopnoea index despite it being given a priority consensus of 90%. Only one guideline published by the AASM in 2019 mentions 9 of the 12 possible review components identified in previous published guidelines up to

2016. The findings from the e-Delphi study regarding the core components of a CPAP review may inform future guidelines to perhaps focus more on a patient-centred clinical review that includes the prioritised core components identified in the e-Delphi study and what matters to the patient.

DVLA and driving regulations

The e - Delphi identified the requirement to focus on asking about sleepiness when driving as an extremely important component at a clinical review (proritised at 95%), for patients with OSAHS. This is supported by the DVLA website advice on excessive sleepiness and driving, the BTS position statement on driving in OSAHS 2018, and European task force findings 2020 (45, 90). The importance of discussing driving-related issues was also prioritised as a core component of the review in the implementation study where this field was completed in 98% of the 219 returned templates. In combing the results of the e-Delphi and the Implementation study giving driving issues a high priority (95% and 98% respectively), then future guidelines may wish to take this into account and provide more clarity for clinicians conducting CPAP reviews. The ERS Statement on Sleep Apnoea, Sleepiness and Driving Risk published in 2020 does provide supporting statements to assist clinicians in assessing fitness to drive (45).

Role of templates

The use of templates within both primary and secondary care to support structured review is not a new concept and there are pros and cons in their use (233, 234). The cons are that template-based reviews may override the patient agenda with the risk of becoming a tick box exercise that does not necessarily support a patient-centred review about what matters to them and may inhibit communication (237). The pros of using templates are the review can be more structured for the healthcare clinician conducting the review, supporting more consistency and reducing variation in practice whilst also focusing on disease-specific guideline-based care (233, 235). In taking a balanced approach to using a structure CPAP template to conduct a clinical

review that includes an opening question regarding what the patient wants to discuss in the review can support both a patient-centred review that also includes the most important components to include in the review based on the e-Delphi consensus study finding and current guidelines. The specialist nurses that used the template in the study agreed that it did support a structured review, that did not affect patient-centred care or their clinical autonomy. Other Centres may use their own templates for CPAP review; however, this is the first CPAP review template to be developed using an international e-Delphi consensus to inform the review content with the clinicians in the implementation study giving further feedback to allow refinement of the template.

Who can do CPAP reviews

A number of respiratory clinicians can do CPAP reviews however this is becoming a primary nurse specialist-led, physiology led or clinical scientist-led service as the respiratory physicians tend to see the more complex cases who may be using NIV and require more medical oversite. There is evidence to support that review outcomes are similar for consultant and nurse-led reviews, and a simplified nurse-led model of care has demonstrated non-inferior results to physician-directed care in the management of symptomatic moderate-severe OSAHS while being less costly (175, 212). This was certainly the case in the three Centres in the implementation study. The increased prevalence of OSAHS means more people are using CPAP and sleep medicine services need to find innovative ways to continue to support routine reviews. The data from the implementation study suggest that a template enables nurses and other professionals to complete uncomplicated CPAP reviews.

Use of templates and training for health care professionals

The use of templates to support an evidence-based review that also supports training and education for new specialist nurses and more junior staff is an important consideration for healthcare services (269). Electronic medical records that use standardised templates for clinical reviews are replacing paper-based systems globally and may offer clinical educators and healthcare trainees a platform to support effective healthcare education (269, 270). Implementation of standardised guideline-based structured templates during the training of junior medical staff and other MDT members are likely to improve compliance to guidelines and promote and protect patient safety and person-centred care (269). The refined structured CPAP review template will require further evaluation at a wider service level to determine if it supports training and education in the delivery of sleep medicine services.

6.18 Summary of Chapter 6

This chapter describes the results of a mixed-methods implementation study of a structured sleep clinic review template in three diverse clinical settings in the UK. The quantitative data from the completed templates indicates that the content and structure of the template and fields completed match well with the findings of the e-Delphi study. The qualitative research interviews facilitated the development of six main themes which allowed a more in-depth understanding of the participants views of the use and potential future use of the template in this speciality. In applying a mixed-methods approach to this implementation study I was able to achieve the study objectives.

Feedback regarding the template is generally positive, however, its structure, content, and layout need to be refined utilising the feedback and comments from the participating clinicians. This study is the first of its kind to evaluate the use of such a template in the real-life clinical setting. The use of structured templates may reduce variation and standardise practice in nurse-led sleep clinic reviews and there should be further studies conducted with a revised template (electronic), based on the results of this study. Within the current context of the global COVID 19 pandemic, the use of a structured sleep clinic review template may assist future service provision and reduce variation in practice. With standardisation of practice, more telephone and teleconsultation will become the norm to reduce face-to-face reviews and reduce the risk of viral spread in the current setting and for the foreseeable future.

Chapter 7: Contributions to clinical practice and implications of PhD findings

This PhD has focused on three distinct but interlinked pieces of research:

1) A systematic review that investigated the role of telemedicine and teleconsultation in people with OSAHS (168).

2) An international e-Delphi Consensus study to determine the most important components to include in a CPAP review (10).

3) A mixed methods Implementation study of a structured clinical review template in CPAP therapy reviews in the real-life clinical setting.

7.1 Systematic Review

The systematic review in chapter four of this thesis concluded that by combining both telemonitoring and teleconsultation no safety concerns were raised in the five studies that met the inclusion criteria, and that adequately powered, well-designed trials were required to establish whether the combination of real-time telemonitoring and remote teleconsultation is a clinically viable and cost-effective option for CPAP therapy users. Since this review was published in 2017, a number of other studies have been published that negated the need to conduct a further RCT combining these interventions. In 2020 a metanalysis by Chen et al of telemonitored care versus usual care demonstrated significantly greater adherence with CPAP therapy when telemonitoring was used (271).

Teleconsultation and telemonitoring solutions in the delivery of sleep medicine reviews have existed for two decades. They have been implemented at pace in the last five years with the advent of more advanced technology, new legislation, and clinical governance to support this model of clinical care (4, The emergence and ongoing global COVID 19 pandemic has forced sleep medicine providers to deliver more teleconsultation clinics and telephone clinics for CPAP therapy users to avoid face-to-face consultation where possible for patient safety (272). This was certainly a factor reported by several

of the participants in the implementation study and the need to sustain the use of remote consultations (Chapter 6). An article published in the BMJ by Greenhalgh et al 2020, has outlined how telephone and teleconsultation can be conducted during COVID 19 (273). The same principles can equally be applied to teleconsultation combined with remote monitoring of CPAP therapy users. There would now appear to be a race for services globally to implement virtual care solutions at pace in preparation for subsequent waves of COVID 19 and beyond to maintain safe, effective, and person-centred services to people using CPAP therapy (274).

On completion of the systematic review in the light of the further published studies supporting the use of telemonitoring and teleconsultation, I decided to focus on defining the most important components to include in a CPAP clinical review (whether face-to-face or remotely).

7.2 The International e - Delphi study

The international e - Delphi study (Chapter 5) was completed by 40 participants who were practicing clinicians (physicians / academics, specialist nurses, physiologists, and service users). This is the first study to seek consensus from an expert panel on what components are considered priorities and the most important to include in the review of CPAP users and how often this should occur. The 17 prioritised consensus components generated from the e-Delphi study were used to develop a prototype structured CPAP review template that was utilised in the final mixed methods implementation study (Chapter 6), in three diverse sleep medicine Centres. There was a clear gap in the published evidence base for the benefits or disadvantages of introducing a structured CPAP review template, and the final implementation study in this PhD aimed to explore this in three diverse UK sleep medicine Centres.

7.3 A mixed methods Implementation study of a structured clinical review template in CPAP therapy reviews in the real-life clinical setting

The implementation study (Chapter 6), combined quantitative and qualitative methods to observe the implementation of the prototype template in CPAP reviews. There was overall good uptake of the templates in the three participating Centres. Completed templates showed that clinicians (mostly nurses), had used most of the fields prioritised by the e-Delphi expert panel. One Centre had a much higher uptake of template use, probably because there was a clinical lead at this site who developed a robust implementation plan supported by an administrative member of the team. The semi-structured interviews showed that the template facilitated a structured, standardised review, reducing variation in practice, and was not perceived to affect clinical autonomy or person-centred care. The use of the template did not impact on how the review was conducted though it extended review time in one Centre where double data entry was required. From a service perspective, the template was perceived as improving efficiency and promoting better patient outcomes whether in face-to-face reviews or teleconsultations. In addition, the template was seen as potentially contributing to multidisciplinary team working and might be useful for education and training purposes. Adaptations to structure, content, and layout were suggested.

7.4 Implications

7.4.1 For people with OSAHS using CPAP therapy

The studies in this PhD have implications for how care is provided for people living with OSAHS who are using CPAP therapy.

- Telemedicine-based options are established as an option for clinical reviews that can reduce the requirement to travel for face-to-face reviews, and by providing care closer to home or from their place of residence, reduce the loss of work productivity and travel costs for both patients and clinicians.

- By establishing the core components of a CPAP review, and the subsequent development and refinement of a structured template in CPAP reviews, people who are using CPAP therapy may benefit from a more person-centered review that includes the core components identified by international consensus, and also what matters to them in a clinical review.

7.4.2 For professionals and clinical services

The findings of these PhD studies also have implications for professionals and providers of clinical services :

- The use of a structured review template reduces variation among clinicians, supports standardised practice and the perceived delivery of person-centred care that is based on current guidelines and best practice.
- Templates may support learning and development in new or less experienced members of staff.
- Templates can be used in paper or electronic format, may reduce administrative processes, and be more time-efficient if it is the sole method for recording the content of a CPAP review.
- From a service perspective, this would support efficiencies within services and free up time to focus on current clinical service pressures.
- By reducing variation in the delivery of healthcare in the NHS this has a key role in the way resources are best used to provide the best value for service users and organisations alike, placing the nursing profession at the forefront in a position to identify and reduce unwarranted variation across services.

I have now refined two templates based on the participants' feedback for future use (Figure 13) and have shared this with the participating sites and the national improvement team working group who are working with clinicians to remobilise services and telemedicine and e-health solutions are central to this work.

7.4.3 For policy-makers

The findings of these PhD studies have implications for policymakers:

- The potential for harnessing and embedding new remote monitoring technologies that enable optimisation of CPAP treatment is an important priority for the NHS and its policymakers to deliver better and safer care pathways and services in patients with underlying OSAHS. The British Thoracic Society has provided detailed guidance for sleep and ventilation services and advises the uses of teleconsultation and telephone review as preferable to face-to-face review in this current climate and beyond. It is apparent that policymakers in the UK and Globally will have to rapidly deploy telemedicine solutions and the evidence base and the technology to do so is already in place in many services nationally and internationally.
- Policymakers are interested in managing variation and quality improvement within healthcare and the development of a structured review template based on the e-Delphi consensus supports this process.
- Templates may facilitate skill-mix, enabling less experienced staff to safely review CPAP users, thereby relieving the pressure on increasingly busy sleep medicine services.
- In refining the template for future use, it remains to be seen if it will be adopted as a potential solution in CPAP review services more widely in Scottish sleep medicine Centres.

7.4.4 For research

The systematic review and subsequent RCTs established that the use of telemedicine (telemonitoring and remote consultation), is a central component within sleep medicine service delivery that can provide person-centred care that is safe and efficient for both service users and clinicians. Collectively, current national and international guidelines do not clearly define what a clinical review of CPAP therapy users should include and how often this should

take place. The core components of a clinical CPAP review established by the e-Delphi consensus study may influence future practice guidelines.
Future research may:

- Evaluate the clinical and cost-effectiveness of using the template in CPAP reviews (face-to-face or remotely).
- Assess the skill mix enabled by using a template with a clinical service.
- Explore the educational potential of the review template for new or less experienced staff.

7.4.5 Objective IV Refining the template

I received enough feedback and comments to meet the final objective IV of the implementation study, which was to refine the template. The feedback from those that used the template was generally positive with additional suggestions. I made two versions of the template, one for CPAP reviews and one for NIV reviews. I added the option to record the diagnosis and the mode of consultation to both templates, and the follow-up plan with additional space for more free-text comments. I also added the machine type, mask interphase, and pressure settings to both templates. I removed the capillary and arterial blood gas options and the transcutaneous carbon dioxide measurement from the CPAP template. I also added a section to record any anticipatory care plan to the NIV template as was suggested by one of the interviewees. Apart from these minor refinements, the two templates are very similar. Figure 13 outlines the refined CPAP and NIV templates. I have shared the refined template with the participating Centres.

Figure 13 CPAP/ NIV revised template

Revised CPAP clinic template

<table>
<tr><td colspan="2">Name/Address label</td><td>Date</td><td>Age</td><td>Gender</td></tr>
<tr><td colspan="3">The patient agenda for the review. What does the patient want to discuss? Any concerns?

Diagnosis:

Face to face review ☐
Telephone review ☐
Video consultation review ☐</td><td colspan="2">Wt:
Ht:
BMI:
Adherence:
Hours per night:
BP:

Settings:</td></tr>
<tr><td>Acceptability of treatment</td><td colspan="4">Are there any side effects from the treatment?
Is the patient happy to continue treatment?

Free text</td></tr>
<tr><td rowspan="2">Technical aspects of CPAP</td><td colspan="4">Does the mask fit correctly?
Is humidification required/ satisfactory?
Is cleaning / filters changing routine satisfactory?</td></tr>
<tr><td colspan="4">Free text</td></tr>
<tr><td rowspan="4">Objective assessment of sleepiness/driving issues

<u>Epworth</u>
1.Sitting
2.TV
3.Lying
4. Reading
5.Talking
6.Public
7.Passenger
8.Driving</td><td colspan="4">Objective assessment of sleepiness (Epworth Sleep Score) :
Are there any problems with sleepiness while driving?
What are they driving: Car/Heavy Goods Vehicle?</td></tr>
<tr><td colspan="4">Free text</td></tr>
<tr><td colspan="4">Have witnessed snoring, apnoeas, choking spells been controlled?
Have initial symptoms improved/resolved?</td></tr>
<tr><td colspan="4">Has NIV resolved the Apnoea Hypopnoea Index?
AHI: Mask type:
Mask Leak: Machine and settings:</td></tr>
</table>

	Free text
Assessing sleep quality/quality of life	What is the quality of sleep? Do they feel refreshed on waking? What are their sleep routines? How much sleep do they get? Quality of life **Free text**
Lifestyle	Ask about work schedule/Shift Pattern **Free text**

Additional optional review components that may be useful in some reviews	
Technical aspects	Factors that help/hinder NIV use. Support system at home/partner engagement Altitude where lives/ Altitude where sleep study was done Electrical safety of NIV unit/ NIV unit noise level
General medical assessment	Review of medical history, medication review, any new comorbidities in relation to symptoms/need for hospitalisation Examination of the nasal passage and throat Chest Auscultation findings: Need to repeat diagnostic study with significant weight loss/weight gain Nocturia/Frequency of getting up to pass urine Cognitive/developmental issues
Sleepiness assessment	Fatigue and depression scale /as interfering with subjective assessment of sleepiness e.g. Epworth Sleep Score Checking partner feedback / quality of life
Lifestyle	Are Vehicle Licensing agencies are aware of the condition? Advice re air travel
Quality of life	
Driving	
Follow up plan: **Additional comments/recommendations** **Signature** **Date**	

Revised NIV clinic template

Name/Address label	Date	Age	Gender

The patient agenda for the review. What does the patient want to discuss? Any concerns? **Diagnosis:** **Face to face review** ☐ **Telephone review** ☐ **Video consultation review** ☐	Wt: Ht: BMI: Adherence: Hours per night: BP: CBG: ABG: R/R MV TCCo2: Settings:

Acceptability of treatment	Are there any side effects from the treatment? Is the patient happy to continue treatment? **Free text**
Technical aspects of CPAP	Does the mask fit correctly? Is humidification required/ satisfactory? Is cleaning / filters changing routine satisfactory?
	Free text
Objective assessment of sleepiness/driving issues **<u>Epworth</u>** **1.Sitting** **2.TV** **3.Lying** **4. Reading** **5.Talking** **6.Public** **7.Passenger** **8.Driving**	Objective assessment of sleepiness (Epworth Sleep Score) : Are there any problems with sleepiness while driving? What are they driving: Car/Heavy Goods Vehicle?
	Free text
	Have witnessed snoring, apnoeas, choking spells been controlled? Have initial symptoms improved/resolved?
	Has NIV resolved the Apnoea Hypopnoea Index? AHI: Mask type: Mask Leak: Machine and settings:
	Free text

Assessing sleep quality/quality of life	What is the quality of sleep? Do they feel refreshed on waking? What are their sleep routines? How much sleep do they get? Quality of life **Free text**
Lifestyle	Ask about work schedule/Shift Pattern **Free text**

Additional optional review components that may be useful in some reviews	
Technical aspects	Factors that help/hinder NIV use. Support system at home/partner engagement Altitude where lives/ Altitude where sleep study was done Electrical safety of NIV unit/ NIV unit noise level Review of medical history, medication review, any new comorbidities in relation to symptoms/need for hospitalisation Examination of the nasal passage and throat Chest Auscultation findings: Need to repeat diagnostic study with significant weight loss/weight gain Nocturia/Frequency of getting up to pass urine Cognitive/developmental issues Fatigue and depression scale /as interfering with subjective assessment of sleepiness e.g. Epworth Sleep Score Checking partner feedback / quality of life Are Vehicle Licensing agencies are aware of the condition? Advice re air travel
General medical assessment	
Sleepiness assessment	
Lifestyle	
Quality of life	
Driving	
Follow up plan: **Additional comments/recommendations** **Anticipatory Care Plan:** **Signature** **Date**	

Conclusion

The three studies outlined in this thesis add to the evidence base for undertaking regular review of CPAP therapy users and telemedicine is an option for conducting a review combined with remote monitoring. The international e Delphi consensus study identified important components that may be recorded in a CPAP review and this informed the development of a structured review template. Implementation of the template was achieved in three Centres and all the objectives of the final study were met. The template was considered to provide structure to both face-to-face reviews and teleconsultations with a potentially important role in training staff and standardising care. One Centre continues to use the refined template in their service in an electronic format and it is hoped that wider adoption will be considered in other services.

Teleconsultation has been centre stage in the midst of a global pandemic for routine CPAP reviews, and electronic templates adapted to fit local services and clinical teams routines could support standardised, evidence based, safe and effective care and promote efficient communication across all healthcare settings.

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Appendix 1 Level 1 systematic review checklist

University of Edinburgh,
Centre for Population Health Sciences
RESEARCH ETHICS SUBGROUP

Self-Audit Checklist for Level 1 Ethical Review for Taught Masters and UG dissertation projects

NOTE to student: *Completion of this form should be under the oversight of your supervisor. A good strategy would be to complete a draft as best you can, then discuss with your supervisor before completing a final copy for your supervisor to sign. (Relevant information is available on Learn VLE.)*

Proposed Project *(State research question and topic area, and briefly describe method/ data. Specify also countries in which data will be collected.):*

Telehealthcare interventions in the management of Obstructive Sleep Apnoea Hypopnoea Syndrome: a systematic review

ABSTRACT

Aim: Telehealth has the potential to offer more convenient care and reduce travel. We aimed to systematically review studies that assessed the effectiveness of telehealth interventions in the review of people with Obstructive Sleep Apnoea Hypopnoea Syndrome (OSAHS) receiving Continuous Positive Airway Pressure (CPAP) therapy versus face-to-face care.

Methods: Following Cochrane methodology, we searched 11 electronic databases (November 2015), scanned included studies reference lists and identified unpublished studies. Included interventions, in any healthcare setting, had to combine remote consultations with telemonitoring of usage/CPAP data. Outcomes measures were; CPAP adherence, proportion reviewed, symptom control, satisfaction/acceptability and cost effectiveness.

Results: From 363 potentially relevant papers, we identified five RCTs (n=269 patients): four from North America and one from Spain. Risk of bias was assessed as moderate in one, and moderate/high in four trials. Proportion reviewed with telehealth (102) versus usual care (99). Four trials reported patients similar (n=2) or greater (n=2) satisfaction with telehealth vs usual care. Two studies, both at moderate to high risk of bias, showed no between group difference in the Epworth Sleepiness Score. One study reported telehealth was more cost effective.

Conclusion: The evidence for telehealth review in CPAP users is limited, however no safety concerns have been raised. Adequately powered, well designed studies are needed to establish whether telehealth is a clinically and cost effective option for people using CPAP therapy living distant to specialist sleep services.

Systematic review registration: PROSPERO 2015:CRD42015019455.

1. Bringing the University into disrepute

Is there any aspect of the proposed research which might bring the University into disrepute? YES / NO

5. Protection of research subject confidentiality

Are there any issues of CONFIDENTIALITY which are NOT adequately handled by normal tenets of confidentiality for academic research? YES/NO

These include well-established sets of undertakings that should be agreed with collaborating and participating individuals/organisations. For example, a 'No' answer is justified *only if*:

(a) There will be no attribution of individual responses;

(b) Individuals (and, where appropriate, organisations) are anonymised in stored data, publications and presentation;

(c) There has been specific agreement with respondents regarding feedback to collaborators and publication.

6. Potential physical or psychological harm, discomfort or stress

(a) Is there a FORSEEABLE POTENTIAL for PSYCHOLOGICAL HARM or STRESS for participants? YES/NO

(b) Is there a FORSEEABLE POTENTIAL for PHYSICAL HARM or DISCOMFORT for participants? YES/NO

(c) Is there a FORSEEABLE RISK to the researcher? YES/NO

Examples of issues/ topics that have the potential to cause psychological harm, discomfort or distress and should lead you to answer 'yes' to this question include, but are not limited to:

relationship breakdown; bullying; bereavement; mental health difficulties; trauma / PTSD; violence or sexual violence; physical, sexual or emotional abuse in either children or adults.

7. Duty to disseminate research findings

Are there issues which will prevent all relevant stakeholders having access to a clear, understandable and accurate summary of the research findings if they wish?* YES/NO

** If, and only if, you answered 'yes' to 3 above, 'stakeholders' includes the participants in the research*

Overall assessment

- If every answer above is a definite NO, the self-audit has been conducted and confirms the ***ABSENCE OF REASONABLY FORESEEABLE ETHICAL RISKS*** – *please tick box* ✓

  This means that regarding this study, as currently self-audited, no further ethical review actions are required within CPHS. However, if in the coming weeks/months there is any change to the research plan envisaged now (and outlined above), the study should be ***re-audited*** *against a Level 1 form, because it may be that the change made negates the absence of ethical risks signed off here.*

- If one or more answers are YES, then risks have been identified and prior to commencing any data collection **formal ethical review** is required - either:
  - by NHS REC (NB copy of ethics application and decision letter to be sent to CPHS Ethics); or
  - if not to be formally reviewed by NHS REC, then CPHS level 2/3 ethical review required. *[If either 4 is 'yes' or 3b is 'vulnerable' then it is possible level 3 review is required.]*

Student Exam Number: s140044

Please see next sheet for names and signatures

Note regarding version of level 1 form bound into MPH dissertation:

The signed hard copy of this Level 1 form has been lodged with the CPHS Research Degree committee. In accordance with usual Level 1 self-audit process, the form was completed and signed by the masters/UG student, with oversight by his/her Supervisor, who countersigned to confirm this.

However, in order to preserve anonymity of students through the marking process, student and supervisor names and signatures (the additional sheet) have been excluded from the dissertation submitted for marking.

The overall assessment shown above is identical with that shown on the signed-off form lodged.

Signed: Postgraduate administrator

2. Data protection and consent

Are there any issues of DATA PROTECTION or CONSENT which are NOT adequately dealt with via established procedures? YES/NO

These include well-established sets of undertakings. For example, a 'No' answer is justified *only if*:

(a) There is compliance with the University of Edinburgh's Data Protection procedures (see www.recordsmanagement.ed.ac.uk);

(b) Respondents give consent regarding the collection, storage and, if appropriate, archiving and destruction of data;

(c) Identifying information (eg consent forms) is held separately from data;

(d) There is Caldicott Guardian approval for (or approval will be obtained prior to) obtaining/ analysing NHS patient-data.

(e) There are no other special issues arising about confidentiality/consent.

3. Study participants

a) Will a study researcher be in direct contact with participants to collect data, whether face-to-face, or by telephone, electronic means or post, or by observation? (eg interviews, focus groups, questionnaires, assessments) YES/NO

b) **Answer this only if qu. 3 above = 'YES':**

*In ethical terms, could any participants in the research be considered to be '**vulnerable**'?*
e.g. children & young people under age of 16, people who are in custody or care (incl. school), a marginalised/stigmatised group

Please tick one: 'vulnerable' ☐ not 'vulnerable' ☐

4. Moral issues and Researcher/Institutional Conflicts of Interest

Are there any SPECIAL MORAL ISSUES/CONFLICTS OF INTEREST? YES/NO

(a) An example of conflict of interest for a researcher would be a financial or non-financial benefit for him/herself or for a relative of friend.

(b) Particular moral issues or concerns could arise, for example where the purposes of research are concealed, where respondents are unable to provide informed consent, or where research findings could impinge negatively/ differentially upon the interests of participants.

(c) Where there is a dual relationship between researcher and participant (eg where research is undertaken by practitioners so that the participant might be unclear as to the distinction between 'care' and research)

5. Protection of research subject confidentiality

Are there any issues of CONFIDENTIALITY which are NOT adequately handled by normal tenets of confidentiality for academic research? YES/NO

These include well-established sets of undertakings that should be agreed with collaborating and participating individuals/organisations. For example, a 'No' answer is justified *only if*:

(a) There will be no attribution of individual responses;

(b) Individuals (and, where appropriate, organisations) are anonymised in stored data, publications and presentation;

(c) There has been specific agreement with respondents regarding feedback to collaborators and publication.

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(a) Is there a FORSEEABLE POTENTIAL for PSYCHOLOGICAL HARM or STRESS for participants? YES/NO

(b) Is there a FORSEEABLE POTENTIAL for PHYSICAL HARM or DISCOMFORT for participants? YES/NO

(c) Is there a FORSEEABLE RISK to the researcher? YES/NO

Examples of issues/ topics that have the potential to cause psychological harm, discomfort or distress and should lead you to answer 'yes' to this question include, but are not limited to:

relationship breakdown; bullying; bereavement; mental health difficulties; trauma / PTSD; violence or sexual violence; physical, sexual or emotional abuse in either children or adults.

7. Duty to disseminate research findings

Are there issues which will prevent all relevant stakeholders having access to a clear, understandable and accurate summary of the research findings if they wish?* YES/NO

** If, and only if, you answered 'yes' to 3 above, 'stakeholders' includes the participants in the research*

Overall assessment

- If every answer above is a definite NO, the self-audit has been conducted and confirms the ***ABSENCE OF REASONABLY FORESEEABLE ETHICAL RISKS*** – *please tick box* ✓

  This means that regarding this study, as currently self-audited, no further ethical review actions are required within CPHS. However, if in the coming weeks/months there is any change to the research plan envisaged now (and outlined above), the study should be ***re-audited*** *against a Level 1 form, because it may be that the change made negates the absence of ethical risks signed off here.*

- If one or more answers are YES, then risks have been identified and prior to commencing any data collection **formal ethical review** is required - either:
  - by NHS REC (NB copy of ethics application and decision letter to be sent to CPHS Ethics); or
  - if not to be formally reviewed by NHS REC, then CPHS level 2/3 ethical review required.

  [If either 4 is 'yes' or 3b is 'vulnerable' then it is possible level 3 review is required.]

Student Exam Number: s140044

Please see next sheet for names and signatures

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However, in order to preserve anonymity of students through the marking process, student and supervisor names and signatures (the additional sheet) have been excluded from the dissertation submitted for marking.

The overall assessment shown above is identical with that shown on the signed-off form lodged.

Signed: Postgraduate administrator

Appendix 2 Systematic review protocol

Telehealth - OSAHS

Telehealthcare interventions in the management of Obstructive Sleep Apnoea Hypopnoea Syndrome

Systematic review protocol

Research team

Phyllis Murphie	*Principal investigator*
Dr Stuart Little	*Co- investigator*
Dr Hilary Pinnock	*Co-investigator*
Professor Brian McKinstry	*Co-investigator*
Library services	*Marshall Dozier*
Statistician	

Table of Contents

Summary

Informed by the preliminary phases of the Medical Research Council's (MRC) framework for the design and evaluation of complex interventions, this systematic review forms essential background work underpinning a programme of research to develop, refine and pilot a telehealthcare intervention to address the clinical review needs of people who are diagnosed with Obstructive Sleep Apnoea Hypopnoea Syndrome and who are using Continuous Positive Airway Pressure (CPAP) therapy and living remotely to centralised Sleep Medicine services.

We will identify, and quality assess and synthesise the published and unpublished evidence for telehealthcare interventions that are designed to deliver clinical care for people with OSAHS remotely compared to usual face to face care.

Introduction

Obstructive Sleep Apnoea Hypopnoea Syndrome (OSAHS) entered the mainstream medical literature approximately 30 years ago and has only been given a higher profile by the general public over the last 10 years. It is one of a number of sleep disordered breathing conditions that is increasingly recognised. It is a major public health problem as an important cause of morbidity and mortality (1-5). The most recent epidemiological data reports the incidence of OSAHS as 3–7% of middle-aged men and 2–5% of women (2-4). Population datasets to estimate OSAHS prevalence in the United States and other countries did not start to emerge until the 1990's (4) and a review by Won et al in 2008 concluded that the accumulated results of several population-based studies across various global regions and ethnic groups show a similar prevalence rate of OSAHS despite differences in study designs and methodologies used (5).

OSAHS is a common condition

The rising prevalence of OSAHS is directly related to obesity (1, 4, 6) with an estimated 25% prevalence of obesity reported in a large cross sectional observational study in England 2011 (7). A significant minority of affected individuals, however, are non obese (2, 6, 8) . Other contributory factors that can narrow the upper airway are; an enlarged tongue, enlarged tonsilar tissue, excess soft tissue in the pharynx and retro or microagnathia (9, 10).

OSAHS is defined as a condition during sleep where there is repeated collapse or closure of the pharyngeal airway resulting in apnoeic and or hyponoeic episodes that can range from mild to moderate to severe depending on how many episodes of these occur per hour of sleep. The symptomatic consequences of OSAHS are numerous and sufferers may report a combination of excessive daytime somnolence, loud and disruptive snoring, a sensation of nocturnal choking and gasping, poor and unrefreshing sleep, mood changes, impaired alertness (sometimes when driving), morning headaches, nocturia and decreased libido (2, 3, 6, 10-13).

OSAHS has associated morbidity and mortality

There are no European collective statistics available on associated morbidity and mortality (2). The quality of life (QOL) of individuals with OSAHS can very often be impaired as a result of these symptoms (14, 15). Excessive daytime somnolence caused by OSAHS can be severe enough to affect concentration and cognitive functioning. Reported adverse effects on work related performance are common, and may include including falling asleep whilst working and work related accidents particularly driving related accidents that may result in loss of employment and earnings (16, 17).

OSAHS has been implicated as a condition that poses an increased risk of road traffic accidents. The European Working Group report on Obstructive Sleep apnoea published in 2013 reported on two recent meta analysis of published literature, that estimate the increased risk for vehicle accidents is 2 to 3 times greater than for the general population (16, 17). The 2014 British Thoracic Society position statement on driving and OSAHS gives clear guidance to specialist teams in secondary care about advising those with sleep disorders (18). The individual who holds a current driving licence is solely responsible for ensuring their fitness to drive; however, clinicians should highlight the risks if excessive sleepiness is impairing driving prior to referral to secondary care for further investigation.

Treatment and the on-going management/follow-up of OSAHS

Following diagnosis of OSAHS the majority of individuals will be established on fixed Continuous Positive Airway Pressure (CPAP) therapy following a trial of auto titration therapy to establish therapeutic pressure setting (1, 11). During the initial CPAP auto titration phase mask fit issues and potential side effects will be resolved. A fixed pressure dedicated CPAP unit will then be supplied to the patient and ongoing management and review will be agreed upon. Some individuals may find fixed CPAP pressure difficult to tolerate and it may be appropriate to leave these patients on auto titration as a long term management strategy. Guidelines recommend that patients using CPAP therapy should be seen periodically for clinical review of their compliance with therapy, replacement of disposables such as masks, filters and hoses and to check that their CPAP units are working correctly. Annual review is offered in many centres however many individuals who are well controlled with established long term compliance can be seen on a less regular basis (3, 11).

Current guidelines recommend that patients using CPAP must have rapid access to skilled clinician follow-up including equipment maintenance; this includes availability of medical advice where services are run primarily by nonmedical personnel. Facilities for appropriate trouble-shooting, such as home overnight monitoring and auto-titrating machines, and telephone support within working hours and answer phone support service are also recommended (1).

The potential of telehealth to support clinical review of people with OSAS using CPAP

Telehealthcare solutions to support the clinical review of patients with a diagnosis of OSAHS who are using CPAP therapy have significant potential to deliver person centred, safe, and effective care that is closer to home (19). There are also potential benefits for patients and clinicians in terms of reduced travel, fuel consumption and environmental impact in terms of reduced carbon emissions (20). It has the potential to deliver equivalent clinical care for people living in remote and rural areas and this can have a significant impact on time taken off work to travel to sleep centres to undergo diagnostic and treatment invent interventions.

Aim of the systematic review

We aim to systematically review the published and unpublished literature for randomised controlled trials to evaluate the application of a telemedicine-based approach in CPAP therapy management, particularly focusing on the utility of teleconsultation, the acceptability, feasibility and clinical effectiveness of using telehealthcare to manage OSAHS compared to traditional face to face care.

Outcomes

Primary outcomes

Process outcomes:

The number of patients with a diagnosis of OSAHS who are using CPAP who have received a teleconsultation review

Recorded adherence with prescribed CPAP therapy via teleconsultation compared to usual care.

Clinical outcomes:

Current control

Improvement in daytime somnolence and self recorded Epworth sleepiness score.

Quality of sleep

Quality of life

Future risk (e.g of RTA's, CV complications)

Secondary outcomes

Satisfaction/acceptability

Patient and clinician acceptability and satisfaction with teleconsultation review in the ongoing management of this condition.
The benefits and drawbacks of the use of Telehealthcare in OSAHS

Health Economics

Health service resource use compared to usual face to face care

Plan of investigation

We will follow the procedures described in the Cochrane Handbook for Systematic Reviews of Interventions.

Identification of studies

We will search electronic databases, scan reference lists of included studies, identify unpublished studies using a PICOS search strategy (see table 1). Search methods for identification of studies will include electronic searches, and trials will be identified using the Cochrane Airways Group Specialised Register of trials, which is derived from systematic searches of bibliographic databases including the Cochrane Central Register of Controlled Trials (CENTRAL,MEDLINE, EMBASE, CINAHL, AMED and British Nursing Index and hand searching of respiratory journals. Search dates will be from 1995-2015. These dates have been chosen as the earliest literature on telehealth interventions in sleep medicine appears in 2000 and we looked back to 1995 to ensure that we captured all of the literature published on the subject. Our search terms are detailed in Appendix 1.

Unpublished and ongoing work and research in progress will be identified by searching key Internet-based relevant databases – UK Clinical Research Network Study Portfolio and the meta Register of Controlled Trials, www.clinicaltrials.gov; www.controlledtrials.com. In addition, to extend our search for published, unpublished and on-going studies, we will contact an international panel of experts in this field.

Only published and unpublished work in the English Language will be considered in this review. In addition, we will search for any qualitative studies associated with included trials to add context to our interpretation of trial data.

All records in the Specialised Register coded as 'sleep apnoea' were searched using the following

terms: telehealth* or telemonitoring* or teleconsultation* or teleclinics* or telecare* or telehealthcare* or remote review. An additional search of CENTRAL will be conducted using the search strategy in Appendix 1. The most recent searches were carried out in March 2015. We searched reference lists from retrieved articles to identify other relevant reports. In addition, we contacted authors of included studies to identify any additional published or unpublished studies which fulfilled the inclusion criteria.

Table 1: PICOS search strategy

		Definitions
Population	People with a diagnosis of OSAHS on CPAP therapy	Sleep apnoea is defined as a condition in which a person experiences repeated episodes of apnoea because of a narrowing or closure of the pharyngeal airway during sleep(1, 13).
Intervention	The application of teleconsultation via video conference link in the management of people with a diagnosis of OSAHS	This will include the use of telehealth to review patients using CPAP therapy remotely: • via video consultation • telemonitoring and remote consultation We will exclude telephone calls with telemonitoring and also telemonitoring without remote consultation
Context	Community based	The patient will be in the community and the Health Care practitioner may be based in primary or secondary care.
Comparator	Usual face to face clinical care	Normally delivered face to face but may include some telephone calls
Outcomes	<u>Primary outcome</u>: Outcomes of interest are clinical (measure of sleepiness, quality of sleep and quality of life), risk of complications (including RTAs) and process outcomes (including number of reviews and adherence to CPAP <u>Secondary outcomes</u>: Patient/clinician acceptability and satisfaction with teleconsultation. The benefits and drawbacks of the use of Telehealthcare in OSAHS Health service resource use compared to usual face to	

	face care	
Study design	Randomised controlled trials (RCTs), quasi-RCTs and controlled clinical trials (CCT).	

Selection of studies

Following an initial sift and rejection of obviously unrelated abstracts the titles and abstracts of trials identified from the searches will be checked by two investigators who will not be masked to study details. In order to ensure that we do not overlook potentially eligible trials we will consider any trial in which OSAHS, CPAP therapy and telehealthcare methods are explicitly mentioned in the abstract.

The full text of all potentially eligible studies will be retrieved and independently assessed against the inclusion criteria (see above) by two reviewers. The reviewers will decide which of the studies fit the inclusion criteria: any disagreements will be resolved by discussion, with a third researcher brought in to arbitrate if needed.

To ensure transparency, the process of selection will be summarised using a PRISMA flow diagram (Appendix 2){Moher, 2009 #2466}.

Dealing with lack of information

If after the full text assessment it is still unclear whether a study fulfils the inclusion criteria, or if we require clarification of any details relating to the intervention or data we will attempt to contact authors by email for further relevant information. If we fail to make contact/retrieve this, we will list the respective study as "potentially relevant study".

Dealing with duplication

Multiple papers may be published for a number of reasons including translations, results at different follow-up period or reporting of different outcomes. We will treat a study with multiple reports as a single study, but draw on and make reference to all the relevant publications.

Assessment of methodological quality

We will assess and document the methodological quality of included controlled trials following the Cochrane approach using the methods detailed in section six of the Cochrane Handbook for Systematic Reviews of Interventions. Intervention studies will be assessed using the Cochrane Effectiveness and Practice Organisation of Care (EPOC) guidelines. We propose to concentrate on using the following seven domain-based parameters to assess quality: adequate sequence generation, allocation concealment; blinding of participants and personnel, blinding of outcomes, incomplete outcome data addressed, free of selective reporting and free of other bias. We will grade each parameter of trial quality: A - low risk of bias; B - moderate risk of bias; C - high risk of bias and an overall assessment for each controlled trial using the same three criteria will be made. We will assess the agreement of reviewers on methodological quality assessment and resolve disagreements by discussion, with a third researcher brought in to arbitrate if needed.

Data extraction

Two reviewers will extract data using a customised data extraction form which will initially be piloted to ensure the form is easily and consistently interpreted and captures all relevant information. We will resolve any disagreements by discussion between reviewers; in the case of consensus not being reached, a third reviewer will become involved and, if necessary, arbitrate.

In order to compile a detailed descriptive summary, two reviewers will independently extract data, using the headings 'telehealth care setting', ' telehealth mode of delivery', 'aspects of holistic care addressed by telehealth intervention', 'duration and components of telehealth intervention'.

Data synthesis

Based on our preliminary scoping work, we anticipate that we will identify a limited number of eligible trials with substantial heterogeneity so that meta-analysis will not be appropriate. We therefore plan to undertake a narrative synthesis by developing a matrix of what has been shown to be effective or ineffective and the elements of the interventions under the headings of setting, mode of tele healthcare delivery, aspects addressed during clinical review.

Project management

This programme of work will carried out by Phyllis Murphie as part of a PhD supervised by Dr Hilary Pinnock and Professor Brian McKinstry.

Research environment and project management

Within the Usher Institute of Population Health Sciences and Informatics of The University of Edinburgh there is an on-going programme of qualitative and quantitative work underway involving social scientists, clinicians, epidemiologists, trialists and statisticians providing access to in-house methodological expertise.

Research team

The research team who will actively work on the review are:

Phyllis Murphie
Dr Stuart Little
Dr Hilary Pinnock
Professor Brian McKinstry

Library facilities will be provided by Marshall Dozier

Statistical advice will be provided by the Usher Institute of Population Health Sciences and Informatics s of The University of Edinburg.

Lay Advisory Group – Lay advisory advice will be sought from the Scottish Association of Sleep Apnoea patients support group.

Timetable

February – March 2015	☑ Develop and agree search strategy ☑ Database searching ☑ Data collection from all sources ☑ Preliminary data analysis
April –May 2015	☑ Selection of papers ☑ Contact authors/experts
June 2015	☑ Data extraction ☑ Quality assessment
July 2015	☑ Final data analysis ☑ Write report ☐ Prepare abstracts and write papers

Implementation potential

The underlying purpose of the study is to establish essential background work for the development, piloting and evaluation of a telehealthcare intervention to address future clinical review of patients who have OSAHS and are being treated with CPAP therapy. This systematic review will clarify the evidence base underpinning the intervention.

Dissemination

We will share our findings with fellow investigators planning intervention studies, present abstracts at international conferences; disseminate findings within our professional spheres of influence. A paper will be published in a peer reviewed journal.

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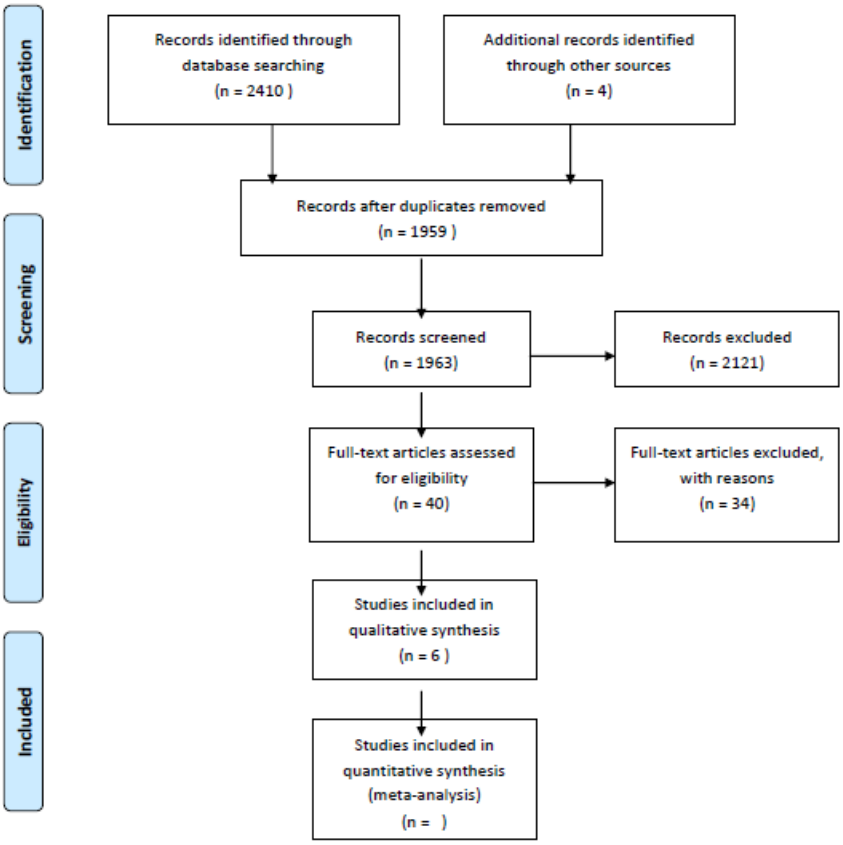
Appendix 1: Search strategy

Search terms for Cochrane library, CINAHL, LILACS, British Nursing Index (BNI), , ISI Web of Science (1995-2015)

("Obstructive Sleep Apnoea Hypopnoea Syndrome" or "Sleep Apnoea Syndrome" or "Sleep disordered breathing" or "CPAP therapy" or "Nasal CPAP")
AND
(telehealth stud* or telecare stud* telemedicine stud* or ehealth stud*or telehealthcare stud* or telemonitoring stud* or clinical trial or controlled clinical trial or randomised controlled trial or quasi-randomised clinical trial)
AND
("quality of life" or "health related quality of life")

Search terms for MEDLINE, EMBASE, AMED, (1995-2015)

Appendix 2 - PRISMA 2009 Flow Diagram



Appendix 3 PRISMA Checklist for Systematic review

PRISMA 2009 Checklist

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	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	1
ABSTRACT			
Structured summary	2	Abstract Introduction: Telehealth has the potential to offer more convenient care and reduce travel. We aimed to systematically review studies that assessed the effectiveness of teleconsultation plus telemonitoring in the review of people with obstructive sleep apnoea hypopnoea syndrome receiving continuous positive airway pressure therapy versus face-to-face care. Methods: Following Cochrane methodology, we searched 10 electronic databases (November 2015), trial registries, and reference lists of included studies, for trials testing interventions that combined remote consultations with telemonitoring of usage/continuous positive airway pressure data. Outcomes measures were: proportion reviewed, continuous positive airway pressure adherence, symptom control, and satisfaction/acceptability and cost effectiveness. Results: From 362 potentially relevant papers, we identified five randomised controlled trials (n 269 patients): four from North America and one from Spain. Risk of bias was moderate in one, and moderate/high in four trials. Two trials reported number/duration of reviews with inconsistent results. The teleconsultation/telemonitoring improved continuous positive airway pressure adherence in two trials (n 19; n 75); two (n 114 and n 75) reported no between-groups differences. Two studies, both at moderate/high risk of bias, showed no between-group difference in the Epworth Sleepiness Score. Satisfaction was generally reported positively in all five trials; one trial reported that the teleconsultation/telemonitoring patients were 'more likely to continue' with continuous positive airway pressure therapy treatment. One study reported teleconsultation/telemonitoring as cost effective. Discussion: The evidence for teleconsultation/telemonitoring in continuous positive airway pressure users is limited; however, no safety concerns have been raised. Adequately powered, well-designed trials are needed to establish whether real-time telemonitoring and remote teleconsultation is a clinically and cost-effective option for people using continuous positive airway pressure therapy.	1
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	1-2
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	3

PRISMA 2009 Checklist

METHODS			
Protocol and registration	5	PROSPERO international prospective register of systematic reviews (CRD42015019455).	2
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	2
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	2
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	2
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	2
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	3
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	3
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	5
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	3
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	4

Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	7
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	NA
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	4
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	Table P5
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	7

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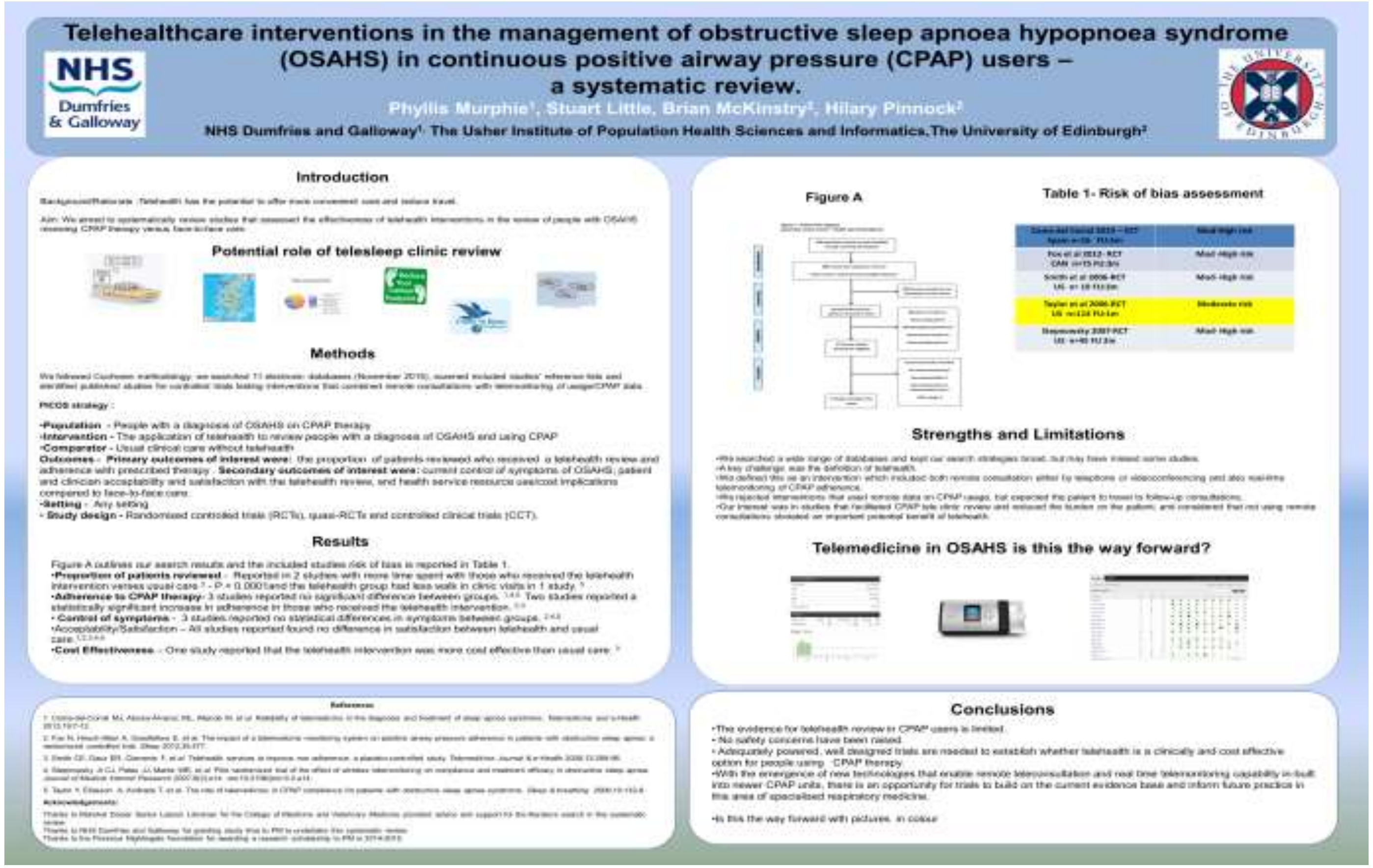
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	5-6
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	4-6
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	7
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	N/A
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	7
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	7
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	8
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	8

From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097

For more information, visit: www.prisma-statement.org.

Appendix 4 Systematic Review Poster presented at ERS 2016



Appendix 5 Published systematic review in Journal of Telemedicine and Telecare 2019

RESEARCH/Original Article

Journal of Telemedicine and Telecare
2019, Vol. 25(1) 17–25

DOI: 10.1177/1357633X17735618
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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

Phyllis Murphie[1,2], Stuart Little[2], Brian McKinstry[1] and Hilary Pinnock[1]

Abstract

Introduction: Telehealth has the potential to offer more convenient care and reduce travel. We aimed to systematically review studies that assessed the effectiveness of teleconsultation plus telemonitoring in the review of people with obstructive sleep apnoea hypopnoea syndrome receiving continuous positive airway pressure therapy versus face-to-face care.

Methods: Following Cochrane methodology, we searched 10 electronic databases (November 2015), trial registries, and reference lists of included studies, for trials testing interventions that combined remote consultations with telemonitoring of usage/continuous positive airway pressure data. Outcomes measures were: proportion reviewed, continuous positive airway pressure adherence, symptom control, and satisfaction/acceptability and cost effectiveness.

Results: From 362 potentially relevant papers, we identified five randomised controlled trials ($n = 269$ patients): four from North America and one from Spain. Risk of bias was moderate in one, and moderate/high in four trials. Two trials reported number/duration of reviews with inconsistent results. The teleconsultation/telemonitoring improved continuous positive airway pressure adherence in two trials ($n = 19$; $n = 75$); two ($n = 114$ and $n = 75$) reported no between-groups differences. Two studies, both at moderate/high risk of bias, showed no between-group difference in the Epworth Sleepiness Score. Satisfaction was generally reported positively in all five trials; one trial reported that the teleconsultation/telemonitoring patients were 'more likely to continue' with continuous positive airway pressure therapy treatment. One study reported teleconsultation/telemonitoring as cost effective.

Discussion: The evidence for teleconsultation/telemonitoring in continuous positive airway pressure users is limited; however, no safety concerns have been raised. Adequately powered, well-designed trials are needed to establish whether real-time telemonitoring and remote teleconsultation is a clinically and cost effective option for people using continuous positive airway pressure therapy.

Keywords

Obstructive sleep apnoea hypopnoea syndrome, continuous positive airway pressure, telehealthcare, telemonitoring, remote consulting

Date received: 24 October 2016; Date accepted: 15 March 2017

Introduction

Obstructive sleep apnoea hypopnoea syndrome (OSAHS) is a major public health concern and an important cause of morbidity and mortality.[1–7] Epidemiological data reports its prevalence as 3–7% of middle-aged men and 2–5% of women.[2,4] Globally, prevalence rates are increasing and this is directly related to obesity[2,8,9] though a minority of individuals are non-obese.[2,10,11] Other contributory factors are; enlarged tongue/tonsillar tissue, excess pharyngeal soft tissue and retro- or micrognathia.[12,13] Quality of life (QoL) is adversely affected.[2,7] The symptomatic consequences of OSAHS include; excessive daytime sleepiness, loud and socially disruptive

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snoring, nocturnal choking or gasping, poor unrefreshing sleep, mood changes, impaired alertness (sometimes when driving), morning headaches, nocturia and decreased libido.[2,8,11,13,14] Driving-related accidents are a particular concern.[1] Work-related performance issues are common, and include incidents or accidents that may result in loss of employment.[1,7] The increased prevalence of OSAHS in the last two decades has seen an exponential rise in referral rates for investigation and treatment of this condition.[2,15]

After diagnosis the majority of individuals are established on fixed continuous positive airway pressure (CPAP) therapy (usually following auto-titration to establish therapeutic pressure settings). CPAP can improve morbidity and current guidelines recommend that patients using CPAP should be reviewed regularly to assess adherence, replace disposables such as masks, filters and hoses, manage side effects and maintain CPAP devices.[11,14] CPAP adherence is problematic for a number of reasons such as mask fit, comfort, prescribed CPAP pressure tolerance, oral-nasal dryness, claustrophobia, abdominal bloating, psychological and social factors. A National Institute for Health and Clinical Excellence technology appraisal reported adherence rates to CPAP therapy as 71% (range 64–83%) in the first year and 79% (range 68–90%) for those who persisted for more than 12 months.[15] However, the definition of CPAP adherence is inconsistent and reports of CPAP adherence range from as low as 28% to more than 83% in more recent studies.[16,17] Modern CPAP devices are now able to measure CPAP wear time and efficacy at effective pressure.[17] Low adherence to CPAP limits its therapeutic benefit,[17] and encouraging optimal – or at least adequate - CPAP use is a clinically important[6,18] but an on-going time consuming problem globally.

Telehealth to support clinical review of CPAP users has the potential to deliver effective and convenient care,[6,19–23] with possible benefits in terms of therapy adherence, user satisfaction and reduced time taken from employment to attend a specialist sleep centre review.[6] For those living remotely, telehealth can reduce travel and can have a significant environmental impact in terms of decreased carbon emissions.[20,21,23]

We aimed to systematically review the literature on the effectiveness, acceptability and health service resource implications of using telehealth (i.e. telemonitoring of CPAP usage data plus remote consultations) to undertake routine reviews of patients using CPAP compared to face-face care in any healthcare setting.

Methods

Our systematic review is registered with the PROSPERO international prospective register of systematic reviews (CRD42015019455).[20] We did not make any changes to the protocol and we adhered to the procedures described in the *Cochrane handbook for systematic reviews of interventions*,[24] and the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (Prisma) statement for reporting on systematic reviews.[25] An ethics checklist was submitted to the University of Edinburgh, Centre for Population Health Sciences Research Ethics Group.

Inclusion criteria

Our population, intervention, outcomes, setting and study design (PICOS) inclusion/exclusion criteria are summarised in Table 1. We searched for randomised controlled trials (RCTs), quasi-RCTs and controlled clinical trials (CCTs) in those using CPAP therapy that compared teleconsultation combined with daily telemonitoring of CPAP data with face-to-face care. The interventions had to include both a consultation remote to the main healthcare facility (video or telephone consultation) and the facility for reviewing real-time telemonitoring of data from the CPAP device (defined as data transmitted and monitored by, or available to the reviewing clinical team via a web based platform at any time). We excluded telephone calls without telemonitoring and also telemonitoring without remote consultation. Our interest was in routine reviews of people established on CPAP, as opposed to the coaching and additional support associated with initiation of treatment.

Search strategy

We searched 10 electronic databases; AMED; British Nursing Index; CINAHL; Cochrane Library; DARE; EMBASE; LILACS; MEDLINE; Web of Science and ZETOC (our detailed search strategy is given in Supplementary Material, File 1). The bibliographies of included studies were scrutinised to identify possible additional studies and we also hand searched relevant sleep medicine and respiratory journals (*Sleep Medicine, Journal of Clinical Sleep Medicine, Sleep, Sleep Medicine Reviews, Thorax, European Respiratory Journal, Breathe, BMJ Open Respiratory Research, Respiratory care, American Journal of Respiratory and Critical Care Medicine, Chest, European Respiratory Review, Respiratory Medicine, NPJ Primary Care Respiratory Medicine*). Search dates were not limited, but we only included studies published in English as our resources did not permit translation. The search was carried out in November 2015.

Selection of studies

An initial sift and rejection of obviously unrelated abstracts was conducted by PM. Titles and abstracts were then screened by PM with 25% checked by HP or BMcK (agreement 100%). In order to ensure that we did not overlook potentially eligible trials, we included all abstracts in which any form of telehealth and OSAHS and/or CPAP were explicitly mentioned. The full text of all potentially eligible studies were retrieved and independently assessed against the inclusion criteria by two reviewers (PM, and SL or HP); any disagreements were

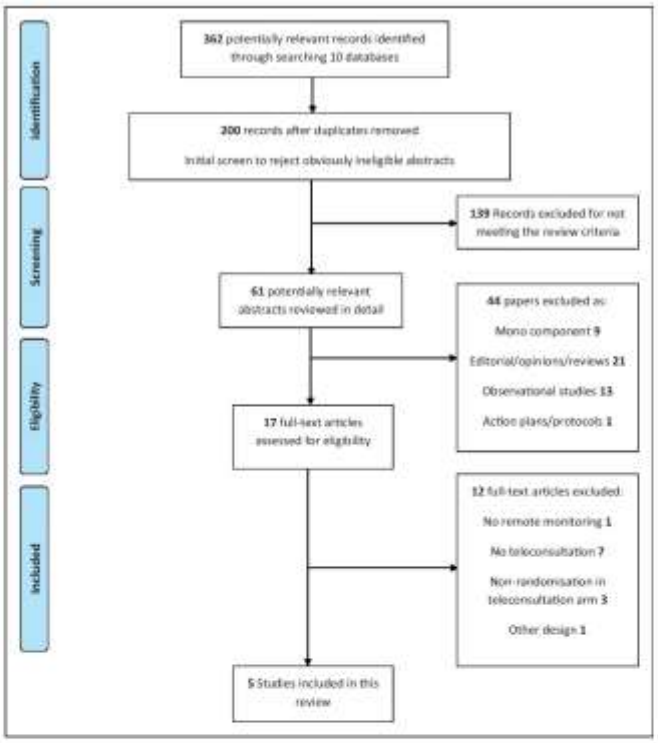

Figure 1. PRISMA flow diagram. Systematic review search – obstructive sleep apnoea hypopnoea syndrome (OSAHS) and telehealthcare.

the epworth sleepiness scale), patient and clinician acceptability and satisfaction with the teleconsultation/telemonitoring review, and health service resource use/cost compared to face-to-face care.

Data synthesis

We expected that we would identify a limited number of eligible studies (based on our preliminary scoping work) with substantial heterogeneity so that meta-analysis would not be appropriate. We therefore undertook a narrative synthesis of the data.

Results

Three hundred and sixty-two potentially relevant publications were identified, from which 17 full text articles were reviewed with 12 being excluded[6,19,22,23,27–34] as they did not match our inclusion criteria which is explained in Supplementary Material, File 4. Five papers fulfilled our inclusion criteria (Table 2). Details of the selection process are given in the Figure 1 PRISMA flow diagram. Four studies were from North America,[3,35–37] and one from Spain,[38] and all were based in community settings. The five studies included a total of 269 participants. All the trials included telemonitoring of CPAP adherence; three utilised telephone consultations,[3,36,37] the other two utilised teleconferencing for remote consultations.[35,38]

Description of the study designs

Four of the included studies were RCTs;[3,35–37] the other was a CCT.[38] The follow-up period in the studies ranged from 1–6 months. Four of the trials reported proportion of reviews achieved; outcome measures included CPAP adherence,[3,35–37] Epworth sleepiness scale,[3,36] patient/clinician satisfaction/acceptability,[3,35–38] and costs of providing the telemonitoring/teleconsultation intervention.[35]

Methodological quality

The results of the methodological quality assessment are detailed in Supplementary Material, File 2. One study was

Table 2. Summary of included studies.

Reference, country, risk of bias (RoB)	Study design, number of participants, FU	Intervention and comparator	Target group participants	Results						Comments
				Proportion reviewed	Adherence to CPAP	Symptom control	Satisfaction acceptability	Costs		
Correia del Corral et al. 2013[28] Spain Mod/high risk RoB	CCT n = 16 FU: 6m	TH (video consultation + telemonitoring of CPAP usage) vs UC (with CPAP telemonitoring)	Adults with OSAHS Mean age 53 years 63% male	Not measured	There was no between group difference in adherence to CPAP: Telehealth 73% vs UC 85% (significance not reported)	Not measured	Visual analogue scale used (1–10) reported as 9.5 for both groups Clinician satisfaction not reported	Telemedicine / teleconsultation costs not reported for CPAP users		TH to remote sites feasible; may provide specialised sleep medicine services to geographically distant populations
Fox et al. 2012[2] Canada Mod/high risk RoB	RCT n = 75 FU: 3m	TH (telemonitoring + telephone support if needed) vs UC (with telephone support where needed)	Adults with OSAHS Mean age 53 years 80% male	Compared with UC, more time was spent with the TH patients: TH: 2.10 min (SD 42) vs UC: 143 min (SD 48) $p < 0.0001$	Adherence greater in TH group: mean hours night: TH 3.2 vs UC: 1.8 ($p = 0.006$)	Epworth sleepiness score: no significant between group differences (TH 5.1 vs UC 5.2)	Not measured	Not measured		TH may improve adherence to CPAP at initiation of therapy
Smith et al. 2008[35] USA Mod/high risk RoB	RCT n = 19 FU: 3m	TH (CPAP) via telehealth equipment with inbuilt camera enabling clinician consultation with patients v TH (W-B = well-being advice)	Adults initiating CPAP for OSAHS Mean age 63 years 5% male not reported	Not measured	TH (CPAP) improved proportion achieving 80% use rate. TH (CPAP): 90% vs TH (W-B): 44% ($p = 0.033$)	Not measured	Proportion rating service 'positively' TH (CPAP) 100% vs TH (W-B) 75% (significance not reported)	TH (CPAP) costs were less than TH (W-B) costs: TH (CPAP): US $420 vs TH (W-B) US $1 500 (significance not reported)		Structured video-consultations may facilitate problem solving strategies via visual camera observation and overcome barriers to adherence
Stepnowsky et al. 2007[36] USA Mod/high risk RoB	RCT n = 45 FU: 2m	TH (regular telemonitoring and telephone calls) vs UC supported by telephone calls	Adults initiating CPAP for OSAHS Mean age 59 years 98% male	Not measured	Non-significant trend to greater adherence in TH group. Hours/night: 4.1 (SD 1.8) vs UC: 2.8 (SD 2.2) ($p = 0.07$)	Epworth sleepiness score. No significant between group differences: TH 9.2 (SD 4.6) vs UC 9.9 (SD 5.3) $p = 0.72$	Likelihood of continuing CPAP higher in TH than UC group on score of 1 to 5 (4.8 vs 4.3; $p = 0.05$)	Not measured		TH monitoring of CPAP adherence with rapid clinician support to guide management may improve adherence and commitment to using CPAP
Taylor et al. 2006[37] USA Mod risk RoB	RCT n = 114 FU: 1m	TH via Health Buddy + daily automated feedback v UC Both groups had telephone support if needed	Adults initiating CPAP for OSAHS Mean age 45 years 71% male	TH group had fewer 'walk-in visits' than the UC group (1/week vs 3/week)	No between group differences in adherence. Hours/night: TH: 4.29 (SD 2.15) vs UC: 4.32 (2.05) $p = 0.87$	Modified Functional Outcome of Sleep Q (reported - no differences between TH and UC group	No between group differences in client Satisfaction Questionnaire. TH: 28.5 (SD 3.1) vs UC 28.0 (SD 3.5) $p = 0.43$	Not measured		Very short follow-up (28 days)

CCT: controlled clinical trial; CPAP: continuous positive airway pressure; m: month; OSAHS: obstructive sleep apnoea/hypopnoea syndrome; RCT: randomised controlled trial; RoB: risk of bias; TH: telehealth; UC: usual care.

assessed as moderate risk of bias,[37] and four were at moderate-to-high risk of bias.[3,35,36,38] The commonest sources of bias were lack of information regarding sequence generation, allocation concealment, completeness of reporting, no published study protocol, and the inability to blind study personnel to the study intervention.

Description of the interventions

The characteristics of the included studies are described in detail (Supplementary Material, File 3), with a summary in Supplementary Material, File 5. All of the interventions were conducted by trained nurses, respiratory or CPAP therapists and physicians. The participants in all five studies had received face-to-face consultations to initiate CPAP therapy prior to entry to the trial.

Telemonitoring of CPAP usage data

Real time telemonitoring (via a modem) of CPAP adherence and efficacy was included in three studies.[3,36,38] Two studies used a home Web-connected telemonitoring system (participant activated) with telephone support.[35,37] In all studies, clinical personnel could access and monitor participants and intervene where indicated using predefined pathways.

Teleconsultation for remote review

Telephone support by the clinical research staff was used to recommend appropriate action where necessary in four studies.[3,35–37] Comma del Corral et al.[38] used a video conference system at a remote site hospital with the central base of the Sleep Research Unit 80 km away. Smith et al. used a home-based telemedicine system with a built-in modem and two-way camera to allow the study nurses to deliver a structured 12-week CPAP education/support programme.[35] In order to mimic the materials and activities of the intervention group, the control group received a 12-week programme on a neutral health topic which discussed the importance of daily vitamin intake. This method was chosen to control for the potential influence of having telehealth in the home and to equalise the Hawthorne effect of being observed through telehealth.[35]

Effectiveness of interventions

The findings of the studies are summarised in Table 2, with further details in Supplementary Material, File 4, and synthesised below.

Proportion reviewed

Two studies reported on number and/or duration of reviews reviewed. Fox et al. found that an additional hour over the three-month trial was spent with the telehealth patients compared with the usual care group.[3] In contrast, the teleconsultation/telemonitoring patients in Taylor et al.'s study in new users of CPAP had fewer 'walk-in visits' than the usual care group.[37]

Adherence to CPAP

In the study at moderate risk of bias[37] which recruited patients with a range of OSAHS severity, adherence to CPAP with teleconsultation/telemonitoring was similar to usual care even though the authors reported that problems with delivery of the telehealth (telephonic communication delays; and delays in delivery of equipment) may have diluted the intervention. Three lower quality studies reported improved CPAP adherence after the teleconsultation/telemonitoring intervention,[3,35,36] although two were relatively small pilot studies.[35,36] The CCT reported adherence as 75% in the teleconsultation/telemonitoring intervention versus 85% in those who received usual care.[38]

Control of symptoms

Two studies (both at moderate to high risk of bias) used the Epworth sleepiness scale to record symptom control[3,36] and the authors found no statistical difference between intervention and control groups. One study also used the Functional Outcome of Sleep Questionnaire (FOSQ) and found no statistically significant differences between intervention and usual care group for any of the component measures of this tool.[36] Two studies did not measure symptom control.[35,38]

Patient/clinician acceptability/satisfaction

All five studies reported on satisfaction with teleconsultation/telemonitoring verses face-face care. Four studies, using visual analogue scales[3,38] or questionnaire ratings[36–38] found no difference in satisfaction between teleconsultation/telemonitoring and usual care. One study[35] (at moderate/high risk of bias) reported on patient acceptability and satisfaction of telemonitoring compared to the control group. The authors undertook thematic analysis of the free text comments on their satisfaction survey; 100% of the surveys in teleconsultation/telemonitoring and 75% in usual care group rated the telehealth sessions positively (no statistical analysis reported); a key theme was that teleconsultation/telemonitoring was helpful in reinforcing the importance of CPAP adherence and they felt more supported with this intervention. None of the studies reported on acceptability to clinicians.

Costs of telehealth

Smith et al. reported that the cost of delivering 14 teleconsultation/telemonitoring sessions for a single participant was US $420 compared to US $1500 for face-face visits.[35] Coma-del-Corral et al. reported on the overall teleconsultation/telemonitoring costs (which included

diagnostic testing) but did not report the direct costs associated with delivering the CPAP telehealth review service.[38]

Discussion

Summary of findings

The evidence base for the effectiveness of remote consultations with telemonitoring in the clinical review of those using CPAP therapy is limited to two small studies[35,38] and three larger studies,[3,36,37] four of which were at moderate to high risk of bias[3,35,36,38] and one at moderate risk if bias.[37] These studies do not provide definitive evidence of effectiveness (in terms of adherence and symptom control) of teleconsultation/telemonitoring in CPAP users; however, there is no suggestion of any harms. The free text comments from a single study suggest that teleconsultation/telemonitoring was well received and perceived as being supportive.[35]

Strengths and limitations

We searched a wide range of databases and kept our search strategies broad, but we may have missed some studies. A key challenge for the review was our definition of telehealth. We defined this for the purposes of this review as an intervention which included both remote consultation either by telephone or videoconferencing and also the facility for real-time telemonitoring of CPAP data adherence. This meant we rejected interventions (for example) that used remote data on CPAP usage, but expected the patient to travel to follow-up consultations. We recognise, however, that by including the dual requirements of the facility for real-time telemonitoring and remote consultations, we have excluded a number of important studies. Our interest was in studies that facilitated CPAP clinical review and reduced the burden on the patient, and considered that not using remote consultations obviated an important potential benefit of telehealth. Limited resources meant we were unable to arrange duplicate independent screening at all stages of the selection process, but we undertook duplicate screening of a proportion of the abstracts and after discussion achieved 100% agreement.

Interpretation in the light of published literature

Telehealth is a rapidly developing field and since we undertook our searches, two potentially relevant studies have been published. The Fields et al. study is a prospective, parallel group pilot RCT,[39] which randomised 60 participants to telehealth (real time telemonitoring with telephone review) or usual (face-face) care. This study found no statistical between group differences in CPAP adherence, patient satisfaction, functional outcomes and dropout rates; though those randomised to telemedicine showed greater improvements in mental health scores. Their formal and verbal feedback with all parts of the study were overwhelmingly positive. Reinforcing the findings of this review, the authors concluded that larger scale trials are warranted in order to establish the benefits (or not) of telehealth for OSAHS.

An RCT by Mufano et al. ($n = 122$) compared a telehealth programme (with real time telemonitoring and telephone support from sleep therapists triggered by responses to automated texts/e-mails) compared to usual care in people newly diagnosed with OSAHS.[40] Reflecting the findings of this review, they found no statistical between group difference in adherence to CPAP therapy or change in Epworth sleepiness scale. However, the telehealth significantly reduced the time spent providing education and coaching making it a time-efficient option.

A number of studies are not included in our systematic review because they did not meet our inclusion criteria which required both the facility for real-time telemonitoring and teleconsultation (see Supplementary Material, File 2 for details). We regarded both features as important in maximising the benefits of telehealth in OSAHS. Nevertheless these studies may have some lessons of relevance to this review.

Six studies did not have the facility for real-time telemonitoring of CPAP,[6,19,22,23,29,34] though two retrieved the data at the end of the study.[6,23] This may have reduced the potential for the clinicians to detect real-time problems in remotely monitored participants such as poor/non adherence, poor mask fit/leak, and treatment efficacy that could have resulted in clinician intervention to resolve any detected problems. Remote consultations were generally well-received,[6,19,23] and one trial found the teleconsultations to be cost-effective (travel costs and lost work time were the most important sources of savings).[23]

Adherence remains a major clinical issue for users of CPAP therapy. Echoing the findings of this review, three of the teleconsultation RCTs showed no benefit in terms of adherence.[6,19,34] daytime sleepiness,[6] or quality of life.[6] In contrast, Sparrow et al., tested a telephone delivered theoretically based motivational intervention and showed a significant improvement in adherence, symptoms and functional status.[22]

Reasons for non-adherence are complex. In an overview of the many factors that influence adherence to CPAP therapy, Shapiro et al.[16] cite features of the CPAP device and side effects of treatment; as well as individual patient factors (such as clinical condition, family/home context, socio-economic status, personality, cognitive functioning), the attitude and communication skills of healthcare professionals, availability and efficiency of healthcare services, and national policy and funding of services.[16] More directly, the requirements of driving licensing agencies may have an important impact on adherence.[7] To be successful in promoting long-term adherence, sleep medicine clinicians need to monitor adherence, educate and coach their patients, and reinforce this education at every clinical review.

Access to real-time usage data is a tool to inform discussions about adherence.

Implications for research and future practice

Based on the results of the five trials included in this systematic review the evidence for remote teleconsultation with daily telemonitoring of CPAP usage in OSAHS is far from clear. Well-designed, adequately powered studies are required to clarify the role of teleconsultation combined with real time telemonitoring in the clinical review of people using CPAP therapy. Reflecting the free-text comments in one of the studies, telemonitoring was perceived as reinforcing the importance of adherence, whilst remote consulting has the potential to facilitate convenient reviews especially for those living at a distance from sleep centres. With the emergence of new technologies that enable remote teleconsultation and real time telemonitoring capability in-built into newer CPAP units, there is an opportunity for trials to build on the current evidence base and inform future practice in this area of specialised respiratory medicine.

Conclusions

The combination of remote consulting and real-time telemonitoring in CPAP therapy users has the potential to offer equivalent care that is more convenient, reduces travel, and is thus environmentally friendly. The technology to implement this model of sleep medicine service delivery is already available and being utilised in many healthcare settings, and driving research in this area. The limited evidence to date from published trials that have included these interventions has not raised any safety concerns, but adequately powered trials at low risk of bias will be needed to establish whether telehealth (combining remote consultation and real time telemonitoring) is a clinically viable, acceptable and cost effective option for people with OSAHS using CPAP therapy.

Acknowledgements

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Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

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Appendix 6 e-Delphi study protocol

Defining the essential components of a clinical review of people using Continuous Positive Airway Pressure (CPAP) therapy: protocol for an e-Delphi consensus exercise

Murphie P, Little S, McKinstry B, Pinnock H.

Introduction

Obstructive Sleep Apnoea Hypopnoea (OSAHS) is a major public health issue and is an important cause of morbidity and mortality (1-7). Epidemiological data reports its prevalence as 3–7% of middle-aged men and 2–5% of women (2-4). Globally, prevalence rates are increasing and this is directly related to obesity (2, 8-9) though a minority of individuals are non-obese (1, 2, 8). Other contributory factors are; enlarged tongue /tonsillar tissue, excess pharyngeal soft tissue and retro- or micro-gnathia (9, 10). Quality of life (QoL) is adversely affected (2, 7). The symptomatic consequences of OSAHS include; excessive daytime sleepiness, loud and socially disruptive snoring, nocturnal choking or gasping, poor unrefreshing sleep, mood changes, impaired alertness (sometimes when driving), morning headaches, nocturia and decreased libido (1, 2, 10-12). Driving-related accidents are a particular hazard (1) . Work-related performance issues are common, and include work-related incidents or accidents that may result in loss of employment (1, 7). The increased prevalence of OSAHS in the last two decades has seen an exponential rise in referral rates for investigation and treatment of this condition (2, 13).

After diagnosis the majority of individuals are established on fixed CPAP therapy (usually following auto-titration to establish therapeutic pressure settings). CPAP can improve morbidity and current guidelines recommend that patients using CPAP should be reviewed to assess adherence, replace disposables such as masks, filters and hoses, manage side effects and maintain CPAP devices (1, 12). CPAP adherence is problematic for reasons such as mask fit, comfort, prescribed CPAP pressure tolerance, oral-nasal dryness, claustrophobia, abdominal bloating, psychological and social factors (14, 15). Low adherence to CPAP limits its therapeutic benefit, (14) and addressing optimal – or at least adequate - CPAP use is a clinically important (16, 17) but an on-going time consuming problem in many sleep centres.

Current recommendations about clinical reviews of people using CPAP (Summarised in Table1)

National and International Guidelines in the Management and review of Adults with OSAHS do not provide clear recommendations on the essential components of a clinical review of a patient using CPAP and how often a review should take place. The (now withdrawn) SIGN guideline 73 did not include a specific section on the clinical review of patients using CPAP therapy (11).

The NICE Technology appraisal 139 –2008 - Continuous positive airway pressure for the treatment of obstructive sleep apnoea/hypopnoea syndrome - recommends that mask interfaces should be replaced annually (in clinical practice mask replacement parts are often required every 6-8 months) and patients on CPAP should be followed up long term. It makes no specific mention of clinical review and what should be assessed (12).

IMPRESS 2009 recommends regular follow-up of patients, monitoring and recording of patient compliance, symptoms and side-effects of treatment with frequent contact/access in the initial phase of CPAP acclimatisation to manage any side effects or problems (1). IMPRESS also recommends that once CPAP treatment has been established satisfactorily, the patient should remain under periodic review usually annually however for well controlled individuals this may be less frequent, combining clinical contact with a check on the machine (interface, headgear, filters,

and compliance). In the case of professional drivers, the DVLA require annual confirmation of symptom resolution and compliance with CPAP. There is no specific mention in IMPRESS of what the components of the clinical review should consist of e.g., weight, blood pressure, Epworth sleep score, mask fit, pressure setting comfort, chest auscultation (1).

The Australasian Sleep Association 2009 recommends that long-term follow-up for CPAP-treated patients should be conducted by appropriately trained health care providers and is indicated yearly and as needed to troubleshoot mask, machine, or usage problems (18).

The Spanish Society of Pulmonology and Thoracic Surgery 2011 do outline the timescale for when a clinical; review should take place following CPAP therapy commencement however do not describe the components of a clinical review (19).

The European Respiratory Society (ERS) European Lung White book 23 – 2014 - Sleep Breathing disorders does not include any recommendation on clinical follow up (2, 17). Nor does the Canadian Thoracic society guideline 2011 - Diagnosis and treatment of sleep disordered breathing (20).

The BMJ Best practice statement- Obstructive sleep apnoea in adults recommends patients with OSAHS receiving CPAP therapy should undergo periodic assessment of symptoms and CPAP adherence after initial regular use has been established (21).

The American Academy of Sleep Medicine provide a treatment and follow up algorithm which describes in some detail a general outcomes assessment following initiation of CPAP therapy and is more specific than other guidelines regarding what should be carried out at clinical review (22).

The American College of Physicians guideline 2015 on the management of OSAHS in adults does not address clinical review

The Consensus and evidence-based Indian initiative on obstructive sleep apnea guidelines published in 2015 do not adress the clinical review or follow up of people with OSAHS using CPAP therapy (23).

The recent NICE clinical knowledge summary Obstructive Sleep Apnoea syndrome 2015 includes a section on management in primary care, however does not specifically mention clinical review of people using CPAP therapy. It does however have a detailed paragraph on advice regarding driving for those with OSAHS (24).

	Weight	BP	Epworth Sleep Score	Reported symptoms	CPAP adherence	Mask interphase issues	Pressure setting/ comfort	Reported side effects	Chest auscultation	CPAP /issues /safety check	Driving/DVLA guidance
Sign 73											
Nice Technology appraisal 2008											
IMPRESS 2009				√	√			√		√	√
American Academy of Sleep Medicine 2009			√	√	√	√	√	√		√	
Australasian Sleep Association 2009					√	√				√	
Spanish Pulmonology Society /Thoracic surgery 2011											
Canadian thoracic society 2011											
BMJ Best practice statement				√	√						
American College of Physicians 2013											
ERS White book 2014											
Consensus and evidence based Indian Initiative 2015											
NICE 2015											√

Table 1- Elements included of a clinical review in current guidelines

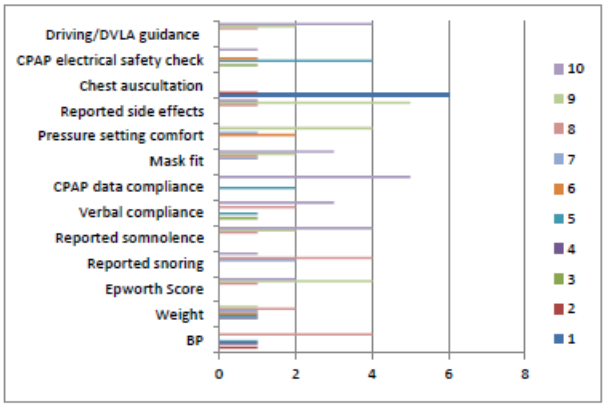
The need for consensus and potential components of a clinical review in those using CPAP therapy

It is evident from our literature review that current guidelines are not sufficiently explicit about what a clinical review of people with OSAHS using CPAP therapy should consist of and how often this should take place. There is thus a need to gain a consensus in order to inform future service delivery. With the recent publication of the UK Driver and Vehicle Licensing Agencies (DVLA) Assessing fitness to drive – a guide for medical professionals (25), it would appear that a consensus on the necessary components of a clinical review would assist clinicians in advising people with OSAHS on their requirements and obligation by law to notify the DVLA of this condition.

Possible components of a clinical review from current guidelines are: measures of adherence to CPAP, symptom control/resolution, side effects of therapy such as oro/nasal dryness, nasal congestion, nasal bridge ulceration and persisting sleepiness, mask fit, CPAP functionality, filter changes, timing of periodic review, OSAHS specific quality of life measures, satisfaction with therapy, avoidance of factors that worsen OSAHS, good sleep hygiene, weight loss where overweight/obese, recording of blood pressure and establishing if the patient has met DVLA requirements.

Preliminary pilot work on planning the clinical components of the e-Delphi study has been undertaken to inform the process. (Figure 1) 7 respondents out of 10 were asked to rate the importance of the components of the clinical review and results are outlined in the following table with 1 being least important and 10 being very important. Those clinicians who were consulted were respiratory physicians and specialist nurses who are delivering sleep medicine services. Their responses will form the basis of the initial list of components of a patient's CPAP clinical review in the formal e-Delphi study.

Figure 1



E-Delphi consensus technique

The Delphi method is a technique originating from the RAND Corporation in the 1950s, for reaching consensus amongst an expert panel in a systematic manner (26). An expert panel is recruited who contribute ideas, and then rank suggestions in successive rounds until a pre-defined consensus is reached. The panellists work independently and their contributions are anonymous, but in each round responses are influenced by summary feedback from previous rounds. As face-to-face discussion is not required, the exercise can be administered by e-mail.

Aim

To use an e-Delphi consensus process to identify and prioritise the essential components of a clinical review of a patient who uses CPAP therapy to treat OSAHS and how often a clinical review should take place.

Methods

We will follow standard consensus methodology to undertake a national and international Delphi exercise (26, 27).

Recruitment of an expert panel.

We will invite up to 50 participants and we would aim to recruit approximately 30 participants with up to 5 being expert patients recruited from a patient public involvement panel which reflects the aims of the Delphi exercise (i.e. to gain a consensus on the essential components of a clinical review in people with a diagnosis of OSAHS on CPAP therapy). It will therefore be important to include representation that encompasses international perspectives from clinicians who are actively managing people with OSAHS, with relevant academic expertise in this condition and service users.

Our recruitment strategy will therefore be to invite:

Healthcare professionals (e.g Clinical academics working in this field

(Respiratory Physicians, General Practitioners, Clinical Nurse Specialists, Nurse Practitioners, Physiologists who are actively reviewing people with OSAHS on CPAP therapy to participate in the study.

- We will e-mail potential members of the expert panel (using e-mails in the public domain for health care professionals) and we will also include a participant information sheet regarding the study with this email.

People with OSAHS recruited from patient organisations

- We will access expert patients via the European Patient Ambassador Programme (EPAP) and European Lung Foundation (ELF). EPAB will place information on their facebook page and website page regarding the purpose of the e-Delphi study and invite potential participants. The European Lung Foundation Director will assist with recruitment of expert patients to the study.

- The initial invitation email will include the Participant Information Sheet and an electronic consent statement. Potential participants will be asked to complete and return this to the study team to indicate their willingness to participate in the e-Delphi process.

Confidentiality of study data, including the identity of participants

- All study participants' comments, suggestions and data will be used anonymously and will not be attributed to named members of the expert panel. Summary statistics about the professional/lay backgrounds will be used to describe the characteristics of the expert panel.

- Participants may voluntarily waive anonymity if they would like to be acknowledged in publications as having contributed to the Delphi process.

- All respondents will be identified by a code number on all data. No hard copies of the study data will be kept.

- Only the research team will have access to the unique code number and identity of the participants.
- The unique code number for each participant will be allocated to them on return of their completed consent form and used throughout the study.
- This will be entered at the bottom of the form in the line 'for research office use only' by PM then this will be used to create the master list of participant names, ID's and email address.
- The unique study ID will also be entered into the participant's data collection spreadsheet before emailing each round to ensure that the correct ID matches the participant name in the master list.

- All electronic study data will be stored in a secure folder on the UoE shared space only accessible by the staff named above.

The three rounds of the e-Delphi will be:

Round 1: ***Open round - to compile a list for prioritisation of components of a clinical review in CPAP users.***

- We will start the e - Delphi process with a list of potential clinical review components from our literature review of current guidelines (table 1) and the local feedback colleagues outlined in Figure 1.
- We will ask the participants to add any additional components and/or comment on existing suggestions.
- We will also ask how often a clinical review should take place.

Panel members will be asked to return their completed responses within 2 weeks of receiving the initial email at the start of each round. A reminder will be sent to non-responders 2 weeks after the initial email.

Results will be collated on an Excel spreadsheet.

Round 2: First scoring round

- This round will be the first scoring round and will ask respondents to look across the clinical review components and prioritise them.
- Participants will be sent a list of potential components for the review (listed in alphabetical order) as collated from the open round and asked to score each component using a scale of 1-5 where 1 is unimportant and 5 is very important.
- Panel members will be asked to return their completed round 2 responses within 2 weeks, with a reminder being sent a few days before the deadline. A second reminder will be sent the day after the due date with a 3 day deadline.
- Results will be collated on an Excel spreadsheet, and an overall median score calculated for each research question.

Round 3: Second consensus scoring

The overall median scores will be entered onto the round 3 spreadsheet and fed back to individual panel members along with their own score. Panel members are then given the opportunity to revise their opinions on the overall priority of the clinical review components in the light of the findings of the previous round by again ranking each research question on a score of 1 to 5 again where 1 is unimportant and 5 is very important.

Panel members will be asked to return their completed spreadsheets within 2 weeks, with a reminder being sent a few days before the deadline. A second reminder will be sent the day after the due date with a 3 day deadline.

Reaching agreement

We anticipate that three rounds will allow an acceptable degree of agreement on the clinical review priorities, but if not a final fourth round, using the format of round 3, will be requested.

Definition of consensus

Consensus will be defined as 75% agreement with priority score of 4-5.

Piloting

There are three aspects to this:

- Piloting the participant invitations and instructions. Each member of the research team will invite a colleague (not an intended member of the expert panel) to comment on the invitation and instructions about the process.
- Timing the process. The members of the research team will time how long it takes them to complete the spreadsheets.

- Piloting the organisational procedures and data entry. By working through the process with the research team, I will be able to set up the required processes and identify any problems.

Organisation

The research team, led by Phyllis Murphie, will oversee the process and she will co-ordinate the exercise and will be responsible for reporting the results.

She will also co-ordinate sending out e-mail rounds, collating responses, setting up a database, preparing the second and third round spreadsheet.

Dissemination

The intention is to publish the results of the e - Delphi study in a peer reviewed journal.

All participants' comments, suggestions and scoring will be used anonymously however they may voluntarily waive anonymity regarding participation, if they would like their contribution to the Delphi process recognised in publications.

Timeline

Round 1 – April- May 2017	Round 2- May- June 2017	Round 3- June- July 2017
Up to 50 participants will be invited to participate (the reliability of the Delphi technique increases with the size of the group: we aim to recruit approximately 30 people) (26). They will be given an initial list of the possible components of a sleep medicine clinical review and will be asked to add any additional components. There will be an opportunity to add free text comments.	The suggestions and comments from round 1 will be collated and the candidate components listed in alphabetical order Participants will be asked to rank the components of the clinical review in order of importance for inclusion in a clinical review of a patient using CPAP therapy should include.	The median scores from round 2 will be calculated and spreadsheets prepared for round 3. The scoring process will be repeated in a third round to refine the components of the clinical review to gain a consensus from the expert panel.

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Appendix 7 e-Delphi study ethics approvals

University of Edinburgh,
Usher Institute of Population Health Sciences and Informatics
RESEARCH ETHICS SUBGROUP

Self-Audit Checklist for Level 1 Ethical Review for PGR projects

See **Intra** website for further information: http://www.cphs.mvm.ed.ac.uk/intra/research/ethicalReview.php

NOTE to student: *Completion of this form should be under the **oversight** of your supervisor. A good strategy would be to complete a draft as best you can, then discuss with your supervisor before completing a final copy for your supervisor to sign.*

***Proposed Project** (State research question and topic area, and briefly describe method/ data. Specify also countries in which data will be collected.):*

Defining the essential components of a clinical review of people using Continuous Positive Airway Pressure (CPAP) therapy: protocol for an international e-Delphi consensus exercise

Murphie P, Little S, McKinstry B, Pinnock H.

The need for consensus and potential components of a clinical review in those using CPAP therapy

National and international guidelines on the management of people with OSAHS using CPAP recommend regular reviews. However, we conducted a literature review in August 2016, and it is evident that current guidelines do not state explicitly what are the essential components of a clinical review of people with OSAHS using CPAP therapy, and how often a review should take place. There is a need to gain a consensus in order to inform future service delivery.

Aim

To use an international e-Delphi consensus process to identify and prioritise the essential components of a clinical review of a patient who uses CPAP therapy to treat OSAHS, and to agree how often this review should take place.

Methods

We will follow standard consensus methodology to undertake a national and international e-Delphi exercise.

Recruitment of an expert panel.

We will recruit an international expert panel of approximately 30 people to achieve the aims of the e-Delphi exercise (i.e. to gain a consensus on the essential components of a clinical review in people with a diagnosis of OSAHS on CPAP therapy). It will be important to include representation which encompasses international perspectives from clinicians who are actively managing people with OSAHS, as well as those with academic expertise in this condition and people using CPAP.

Our recruitment strategy will therefore be to invite:

- Healthcare professionals (e.g. Respiratory Physicians, General Practitioners, Clinical Nurse Specialists, Nurse Practitioners, Physiologists who are actively reviewing people with OSAHS on CPAP therapy) to participate in the study.
- Clinical academics working in this field
- People with OSAHS recruited from patient organisations

We will e-mail potential members of the expert panel (using e-mails addresses in the public domain for professionals; via patient organisations for expert patients), inviting them to participate. The invitation will include a description of the process, the anticipated timescale, and estimated commitment. The three rounds of the e-Delphi will be:

1. An open round in which participants will suggest components they consider to be important in a review
2. A first scoring round in which participants will score the importance of the candidate components
3. A second scoring round in which the median scores from the second round are fed back and participants are invited to re-consider their score.

We will request participants to contribute to all three rounds as this forms the basis of consensus methodology.

1. Bringing the University into disrepute

Is there any aspect of the proposed research which might bring the University into disrepute? NO

2. Data protection and consent

Are there any issues of DATA PROTECTION or CONSENT which are NOT adequately dealt with via established procedures? NO

These include well-established sets of undertakings. For example, a 'No' answer is justified *only if*:

a. There is compliance with the University of Edinburgh's Data Protection procedures (see www.recordsmanagement.ed.ac.uk);
b. Respondents give consent regarding the collection, storage and, if appropriate, archiving and destruction of data;
c. Identifying information (eg consent forms) is held separately from data;
d. There is Caldicott Guardian approval for (or approval will be obtained prior to) obtaining/ analysing NHS patient-data.
e. There are no other special issues arising about confidentiality/consent.

3. Study participants

a) Will a study researcher be in direct contact with participants to collect data, whether face-to-face, or by telephone, electronic means or post, or by observation? (eg interviews, focus groups, questionnaires, assessments) YES

b) **Answer this only if qu. 3 above = 'YES':**
*In ethical terms, could any participants in the research be considered to be '**vulnerable**'?*
e.g. children & young people under age of 16, people who are in custody or care (incl. school), a marginalised/stigmatised group

Please tick one: 'vulnerable' ☐ not 'vulnerable' ☐

4. Moral issues and Researcher/Institutional Conflicts of Interest

Are there any SPECIAL MORAL ISSUES/CONFLICTS OF INTEREST? NO

a. An example of conflict of interest for a researcher would be a financial or non-financial benefit for him/herself or for a relative of friend.
b. Particular moral issues or concerns could arise, for example where the purposes of research are concealed, where respondents are unable to provide informed consent, or where research findings could impinge negatively/ differentially upon the interests of participants.
c. Where there is a dual relationship between researcher and participant (eg where research is undertaken by practitioners so that the participant might be unclear as to the distinction between 'care' and research).

5. Protection of research subject confidentiality

Are there any issues of CONFIDENTIALITY which are NOT adequately handled by normal tenets of confidentiality for academic research? NO

These include well-established sets of undertakings that should be agreed with collaborating and participating individuals/organisations. For example, a 'No' answer is justified *only if*:

a. There will be no attribution of individual responses;
b. Individuals (and, where appropriate, organisations) are anonymised in stored data, publications and presentation;
c. There has been specific agreement with respondents regarding feedback to collaborators and publication.

6. Protection of research subject confidentiality

Are there any issues of CONFIDENTIALITY which are NOT adequately handled by normal tenets of confidentiality for academic research? NO

These include well-established sets of undertakings that should be agreed with collaborating and participating individuals/organisations. For example, a 'No' answer is justified *only if*:

a. There will be no attribution of individual responses;
b. Individuals (and, where appropriate, organisations) are anonymised in stored data, publications and presentation;
c. There has been specific agreement with respondents regarding feedback to collaborators and publication.

1. Protection of research subject confidentiality

Are there any issues of CONFIDENTIALITY which are NOT adequately handled by normal tenets of confidentiality for academic research? NO

These include well-established sets of undertakings that should be agreed with collaborating and participating individuals/organisations. For example, a 'No' answer is justified *only if*:

a. There will be no attribution of individual responses;

b. Individuals (and, where appropriate, organisations) are anonymised in stored data, publications and presentation;

c. There has been specific agreement with respondents regarding feedback to collaborators and publication.

2. Potential physical or psychological harm, discomfort or stress

a. Is there a FORSEEABLE POTENTIAL for PSYCHOLOGICAL HARM or STRESS for participants? NO

b. Is there a FORSEEABLE POTENTIAL for PHYSICAL HARM or DISCOMFORT for participants? NO

c. Is there a FORSEEABLE RISK to the researcher? NO

Examples of issues/ topics that have the potential to cause psychological harm, discomfort or distress and should lead you to answer 'yes' to this question include, but are not limited to:

relationship breakdown; bullying; bereavement; mental health difficulties; trauma / PTSD; violence or sexual violence; physical, sexual or emotional abuse in either children or adults.

3. Duty to disseminate research findings

Are there issues which will prevent all relevant stakeholders having access to a clear, understandable and accurate summary of the research findings if they wish?* NO

** If, and only if, you answered 'yes' to 3 above, 'stakeholders' includes the participants in the research*

Overall assessment

- If every answer above is a definite NO, the self-audit has been conducted and confirms the ***ABSENCE OF REASONABLY FORESEEABLE ETHICAL RISKS*** – *please tick box* ☐

  *This means that regarding this study, as currently self-audited, no further ethical review actions are required within Usher. However, if in the coming weeks/months there is any change to the research plan envisaged now (and outlined above), the study should be **re-audited** against a Level 1 form, because it may be that the change made negates the absence of ethical risks signed off here.*

- If one or more answers are YES, then risks have been identified and prior to commencing any data collection **formal ethical review** is required - either:

  ~ by NHS REC (NB copy of ethics application and decision letter to be sent to Usher Ethics); or

  ~ if not to be formally reviewed by NHS REC, then Usher level 2/3 ethical review required.

  [If either 4 is 'yes' or 3b is 'vulnerable' then it is possible level 3 review is required.]

Two copies of this form should be taken for inclusion in the final dissertation/thesis and the original should be returned to the Usher Ethics administrator.

Phyllis Murphie	Hilary Pinnock
Student Name	Supervisor Name
Student Signature	Supervisor Signature *

*** NOTE to supervisor:** *The Usher Ethics Subgroup will not check this form (the light touch Level 1 form means we have insufficient detail to do so). By counter-signing this check-list as truly warranting all 'No' answers, **you** are taking responsibility, on behalf of Usher and UoE, that the research proposed truly poses no potential ethical risks. Therefore, if there is any doubt on any issue, it would be a wise precaution to mark it as 'uncertain' and contact the Ethics Subgroup as to whether a level 2 form might be required as well. (See Intra Ethics website – URL at top of form)* *13 Dec 2016*

Appendix 7 Continued

CENTRE FOR POPULATION HEALTH SCIENCES

Research Ethics Application - Level 2/3

Due ethical process/'approval' is required for *all research projects* carried out by staff or students in the Centre for Population Health Sciences (CPHS).

Prior to completion of any forms, applicants should familiarise themselves with the ethical review process within CPHS - http://www.cphs.mvm.ed.ac.uk/intra/research/ethicalReview.php

All research projects require at least a completed and signed Level 1 form. In the case of a PG student applicant, any form required needs to be completed under the oversight of the student's supervisor.

This Level 2/3 form should be used for all projects that have been identified, by the Level 1 Self-Audit form, as requiring ***formal ethical review*** - i.e. level 2 / 3, within the three-tier system of ethics approval set out by the **School of Health in Social Science Research Ethics Committee**.

The completed form should be submitted to the CPHS Subject Area Research Ethics Group-

- All level 2/3 applications should be submitted via the CPHS Ethics administrator - cphs.ethics@ed.ac.uk
- For PG research and masters students the application should also be copied to the PG Administrator - cphs.pg@ed.ac.uk

Research must not proceed before ethical approval has been granted. For this reason you are advised to: a) Submit your application well in advance of any required date of approval, particularly for level 3 research;

b) Acknowledge any potential for ethical concern, even minimal, and use your responses on the form, and additional documentation submitted with the form, to address this, detailing steps to be/being taken to minimise discomfort/ distress.

Name of Applicant:	Phyllis Murphie
If applicant is a PG student: Name of Supervisor	Professor Hilary Pinnock
Confirm supervisor has checked completion of form	*Please tick if yes*
If UG/MPH, also give Student Exam number	
Project Title:	Defining the essential components of a clinical review of people using Continuous Positive Airway Pressure (CPAP) therapy: protocol for an e-Delphi consensus exercise
Funding body:	Self-funded
Level of review requested (2 or 3)	2
Expected timescale for the research	6 months
Date this ethics application submitted:	18-1-17

List those who will be involved in conducting the research, including names and positions (e.g. 'PhD student')	Phyllis Murphie - PhD Student Dr Stuart Little - Respiratory Physician Professor Brian McKinstry - Professor Primary Care EHealth Professor Hilary Pinnock - Professor of Primary Care Respiratory Medicine

LEVEL 2 / 3 ETHICAL REVIEW

OVERVIEW of documentation provided in support of your application *

You are advised to submit copies of: consent form, participant information, questionnaire, interview guide, vignettes, disclosure, GP letters etc

For any such additional documents submitted, please enter below the document names - *Please ensure 'names' listed below match the actual names of the doc-files. It us helpful to admin / review if document names indicate content in their initial text (e.g. staff interview guide, participant interview guide), with additional detail coming towards the end (e.g. version number, date etc).*

Document name	**Tick ✓**	**Document name** *(enter – & add rows if needed)*	**Tick ✓**
Level 1 Ethics Form - e - Delphi - 181-17 -Rev1	√	e-Delphi excel spreadsheet - Ver - 1	√
Level 2 ethics form -e-Delphi Rev 1 18-1-17	√	Participant invitation email	√
e-Delphi protocol - Rev1 18-1-17	√	Ethics resubmission letter -18-1-17	√
Letter to participants Rev 1-highlighted 18-1-17	√		

***** We *do not accept paper documentation so, if necessary, please scan all documents into electronic format for submission to CPHS Ethics Group. Please keep a copy of all documentation for your records.*

STRUCTURE OF REST OF FORM Sections: Item numbers

Page *(on blank form!)*

Risks to, and safety of, researchers	1 – 3	2
Risks to, and safety of, participants	4 – 7	3
Research design	8 – 22	3
Data protection	23 – 33	5

How to complete form:

Please give some response to every item. The response boxes will expand as you complete them.

Where a Yes/No response choice is offered, please delete as applicable. If the answer to such a question is '**Yes**', you ***must*** elaborate i.e. give details as to how this issue will be addressed to ensure that ethical standards are maintained, and/or provide a case as to why the concern does not prevent ethical approval being given. [For information/background about ethical issues in research, please refer to ethics resources made available on CPHS Ethics website *(URL on page 1)* and on H drive *(path H:\Research support docs\Governance\Ethics).*]

The more comprehensive the detail/ explanation given in and with the form, in particular regarding the key ethical issues *(vulnerable participants, recruitment/consent process, data collection method, confidentiality/anonymity)*, the quicker should be your receipt of ethical opinion. Forms that do not contain sufficient detail will be returned, incurring delay.

RISKS TO, AND SAFETY OF, RESEARCHERS NAMED IN THIS APPLICATION
Do any of those conducting the research named above need appropriate training to enable them to conduct the proposed research safely and in accordance with the ethical principles set out by the College?
NO PM has read the ethical principles set out by the College to enable conducting the proposed research in accordance with these ethical principles. PM has had previous experience with conducting research within her NHS role, however her previous GCP training has expired. Although not required for this research project PM has enrolled for a GCP training course on the 23rd February 2017 at Edinburgh University as this will be required for future research in her PhD.
Are any of the researchers likely to be sent or go to any areas where their safety may be compromised, or they may need support to deal with difficult issues?
NO
Could researchers have any conflicts of interest?
NO

RISKS TO, AND SAFETY OF, PARTICIPANTS	
4. Regarding whether any participants are children or protected adults: (protected adults are those in receipt of registered services - i.e. care, health, community care or welfare services) Note: Any researcher who will have contact in Scotland with children, or protected adults, requires approval from Disclosure Scotland – *see* http://www.disclosurescotland.co.uk/index.htm	
Does this study require Disclosure Scotland approval for any of its researchers? *If yes, please* ✓ If *yes*, ethical approval will be *subject to documentation confirming Disclosure Scotland approval*.	
5. Could the research induce any psychological stress or discomfort in participants?	
NO	
6. Does the research involve any physically invasive or potentially physically harmful procedures?	
NO	
7. Could this research adversely affect participants in any other way?	
NO	

RESEARCH DESIGN
8. Does the research involve living human subjects specifically recruited for this research project If **'no'**, skip items 9 to 22 i.e. go to next section (**Data Protection**, item 23 onwards)
YES
9. How many participants will be involved in the study? (please give justification of number proposed)

We aim to recruit approximately 30 participants to complete an e-Delphi consensus process. To achieve this we will invite up to 50 international experts, including up to five expert patients recruited from a patient public involvement panel.

This sample size has been selected because Delphi panels are generally under 50;[1] and the majority of Delphi studies have included between 20-30 respondents. To obtain an international consensus with a broad range of participants we will invite 50 people with a view to recruiting at least 30 to the process.

1 Avella, J. R. (2016). Delphi panels: Research design, procedures, advantages, and challenges. International Journal of Doctoral Studies, 11, 305-321. Retrieved from http://www.informingscience.org/Publications/3561

10. What criteria will be used in deciding on inclusion/exclusion of participants?

Participants will be an international range of policymakers, guideline writers, leading academics in the field, expert patients, and specialist healthcare professionals who are involved in the clinical review of people with obstructive sleep apnoea hypopnoea syndrome who are using continuous positive airway pressure treatment. Participants will be identified by their publications, prominent role in conferences, professional bodies or guideline committees. We will approach patient groups (e.g. the European Lung Foundation – European Patient Ambassador Participation group) to identify lay advisors for the expert group.

11. How will the sample be recruited? (e.g. posters, letters, a direct approach- specify by whom)

Professional participants will be recruited by a covering email (from PM – principal investigator) with information regarding the purpose of the e-Delphi study and an attached word document that will outline the study requirements for potential participants. All professional participants' emails and contact details are in the public domain.

Patients will be recruited via their organisation.

Reminder e-mails will be sent within 2 weeks for non-respondents.

12. Will the study involve groups or individuals who are in custody or care? - such as students at school, self-help groups, residents of nursing home

NO

13. Will there be a control group?

NO

14. What information will be provided to participants prior to their consent? (e.g. information leaflet, briefing session)

YES An email letter will be sent to all participants explaining the purpose of the e-Delphi study, and exactly what participation will involve. Contact details (for PM) will be provided to enable potential participants to request further information.

15. Participants have a right to withdraw from the study at any time.

Please ✓ to confirm that participants will be advised of their rights, including the right to continue receiving services if they withdraw from the study.

16. Will it be necessary for participants to take part in the study without their knowledge and consent? (e.g. covert observation of people in non-public places)

NO

17. Where consent is obtained, what steps will be taken to ensure that a written record is maintained?

The e-Delphi is an electronic format, so there will not be any paper records. The first part of the first round spreadsheet will include a section that requires the participants to indicate that they have received information and consent to participate in the e-Delphi study by entering 'Yes' in the appropriate column.

18.	**In the case of participants whose first language is not English, what arrangements are being made to ensure informed consent?**
	The e-Delphi will be conducted in English, the international language of academic conferences and publications. It will therefore not disadvantage the professional members of the team. The expert patients will need to be able to communicate in English to complete the process. This is a PhD project and we do not have budget to pay for translation.
19.	**Will participants receive any financial or other benefit from their participation?**
	NO
20.	**Are any of the participants likely to be particularly vulnerable, such as elderly or disabled people, adults with incapacity, your own students, members of ethnic minorities, or in a professional or client relationship with the researcher?**
	NO
21.	**Will any of the participants be under 16 years of age?**
	NO
22.	**Will any of the participants be interviewed in situations which will compromise their ability to give informed consent, such as in prison, residential care, or the care of the local authority?**
	NO N/A: there are no interviews planned

DATA PROTECTION
23. Will any part of the research involve audio, film or video recording of individuals?
NO
24. Will the research require collection of personal information from any persons without their direct consent?
NO The only personal information collected will be contact details (for use in conducting the process) and information about professional role as provided by the participant.
25. How will the confidentiality of data, including the identity of participants (whether specifically recruited for the research or not) be ensured?
Only the research team named above will have access to the identity of the participants. This is required in order to a) send reminders to non-responders; b) feedback individual's median scores for the second scoring round; c) send a copy of the publication to the participants. All electronic data will be in a secure folder on the UoE shared space only accessible by the staff named above. All respondents will be identified by a code on all data. No hard copies of the study data will be kept. The participants will not be identified by name – only their professional/lay background will be reported in results.
26. Who will be entitled to have access to the raw data?
Only the research team (PM and her supervisors HP, BMcK).
27. How and where will the data be stored, in what format, and for how long?
Data will be stored electronically in a secure UoE folder only accessible to PM, and HP, BMcK. It will be retained for the duration of the study, and thereafter be archived according to University regulations for 5 years.
28. What steps have been taken to ensure that only entitled persons will have access to the data?
MVM IT staff have permitted electronic access only to those staff named above. Access to a folder on the UoE shared drive will be set up which is accessible to PM, HP and BMcK.
29. How will the data be disposed of?
In accordance with the UoE Research Data Management policy May 2011.
30. How will the results of the research be used?

This e-Delphi is a crucial component of PM's PhD, in which she is developing a telehealth intervention to deliver routine reviews for people using CPAP for OSAHS. In the absence of clear guideline recommendations for what should be included in a review, the e-Delphi will provide the required consensus to inform a telehealth review. We will present abstracts, publish the results in a peer reviewed journal. The findings will be of interest to guidelines which currently to not make clear recommendations about CPAP/OSAHS reviews.
31. What feedback of findings will be given to participants?
A copy of the peer reviewed publication will be emailed to the participants, with a lay summary.
32. Is any information likely to be passed on to external companies or organisations in the course of the research?
NO
33. Will the project involve the transfer of personal data to countries outside the European Economic Area?
NO

THE UNIVERSITY *of* EDINBURGH

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05 June 2017

Ms P Murphie

Dear Phyllis

Re: Defining the essential components of a clinical review of people using Continuous Positive Airway Pressure (CPAP) therapy: protocol for an e-Delphi consensus exercise

The Usher ethics committee have reviewed the additions to the documentation for your ethics application for the above study, to cover Round 2 of the Delphi process planned.

Ethics approval for the above study is hereby extended up to *and including the* ***second*** round of the Delphi survey process.

However, as you know, the Ethics committee have not seen the survey forms for subsequent stages (or what is most likely, the third and final stage). Therefore you are granted only ***conditional approval*** for the subsequent stage(s). Once each of these survey forms in turn is ready, and you submit same to the Committee, full approval of these stages should be a formality, and speedy, since it will be only the form that needs to be checked. There should be no data collection for any of the subsequent stages until the relevant survey form has been submitted and approval given.

Please be aware that this ethical approval is in respect of the protocol and methods as described in the documents submitted to the committee (with amended documents superseding predecessors). If there is in the future *a change* to the study design/protocol/methods, you should check whether this means your level 2 application form needs to be revised, and submit to the committee (via me), any documents that have been revised (study materials/protocol/level 2 form), using tracked changes. You should make clear in your covering email whether:

(i) you are requesting ethical review of a study amendment; or

(ii) you are not sure whether such is needed and, in the first instance, would like the committee's opinion on whether a formal approval is needed of the amended design/methods.

Yours sincerely

Diane White
Ethics Review Group Administrator

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01 August 2017

Ms P Murphie

Dear Phyllis

Re: Defining the essential components of a clinical review of people using Continuous Positive Airway Pressure (CPAP) therapy: protocol for an e-Delphi consensus exercise

The Usher ethics committee have reviewed the additional documentation that you have submitted for your ethics application for the above study, to cover Round **3** of the Delphi process planned.

Ethics approval for the above study is hereby extended up to *and including the **third*** round of the Delphi survey process, that is for the **full study**.

Please be aware that this ethical approval is in respect of the protocol and methods as described in the documents submitted to the committee (with amended documents superseding predecessors). If there is in the future *a change* to the study design/protocol/methods, you should check whether this means your level 2 application form needs to be revised, and submit to the committee (via me), any documents that have been revised (study materials/protocol/level 2 form), using tracked changes. You should make clear in your covering email whether:

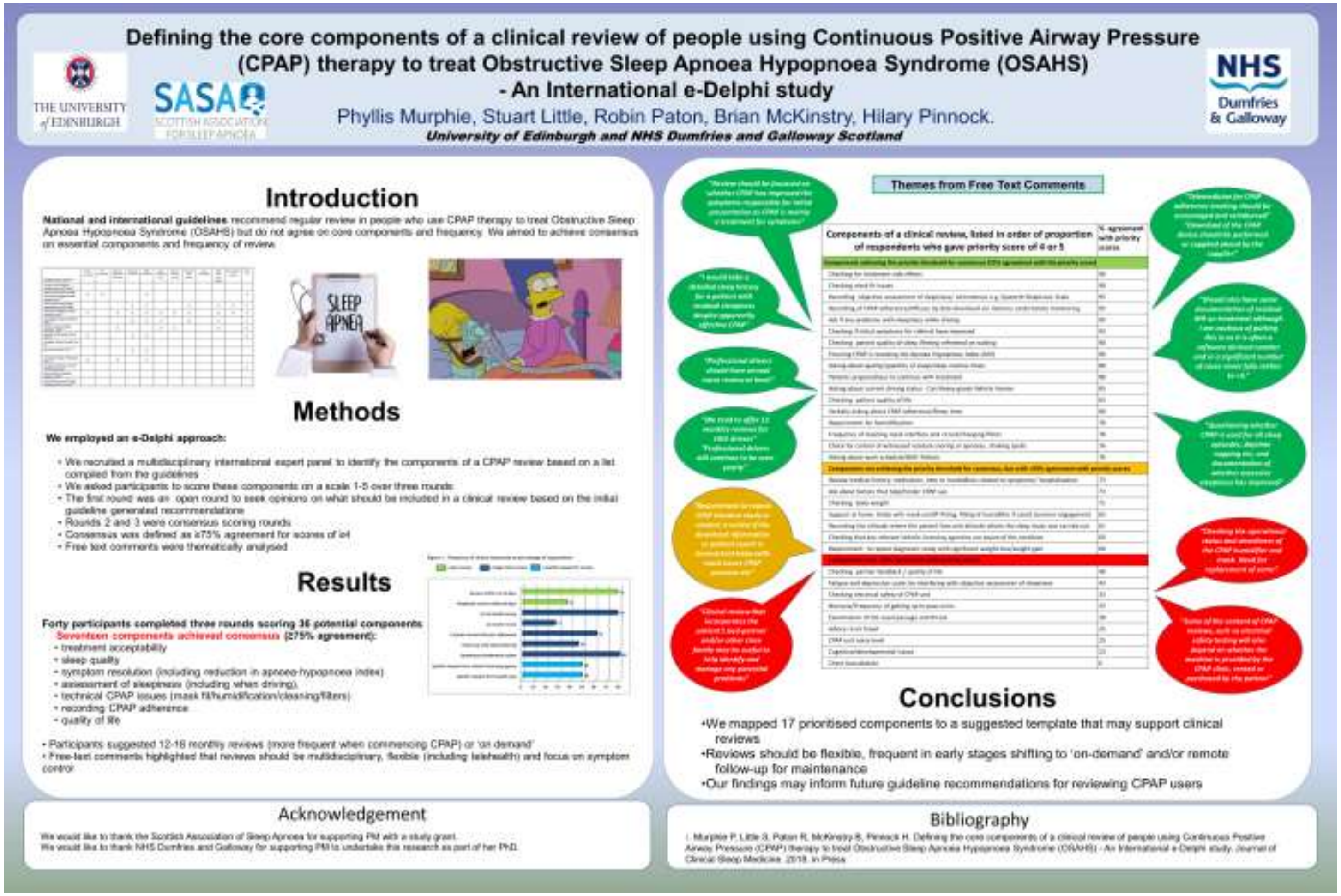
(i) you are requesting ethical review of a study amendment; or

(ii) you are not sure whether such is needed and, in the first instance, would like the committee's opinion on whether a formal approval is needed of the amended design/methods.

Yours sincerely

Diane White
Ethics Review Group Administrator

Appendix 8 e- Delphi study poster presented at ERS Paris 2018



Appendix 9 e-Delphi study published in the Journal of Clinical Sleep medicine

pii: jc-18-00151 http://dx.doi.org/10.5664/jcsm.7372

JCSM
Journal of Clinical Sleep Medicine

SCIENTIFIC INVESTIGATIONS

Defining the Core Components of a Clinical Review of People Using Continuous Positive Airway Pressure Therapy to Treat Obstructive Sleep Apnea: An International e-Delphi Study

Phyllis Murphie, RGN, MSc[1,2]; Stuart Little, MD[1]; Robin Paton, BN[1]; Brian McKinstry, MD, PhD[3]; Hilary Pinnock, MD, PhD[3]

[1]Allergy and Respiratory Research Group, Usher Institute of Population Health Sciences and Informatics, The University of Edinburgh, United Kingdom; [2]Department of Respiratory Medicine, NHS Dumfries and Galloway, United Kingdom; [3]e-Health Research Group, Usher Institute of Population Health Sciences and Informatics, The University of Edinburgh, United Kingdom

Study Objectives: Guidelines recommend regular review of individuals using continuous positive airway pressure (CPAP) to treat obstructive sleep apnea but do not agree on the core components and frequency. We aimed to achieve consensus on essential components and frequency of review.

Methods: We used an e-Delphi approach, recruiting a multidisciplinary international expert panel to identify components based on a list compiled from guidelines and to score these on a scale 1 to 5 over three rounds. Consensus was defined as ≥ 75% agreement for scores of 4 or higher. Free-text comments were thematically analyzed.

Results: Forty participants completed 3 rounds scoring 36 potential components. Seventeen components achieved consensus: treatment acceptability, sleep quality, symptom resolution (including reduction in apnea-hypopnea index), assessment of sleepiness (including when driving), technical CPAP issues (mask fit/humidification/cleaning/filters), recording CPAP adherence, and quality of life. Participants suggested 12 to 18 monthly reviews (more frequent when commencing CPAP) or "on demand." Free-text comments highlighted that reviews should be multidisciplinary, flexible (including telehealth), and focus on symptom control.

Conclusions: We mapped 17 prioritized components to a suggested template that may support clinical reviews. Reviews should be flexible, frequently in the early stages of commencing CPAP, shifting to "on demand" and/or remote follow-up for maintenance. Our findings may inform future guideline recommendations for reviewing CPAP users.

Keywords: continuous positive airway pressure, CPAP, obstructive sleep apnea, OSA, routine review, sleep apnea

Citation: Murphie P, Little S, Paton R, McKinstry B, Pinnock H. Defining the core components of a clinical review of people using continuous positive airway pressure therapy to treat obstructive sleep apnea: an international e-Delphi study. *J Clin Sleep Med*. 2018;14(10):1679–1687.

BRIEF SUMMARY

Current Knowledge/Study Rationale: Current sleep medicine guidelines recommend regular review in individuals using continuous positive airway pressure (CPAP) and who have obstructive sleep apnea. However, they do not collectively define the core components and frequency of such a review. We aimed to achieve consensus on essential components and frequency of review.

Study Impact: This is the first study to provide an international consensus on the most important components that may be considered when reviewing people using CPAP therapy. Our findings may inform future guideline recommendations for reviewing individuals using CPAP.

INTRODUCTION

Obstructive sleep apnea (OSA) is a common, treatable condition that represents a major public health issue globally and is an important cause of morbidity and mortality.[1–8] It has been recognized as an important respiratory condition for more than 36 years since the groundbreaking publication by Sullivan et al. in *The Lancet* in 1981.[9] OSA is usually a lifelong condition that requires long-term treatment such as continuous positive airway pressure (CPAP) in order to relieve symptoms, improve quality of life, mitigate the effect of daytime sleepiness on work-performance and driving-related accidents, and reduce the risk of cardiovascular comorbidity.[1,10–12]

Current guidelines recommend regular review for individuals undergoing CPAP therapy and suggest a range of components that could be included such as assessment of daytime sleepiness using the validated Epworth Sleepiness Scale, adherence to CPAP, practical issues with masks and equipment, guidance regarding driving privileges, and weight and blood pressure monitoring. However, the guidelines give conflicting advice about the core components of a clinical review (**Table 1**) and how often this should take place, and there is therefore a need for consensus.

An e-Delphi is a method that can be used for reaching consensus among a panel of experts where there is limited evidence on the priority attached to a range of items.[13,14] Communication can take place by email, enabling participation by national

and international participants over a short period of time.[14] We conducted an international e-Delphi study to reach consensus on the important core components and optimal frequency of a clinical review in people using CPAP therapy for OSA.

METHODS

Ethics

We obtained ethical approval from the Usher Institute of Population Health Sciences and Informatics, University of Edinburgh (application number 1700).

e-Delphi Methodology

Stemming from the RAND Corporation in the 1950s,[15] an e-Delphi delivers a series of questionnaires over (typically) three rounds in which expert panelists contribute their ideas independently and anonymously. In subsequent rounds individual responses may be influenced by feedback of the collective participant responses from previous rounds facilitating consensus.

Guideline Review and Pilot Work

We identified current guidelines, position statements, best practice statements/recommendations, and consensus statements for the management of adults with OSA therapy, searching Medline, Turning Research Into Practice databases, and Google Scholar using the following search terms "sleep apnoea/apnea syndrome," "CPAP therapy," "national/international guidelines," "clinical review," and "follow up." We scrutinized these documents for recommendations about the components of regular review of CPAP users and extracted all suggested elements of a review to form an initial list of possible components. Follow-up was explicitly stated as within scope for 7 of the 13 included publications in our study. We then piloted the e-Delphi process with 10 local sleep medicine clinicians who were asked to "sense-check" the review components from the literature review; any additional components they considered to be important would be added to the initial list.

Recruitment of an Expert Panel

Delphi panels are generally fewer than 50 participants; and most Delphi studies have included 20 to 30 respondents.[16] We therefore invited, by email, 80 international experts with a view to recruiting approximately 30 participants to the study. The clinicians who were involved in the pilot work were excluded from the expert panel. Our recruitment strategy was to invite health care professionals who were actively involved in the review of people with OSA who were using CPAP therapy (eg, clinical academics, respiratory physicians, general practitioners, clinical nurse specialists/nurse practitioners, respiratory therapists, respiratory physiologists—health care scientists trained to support people using CPAP). We also invited up to five individuals with OSA using CPAP from a local service. Our aim was to encompass both clinical experiences with relevant academic expertise as well as individuals undergoing CPAP therapy. All suggestions, comments, and data were anonymized but participants were offered the option of being acknowledged in publications.

The Three Rounds of the e-Delphi

We followed recommended consensus methodology,[17,18] and anticipated that it would require up to three rounds to reach consensus with a fourth round if required. The data collection sheets for the three rounds are in **Appendix 1** in the supplemental material.

Round 1: Open Round

Initially we developed a data collection sheet (using an Excel spreadsheet) of potential clinical review components from our literature review of current guidelines combining any additional suggestions from our pilot work. The Round 1 data collection sheet was then emailed to the expert panel requesting any additional review components and/or free-text comments on the existing suggestions. Opinions were also sought regarding the importance of the timing and suggested frequency of clinical review. We collated all the additional suggested components to create the final list for prioritization in Round 2.

Round 2: First Scoring Round

This was the first scoring round and the panel were asked to review the list generated from Round 1 and identify the components that should be prioritized in a review using a scale of 1 to 5 (1 = unimportant and 5 = very important). We avoided ranking because the importance of specific prioritized components would depend on individual clinical context. The results were collated and a median score calculated for each component in preparation for Round 3.

Round 3: Second Scoring Round

The Round 3 data collection sheet included the median scores from Round 2 along with individuals' own Round 2 score. In Round 3 the participants were given the opportunity to revise their opinions (or not) on the priority of the clinical review components in light of the median findings of the previous round by again ranking each research question on a score of 1 to 5 (where 1 = unimportant and 5 = very important). We predicted an acceptable level of agreement on priority components with three rounds; however, a final fourth round (following the method of the Round 3) could be conducted if required.

Analysis

We calculated the median scores for each component of the clinical review and the proportion of respondents scoring each item as 4 or 5. In discussion with the multidisciplinary team, consensus was defined as ≥ 75% agreement for the priority scores of 4 or 5. Prioritized components were grouped (eg, treatment acceptability, technical CPAP issues, sleepiness assessment, adherence, symptom resolution, assessing sleep quality, driving issues, quality of life, lifestyle issues/sleep hygiene) and mapped to a template that could be used to facilitate a standardized review.

Free-Text Comments From Participants

Participants were invited to contribute their additional free-text comments in all the rounds. We used an inductive approach to thematically analyze the free-text comments to identify the key issues from the perspective of the individual participants.

Table 2—Components of a clinical review, listed in percentage order of proportion of respondents who gave priority score of 4 or 5.

Components of a Clinical Review, Listed in Order of Proportion of Respondents Who Gave Priority Score of 4 or 5	% Agreement With Priority Scores	Grouped Into Key Categories
Components achieving the priority threshold for consensus (75% agreement with the priority score)		
Checking for treatment side effects	98	Acceptability
Checking mask fit issues	98	Technical
Recording validated assessment of sleepiness/ somnolence (eg, Epworth Sleepiness Scale)	95	Sleepiness assessment
Recording of CPAP adherence/efficacy by data download via memory card/remote monitoring	95	Adherence check
Ask if any problems with sleepiness while driving	95	Sleepiness assessment
Checking if initial symptoms for referral have improved (eg, tiredness/sleepiness/ hypersomnolence/concentration/memory)	93	Symptom resolution
Checking patient quality of sleep/feeling refreshed on waking	90	Assess sleep quality
Ensuring CPAP is resolving the apnea-hypopnea index	90	Symptom resolution
Asking about quality/quantity of sleep/sleep routine times	88	Assess sleep quality
Patients preparedness to continue with treatment	88	Acceptability
Asking about current driving status - car/heavy goods vehicle license	85	Driving
Checking patient quality of life	83	Quality of life
Verbally asking about CPAP adherence/sleep time	80	Adherence check
Requirement for humidification	78	Technical
Frequency of cleaning mask interface and circuit/changing filters	78	Technical
Check for control of witnessed residual snoring or apneas, choking spells	76	Symptom resolution
Asking about work schedule/shift pattern	76	Lifestyle issues/seep hygiene
Components not achieving the priority threshold for consensus, but with > 50% agreement with priority scores		
Review of medical history, medication, any new comorbidities in relation to symptoms/ need for hospitalization	73	General medical assessment
Ask about factors that help/hinder CPAP use	73	Technical
Checking body weight	71	General medical assessment
Support system at home (help with mask on/off fitting and filling of humidifier if used)/ partner engagement	65	Technical
Recording the altitude where the patient lives and the altitude where the sleep study was carried out	61	Technical
Checking that any relevant vehicle licensing agencies are aware of the condition	60	Driving
Requirement to repeat diagnostic study with significant weight loss/weight gain	60	General medical assessment
Components with < 50% agreement with priority scores		
Checking partner feedback/quality of life	48	Quality of Life
Fatigue and depression scale/as interfering with validated assessment of sleepiness (eg, Epworth Sleepiness Scale)	43	Sleepiness assessment
Checking electrical safety of CPAP unit	33	Technical
Nocturia/frequency of getting up to pass urine	33	General medical assessment
Examination of the nasal passage and throat	28	General medical assessment
Advice regarding air travel	25	Lifestyle issues/seep hygiene
CPAP unit noise level	25	Technical
Cognitive/developmental issues	23	General medical assessment
Chest auscultation	0	General medical assessment

CPAP = continuous positive airway pressure

and in the early months of use. Opinions on the frequency of review after CPAP was established was more varied with most participants suggesting 12- to 18-month follow-up, and more frequent reviews targeted to those with poor adherence. A flexible approach that offered "open access" or follow-up "on demand" was prioritized by 80% of participants. Almost half the respondents highlighted contexts (such as a specific request from a traffic agency, or before elective surgery) that might

Table 3—Themes from free-text comments.

Assessment of Symptom Resolution
"Assessment should be focussed on whether CPAP has improved the symptoms responsible for initial presentation as CPAP is mainly a treatment for symptoms." (Respiratory Physician and Academic) "Should also have some documentation of residual AHI on treatment although I am cautious of putting this in as it is often a software derived number and in a significant number of cases never fully settles to < 5." (Respiratory Physician)
Assessment of Sleepiness and its Impact on Activities Including Driving
"Questioning whether CPAP is used for all sleep episodes, daytime napping etc, and documentation of whether excessive sleepiness has improved." (Respiratory Physician) "I would take a detailed sleep history for a patient with residual sleepiness despite apparently effective CPAP" (Respiratory Physiologist) "Professional drivers should have annual nurse review at least." (Respiratory Physician) "We tend to offer 12 monthly reviews for HGV drivers." (Specialist Nurse) "Professional drivers will continue to be seen yearly." (Respiratory Nurse Specialist)
The Importance of Monitoring Adherence to CPAP Therapy
"Telemedicine for CPAP adherence tracking should be encouraged and reimbursed." (Respiratory Physician) "Download of the CPAP device should be performed or supplied ahead by the supplier." (Respiratory Physician)
Technical Issues Regarding CPAP Therapy
"Checking the operational status and cleanliness of the CPAP humidifier and mask. Need for replacement of same." (Respiratory Physician) "Some of the content of CPAP reviews, such as electrical safety testing will also depend on whether the machine is provided by the CPAP clinic, rented or purchased by the patient." (Respiratory Physician)
Checking Partner Feedback and Quality of Life
"Clinical review that incorporates the patient's bed-partner and/or other close family may be useful to help identify and manage any potential problems." (Specialist Nurse)
The Requirement for General Medical Assessment
"Requirement to repeat CPAP titration study or conduct a review if the download information or patient report is inconsistent helps with mask issues CPAP pressure, etc." (Respiratory Physician)
Frequency and Mode of Review
"An early review after the first visit to initiate CPAP is in my view crucial for increasing the chance of long term CPAP compliance." (Respiratory Physician) "The frequency of a clinical review varies globally depending on local health care systems and providers, and professional drivers should have annual review." "Clinical review provides the opportunity for education/support and reinforcement of treatment." (Respiratory Nurse Specialist)

AHI = apnea-hypopnea index, CPAP = continuous positive airway pressure, HGV = heavy goods vehicle.

determine the need for a review. Follow-up via a telemonitoring option, where available, was acceptable. There was general agreement that the timing of the review should be flexible to meet the clinical and support needs of the patient as well as being compatible within the health care delivery context.

DISCUSSION

Main Findings

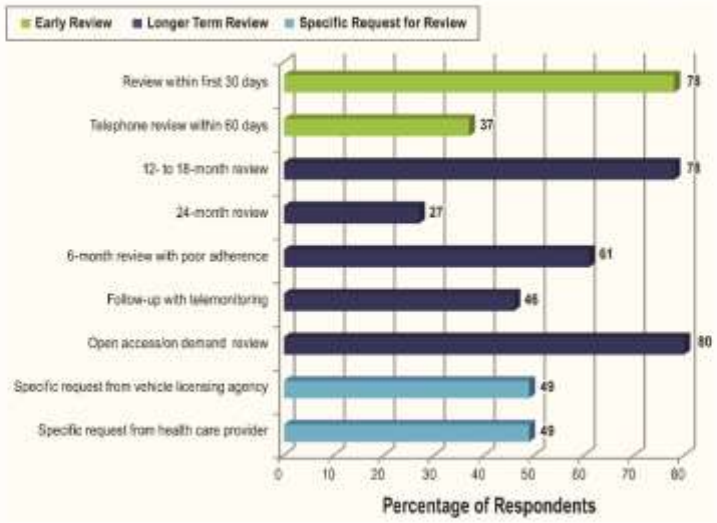
This is the first study to provide an international consensus on the most important components that should be considered when reviewing people using CPAP therapy. From a list of 36 components, 17 reached consensus (≥ 75%) and were considered the most important to include during a CPAP therapy review. The components identified have been grouped into key categories: technical aspects (n = 8 components); general medical assessment (n = 7); sleepiness assessment (n = 3); symptom resolution (n = 3); acceptability of treatment (n = 2); adherence check (n = 2); assessing sleep quality (n = 2); driving issues (n = 2); quality of life (n = 2); and lifestyle issues/sleep hygiene (n = 2). The need for flexible follow-up arrangements was highlighted by the free-text comments indicating that clinical review arrangements should focus on individual patient needs.

Interpretation in Light of Other Literature

Current clinical guidelines regarding the review of CPAP users collectively suggest 10 components that should be included in a CPAP review with the guideline from the American Academy of Sleep Medicine being the most comprehensive and identifying 8 of 12 prioritized components.[17] There is, however, wide variation in the guideline recommendations published between 2003[26] to 2016,[26] highlighting the need for an international consensus on what is important to include in a clinical review and how often this should occur.

Some components (eg, asking if any problems with sleepiness while driving, self-reported assessment of sleepiness [eg, Epworth Sleepiness Scale], checking for any mask interphase issues) were strongly prioritized in our e-Delphi study but were not always recommended by the guidelines. Specifically, current guidelines do not highlight checking the apnea-hypopnea index (AHI) in the downloaded CPAP data despite the priority accorded by most e-Delphi participants to assessing improvements in AHI. In the United Kingdom, this priority may reflect

Figure 1—Frequency of review.



the recently updated guidance from the Driving and Vehicle Licensing Agency, which states that in moderate to severe OSA subsequent licensing will require control of the condition; improved sleepiness; and treatment adherence.[27] This is an important finding of our e-Delphi study and driving-related issues need to be given higher priority in future clinical review and guideline development.

Our study identified a number of components that were considered as being less important (< 75%) suggesting these may be optional and included according to clinical judgement (**Table 2**). For example, "asking about comorbidities", which achieved a rating just under the priority threshold, will be important in some clinical contexts. Ongoing/long-term review also provides the opportunity for education/support and reinforcement of treatment rationale in individuals undergoing CPAPs.

Asking about quality and quantity of sleep, sleep routine times and work schedules/shift patterns, measuring patients' quality of life, and reviewing patients' preparedness to continue with treatment all reached a priority consensus of > 75% although few current guidelines specifically recommend these components. The consensus gained from our e-Delphi study has highlighted the importance of considering including these components in a review. The priority attached to checking practical maintenance of CPAP equipment depended on the organization of sleep medicine services; in some systems this was not part of a clinical review.

One option for implementing these clinical review components in CPAP users is to provide a clinical review template. Although opinions vary on the use of templates in clinical practice, they can facilitate a structured process and improve consistency of care.[28] However, there are limitations to the use of clinical templates; for example, they may not address all issues with CPAP usage from a patient's perspective so some flexibility in their use is important. Building on our findings we have outlined a suggested clinic review template based on the components prioritized by the e-Delphi respondents that could assist sleep medicine clinicians to provide a structured review (**Table 4**).

A key finding from our e-Delphi study is that there needs to be flexibility in the delivery of services—both in frequency and mode. Early and frequent review is recommended as a priority for new CPAP users and those having difficulty with adherence, or practical problems such as treatment side effects, and then reducing to annual/ biannual review when stable. The option of offering an "open access" service in which the patient could determine his or her need for review appealed to 80% of the respondents in our study. With the ever-increasing demand for sleep medicine services globally this may be seen as an attractive option for health care providers; however, there is currently no published literature to inform this practice. Furthermore, with the rise in the implementation of sleep telemedicine services CPAP review can be facilitated with telemonitoring and overseen remotely.

Strengths and Limitations

We generated an extensive list of potential components of a CPAP therapy clinical review by amalgamating recommendations from current guidelines, best practice, and position statements with suggestions from an international multidisciplinary

Table 4—Outline of a sleep clinic review template.

Acceptability of Treatment
Checking for treatment side effects
Preparedness to continue with treatment
Technical Aspects of Therapy
Checking mask fit issues
Requirement for humidification
Frequency of cleaning mask interface and circuit/changing filters
Objective Assessment of Sleepiness
Recording a validated assessment of sleepiness/somnolence (eg, Epworth Sleepiness Scale)
Ask if any problems with sleepiness while driving
Measurement of Adherence to CPAP Therapy
Recording of CPAP adherence/efficacy by data download via memory card or remote monitoring
Verbally asking about CPAP adherence/sleep time
Resolution of Symptoms
Checking if initial symptoms for referral have improved (eg, tiredness, sleepiness, hypersomnolence, concentration, memory)
Ensuring CPAP is resolving the apnea-hypopnea index
Check for control of witnessed residual snoring or apneas, choking spells
Assessing Sleep Quality
Checking patient quality of sleep /feeling refreshed on waking
Asking about quality/quantity of sleep/sleep routine time
Driving/Vehicle Licensing Agency Issues
Asking about current driving status - car/heavy goods vehicle license
Quality of Life
Checking patient quality of life
Lifestyle Issues/Sleep Hygiene
Asking about work schedule/shift pattern

This table lists the prioritized components of a review. The vision is that this might be used as the basis of a (potentially computerized) template which would start with an open question setting the agenda for the review (what does the patient wish to discuss?); act as a checklist to prompt delivery of important components of a review include space for free-text entries; and be followed by a second page with the components that did not reach consensus, but which will be important in some contexts. CPAP = continuous positive airway pressure.

panel of participants of clinicians/academics involved in this field and also individuals undergoing CPAP therapy. Interpretation and development of the outline review template followed a structured mapping of components. Forty participants (exceeding our recruitment target) completed all three rounds of our study electronically; only one person withdrew between the two scoring rounds enabling the consensus process. An important strength of our study is that the participants were from a range of health care backgrounds involved in the delivery of sleep medicine services, representing 21 countries with a broad range of economic backgrounds and health care systems. Although the number of participants recruited to our expert panel is larger than in other e-Delphi studies[21,24] they may not represent the full range of perspectives from sleep medicine clinicians/providers as the delivery of sleep medicine services varies widely in health care systems globally. Providing an initial list of components derived systematically from guidelines would have helped clarify the process for participants but will have influenced their suggestions, though the international expert panel of clinicians/academics and CPAP therapy users trebled the list of components. A consensus conference would have allowed a nuanced discussion but, for logistical reasons, would have restricted the number of participants, in particular reducing the international perspective. However, our e-Delphi actively encouraged free-text comments throughout all rounds of the study, which we analyzed thematically to provide insights into the results of scoring.

CONCLUSIONS

Our international expert panel agreed that the most important components of a clinical review of people using CPAP therapy to treat OSA were assessing: treatment acceptability; technical aspects of therapy; use of a validated sleepiness assessment tool; recording adherence/efficacy verbally or by data download via memory card/remote monitoring; symptom resolution; driving issues; sleep quality; quality of life and lifestyle issues/sleep hygiene. We have mapped these components into a suggested sleep medicine review template that may assist clinicians to conduct a patient centered, structured, and evidence-based clinical review. There were diverse opinions on the optimal frequency of review but general agreement that relatively frequent review should be undertaken in patients with

a new diagnosis. Long-term follow-up may be less frequent, or potentially "on demand," and can be provided by a range of professionals with the option of using telemonitoring where available. Feedback on the utility of the template is welcomed, so that our findings can be refined to inform future guideline recommendations and the delivery of care for people with OSA using CPAP.

ABBREVIATIONS

AHI, apnea-hypopnea index
CPAP, continuous positive airway pressure
OSA, obstructive sleep apnea

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Appendix 10 Implementation study protocol

The use of a structured sleep medicine clinical review template in clinical practice - A mixed methods implementation study

An Implementation study protocol.

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Sponsor	NHS Dumfries and Galloway
Co-sponsors	University of Edinburgh and ACCORD
Funder	No funding
Protocol Version and date	V1 10/03/19

List of abbreviations

BMcK – Brian McKinstry

CPAP - Continuous Positive Airway Pressure

HP - Hilary Pinnock

OSAHS - Obstructive Sleep Apnoea Hypopnoea Syndrome

PM - Phyllis Murphie

REC – Research ethics committee

StaRI - Standards for reporting implementation studies of complex interventions

SL- Stuart Little

TJ – Tracey Jackson

1. Summary

Current guidelines, best practice and position statements recommend regular review of people with Obstructive Sleep Apnoea Hypopnoea Syndrome (OSAHS) who are using Continuous Positive Airway Pressure (CPAP) therapy and suggest a varied range of components that may be included in the clinical review (such as subjective assessment of sleepiness and resolution of presenting symptoms, recording of adherence to CPAP, as well as resolving practical issues with masks and equipment, driving guidance/vehicle license advice, blood pressure and weight monitoring) but with no consensus on which to base standards. We conducted an international consensus study to define the most important core components of a clinical review (1). Following this research we have devised a structured sleep medicine clinic review template (based on an international consensus of sleep medicine clinicians). We now wish to evaluate implementation of the template in the routine care of people using CPAP for OSAHS using a mixed methods research design. To inform more generally the impact of implementation of the template in consultations, we will explore perceptions of the impact on the dynamics of the consultation and specifically potential disadvantages to patient centeredness and benefits to completeness of review. This is a novel approach; no previous studies have been published regarding the use of a structured sleep medicine review template.

2. The position of this study within a PhD programme

The first research output from this PhD candidate (PM) was a systematic review that explored the use of remote consulting with telemonitoring of continuous positive airway pressure usage data for the routine review of people with obstructive sleep apnoea hypopnoea syndrome (2). We concluded that the evidence for teleconsultation/telemonitoring in CPAP users is limited; however, no safety concerns have been raised. Adequately powered, well-designed trials are needed to establish whether real-time telemonitoring and remote teleconsultation is a clinically and cost effective option for people using continuous positive airway pressure therapy. The next research project within the PhD was the international e-Delphi study described above (1). This implementation study is the final piece of research in a programme of study towards a Population Health Sciences (PhD) (Part time) with the University of Edinburgh. Its aims to evaluate the use of a structured sleep medicine review template in the real life clinical setting (face to face/teleconsultation) using mixed methods research. The finding from this final study will be included in PM's thesis titled 'Routine reviews of people with OSAHS: telehealth and templates'.

3. Introduction

3.1 Background

Reviewing people with Obstructive Sleep Apnoea Hypopnoea Syndrome

Obstructive Sleep Apnoea Hypopnoea (OSAHS) is a very common; treatable condition that represents a major global public health issue and is an important cause of morbidity and mortality (3-5). A recent systematic review has reported that the prevalence of OSAHS in the general adult population ranges from 6% to 17%, but may be as high as 49% in older people (2). With its associated burden of symptoms and complications OSAHS is emerging as a major challenge for healthcare services, particularly in high income countries (6). People who are diagnosed with OSAHS, and who are using Continuous Positive Airway Pressure (CPAP) therapy require regular long term follow up (3, 7). Modes of CPAP review are traditionally face to face, but the advent of remote monitoring of CPAP data has stimulated considerable interest in telephone or video-consultations. The evidence supporting this pragmatic approach to delivering care is growing and increasingly these remote modes of review are becoming the norm in routine clinical care. (2-4, 8).

Past and current research

Current guidelines, best practice and position statements recommend regular review and suggest a varied range of components that may be included (such as subjective assessment of sleepiness and resolution of presenting symptoms, recording of CPAP adherence, as well as resolving practical issues with masks and equipment, driving guidance/vehicle license advice, blood pressure and weight monitoring) but with no consensus on which to base standards. (3, 9-12). We have therefore undertaken an e-Delphi to reach consensus on the core components of a review (whether face-to-face and remote) (1). Based on this, we have developed a template to guide the content of a review (Appendix 1). This is a novel approach; no previous studies have been published regarding the use of a structured sleep medicine review template. We therefore wish to evaluate implementation of the template in routine care of people using CPAP for OSAHS.

Templates and their role in clinical reviews

The use of templates as tools to assist with the delivery of healthcare can facilitate a structured review process and support consistency of care and have been used in clinical practice for more than two decades (13, 14). The use of electronic templates is widespread in general practice and was introduced to implement improvements in the quality of care delivered for people with long-term conditions, as well as to standardise data collection linked to the Quality and Outcome Framework (15, 16). However, by directing the content of the consultation, templates risk overriding the patient's agenda and preventing them expressing their worries. Including an initial question in a standardised review template about the patient's main concerns has been suggested as a way to avoid this potential disadvantage and facilitate a patient focused review (17).

Implementation Research

The field of implementation research has developed over recent years as a methodology to understand and evaluate strategies for implementing evidence based interventions into the real life clinical setting (18). A core aim is to shorten the time it takes to translate evidence-based practice into routine clinical care (19, 20). A decade ago Bhattacharyya et al 2009 defined implementation research as the study of methods to promote the uptake of research findings into routine practice (21). A core concept enunciated by the StaRI reporting standards and reflected in the terminology used in this proposal, is the distinction between the evidence-based intervention (e.g. core components of a OSAHS/CPAP review) and implementation strategy which is used to promote the use of the intervention (e.g. the use of a template integrated within the existing routines of the clinical setting. (22). In an implementation study, it is the implementation strategy that is the primary focus of the evaluation (e.g. uptake and use made of the template). Implementation strategies are context-specific (e.g. a computerised service will need computerised templates; a paper-based service will need the templates on paper, and may need to indicate codes for subsequent coding).
Preparatory phases of implementation studies involve understanding existing routines and adapting the intervention and the strategies to improve local conditions in a specific health care delivery setting. Implementation research (in distinction to local quality improvement studies) seek to generate generalisable findings by adopting a theoretical approach and/or implementing in a theoretically determined range of settings (e.g. computerised/non-computerised clinics; mostly face-to-face/remote reviews; rural/urban).

Need for research in this area

Our international e-Delphi study used consensus methodology to define the core components of a clinical review of people using CPAP therapy to treat OSAHS and to determine how often such a review should take place (1). Based on this consensus work we have developed a CPAP clinic review template (Appendix 1). We now wish to test the feasibility of implementing the sleep clinic review template in three diverse clinical contexts. To inform more generally the impact of implementation of the template in consultations, we will explore perceptions of the impact on the dynamics of the consultation and specifically about potential disadvantages to patient centeredness and benefits to completeness of review.

Mixed methods research

This study will assess the implementation of the sleep apnoea review template over a 2-month period into the routine clinical care of people with OSAHS who are using CPAP in three diverse settings (an urban based large sleep medicine service, a remote and rural sleep medicine service and a combined urban/rural sleep medicine service). A convergent mixed methods design will be used whereby quantitative and qualitative data will be collected in parallel, analysed separately and then merged (23). Quantitative data will assess the effectiveness of the implementation strategy (e.g. uptake of the template by site and by clinician), the feasibility of using the template (e.g. time taken to complete) and its utility as an intervention (fields completed, additional fields required). The qualitative data will explore the perceptions of sleep medicine clinicians on the utility of the template in patient

consultations (face to face, teleconsultation, telephone review). We have chosen to use mixed methods in our study as the use of differing approaches has the potential to provide a greater depth and breadth of information which is not possible utilising singular research approaches in isolation.

4. Implementation research objectives

I. Development of an implementation strategy

a) To create an implementation strategy with clinical and administrative staff from each of the participating centres.

b) To qualitatively explore the perceptions of sleep medicine clinicians (respiratory physicians, physiologists, specialist nurses) from participating centres on:

- practical strategies for implementing the use of the template in their centre (e.g. electronic or paper, reminders, attached to records etc)
- their perceptions on how using a structured sleep medicine review template may affect patient professional interactions during the clinical review and how negative effects may be avoided

II. Implementation

To implement the review template in routine clinical practice in the participating centres over 2 months.

III. Evaluation: assessment of effectiveness and exploration of perception of template use

We will use a mixed methods approach to:

a) assess the effectiveness of the implementation strategy (e.g. uptake of the template by site and by clinician), the feasibility of using the template (e.g. time taken to complete) and its utility as an intervention (fields completed, additional fields required).

b) explore the perceived effectiveness of the template, perceptions of using the template in the clinical setting (advantages and disadvantages), impact on the consultation, and suggestions for refinement, from the perspective of healthcare professionals and coders.

IV. Refine the intervention and implementation strategy

To further refine the structured sleep medicine review template and develop recommendations for future implementation.

5. Study design

5.1 Type of study

This multicentre study will use both qualitative and quantitative methods to develop and evaluate implementation strategies and explore the utility/applicability of the sleep medicine review template in established CPAP therapy users. We will use the Standards for Reporting implementation Studies (StaRI) checklist to assist in accurately reporting the findings of this implementation study (24, 25).

6. Recruitment of sites

Three sleep medicine clinicians/centres have expressed an interest in participating in this research study and they are listed as collaborators on page 1 of this study protocol. These sites have been selected to represent different contexts: urban/rural (Dundee and Edinburgh) rural (Inverness).

6.1 Study population

All clinicians from the participating centres will be invited to use the sleep medicine review template in all consultations, and this will be facilitated (e.g. a blank template attached to each set of records) though they have the choice whether or not they use the template in their consultations. In addition, whether the template is used for any individual consultation or whether specific components are addressed will be entirely at the discretion of the clinician.

Between 5 and 7 clinicians (respiratory physicians, physiologists, specialist nurses) from each site (in total up to 20), who are actively involved in reviewing people who are using CPAP therapy to treat OSAHS will be invited to consent to participate in the final visit qualitative interviews. Interviewees will be selected to offer a maximum variation sample of professional background, frequent/occasional/non-users of the template.

7. Study procedures

The study intervention involves asking the participating clinicians to use and complete a structured sleep clinic template when they are conducting a review of CPAP therapy users over an 8-week period in the clinical review that they conduct (see Appendix 1).

In line with the four objectives (see above), the study procedure will follow four phases:

I. **Development of an implementation strategy** (Objective 1)
   - The implementation strategy for each site will be developed during a series of site visits in discussion with the participating clinicians and the principal investigator. Each site will have different routines and will develop different strategies for ensuring a template is available for every consultation (e.g. attached to the clinic documentation)
   - A specific consideration will be the choice of a paper or electronic template. The preference of the participating site teams will be identified and materials prepared accordingly

- A process of secure collection of anonymised copies of the template for analysis will be agreed

Data collection: Field notes will be kept of all visits (including minutes of any formal discussions/meetings), telephone calls, e-mails etc. Copies of existing materials will be collected (e.g. recording sheets, format of computer records etc). All decisions about implementation strategies will be documented. At one of these initial visits, we will convene a group discussion of participating clinicians during which we will explore perceptions of current arrangements, the components that they usually include in the clinical review of CPAP therapy users and any benefits or concerns they may have about the use of a template.

II. **Implementation** (Objective 2)

a. Baseline visit to set up the study. Each site will be provided with anonymised Sleep Medicine review templates to suit their service (all sites have indicated their preference for a paper version) – training, explanation on completion of the template
b. Mid-study visit to check for any problems (and potentially make any minor amendments if feasible). Potentially this 'visit' may be done by telephone or teleconference
c. Final visit to undertake evaluation data collection (see phase III)

Data collection: Field notes will be kept of all visits (including minutes of any formal discussions/meetings), telephone calls, e-mails etc.

III. **Evaluation** (Objective 3)

a. *Quantitative assessment of effectiveness of the implementation strategy.* Anonymised copies of completed templates will be collected at each site visit. Paper copies will be anonymised by providing a carbon copy of the template with the clinician's profession details, age and gender of the patient being reviewed. (participants will be asked to remove any patient specific identifiable information from the completed templates) and will be collected as a hard copy printed paper version at subsequent site visits or scanned and retuned by email within NHS email. The templates will indicate the site, the clinician's profession, and mode of consultation (face-to-face, teleconsultation, telephone) but have no personal details about the patient. In addition we will collect the total number of clinical reviews by clinician and mode of consultation (face to face, telephone review, teleconsultation), time to complete the template (by asking the participants the average time required to complete the template), and the number of templates and the components completed during the study.
b. *Qualitative exploration of clinical perception of template use.* Between 5 and 7 clinicians (respiratory physicians, physiologists, specialist nurses) from each site (in total up to 20), will be invited to participate in semi structured interviews at the final visit. These interviews will explore how the templates were used in the clinical setting. The discussion will cover the following questions (see Appendix 2) for draft detailed topic guide):

- What were the advantages/disadvantages of using the template on clinical practice?
- How was the template used (during the consultation as a prompt; after the consultation as a means of recording)
- What is the impact of the template on
    - the way the clinical review is conducted?
    - clinical autonomy?
    - patient/professional relationships?
    - patient centred care?
- Are there any components that should be added or removed from the template?

All interviews will audio recorded and transcribed verbatim into nVivo software program for analysis.

IV. **Refining** (Objective 4)

Informed by the evaluation, further refinement of the structured sleep medicine review template will be undertaken for future implementation.

8. Data analysis

8.1 Quantitative data analysis

The analysis of the quantitative data will be limited to descriptive analysis and summary statistics and key outcomes will be effectiveness of the implementation strategy as assessed by:

- The uptake of the template by the participating clinicians and sites (the number of templates that are completed at each site by the clinicians and the proportion of all clinical reviews)
- The feasibility of using the template in terms of perceived time taken to complete the template; participants will be asked to record this assessed timing at the end of the template.
- The utility of the template as an intervention in terms of the number of fields completed and any additional fields that are suggested by participants

  Subgroup analysis will include the use of the template in relation to the mode of clinical review (face to face, telephone, teleconsultation) and the background of the clinician who is conducting the review

8.2 Qualitative data analysis

Clinician group discussion (at initial visit) and clinician semi structured interviews (at final visit):

- We will undertake a thematic analysis of the initial group discussion (field notes will be taken) with participating clinicians at all sites at visit 1 regarding which components of the clinical review they normally include in their clinical practice
    - the group discussion will be audio-recorded and transcribed and coded thematically, cognisant of unexpected themes which may arise

  - we will analyse these initial group discussions at the beginning of the study in order to iteratively inform the topic guide for the final interviews
- We will use the framework method for analysis of the qualitative data to undertake a thematic analysis and interpretation of the semi structured recorded interviews from up to 20 participating clinicians (2-10 from each site) at the end of the study regarding their views on the application and utility of the template in the clinical review process (26).
- Steps in the thematic analysis will follow:
  - Stage 1- Transcription
  - Stage 2- Familiarisation with the interviews
  - Stage 3- Coding
  - Stage 4- Developing an analytical framework
  - Stage 5- Applying the analytical framework
  - Stage 6- Charting data into the framework matrix
  - Stage 7- Interpreting the data

- The Normalisation Process Theory (NPT) is a sociological toolkit that can be utilised to assist researchers to understand the processes of developing, implementing, embedding, and integrating some new technology or complex intervention in healthcare (27-29). The NPT proposes four conceptual elements or constructs that represent different methods of working patterns around implementing a new practice: Coherence, Cognitive Participation, Collective Action, and Reflexive Monitoring. We will use the NPT as a framework to inform or guide the way in which we code, analyse and interpret the data from our qualitative interviews. We will code the qualitative data of the thematic analysis and then group the codes under the various NPT constructs and components. We will also use NPT on-line resource to promote discussion within the multidisciplinary team about the findings, their interpretation, and to inform conclusions of the implementation study and future recommendations.

- We will use constant comparison seeking a wide range of views particularly seeking out contradictory views and will recruit participants until no new views related to our objectives are being expressed i.e. data saturation

- More than one researcher will extract 5% of the data to ensure that there is attention to the potential for and control for interviewer bias

- The principal investigator will keep a research diary, where they record reflexive notes, impressions of the data and thoughts about analysis throughout the process. I will apply reflexivity to my position as both the chief investigator/designer of the study and as a practicing sleep medicine clinician to provide more effective and impartial analysis of the data (26).

8.3 Synthesis of quantitative and qualitative research data

We will use triangulation to analyse the quantitative and qualitative data separately and then compare the research findings in order to develop a complete understanding of the clinical effectiveness or

otherwise of the template and the clinicians perceptions on how the use of template affected (positively or negatively) on the efficiency, completeness and/or patient centeredness of the review consultation. Triangulation provides opportunities for convergence and corroboration of results that are derived from different research methods. Each component of the study data will be assessed for agreement (convergence), complementarities or contradictions (dissonance). The interpretation of the qualitative data from the thematic framework analysis will be compared with the quantitative data to corroborate or contextualise themes that emerge from the qualitative finding that may inform further refinement of the structured sleep medicine review template for future implementation.

8.4 Critical distance and impartiality in data validation

The research team will incorporate methodological strategies within the conduct of the implementation study to ensure the validity, 'trustworthiness', reliability and generalisation of the research findings:

- Take into account any personal biases which may influence the research findings (we will conduct an end of study discussion with clinicians who are unconnected to the study to facilitate an unbiased interpretation of the research findings)
- Recognise the ongoing potential for bias as the study progress and apply critical reflection to ensure the relevance of the data collected and analysis methods
- Good record keeping at all stages of the study will provide a transparent and consistent view of data interpretation
- Compare and contrast similarities and differences in the responses from interviewees to ensure differing opinions/views are captured reliably
- Continuously engage with other co-investigators to reduce potential for research bias
- Triangulation of the mixed methods research data in order to produce a more comprehensive set of research findings

9. Dissemination and Publication Strategy

The intention is to publish the results of the implementation study in a peer reviewed journal and to present research findings to the study participants and professional groups at national and international meetings and conferences, social media and patient interest groups.

10.Timetable for study

Month	2019
March-April 2019	Ethics submission/Approval
April- May 2019	Visit 1 Start up visit – all sites

May - June 2019	**Visit 2- Mid study site visits**
June July 2019	**Visit 3- Final site visit**
July – September 2019	**Analysis of data**
September 2019 onwards	**Reports and Publications**

11. Study management

The research team, led by PM, will oversee the implementation study and she will co-ordinate the data collection and will be responsible for reporting the results. HP and BMcK and TJ will support PM with the data analysis and PM, HP, BMcK, and SL will write up the results of the implementation study.

12. Research environment and available expertise

Resources will be provided by the Usher Institute of Population Health Sciences and Informatics at the University of Edinburgh, including the full range of support services (e.g. library facilities and computing support) as well as office space.

12.1 Implementation study project team

The research study team bring a broad range of expertise in health service research and professional influence to underpin the methodology and address the challenges of this programme of research including clinical and academic expertise in the management and review of people with OSAHS who are using CPAP therapy, professional education and behavioural change, methodological expertise in complex interventions and implementation science, use of routine data and pragmatic trials, health economics and qualitative research.

12.2 Principal Investigator

Sister Phyllis Murphie is a Respiratory Nurse Consultant and PhD candidate within the UOE and is the principal investigator who will take responsibility for the day to day conduct of the study. She has 21 years of clinical experience in leading the sleep medicine service in NHS Dumfries and Galloway.

12.3 Researchers

Dr Stuart Little is a Respiratory Physician (MD) who is based at the Dumfries and Galloway Royal Infirmary and has mentored and supported Phyllis Murphie for the duration of her PhD. He is co author on two of the publication outputs from her PhD and is a co investigator in this study.

Professor Brian McKinstry is a GP and Professor of Primary Care E- health at the University of Edinburgh. He is Phyllis Murphie second PhD supervisor. He leads the Telescot Research Programme, as well as the Edinburgh Health Research Unit and SHARE (the Scottish Health Research register.

Professor Hilary Pinnock is a GP with an interest in respiratory care and experience of developing and evaluating complex interventions. She is Phyllis Murphie's principal PhD supervisor. She is Professor of Primary care Respiratory Medicine at the University of Edinburgh. She is head of Assembly 1- General Pneumonology of the European Respiratory Society.

12.4 Collaborators

Dr Tom MacKay is Director of the Edinburgh Royal Infirmary Sleep Medicine Service and is the Vice President of the Royal College of Physicians Edinburgh.

Dr Will Anderson, MD is a Consultant Respiratory Physician based at Ninewells Hospital, Dundee. He is also Honorary Senior Clinical Lecturer University of Dundee.

Dr Lorna Murray PhD is a Respiratory Physician and Lead for the Sleep medicine service in NHS Highland, Raigmore Hospital Inverness

Dr Tracy Jackson is a Research Fellow with Asthma UK Centre for Applied Research at the Usher institute University of Edinburgh.

12.5 Patient and public involvement

I have been working closely with the Scottish Association of Sleep Apnoea (SASA) since I commenced my research studies in 2014 and have presented my research to them at their annual conference since commencing my PhD in 2014. SASA is a charity that consists of members who have OSAHS and who use CPAP therapy. Prior to conducting our international e-Delphi study (1), I was a speaker at their annual conference in 2016 where I asked them to share their views on what they considered were the most important components of a clinical review. Their responses helped to inform a local pilot study before conduction of our international study. Several members of the group have been recruited to be advisors to the project and I have been invited to present my finding from this implementation study at their annual conference on the 5th of October 2019.

13. Oversight arrangements

13.1 Inspection of records

Investigators and institutions involved in the study will permit study related monitoring and audits on behalf of the sponsor, REC review, and regulatory inspection(s). In the event of audit or monitoring, the Investigator agrees to allow the representatives of the sponsor direct access to all study records and source documentation. In the event of regulatory inspection, the Investigator agrees to allow inspectors direct access to all study records and source documentation.

13.2 Risk assessment

There are no risks to clinical care as clinicians will continue to provide their usual care. The template does not seek to change clinical decisions with regards to care, only to standardise the routine assessment of people with OSAHS in line with guideline-based practice.

There are a number of risks to the implementation study:

- potential for consenting clinicians not to participate in the study
- potential for increase in time for clinical consultation
- potential for only a small number of templates to be completed
- potential for loss of completed templates (at the baseline visit the researcher will identify a clinician on each site who will be responsible for collecting all the completed templates following each CPAP review clinic and ensuring that the completed templates are returned to the principal investigator at each site visit)

13.3 Study monitoring and audit

The ACCORD Sponsor Representative will assess the study to determine if an independent risk assessment is required. If required, the independent risk assessment will be carried out by the ACCORD Quality Assurance Group to determine if an audit should be performed before/during/after the study and, if so, at what frequency.

Risk assessment, if required, will determine if audit by the ACCORD QA group is required. Should audit be required, details will be captured in an audit plan. Audit of Investigator sites, study management activities and study collaborative units, facilities and 3rd parties may be performed.

14. Good clinical practice

14.1 Ethical conduct

The study will be conducted in accordance with the principles of the International Conference on Harmonisation Tripartite Guideline for Good Clinical Practice (ICH GCP).

Before the study can commence, all required approvals will be obtained and any conditions of approvals will be met.

14.2 Investigator responsibilities

The Investigator is responsible for the overall conduct of the study at the site and compliance with the protocol and any protocol amendments. In accordance with the principles of ICH GCP, the following areas listed in this section are also the responsibility of the Investigator. Responsibilities may be delegated to an appropriate member of the research team/study site staff.

14.2.1 Informed consent

The Investigator is responsible for ensuring informed consent is obtained before any protocol specific procedures are carried out. The decision of a participant to participate in clinical research is voluntary and should be based on a clear understanding of what is involved.

Participants must receive adequate oral and written information – appropriate Participant Information and Informed Consent Forms will be provided. The oral explanation to the participant will be performed by the researchers or qualified delegated person, and must cover all the elements specified in the Participant Information Sheet and Consent Form.

The participant must be given every opportunity to clarify any points they do not understand and, if necessary, ask for more information. The participant must be given sufficient time to consider the information provided. It should be emphasised that the participant may withdraw their consent to participate at any time without loss of benefits to which they otherwise would be entitled.

The researcher or delegated member of the study team and the participant will sign and date the Informed Consent Form(s) to confirm that consent has been obtained. The participant will receive a copy of this document and a copy filed in the Study Master File (SMF).

14.2.2 Study staff

The Investigator must be familiar with the protocol and the study requirements. It is the Investigator's responsibility to ensure that all staff assisting with the study are adequately informed about the protocol and their study related duties.

14.2.3 Data recording

The Principal Investigator is responsible for the quality of the study related data recorded.

14.2.4 GCP training

For non-CTIMP (i.e. non-drug) studies all researchers are encouraged to undertake GCP training in order to understand the principles of GCP. However, this is not a mandatory requirement unless deemed so by the sponsor. GCP training status for all investigators should be indicated in their respective CVs.

14.2.5 Confidentiality

All records must be identified in a manner designed to maintain participant confidentiality. All records must be kept in a secure storage area with limited access. Clinical information will not be released without the written permission of the participant. The Investigator and study site staff involved with

this study may not disclose or use for any purpose other than performance of the study, any data, record, or other unpublished, confidential information disclosed to those individuals for the purpose of the study. Prior written agreement from the sponsor or its designee must be obtained for the disclosure of any said confidential information to other parties.

14.2.6 Data protection

All Investigators and staff involved with this study must comply with the requirements of the Data Protection Act 2018 (and GDPR) with regard to the collection, storage, processing and disclosure of personal information and will uphold the Act's core principles. Access to collated participant data will be restricted to individuals from the research team interviewing the participants, representatives of the sponsor(s) and representatives of regulatory authorities.

Computers used to collate the data will have limited access measures via user names and passwords.

Published results will not contain any personal data that could allow identification of individual participants.

15. Study conduct responsibilities

15.1 Protocol amendments

Any changes in research activity, except those necessary to remove an apparent, immediate hazard to the participant in the case of an urgent safety measure, must be reviewed and approved by the Chief Investigator.

Amendments will be submitted to a sponsor representative for review and authorisation before being submitted in writing to the appropriate REC, and local R&D for approval prior to participants being enrolled into an amended protocol.

15.2 Management of protocol non compliance

Prospective protocol deviations, i.e. protocol waivers, will not be approved by the sponsors and therefore will not be implemented, except where necessary to eliminate an immediate hazard to study participants. If this necessitates a subsequent protocol amendment, this should be submitted to the REC, and local R&D for review and approval if appropriate.

Protocol deviations will be recorded in a protocol deviation log and logs will be submitted to the sponsors every 3 months. Each protocol violation will be reported to the sponsor within 3 days of becoming aware of the violation. All protocol deviation logs and violation forms should be emailed to QA@accord.scot

Deviations and violations are non-compliance events discovered after the event has occurred. Deviation logs will be maintained for each site in multi-centre studies. An alternative frequency of deviation log submission to the sponsors may be agreed in writing with the sponsors.

15.3 Serious breach requirements

A serious breach is a breach which is likely to effect to a significant degree:

a. the safety or physical or mental integrity of the participants in the study; or

b. the scientific value of the study

If a potential serious breach is identified by the Chief investigator, Principal Investigator or delegates, the co-sponsors (seriousbreach@accord.scot) must be notified within 24 hours. It is the responsibility of the co-sponsors to assess the impact of the breach on the scientific value of the study to determine whether the incident constitutes a serious breach and report to research ethics committees as necessary.

15.4 Study record retention

All study documentation will be kept for a minimum of 3 years from the protocol defined end of study point. When the minimum retention period has elapsed, study documentation will not be destroyed without permission from the sponsor.

15.5 End of study

The end of study is defined as the last participant's last interview or the last data collection. The Investigators or the co-sponsor(s) have the right at any time to terminate the study for clinical or administrative reasons.

The end of the study will be reported to the REC, and R+D Office(s) and co-sponsors within 90 days, or 15 days if the study is terminated prematurely. The Investigators will inform participants of the premature study closure and ensure that the appropriate follow up is arranged for all participants involved. End of study notification will be reported to the co-sponsors via email to resgov@accord.scot.

A summary report of the study will be provided to the REC within 1 year of the end of the study.

16. Insurance and Indemnity

The co-sponsors are responsible for ensuring proper provision has been made for insurance or indemnity to cover their liability and the liability of the Chief Investigator and staff.

The following arrangements are in place to fulfil the co-sponsors' responsibilities:

The Protocol has been designed by the Chief Investigator and researchers employed by the University and collaborators. The University has insurance in place (which includes no-fault compensation) for

negligent harm caused by poor protocol design by the Chief Investigator and researchers employed by the University. Sites participating in the study will be liable for clinical negligence and other negligent harm to individuals taking part in the study and covered by the duty of care owed to them by the sites concerned. The co-sponsors require individual sites participating in the study to arrange for their own insurance or indemnity in respect of these liabilities.

Sites which are part of the United Kingdom's National Health Service will have the benefit of NHS Indemnity.

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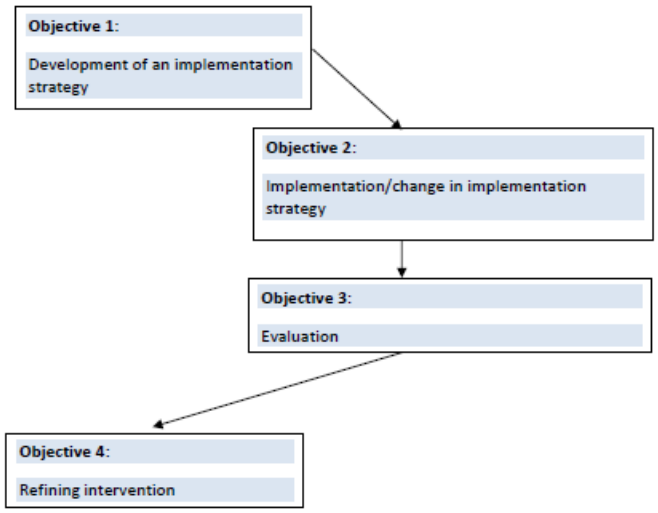
Implementation study diagram



Appendix 1 - Sleep clinic review template

This template was devised using the results of an international e-Delphi consensus.

1. The review starts with an open question to find out what the patient wants to discuss.
2. The items on this page are the components that 75% of the international experts considered should be included in every sleep clinic review.
3. Other items that may be important in some consultations are listed on page 2.

Name /address – Will be anonymised	Date	Age	Gender
The patient agenda for the review. What does the patient want to discuss? Any concerns?			

Acceptability of treatment	Are there any side effects from the treatment? Is the patient happy to continue treatment? Free text
Technical aspects of CPAP	Does the mask fit correctly? Is humidification satisfactory? Is cleaning / filters changing routine satisfactory?
	Free text
Objective assessment of sleepiness/driving issues	Objective assessment of sleepiness (Epworth Sleep Score) Are there any problems with sleepiness while driving? What are they driving: Car/Heavy Goods Vehicle?
	Free text
	Have initial symptoms improved?
	Has CPAP resolved the Apnoea Hypopnoea Index? Have witnessed snoring, apnoeas, choking spells been controlled?
	Free text
Assessing sleep quality/quality of life	What is the quality of sleep? Do they feel refreshed on waking? What are their sleep routines? How much sleep do they get? Quality of life Free text
Lifestyle	Ask about work schedule/Shift Pattern Free text

Additional optional review components that may be useful in some reviews	
Technical aspects	Factors that help/hinder CPAP use. Support system at home/partner engagement
	Altitude where lives/ Altitude where sleep study was done
	Electrical safety of CPAP unit/ CPAP unit noise level
General medical assessment	Review of medical history, medication, any new co morbidities in relation to symptoms/need for hospitalisation
	Examination of the nasal passage and throat
	Body weight/Blood pressure Chest Auscultation
Sleepiness assessment	Need to repeat diagnostic study with significant weight loss/weight gain
	Nocturia/Frequency of getting up to pass urine
	Cognitive/developmental issues
Lifestyle	Fatigue and depression scale /as interfering with subjective assessment of sleepiness e.g. Epworth Sleep Score
Quality of life	Checking partner feedback / quality of life
Driving	Are Vehicle Licensing agencies are aware of the condition?
	Advice re air travel

We are assessing the use of this template, and would value any feedback you can give us.

Did you find the template useful in this review? Yes ☐ No ☐ Not sure ☐

Did use of the template increase the length of the consultation Yes ☐ No ☐

Time taken to complete the template during the consultation ----------

Time taken to complete the template after the consultation ----------

Review: Face to face ☐ telephone ☐ videoconsultation ☐

Do you have any feedback about how the template worked (or didn't) in this review?

Please tick box that represents your position

☐ Respiratory Physician/doctor ☐ Physiologist ☐ Specialist Nurse

Thank you for your feedback

Appendix 2 – Topic guide for semi-structured interviews

Main question	Additional questions	Clarifying questions
1. Do you perceive any differences in the way that you carried out the consultation as a result of using the template? 2. What is the impact of the template on the way the clinical review is conducted regarding: • your clinical autonomy? • your patient/professional relationships? • patient centred care? 3. Are there any additional components that should be added or removed from the template?	Clarify what perceived differences using the template has made to the consultation process. Clarify the impact of the template on the clinical review regarding: • Autonomy • Patient/profession relationship • Patient centred care Clarify any additional component you would like to see added/removed from the template	• Can you please clarify what you mean by ....? • Can you please expand a little on ...? • Can you please give some examples of ...? • In particular what do you think of...?

Appendix 11 Health Research Authority Approval for the Implementation study

Health Research Authority

North West - Liverpool Central Research Ethics Committee
3rd Floor
Barlow House
4 Minshull Street
Manchester
M1 3DZ
Telephone: 0207 104 8196

23 July 2019

Mrs Phyllis Murphie
Dumfries and Galloway Royal Infirmary
Garroch
Dumfries
DG2 8RX

Dear Mrs Murphie

Study title:	**The use of a structured sleep medicine clinical review template in clinical practice - A mixed methods implementation study.**
REC reference:	**19/NW/0410**
Protocol number:	**AC19042**
IRAS project ID:	**251231**

Thank you for your response. I can confirm the REC has received the documents listed below and that these comply with the approval conditions detailed in our letter dated 10 July 2019

Documents received

The documents received were as follows:

Document	*Version*	*Date*
Other [Participant consent form 2]	V2	15 July 2019
Other [Ethics review amendments]	V1	15 July 2019
Participant consent form [Participant consent form 1]	V2	15 July 2019
Participant information sheet (PIS) [Participant Information Sheet]	V2	15 July 2019

Approved documents

The final list of approved documentation for the study is therefore as follows:

Document	*Version*	*Date*
Evidence of Sponsor insurance or indemnity (non-NHS Sponsors only) [Combined insurance doc]		31 July 2018
Interview schedules or topic guides for participants [Topic guide group discussion]	V1	19 May 2019
IRAS Application Form [IRAS_Form_30052019]		30 May 2019
Other [Topic guide semi-structured interviews]	V1	19 May 2019
Other [Data protection information]	V1	19 May 2019
Other [Response to validation]		25 June 2019
Other [Participant consent form 2]	V2	15 July 2019
Other [Ethics review amendments]	V1	15 July 2019
Participant consent form [Participant consent form 1]	V2	15 July 2019
Participant information sheet (PIS) [Participant Information Sheet]	V2	15 July 2019
Research protocol or project proposal [Implementation study protocol]	V1	19 May 2019
Summary CV for Chief Investigator (CI) [Summary CV CI]	V1	25 March 2019
Summary CV for supervisor (student research) [Summary CV -HP]		07 March 2019

You should ensure that the sponsor has a copy of the final documentation for the study. It is the sponsor's responsibility to ensure that the documentation is made available to R&D offices at all participating sites.

19/NW/0410	**Please quote this number on all correspondence**

Yours sincerely

Damilola Odunlami
Approvals Officer

E-mail: nrescommittee.northwest-liverpoolcentral@nhs.net

Copy to: Mrs Phyllis Murphie
Ms Melissa Taylor, NHS Lothian Research & Development Office

Appendix 12 Standards for Reporting Implementation Studies: the StaRI checklist for completion

The StaRI standard should be referenced as: Pinnock H, Barwick M, Carpenter C, Eldridge S, Grandes G, Griffiths CJ, Rycroft-Malone J, Meissner P, Murray E, Patel A, Sheikh A, Taylor SJC for the StaRI Group. Standards for Reporting Implementation Studies (StaRI) statement. *BMJ* 2017;356:i6795

The detailed Explanation and Elaboration document, which provides the rationale and exemplar text for all these items is: Pinnock H, Barwick M, Carpenter C, Eldridge S, Grandes G, Griffiths C, Rycroft-Malone J, Meissner P, Murray E, Patel A, Sheikh A, Taylor S, for the StaRI group. Standards for Reporting Implementation Studies (StaRI). Explanation and Elaboration document. *BMJ Open* 2017 2017;7:e013318

Notes: A key concept of the StaRI standards is the dual strands of describing, on the one hand, the implementation strategy and, on the other, the clinical, healthcare, or public health intervention that is being implemented. These strands are represented as two columns in the checklist.

The primary focus of implementation science is the implementation strategy (column 1), and the expectation is that this will always be completed.	The evidence about the impact of the intervention on the targeted population should always be considered (column 2) and either health outcomes reported, or robust evidence cited to support a known beneficial effect of the intervention on the health of individuals or populations.

The StaRI standards refers to the broad range of study designs employed in implementation science. Authors should refer to other reporting standards for advice on reporting specific methodological features. Conversely, whilst all items are worthy of consideration, not all items will be applicable to, or feasible within every study.

Checklist item		Reported on page #	Implementation Strategy	Reported on page #	Intervention
			"Implementation strategy" refers to how the intervention was implemented		"Intervention" refers to the healthcare or public health intervention that is being implemented.
Title and abstract					
Title	**1**	123	Identification as an implementation study, and description of the methodology in the title and/or keywords		
Abstract	**2**	123	Identification as an implementation study, including a description of the implementation strategy to be tested, the evidence-based intervention being implemented, and defining the key implementation and health outcomes.		
Introduction					
Introduction	**3**	125	Description of the problem, challenge or deficiency in healthcare or public health that the intervention being implemented aims to address.		
Rationale	**4**	125	The scientific background and rationale for the implementation strategy (including any underpinning theory/framework/model, how it is expected to achieve its effects and any pilot work).	125	The scientific background and rationale for the intervention being implemented (including evidence about its effectiveness and how it is expected to achieve its effects).
Aims and objectives	**5**	130	The aims of the study, differentiating between implementation objectives and any intervention objectives.		
Methods: description					
Design	**6**	131	The design and key features of the evaluation, (cross referencing to any appropriate methodology reporting standards) and any changes to study protocol, with reasons		
Context	**7**	132	The context in which the intervention was implemented. (Consider social, economic, policy, healthcare, organisational barriers and facilitators that might influence implementation elsewhere).		
Targeted 'sites'	**8**	132	The characteristics of the targeted 'site(s)' (e.g. locations/personnel/resources etc.) for implementation and any eligibility criteria.	132	The population targeted by the intervention and any eligibility criteria.
Description	**9**	134	A description of the implementation strategy	134	A description of the intervention
Sub-groups	**10**	NA	Any sub-groups recruited for additional research tasks, and/or nested studies are described		
Methods: evaluation					
Outcomes	**11**	135	Defined pre-specified primary and other outcome(s) of the implementation strategy, and how they were assessed. Document any pre-determined targets	135	Defined pre-specified primary and other outcome(s) of the intervention (if assessed), and how they were assessed. Document any pre-determined targets
Process evaluation	**12**	135	Process evaluation objectives and outcomes related to the mechanism by which the strategy is expected to work		
Economic evaluation	**13**	NA	Methods for resource use, costs, economic outcomes and analysis for the implementation strategy	NA	Methods for resource use, costs, economic outcomes and analysis for the intervention

Sample size	**14**	NA	Rationale for sample sizes (including sample size calculations, budgetary constraints, practical considerations, data saturation, as appropriate)		
Analysis	**15**	137- 138	Methods of analysis (with reasons for that choice)		
Sub-group analyses	**16**	NA	Any a priori sub-group analyses (e.g. between different sites in a multicentre study, different clinical or demographic populations), and sub-groups recruited to specific nested research tasks		
Results					
Characteristics	**17**	149 and 153	Proportion recruited and characteristics of the recipient population for the implementation strategy	158	Proportion recruited and characteristics (if appropriate) of the recipient population for the intervention
Outcomes	**18**	149-181	Primary and other outcome(s) of the implementation strategy	149-181	Primary and other outcome(s) of the Intervention (if assessed)
Process outcomes	**19**	182 and 186	Process data related to the implementation strategy mapped to the mechanism by which the strategy is expected to work		
Economic evaluation	**20**	NA	Resource use, costs, economic outcomes and analysis for the implementation strategy	NA	Resource use, costs, economic outcomes and analysis for the intervention
Sub-group analyses	**21**	NA	Representativeness and outcomes of subgroups including those recruited to specific research tasks		
Fidelity/ adaptation	**22**	186	Fidelity to implementation strategy as planned and adaptation to suit context and preferences	186	Fidelity to delivering the core components of intervention (where measured)
Contextual changes	**23**	148	Contextual changes (if any) which may have affected outcomes		
Harms	**24**	187	All important harms or unintended effects in each group		
Discussion					
Structured discussion	**25**	182	Summary of findings, strengths and limitations, comparisons with other studies, conclusions, and implications		
Implications	**26**	190	Discussion of policy, practice and/or research implications of the implementation strategy (specifically including scalability)	190	Discussion of policy, practice and/or research implications of the intervention (specifically including sustainability)
General					
Statements	**27**	Appendix 7	Include statement(s) on regulatory approvals (including, as appropriate, ethical approval, confidential use of routine data, governance approval), trial/study registration (availability of protocol), funding and conflicts of interest		

Appendix 13 Site visit schedule and fieldnotes

Site 1 - start-up visit - 4/10/20 - Reflective diary regarding implementation study visit Site one - 4th of October 2019. Large Urban sleep medicine centre.

The group discussion on the dedicated sleep medicine Unit involved asking the participants what you normally do within a sleep medicine review. I wanted to learn about the routine within the department and how they normally structure a clinical review.

I was invited to present my research protocol to the entire department at the multidisciplinary meeting and there were a number of people who indicated they were willing to participate in the study. I was given by e-mail addresses of the interested participants are I arranged to provide the study documentation electronically. 8 clinicians indicated they would participate in the study, 3 consultants, and 5 nurses.

Following the multidisciplinary meeting, I met with four of the nurses from their sleep medicine department for an initial group discussion. These members of staff are based within the sleep medicine unit and they conduct sleep medicine clinical reviews regularly. Four of the nurses consented to participate in the implementation study at this first start-up visit.

We discussed the best way to facilitate the completion of the templates at the time of the clinical review and how to collect these anonymised templates after study completion. The nurse would use the templates in ward-based clinical review and keep the templates in a secure office until I was able to collect them. I felt that this was a good start to the implementation study however I had very little time with the consultant medical staff due to clinic workload and I knew that I would need a further site visit to facilitate their participation.

I was able to conduct an initial discussion with one of the Consultants on 4/10/20 by telephone and he also consented to participate in the study however he was then redeployed to another service and did not participate further.

5/11/19 - Site 1 – second visit – I attended a second site visit where I sat in on a morning clinic with one of the consultants to understand the medical clinic routine and spoke to the clinic outpatient staff to see how they may assist with attaching the templates to the case notes for completion and this was agreed, however, no templates were completed and there was no medical participation.

22/11/20 Site 1 – third visit – I attended the ward team meeting again requesting further medical and nursing staff participation and several medical staff members and several morse nurses agreed to participate.

14/1/20 – Site 1 – forth - final visit for two nurses who completed the study. Final face-to-face recorded semi-structured interviews with 2 of the specialist nurses who completed the study.

At this visit, I also met with two of the Respiratory Clinician and further team members requesting their participation in the study as I had already extended the study due to slow recruitment at site 1. Both consultants agreed to participate and further copies of the templates were provided to be attached to the clinic notes sheets for completion during the clinics.

8-2-20 – Site 1 visit 5 - A further site visit was conducted 8/2/20 again meeting with the department manager and several of the extended members of the nursing team who agreed to participate – however no further templates were completed and returned. And at the end of February 2020, COVID 19 appeared and there was no further participation from site 1.

4/11/2019 – Site 2

Start-up visit- Group discussion was undertaken with Specialist Nurse and Respiratory Consultant. The specialist nurse completes all the clinical reviews for CPAP and NIV patients and the consultants mostly just see the new patients. The specialist nurse uses her own template in her clinics however was happy to use the template for the duration of the study. The meeting was cut short due to train delays therefore the initial group discussion was too short to have a full discussion about what was included in the clinical review and we agreed that we could continue this conversation at a later date in the study.

12/2/20 – Site 2 – Final Telephone visit

Site 2 – Final structured recorded interview with 1 Respiratory Nurse specialist who completed the study was conducted by telephone as winter work pressures prevented me from visiting the site in person. The quality of the interview was much reduced by not being able to a video consultation that day as the technology was not working properly and attend anywhere was not working properly that day. I did take extensive field notes as well as recording the interview which helped to capture most of the content of the semi-structured interview.

Site 3 - 4-12-19 Start-up visit

This was conducted by teleconsultation using NHS near me with 2 Respiratory Physicians and two specialist nurses who consented to participate in the study. A member of the administrative team was tasked with providing a template for the consultations and collating the anonymised templates for return by registered post at the end of the study. It was agreed by the Lead Respiratory Consultant that they would commence the study the first week in January for a period of 8 weeks. Email correspondence from the Lead Respiratory physician from the 13th March 2020 confirmed that they completed the study as planned however due to the arrival of COVID 19 they were asked by the NHD board to cancel non-urgent visitors to the hospital so a final face to face visit to undertake the semi structured interviews was not possible. It was suggested in this email correspondence that the final interviews could be conducted by video consultation also however this was not possible due to redeployment of myself to the respiratory ward in our hospital due to COVID 19. The completed templates were returned to me by registered post for the quantitative data analysis. I sent the final semi-structured interviews topic guide to the participants on the 4th April 2020 with a request for online completion if they could find time to do this however this has not been possible to complete again due to COVID 19.

Appendix 14 Awards during PhD

From: RCN Foundation <RCN.Foundation@rcn.org.uk>
Date: 21 September 2018 at 16:11:52 BST
To: "phyllis.murphie@me.com" <phyllis.murphie@me.com>
Cc: RCN Foundation <RCN.Foundation@rcn.org.uk>
Subject: RCN Foundation Awards Night 2019 - Save the Date

Dear Phyllis,

Having received an education grant from the RCN Foundation, I am delighted to inform you that the Foundation's Trustees have selected you for an Impact Award.

This award is in recognition of what you have been able to achieve with the funding which you received from the Foundation.

The award will be presented at the RCN Foundation Awards Night on 21 March 2019 at 18.15 with carriages at 22.45. The dinner will be held at 20 Cavendish Square, London, W1G 0RN.

Please can you let us know if you want to attend on 21 March by the 1 October 2018. If you are attending the RCN Foundation will reimburse reasonable travel expenses and accommodation costs up to £150.

Once you have accepted this we will send you more information nearer the time.