

The use of nasal continuous positive airway pressure to treat obstructive sleep apnoea

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Aim: To review 66 children with obstructive sleep apnoea (OSA) for whom a trial of nasal continuous positive airway pressure (nCPAP) was proposed.

Methods: Baseline sleep studies were performed to assess OSA severity; a trial of nCPAP was performed where moderate to severe OSA, not relieved by adenotonsillectomy, was found. The nCPAP trial was considered either technically successful (ST), if the child accepted the mask for sufficient time to determine nCPAP efficacy, or a technical failure (FT) if otherwise. Patients with an initial FT were offered a period of home acclimatisation to familiarise them with wearing the mask during sleep. ST patients in whom nCPAP was effective were established on long term therapy.

Results: Nasal CPAP trials were successful (ST) in 49/66 (74%) patients. Nasal CPAP efficacy could not be determined in the remaining 17 FT patients (26%), generally because of their poor nCPAP tolerance. These patients were subsequently considered for other treatment. A total of 42/49 (86%) ST patients were established on long term nCPAP therapy, 2/49 (4%) derived no benefit from nCPAP, while 5/49 (10%) refused long term nCPAP therapy. Of patients on long term nCPAP, the most frequently reported side effects were skin irritation and nasal dryness; however, these were not serious enough to require any patients to discontinue therapy. A period of home acclimatisation was found to be effective in increasing nCPAP acceptance, with 26% of FT children being subsequently successfully reassessed for nCPAP.

Conclusion: The use of nCPAP was feasible in a significant proportion of a paediatric OSA population. Failure was usually because of the child's intolerance of the nCPAP equipment. Nasal CPAP was an effective treatment in the majority of patients where it could be assessed, and was adopted as a long term therapy in most cases. We have successfully used nCPAP to treat OSA across a wide range of ages. Motivated parents and skilled support staff have proved essential for the success of nCPAP in a paediatric setting.

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Nasal continuous positive airway pressure (nCPAP) for the treatment of obstructive sleep apnoea syndrome (OSA) was first described in 1981,¹ and is now widely accepted as the first line therapy in adult patients.² CPAP acts as a pneumatic splint to the upper airway, serving to maintain airway patency, preventing the airway collapse associated with obstructive sleep apnoea. The first reported use in children was in 1984, when Schmidt-Nowara described a single case where nCPAP was successfully used for one year.³ Two years later, Guilleminault *et al* proposed nCPAP as an alternative treatment to tracheostomy for children with medical conditions such as craniofacial anomalies or neuromuscular disorders in which (OSA) was not abolished by adenotonsillectomy.⁴ Over the intervening years there has been an increase in nCPAP use in children.⁵⁻¹⁵ The consensus of these studies is that nCPAP is both effective and well tolerated in more than 80% of children¹¹⁻¹²; in general, only minor side effects (nasal symptoms, air leaks, and skin breakdown) have been reported.¹²⁻¹⁵ However, there are case reports of patients in whom hypoventilation¹¹ and midface hypoplasia¹⁶ have been found with nCPAP therapy. Follow up nCPAP assessments are recommended every 6-12 months, as mask size and effective nCPAP pressure are likely to change with growth.¹²⁻¹⁷⁻¹⁸

Figure 1 shows a young child with nasal CPAP equipment in situ. Nasal CPAP is primarily used in children with medical conditions such as craniofacial anomalies, neuromuscular disorders, genetic syndromes, skeletal dysplasia, and obesity, in which upper airway obstruction is not cured by adenotonsillectomy.¹⁸ Additionally, nCPAP can be used in the preoperative period to stabilise children at risk of respiratory compromise for adenotonsillectomy, or as an interim treat-

ment while dental and facial growth are completed and craniofacial surgery can be performed.¹⁸⁻²⁰

Compliance in children, described in terms of nCPAP use as a percentage of hours prescribed, has been estimated as 50-100%,²¹ and seems to be related to the routine of home environment and care givers,²² but limited objective data are available. To our knowledge, large studies on nCPAP in children with OSA have been reported mainly from the United States and Australia,⁵⁻¹¹⁻¹²⁻¹⁵ with a larger number of single cases reported or in abstract form only.³⁻⁶⁻¹⁰ There are also a number of large studies referring specifically to the use of nCPAP to treat infantile sleep apnoea.⁵⁻²³⁻²⁵ The purpose of the present report was to describe our experience with nCPAP in children with OSA, as an example of a European experience.

METHODS

The data in this report are from the patient records of the Great Ormond Street Hospital Sleep Disorders Unit from 1994 to 1999. Every child that was diagnosed with moderate to severe OSA and in whom a nCPAP trial was suggested has been included. Treatment with nCPAP was recommended in children where surgical treatment of OSA was not possible. Patients using nCPAP for conditions other than OSA (for example, respiratory failure) have not been included.

Abbreviations: AS, active sleep; CPAP, continuous positive airway pressure; ECG, electrocardiogram; FT, failed treatment; nCPAP, nasal continuous positive airway pressure; OSA, obstructive sleep apnoea; QS, quiet sleep; ST, successful treatment



Figure 1 An infant undergoing a sleep study with nasal CPAP. The CPAP mask consists a rigid plastic frame with a soft silicone nasal cushion. The mask is held in place over the nose by a set of headgear with a three-point fixing to the mask frame using adjustable Velcro straps. The CPAP pressure is applied from a flow generator located at the cot side (out of picture) via the corrugated tubing, seen to the patient's right. Mask pressure is monitored during the CPAP trial via the narrow manometer tubing connected to the CPAP mask. The respiband monitoring respiratory movements is evident round the chest, an oximeter probe is attached to the right foot, and the ECG leads can be seen exiting from the child's clothes.

Sleep studies

Overnight sleep studies were performed on the respiratory ward and commenced at the child's usual bedtime. Studies ranged in duration between 6 and 10 hours and were supervised by an experienced sleep technician or nurse to ensure the quality of the recordings. Diagnostic studies included measurement of: electrocardiogram (ECG), arterial oxygen saturation (SpO_2 ; Ohmeda Biox) from a flexible probe attached to the finger or toe, thoracic and abdominal respiratory movements (strain gauges), and oronasal airflow via thermistor. A video (infrared camera) and sound recording was also made. Obstructive apnoea events, defined by the presence of chest and abdominal wall motion in the absence of airflow longer than two breath cycles, were counted.^{26, 27} The obstructive apnoea index was defined as the mean number of obstructive apnoeas per hour of sleep. Oximetry artefacts in the pulse waveform tracing were discarded manually following visual inspection of the traces. The mean and nadir SpO_2 values were determined. Oxyhaemoglobin desaturation episodes were defined as periods associated with a $\geq 4\%$ reduction from the prior 10 second baseline. Reductions in SpO_2 were reported as indices (for example, the number of events per hour of total sleep time). Because of the absence of electroencephalographic facilities in the polysomnograms used at the time of the survey (Visilab II, SSI Oxford UK, and CARDAS, Oxford UK), sleep stages were defined as either wake, quiet sleep (QS), or active sleep (AS) based on the presence of movements, heart rate variability, and by reference to the video record.

Full polysomnography remains the gold standard for diagnostic investigation, but universally accepted diagnostic criteria for OSA in children are lacking and normative data are limited.²⁶⁻²⁹ Based on our own laboratory criteria, we considered a child having moderate to severe OSA when one or more of the following criteria were met²⁶:

- (1) Obstructive apnoea index ≥ 5 events per hour
- (2) Desaturation index ≥ 4 events per hour with episodes of desaturation longer than 10 seconds and SpO_2 nadir less than 90%.

Other evidence supporting the diagnosis of OSA came from direct observations of the patient or from the review of the videotape and included:

- (1) Pronounced snoring for more than 50% of the night
- (2) Laboured breathing indicated by notable paradoxical inward movement of the chest, sternal retractions, or use of the accessory muscles of respiration
- (3) Frequent awakening from sleep associated with snoring, increasing respiratory effort, and partial obstruction.

Nasal CPAP trial

An nCPAP trial was performed during a second overnight sleep study, under the close supervision of a trained observer, to establish the efficacy of the therapy and to allow any practical problems with mask fitting or attachment to be corrected. The nCPAP trial was a split night study, with the initial part of the night off nCPAP to confirm the previous diagnosis, and the second part of the night with the nCPAP equipment fitted to the child. Titration of the nCPAP pressure commenced at the lowest pressure (4 cm H_2O) and involved gradually increasing the pressure by small (generally 2 cm H_2O) increments until obstructive sleep apnoea and oxyhaemoglobin desaturation were overcome. In practice, there is a notable change in the child's breathing pattern as soon as an effective pressure is reached. Breathing becomes less laboured, snoring stops, arousals are diminished, and SpO_2 stabilises. An nCPAP trial was considered successful (ST) if the child was cooperative in wearing the mask for the time necessary to define nCPAP efficacy; a failed trial (FT) was defined when the child did not tolerate the mask for the necessary time to define efficacy of the therapy. Nasal CPAP could only be classed as successful if it was shown to be effective in both quiet sleep (approximating to deep sleep stages) and active sleep (including REM sleep periods). The time taken to achieve a decision on nCPAP efficacy was 2-4 hours with nCPAP in place and depended on the level of nCPAP required and the pattern of the sleep states of the individual.

Children in whom nCPAP was accepted and found to be effective were sent home with the appropriate equipment. Parents received a detailed explanation about obstructive sleep apnoea, the need for treatment, and how the nCPAP system works. Telephone support, for any problems arising or for equipment replacement parts, was also given to the families. Follow up sleep studies and clinical assessments were performed at one month, six month, and one year intervals in order to evaluate the continued effectiveness of nCPAP, to readjust the mask size, and to change the pressure level where necessary. On each occasion, information regarding problems, side effects, and compliance were obtained from parents.

Home acclimatisation

For patients in whom the nCPAP trial was unsuccessful, a period of home acclimatisation to the nCPAP equipment was necessary before attempting the nCPAP trial a second time. For selected families or patients, a period of acclimatisation to the nCPAP equipment was necessary even before attempting the first nCPAP trial itself. Home acclimatisation involved the progressive introduction, by parents, of nCPAP to the child, using the following stages:

- (1) Use of the nCPAP mask alone (without tubing) with the parents introducing the mask during daytime play until the child was able to wear the open mask without distress.
- (2) Encouraging the child to go to sleep wearing the open mask. To minimise any dangers of hypercapnia from rebreathing, the mask was used with all the ports open (no tubing attached) and was removed by the parents shortly after the child fell asleep.
- (3) Once the child was able to wear the mask to go to sleep, nCPAP was introduced at the lowest pressure setting (4 cm H_2O). Once this stage was reached at home a formal nCPAP trial was arranged in the hospital.

Table 1 Summary of cases in which nCPAP was used

Congenital malformations of the upper airway	n	%
<i>Craniosynostosis syndromes*</i>	24	36.3
Isolated facial defects†	6	9.1
Osteochondrodysplasia‡	4	6.1
<i>Conditions predisposing to OSA</i>		
Mucopolysaccharidosis	8	12.1
Obesity§	4	6.1
Neuromuscular disease	4	6.1
Down's syndrome	2	3.0
Cerebral palsy	2	3.0
<i>Lower airway structural abnormalities</i>		
Laryngotracheomalacia, bronchomalacia	3	4.5
<i>Miscellaneous causes of OSA</i>		
Multiple congenital anomalies	1	1.5
Pickwickian syndrome	1	1.5
Sickle cell disease	1	1.5
Hydrocephalus	1	1.5
Cystic nasopharyngeal hygroma	1	1.5
Idiopathic OSA (no other causes found)	1	1.5
Infantile mixed central obstructive sleep apnoea	1	1.5
Pending adenotonsillectomy	1	1.5
Persisting OSA post-adenotonsillectomy	1	1.5
Total	66	100

*Craniosynostosis syndromes (n=24) of which: Pfeiffer syndrome (n=9), Apert syndrome (n=7), Crouzon syndrome (n=6), cranioleiodysostosis (n=2).

†Isolated facial defects (n=6) including: Treacher–Collins syndrome (n=3), Pierre–Robin syndrome (n=1), hemifacial microsomia (n=1), micrognathia (n=1).

‡Osteochondrodysplasia (n=4) including: achondroplasia (n=3), Jeune syndrome (n=1).

§Obesity (n=4) including: obesity + spastic quadriplegia (n=1), obesity + spina bifida (n=1), obesity + hypothalamic syndrome (n=1), simple obesity (n=1).

nCPAP equipment

The system used in the present study was the Sullivan-III nCPAP system (ResMed; Abingdon, UK) with the machine connected to the mask frame through a 2 m length of flexible tubing (internal diameter 2 cm), which allowed the child to move and to change position during sleep. The mask system used was either the infant or standard paediatric model, depending on the age and size of the individual. A number of sizes of soft silicone rubber nasal masks were available to achieve the optimal fit for each individual. Both the nasal mask and mask frame were held in place by standard head straps (fig 1).

Data analysis

All results are expressed as mean (SD). Differences between groups were compared using the Wilcoxon–Mann–Whitney test for continuous data and χ^2 analysis for categorical data.

RESULTS

Patient details

Data were obtained from 66 patients, 39 (59%) boys and 27 (41%) girls. At the time of the diagnostic sleep study, 18 (27.3%) patients were less than 1 year, 28 (42.4%) were aged 1–5 years, 12 (18.2%) were aged 6–12 years, and eight (12.1%) were aged 13–19 years. Table 1 reports details of patients in whom nCPAP was used.

Nasal CPAP trial history

Figure 2 shows the nCPAP trial histories of the 66 patients.

Of 66 children selected for a nCPAP trial, 64 (97%) underwent a first nCPAP trial in the hospital to define the efficacy of the therapy, while for two patients (3%), a period of home acclimatisation prior to nCPAP trial was chosen by physicians, because of low acceptance of the therapy by the parents. After a period of time, 4–6 months, these two families returned the machine to the hospital, reporting insurmountable difficulties in adapting the child to the therapy. Because

both children were at high risk of respiratory compromise for adenotonsillectomy and too young to have craniofacial surgery performed, the parents were strongly encouraged to continue with a further period of home acclimatisation, but despite this, success was not achieved. One of the two patients underwent adenotonsillectomy one year later, but a following sleep study showed incomplete resolution of obstruction. The other patient is under regular follow up until craniofacial surgery can be performed.

Of the 64 children who underwent the first nCPAP trial, 43 (67.2%) had a successful trial (ST) in that they tolerated the mask and nCPAP was shown to be effective. Twenty one (32.8%) were reluctant to wear the mask during the nCPAP trial, making it impossible to define nCPAP efficacy. For these children a period of home acclimatisation was proposed; this was accepted by 14 of the families (66.7%) and refused by seven (33.3%). Of these seven children, two had a nasopharyngeal tube inserted and were given supplemental oxygen, one underwent craniofacial advancement surgery when facial growth was completed, and three were lost to follow up.

From the 14 patients on home acclimatisation, nine (64.3%) came back for subsequent nCPAP trial when they were able to tolerate the lowest pressure at home, while the remaining five failed to be acclimatised, with parents reporting continuing difficulties adapting the child to the therapy. Of these five children, two were given supplemental oxygen, two were treated with nasal steroids until craniofacial surgery could be performed, and one was lost to follow up.

Of the nine children that underwent a second nCPAP trial, four (44.4%) tolerated the mask; nCPAP was shown to be successful and they went home on therapy. Five subjects (55.6%) still refused to wear the mask during the trial and awoke distressed at any attempts made, making it impossible to verify nCPAP efficacy.

Of 43 children in whom the first nCPAP trial was successful, 36 were started on long term therapy at home. In five cases,

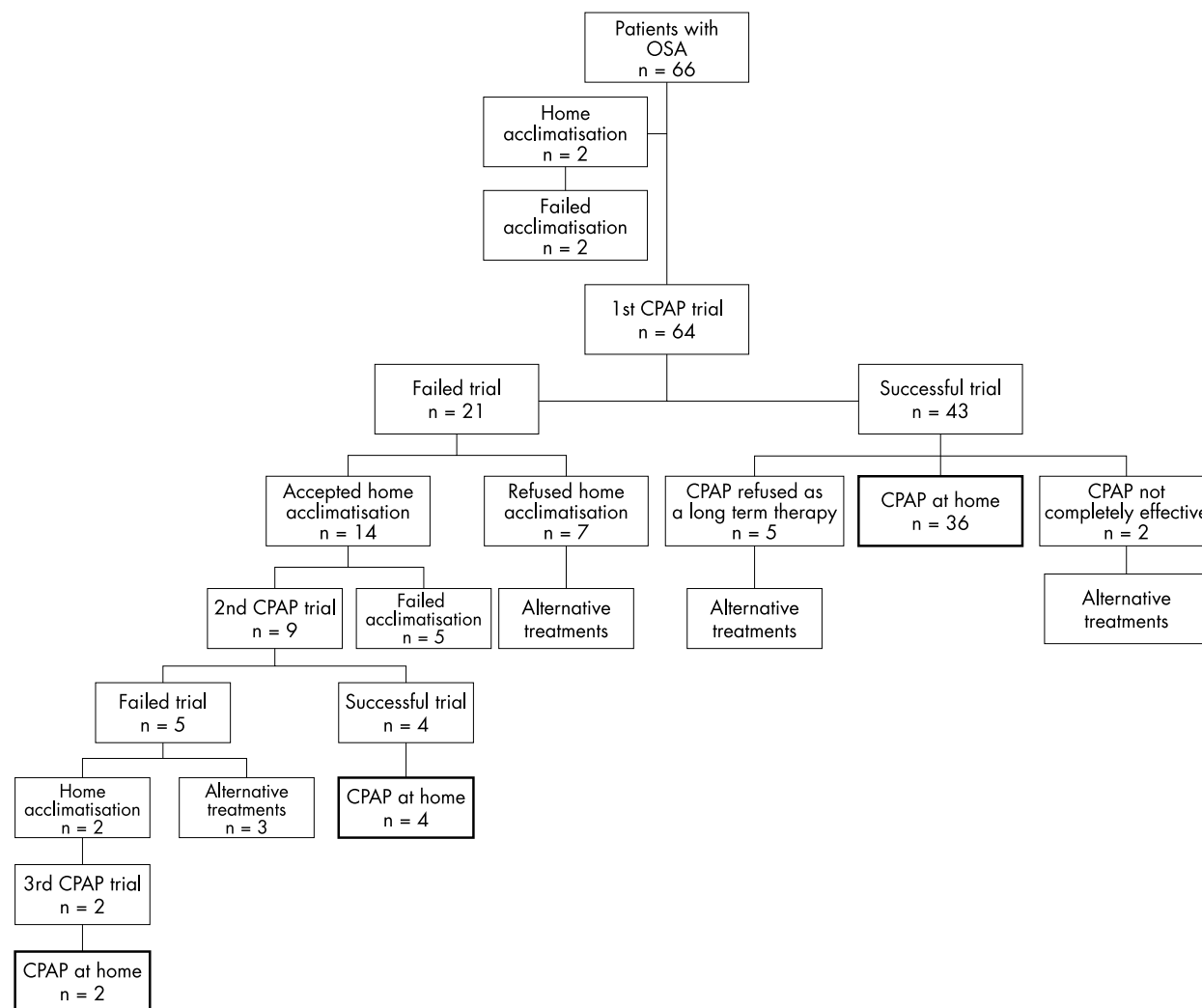


Figure 2 Flow diagram showing the history of 66 patients with OSA in which an nCPAP trial was suggested. The outlined boxes indicate the patients that were established on nCPAP therapy at home. For children that failed to be treated with nCPAP, alternative treatments such as nasopharyngeal prong, supplemental oxygen, and craniofacial surgery were used. One child underwent tracheostomy for acute respiratory failure and was subsequently ventilated with BiPAP via tracheostomy. One child underwent adenotonsillectomy after a period of stabilisation with CPAP.

although nCPAP was effective and the child accepted the mask during the trial, parents subsequently declined to continue the therapy as a long term treatment. Two of these patients had a nasopharyngeal tube inserted while a further three were lost to follow up. In two subjects (4.7%) nCPAP was found to be ineffective in alleviating the obstruction. One of them subsequently died as a result of exacerbation of his primary medical condition; the other was assessed for possible craniofacial surgery.

Excluding the two subjects in whom nCPAP was not effective, no statistically significant differences were found in terms of age, sex, or diagnosis, between the 42 children that finally accepted nCPAP therapy and the 22 children that, even after several attempts, refused to accept the therapy (table 2). Nevertheless, the highest proportion of the successful trials were achieved in children less than 1 year, and those older than 5, with consistently lower success rates in children between 1 and 5 years old (fig 3).

Long term nCPAP use

Of the 49 patients that completed the nCPAP trial successfully, 42 (86%) were established on long term nCPAP therapy. They have used nCPAP for a mean (SD) of 2.5 (1.8) years, ranging from 2 months to 6.5 years, with 69% of children using nCPAP

for more than one year. At the time of the study, 31 patients (73.8%) had continuously used the therapy at home, while 11 (26.2%) had interrupted the therapy. Of these, three had discontinued nCPAP during an admission for craniofacial surgery, one had a tracheostomy for acute respiratory failure but continued to be ventilated with BiPAP via tracheostomy, two subsequently died as a result of exacerbation of their primary medical condition, one had adenotonsillectomy, and one infant improved after one year of treatment. The longest duration of nCPAP therapy has been 6 years and 6 months in a boy who commenced nCPAP at the age of 4 years. Only three patients discontinued their therapy because of poor compliance.

Mean nCPAP pressure, used to abolish upper airway obstruction, was 8.5 (3.2) cm H₂O, ranging from 4 to 16 cm H₂O. The youngest child who was treated with nCPAP was a girl of 2 weeks of age; the oldest was a boy of 19 years and 4 months. None of these patients have had serious complications as a result of the therapy, although frequent minor discomforts were reported, generally related to poor mask fit (skin or eye irritation) or to nasal symptoms such as dryness. None of these problems has necessitated discontinuation of the therapy. Good compliance, with use every night and all night long was reported by 67.7% of parents; the others

Table 2 Comparison between patients that accepted and patients that refused nCPAP therapy

	nCPAP accepted (n=42)	nCPAP refused (n=22)
Age at diagnostic sleep study	5.3 (5.2)	4.3 (5.7)
Age at nCPAP trial	5.9 (5.1)	5.2 (5.5)
Sex	62% male 38% female	54.5% male 45.5% female
<i>Diagnoses</i>		
Congenital malformations of the upper airway	21 (50%)	12 (54.5%)
Conditions predisposing to OSAS	10 (23.8%)	9 (41%)
Lower airway structural abnormalities	3 (7.2%)	0
OSAS	4 (9.5%)	0
Associated medical conditions	4 (9.5%)	1 (4.5%)

Data are expressed as mean (SD) or n (%). The sum of patients is 64 because two patients in which nCPAP was not effective were excluded from this comparison. No statistically significant differences between the groups were found for any of the variables. OSAS, obstructive sleep apnoea syndrome.

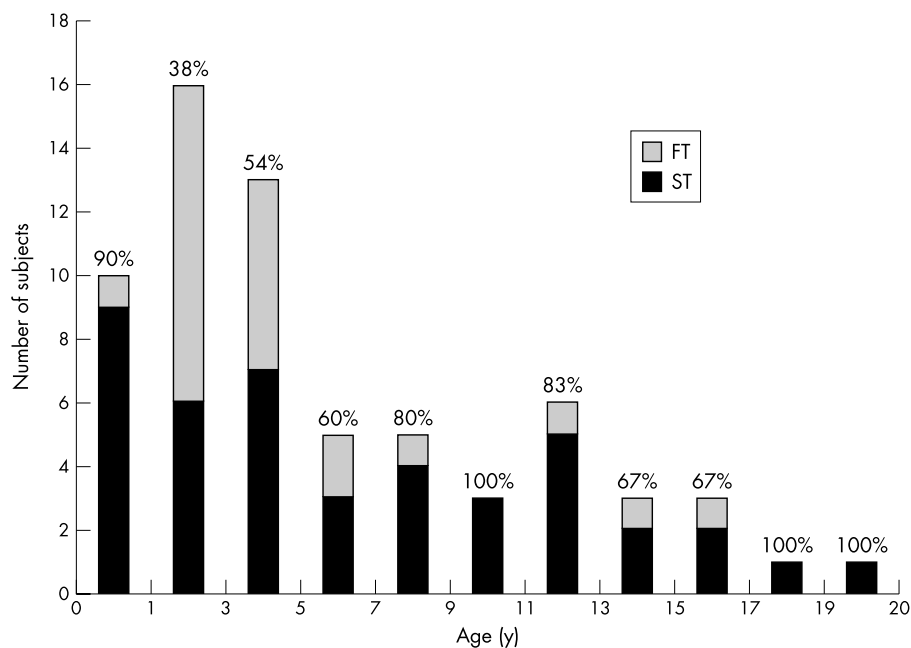


Figure 3 Number of children with successful nCPAP trials (ST) and with failed trials (FT), broken down by the age at outset of their nCPAP trial. Percentages represent success rate for each age group.

reported the child wearing CPAP to bed every night, but sometimes removing the mask during sleep.

DISCUSSION

We have described our experience using nCPAP to treat children with OSA. In our study, nCPAP was effective and tolerated by 86% of children; a period of acclimatisation in the home environment was found to be a useful strategy to achieve success in 26% of patients in whom initial assessments were not possible because of intolerance of nCPAP.

Experiences with nCPAP in children have been published mainly from the United States^{4-8, 12, 15-17} and Australia,^{3, 9, 11} while limited published data are available from Europe.¹³ The only UK report on paediatric patients was published by our group in 1996,¹³ and referred to the use of nCPAP in children with craniofacial dysostoses. In the present study we report our experience with nCPAP on a wider group of paediatric patients. In common with Waters *et al* and Marcus and coworkers,^{11, 12} we found an overall success rate of more than 80%, with the main reason for non-acceptance of the therapy being poor patient compliance. Patients complained of frequent minor discomforts, but they responded to simple

treatments such as a change of mask size or type, in case of forehead soreness or air leaks, the use of skin cream or protective tape to alleviate skin irritation, and passive humidification inserts in the case of nasal dryness. A period of acclimatisation in the home environment was useful to recuperate 37.5% of patients, who would otherwise have been lost as a result of low compliance.

The importance of a training programme to improve acceptance and tolerance to nCPAP in children, has been reported by Rains,²² where four children participated with their parents in a behavioural intervention programme before they were discharged with nCPAP. That intervention resulted in all patients tolerating nCPAP from the second night of treatment. In our study, the majority of failures have been prior to starting the therapy, when parents and children have to adapt to nCPAP use, while few have discontinued nCPAP because of poor compliance after the child has started on therapy. One factor that might have been expected to effect the success of nCPAP trials was the child's age. We found a higher rate of success in infants and in children over 5 years; for the intervening ages we found that the child frequently refused to wear the mask or to tolerate an applied pressure.

Our experience suggests that both parental determination and child cooperation are important to achieve success in this therapy. In 12% of cases, where nCPAP was effective and tolerated by the child during the trial, parents subsequently refused to continue the therapy at home. In these cases, it seems likely that the parents' poor perception of the need for, and benefits of nCPAP for their child, is the determining factor in the successful use of nCPAP in the long term. An advanced training programme for parents regarding (1) the importance of treating OSA to avoid the risk of complications, (2) the practical and simple use of nCPAP equipment, and (3) the behavioural management of the child's resistance to wear the mask, may be a useful strategy to reduce these failures. Further research should evaluate how to improve the rate of success with nCPAP in children.

From the group established on long term therapy, only one patient (who was an infant) has improved sufficiently to allow nCPAP therapy to be discontinued. It has been reported that nCPAP may be easier to discontinue over time in infants than in older children.²⁰ Regular follow up assessments are needed to investigate the possibility of weaning these patients off nCPAP.

Compliance in terms of nCPAP use, as a percentage of hours prescribed, has been estimated at 50–100%, with adolescents the least compliant,^{12 15 21} but objective data are lacking. We found that 67.7% of children were reported to use nCPAP regularly, while the others were reported to wear CPAP every night but sometimes remove the mask during sleep. These data are from parental reports, so need to be verified. Most CPAP devices today have a clock time counter incorporated that allows reading of the hours of use of the machine. Regrettably, the present retrospective study did not have complete records of counter readings. It would be desirable to have further prospective studies to evaluate the objective compliance with nCPAP in children and the reliability of parent reports.

Conclusion

We have been able to achieve similar success rates for the use of nCPAP as a treatment in children with OSA as those reported from outside the UK. It is possible to use nCPAP in a wide range of ages, from young babies through to older children, either as a primary treatment or as a bridge therapy in patients at high risk for surgery. In order to improve the success rate for nCPAP, there needs to be a focus on education and motivation of the parents to convince them of the efficacy and safety of nCPAP in order to reduce the number of failures.

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