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Tracheostomy for paediatric obstructive sleep apnoea: A systematic review

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Review Article

Dr M Camacho takes responsibility for the integrity of the content of the paper

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Abstract

Objective. To search the international literature (any language) for publications reporting outcomes of tracheostomy performed to treat obstructive sleep apnoea in children.

Method. Data sources included: Google Scholar, Cumulative Index to Nursing and Allied Health Literature, Embase, Scopus, and PubMed/Medline. Four authors searched systematically through to 20 January 2018.

Results. A total of 597 studies were screened; 64 were downloaded and 11 met criteria. A total of 196 patients underwent tracheostomy (mean age, 4.2 years; range, newborn to 18 years); 40 had detailed qualitative data and 6 had detailed quantitative data. Apnoea/hypopnoea index showed a 97 per cent reduction ($n = 2$) and apnoea index showed a 98 per cent reduction ($n = 3$). Lowest oxygen saturation showed a 34 oxygen saturation point improvement ($n = 3$). Several patients demonstrated significant improvement in breathing. All identified patients were syndromic, had significant co-morbidities or had severe obstructive sleep apnoea.

Conclusion. Based on reports of children who have undergone a tracheostomy, for whom there are pre- and post-operative data, tracheostomy appears to be a successful treatment for obstructive sleep apnoea. However, additional research is recommended given the small number of patients in the literature.

Introduction

Obstructive sleep apnoea (OSA) is a common sleep disorder in adults and children. In adults, tracheostomies have been used to treat OSA effectively, in both obese[1] and non-obese adult patients.[2] In children, first-line surgery generally consists of adenotonsillectomy.[3] Second-line treatments for children include myofunctional therapy,[4] rapid maxillary expansion (for transverse maxillary deficiency), weight loss, anti-inflammatory agents and positive airway pressure therapy.[5] However, some children may be candidates for tracheostomy as treatment for OSA, such as select children with craniofacial disorders, severe micrognathia (i.e. Pierre Robin sequence), severe microglossia and severe morbid obesity without adenotonsillar hypertrophy.

To our knowledge, a systematic review with meta-analysis for tracheostomy as a treatment for OSA has not been performed. Therefore, our objective was to search the international literature for the following participants, interventions, comparators, outcomes and study design ('PICOS') criteria: (1) the patients were children aged 18 years or younger with OSA; (2) the intervention was tracheostomy; (3) the comparison was pre- versus post-tracheostomy data; (4) the outcomes were sleep study data and qualitative descriptions of sleep-disordered breathing outcomes; and (5) the study design was any design, including randomised trials, cohort studies, case series, case reports, posters and abstracts.

Materials and methods

Search strategy

We performed a search beginning 1 April 2016 through to 20 January 2018. The Preferred Reporting Items for Systematic Reviews and Meta-Analyses ('PRISMA') statement was followed. The literature search and article selection details are shown in Figure 1.

Each database was searched beginning from its inception. The following databases were searched: Google Scholar, PubMed/Medline, the Cumulative Index to Nursing and Allied Health Literature, Embase, and Scopus. An example of one of the search strategies used in PubMed/Medline is: '(((tracheo*) AND (sleep OR apnea OR apnoea)) AND (pediatric OR paediatric OR kid OR kids OR children OR child OR infant OR neonate OR toddler))'.

Study selection

Studies reporting outcomes for respiratory disturbance index, apnoea/hypopnoea index, apnoea index, mean oxygen saturation, lowest oxygen saturation, oxygen desaturation

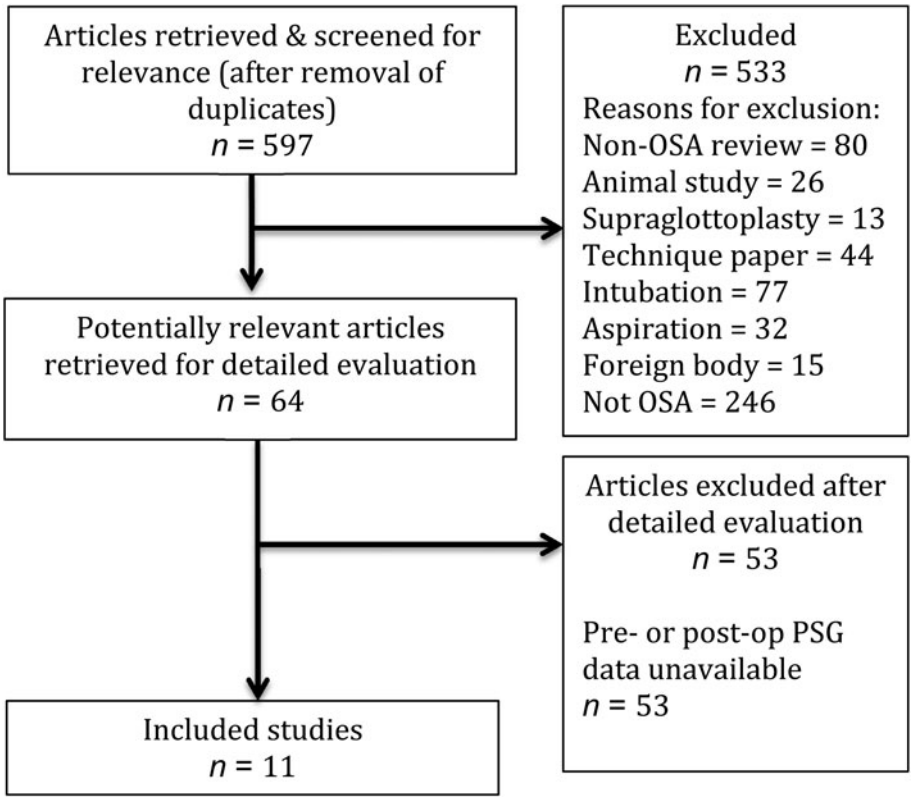

Fig. 1. Literature search and article selection. OSA = obstructive sleep apnoea; PSG = polysomnography

index, complications, quality of life (QoL) outcomes and/or mortality benefit were included. The studies included needed to report outcomes pre- and post-tracheostomy in a quantitative or qualitative fashion. There was no limitation based on language; therefore, the search included English and non-English manuscripts. Studies reporting outcomes for adults only were excluded.

Data abstraction

Four reviewers (MC, AB, JK and KL) independently performed a review of the literature, without regard to language. Abstracted data included the mean ages of children, the publication year, study sample sizes, respiratory disturbance index, apnoea/hypopnoea index, apnoea index, oxygen desaturation index, lowest oxygen saturation, mean oxygen saturation, QoL and/or mortality benefit.

Results

A total of 597 potentially relevant studies were screened. Sixty-four of these were downloaded in full-text form. Eleven of the studies met the criteria.[6–16]

A total of 196 patients identified in the literature underwent tracheostomy as treatment for OSA. The patients' mean age was 4.2 years (range, newborn to 18 years old). Forty patients had detailed qualitative data and six had detailed quantitative data.

The apnoea/hypopnoea index reduced from 34.2 ± 40 to 0.75 ± 0.35 events per hour (97 per cent reduction; $n = 2$), and the apnoea index reduced from 64.1 ± 24.9 to 1.9 ± 0.5 events per hour (98 per cent reduction; $n = 3$). Lowest oxygen saturation improved from 50.3 ± 26.5 to 84.3 ± 2.1 per cent (34 oxygen saturation point improvement; $n = 3$).

Several other patients demonstrated significant improvements in breathing.

Methodological quality

The identified studies included retrospective case series (level 4 evidence) and case reports (level 5 evidence) (Table 1).[6–16] There were no randomised trials identified in the literature.

Quantitative data

The quantitative outcomes identified in the literature are summarised in Table 2.[10–12,14–16]

Kasow *et al.* described a patient with malignant infantile osteopetrosis, who underwent a tracheostomy at 7.5 months of age.[14] The patient's apnoea/hypopnoea index was 5.9 events per hour pre-operatively and 0.5 events per hour post-operatively, corresponding to a 91.5 per cent relative reduction.

Gozal *et al.* described the longitudinal assessment of a Prader–Willi syndrome patient who underwent a tracheostomy.[10] Pre-operatively, the 14-year-old girl had an apnoea index of 42.2 events per hour, with a mean oxygen saturation level of 93 per cent and a lowest oxygen saturation level of 62 per cent. At four and at nine weeks post-tracheostomy, her apnoea/hypopnoea index was 2.4 and 1 event per hour, her mean oxygen saturation level was 96 and 98 per cent, and her lowest oxygen saturation level was 86 and 89 per cent, respectively. The relative reduction in the apnoea/hypopnoea index was 97.6 per cent.[10]

Handford *et al.* described an eight-year-old haemophilic boy, who had an apnoea/hypopnoea index above 62.5 events per hour, with each event lasting 20–40 seconds.[12] His lowest oxygen saturation level was 20 per cent or lower at times. About 60 per cent of the time, the events were associated with oxygen saturations between 40 and 60 per cent. He had frequent arrhythmias as well. The patient underwent a tonsillectomy, which failed. He subsequently underwent a tracheostomy, with a size 4 Shiley tracheostomy tube inserted. Two weeks later, the patient had no OSA with the

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Table 1. General characteristics and quality criteria of included studies

Study (year)	Study design	Study site	Evidence level	Patient co-morbidities	Outcomes analysed
Bannink *et al.*[6] (2010)	Retrospective cohort	Netherlands	IV	Syndromic craniosynostosis	Clinical outcomes, PSG, airway volume
Burstein *et al.*[7] (1995)	Prospective cohort	USA	IV	Cerebral palsy	AI, RDI
Cohen *et al.*[8] (1998)	Prospective cohort	USA	IV	Craniofacial disorders	Caregiver QoL
Cook & Berkowitz[9] (2005)	Case report	Australia	VI	Nemaline core myopathy	Clinical outcomes
Gozal *et al.*[10] (1996)	Case report	USA	VI	Prader–Willi syndrome	AI, MSAT, LSAT
Guilleminault *et al.*[11] (1976)	Case series	USA	VI	Haemodynamic abnormalities	AI
Handford *et al.*[12] (1984)	Case report	USA	VI	Haemophilia, arrhythmias, cor pulmonale	AHI, LSAT
Imataka *et al.*[13] (2016)	Case report	Japan	VI	Coffin–Lowry syndrome, seizures	LSAT, epileptic-like seizures
Kasow *et al.*[14] (2008)	Case series	USA	VI	Infantile osteopetrosis	AHI
Perks *et al.*[15] (1980)	Case report	UK	VI	Scheie syndrome	AI, MSAT
Rizzi *et al.*[16] (2017)	Case series	USA	VI	Severe OSA, craniofacial abnormalities, neuromuscular disorders	AHI, LSAT

PSG = polysomnography; AI = apnoea index; RDI = respiratory disturbance index; QoL = quality of life; MSAT = mean oxygen saturation; LSAT = lowest oxygen saturation; AHI = apnoea/hypopnoea index; OSA = obstructive sleep apnoea

Table 2. Quantitative outcomes identified in literature

			AHI or AI			LSAT (%)	
Study (year)	Cases (*n*)	Age	Pre-op value	Post-op value	Change (%)	Pre-op value	Post-op value
Kasow *et al.*[14] (2008)	1	7.5 mth	AHI = 5.9	AHI = 0.5	−91.5	69	82
Gozal *et al.*[10] (1996)	1	14 y	AI = 42.2	AI = 1.9	−97.6	62	86
Handford *et al.*[12] (1984)	1	8 y	AHI = 62.5	AHI = 1	−98.5	20	85
Perks *et al.*[15] (1980)	1	18 y	AI = 59	AI = 2.4	−95.9	76 apnoeas per hour	98 apnoeas per hour
Guilleminault *et al.*[11] (1976)	1	14 y	AI = 91.1	AI = 1.4	–	–	–
Guilleminault *et al.*[11] (1976)	1	12 y	816 apnoeas per night	–	–	–	–
Rizzi *et al.*[16] (2017) *	6	2 y	60.2 events per hour (95% CI = −15.7, 136.1)	6.6 events per hour (95% CI = −9.9, 23.1)	–	69.6 (95% CI = 46.9, 92.3)	90 (95% CI = 80.2, 99.8)
Total†		11.1 ± 6.1 y	AHI = 34.2 ± 40.0 AI = 64.1 ± 24.9	AHI = 0.75 ± 0.35 AI = 1.9 ± 0.5	AHI = −97.0 AI = −97.8	50.3 ± 26.5	84.3 ± 2.1

*Pre-tracheostomy data based on all patients and post-tracheostomy data based on capped sleep study, and are therefore excluded from total calculations. †Total values are based on studies providing pre- and post-tracheostomy data. AHI = apnoea/hypopnoea index; AI = apnoea index; LSAT = lowest oxygen saturation; pre-op = pre-operative; post-op = post-operative; mth = months; y = years; 95% CI = 95 per cent confidence interval

tracheostomy in place, with a lowest oxygen saturation level between 85 and 89 per cent. One week later, he had resolving cor pulmonale, based on electrocardiographic and X-ray evidence.[12]

Perks *et al.* described an 18-year-old patient with Scheie's syndrome (mucopolysaccharidosis) who had OSA.[15] Pre-tracheostomy, the patient had an apnoea index of 59 events per hour and a mean oxygen saturation level of 76 per cent. Post-tracheostomy, the apnoea index reduced to 2.4 events per hour and the mean oxygen saturation level was 98 per cent. The apnoea index demonstrated a relative reduction of 95.9 per cent.[15]

Guilleminault *et al.* described 2 patients with OSA treated with a tracheostomy: a 14-year-old girl with 91.1 apnoea index and a 12-year-old boy with 816 apnoeas per night.[11] Both patients normalised their breathing during sleep with the tracheostomy in place. Both patients also had a dramatic reversal of haemodynamic abnormalities and clinical symptoms within 48 hours of the surgery.[11]

Qualitative data

Burstein *et al.* described two children with severe cerebral palsy with flaccidity, who underwent upper airway surgery.[7]

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Extubation had failed and a tracheostomy was subsequently used for salvage surgery.

Bannink *et al.* described children with syndromic craniosynostosis and severe OSA.[6] Five patients underwent tracheostomy. Four children were successfully decannulated after midface advancement, while one patient required the tracheostomy in order to eliminate OSA.

Cohen *et al.* described sleep apnoea surgery versus tracheostomy, and evaluated QoL outcomes.[8] A 76-item questionnaire was developed and used. Overall, the tracheostomy patients' parents ranked 95 per cent of all items on the questionnaire as being worse than the sleep apnoea surgery patients' parents' rankings. The QoL data assessed in the parents' questionnaires included hours per day spent on respiratory care, average medical visits per year, estimated costs per month and a psychosocial subscale; all items were scored worse in the tracheostomy group versus the sleep apnoea surgery group.[8]

Cook and Berkowitz described a tracheostomy in a child with nemaline core myopathy.[9] Following the failure of adenoidectomy performed as treatment for OSA (lowest oxygen saturation level of 55 per cent), the patient underwent a tracheostomy. Later, the patient developed lobar collapse and sepsis, and was placed on bilevel positive airway pressure therapy. A repeat sleep study conducted six months later showed adequate ventilation.[9]

Imataka *et al.* described a 12-year-old boy with Coffin–Lowry syndrome, who had epileptic-like seizures induced by OSA syndrome.[13] The patient had a lowest oxygen saturation level down to 60 per cent. He underwent a tracheostomy; the OSA subsequently improved and the epileptic-like seizures were eliminated.

Rizzi *et al.* described 29 paediatric patients who underwent a tracheostomy for severe OSA.[16] The mean age at time of surgery was two years. Forty-five per cent of the patients had an associated craniofacial abnormality and 34 per cent had a neuromuscular disorder. The majority of children requiring tracheostomy for OSA remained tracheostomy-dependent for more than 24 months; there were no long-term complications following tracheostomy placement. Over the follow-up period, 6 of 29 patients (21 per cent) underwent a capped sleep study; 5 of these patients were decannulated. The average apnoea/hypopnoea index at the time of the capped study was 6.6 events per hour (95 per cent confidence interval (CI) = 29.9–23.1). The mean blood oxygen saturation nadir on post-operative polysomnography was 90.0 per cent (95 per cent CI = 80.2–99.8). This represented a mean apnoea/hypopnoea index decrease of 89 per cent and a mean blood oxygen saturation nadir increase of 29.3 per cent. Decannulation was achieved in 5 of 29 patients (17 per cent), including 1 self-decannulation. Mean time to decannulation was 40.8 months (95 per cent CI = 7.9–73.7). Of the 16 patients followed up for 24 months, only 1 (6 per cent) had been decannulated at the 24-month mark.[16]

Discussion

This systematic review has three main findings. First, the paediatric patients who underwent a tracheostomy consistently had a significant improvement in OSA outcomes. In the three patients who had pre- and post-tracheostomy apnoea/hypopnoea index outcomes reported, the apnoea/hypopnoea index decreased by 95 per cent. Several other paediatric patients were reported to have normalised their breathing, but did not have the sleep study outcomes reported quantitatively. Additionally, oxygen saturation also improved significantly after surgery in those for whom it was quantified. It is logical that bypassing the upper airway with a tracheostomy provides significant improvement, and even cure, in the majority of patients. Patients with residual OSA could have apnoeas or hypopnoeas secondary to neck soft tissue obstructing the lumen of the tracheostomy tube; in other patients, there may be tracheomalacia contributing to the OSA. In addition, patients with co-morbid obesity hypoventilation syndrome may need positive airway pressure therapy through the tracheostomy tube, in order to eliminate OSA.

Second, the patients who underwent tracheostomy for paediatric OSA were syndromic children, had significant co-morbidities or had severe OSA. We did not identify any mild or moderate OSA patient without significant co-morbidities or syndromes in the literature. This is probably because most of the patients would be treated with traditional adenotonsillectomy, and secondary surgical procedures (e.g. uvulopalatopharyngoplasty, tongue surgery or mandibular advancement surgery) would be attempted before a tracheostomy was placed. Syndromes that predispose patients to OSA are those that cause a small mandible or retrodisplaced mandible, such as Pierre Robin sequence, which can be treated with a temporary tracheostomy while the patient undergoes mandibular advancement surgery.

Third, additional research is needed. Currently, there have been many studies reporting OSA outcomes for adults.[2] There are very few studies reporting outcomes in children (11 manuscripts with 46 patients). Because many children undergoing a tracheostomy for OSA are diagnosed clinically, the majority of studies did not report quantitative data, but rather provided qualitative data. Although we value research and quantitative sleep study data, we also have to be reasonable; we are not suggesting that all patients should undergo a sleep study. Given the importance of stabilising the airway of a child in respiratory distress due to severe OSA, it may be inappropriate to wait to perform a tracheostomy in these situations. Additionally, the smallest tracheostomy diameter that can be used to provide relief of OSA is currently not known; however, a systematic review found that mini-tracheostomies as small as 4 mm have been used successfully in the short term to treat acute upper airway obstruction.[17]

Limitations

There are few studies reporting outcomes in children. Future studies could perform pre- and post-operative sleep study outcomes when it is reasonable. It is unlikely that patients will be randomised between no treatment or tracheostomy groups; however, to increase the level of evidence, the use of prospective studies, and in some cases randomisation to positive pressure therapy or tracheostomy groups, could be considered.

Conclusion

This paper describes a level of evidence 3a study. Based on reports of children who have undergone a tracheostomy, for whom there are pre- and post-operative data, tracheostomy appears to be a successful treatment for OSA. However, given the small number of patients in the literature, additional research is recommended.

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Competing interests. The views expressed in this article are the private views of the authors and do not necessarily reflect the official views of the Department of the Army, the Department of Defense, or the US Government.

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