

Main Article

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Corresponding author:

Aikaterini Dritsoula;
Email: adritsoula@yahoo.co.uk

The role of rigid laryngo-tracheo-bronchoscopy in children with obstructive sleep apnoea: a case series of 65 children

Aikaterini Dritsoula^{1,2} , Raymond Clarke³, Elpis Hatziagorou⁴, Stefanos Triaridis⁴, Persefoni Talimtzis⁴ and Heather Elphick⁵

¹ENT Department, Sheffield Children's NHS Foundation Trust, Sheffield, UK, ²Leeds Teaching Hospitals NHS Trust, Leeds, UK, ³ENT Department, Alder Hey Children's NHS Foundation Trust, Liverpool, UK, ⁴School of Medicine, Aristotle University of Thessaloniki, Thessaloniki, Greece and ⁵Paediatric Respiratory and Sleep Medicine, Sheffield Children's NHS Foundation Trust, Sheffield, UK

Abstract

Objective. To assess the role of laryngo-tracheo-bronchoscopy in children with obstructive sleep apnoea by identifying airway abnormalities at surgery, that occur separately or in addition to adenotonsillar hypertrophy, and examining the correlation with respiratory parameters.

Methods. A retrospective study was conducted of children with obstructive sleep apnoea who underwent laryngo-tracheo-bronchoscopy intra-operatively, performed by a single ENT surgeon from February 2016 to July 2019. Pre- and post-operative minimum oxygen saturation, apnoea-hypopnoea index, and oxygen desaturation index were recorded.

Results. Sixty-five children were identified; 34 were aged less than three years and 31 were aged three years or more. 77 per cent and 13 per cent respectively had an airway abnormality; the *t*-test showed a significantly higher mean oxygen desaturation index and lower mean minimum oxygen saturation pre-operatively compared to children without an airway abnormality.

Conclusion. An update of the surgical pathway for children aged less than three years with obstructive sleep apnoea is required to include laryngo-tracheo-bronchoscopy intra-operatively. A *t*-test analysis of the pre-operative respiratory parameters suggests that airway abnormalities contribute to obstructive sleep apnoea severity.

Introduction

The spectrum of respiratory disorders during sleep is described as sleep-disordered breathing. Obstructive sleep apnoea (OSA) is defined as the disruption of normal ventilation and normal sleep patterns during sleep due to prolonged partial and/or intermittent complete upper airway obstruction, and lies at the severe end of the spectrum. Symptoms of OSA include snoring, breathing pauses overnight, restlessness and daytime tiredness. Furthermore, children can have neurocognitive symptoms such as attention deficits, hyperactivity, aggressive behaviour and learning difficulties.¹

The American Academy of Otolaryngology – Head and Neck Surgery, and the American Academy of Paediatrics, recommend adenotonsillectomy as the primary therapy in healthy children aged over two years of age with OSA. Continuous positive airway pressure (CPAP) is a treatment option for children for whom surgery is not suitable or who have persistent OSA after adenotonsillectomy. Lingual tonsillectomy and supraglottoplasty are the most common procedures reported in children with persistent OSA after adenotonsillectomy.²

Rigid laryngo-tracheo-bronchoscopy is the direct visualisation of the upper airway under general anaesthesia with the use of a rigid Hopkins rod endoscope. We propose that when performed in children with OSA, laryngo-tracheo-bronchoscopy can guide airway surgical intervention intra-operatively, and when performed at the same time as adenotonsillectomy it will assist in planning further surgical management in children with persistent OSA after adenotonsillectomy. This is especially important in those who will require CPAP, as this treatment option is not as well tolerated in the paediatric population.³

Our case series allows examination of the role of laryngo-tracheo-bronchoscopy in children with OSA by identifying airway abnormalities, that occur separately or in addition to adenotonsillar hypertrophy, in this group of children during surgery; in addition, the correlation with respiratory parameters is examined.

Materials and methods

The study was a retrospective patient records evaluation. Health Research Authority approval was obtained (Integrated Research Application System 'IRAS' number: 301085).

Children with OSA who underwent laryngo-tracheo-bronchoscopy plus supraglottoplasty or aryepiglottoplasty, or laryngo-tracheo-bronchoscopy with or without tonsillectomy and with or without adenoidectomy, under the care of a single ENT surgeon in Sheffield Children's Hospital, from February 2016 to July 2019, were identified. These children mainly had OSA symptoms, such as snoring, overnight restlessness, breathing pauses and daytime tiredness. Laryngo-tracheo-bronchoscopy plus supraglottoplasty or aryepiglottoplasty was performed in children with OSA and signs of laryngomalacia, such as inspiratory stridor, subcostal recessions and flexible endoscopy findings of laryngomalacia, when feasible. Laryngo-tracheo-bronchoscopy with or without tonsillectomy and with or without adenoidectomy, was performed in children with OSA who also had: a co-morbidity (e.g. congenital syndrome, craniofacial anomaly, neuromuscular disease) with a higher risk of an airway abnormality, or had airway symptoms that could not be attributed to adenotonsillar hypertrophy, such as stridor and recurrent croup or chest infections.

Data on age at surgery, airway abnormalities identified on laryngo-tracheo-bronchoscopy, the surgical procedure (adenotonsillectomy, tonsillectomy, adenoidectomy, supraglottoplasty/aryepiglottoplasty) performed and any co-morbidities were recorded. If the child underwent pre- and post-operative cardiorespiratory polygraphy or a pulse oximetry test, respiratory parameters were also recorded. The values of oxygen saturation nadir for all sleep studies, the apnoea-hypopnoea index in children who underwent polygraphy, and the 4 per cent oxygen desaturation index in children who had a pulse oximetry test were recorded, respectively.

Children were analysed as a whole group and as subgroups according to age – less than three years versus three years or more – because of the high percentage of airway abnormalities in children younger than three years.^{4–6}

Continuous variables are presented as means and standard deviations, while categorical variables are shown as frequencies and percentages. The independent samples *t*-test was used to compare pre-operative oxygen desaturation index or apnoea-hypopnoea index, and oxygen saturation, in: (1) children with an airway abnormality versus those without; (2) children with an airway abnormality who are aged less than three years versus those aged three years or more; and (3) children with a

co-morbidity versus those without. The chi-square test was used to compare children, aged less than three years with those aged three years or more, with airway abnormalities identified on laryngo-tracheo-bronchoscopy, and children with and without co-morbidities. Fisher's exact test was used to compare children, aged less than three years with those aged three years or more, with airway abnormalities identified on laryngo-tracheo-bronchoscopy only in children with co-morbidities. Statistical analysis was performed using IBM SPSS Statistics software, version 27. The statistical significance level was set at 5 per cent.

Results

Sixty-five children underwent laryngo-tracheo-bronchoscopy as part of their surgical management for OSA. Their mean age was 3.5 years (range of 3 months to 15 years and 5 months). Forty of the patients (61.5 per cent) were boys. Thirty-four patients were aged less than three years and 31 were aged three years or more. Statistical analysis was themed, as shown below.

First, the rates of airway abnormalities identified on laryngo-tracheo-bronchoscopy were examined. Laryngomalacia was the prevalent airway abnormality, identified in 18 children (60 per cent). All laryngo-tracheo-bronchoscopy findings are recorded in Table 1.

Second, airway abnormalities identified on laryngo-tracheo-bronchoscopy in children aged less than three years were compared to those in children aged three years or more. One or more airway abnormalities were found on laryngo-tracheo-bronchoscopy in 30 out of the 65 children (46 per cent). Airway abnormalities were found in 26 out of 34 children aged less than three years (77 per cent), and in 4 out of 31 children aged three years or more (13 per cent). When children aged less than three years and three years or more were compared with regard to the presence of airway abnormalities on laryngo-tracheo-bronchoscopy, the difference was statistically significant ($p < 0.05$).

Third, when children with co-morbidities and airway abnormalities identified on laryngo-tracheo-bronchoscopy in those aged less than three years (9 out of 10) were compared with those in children aged three years or more (3 out of 10), the difference was statistically significant ($p = 0.02$) (Table 2).

Table 1. Airway abnormalities identified on laryngo-tracheo-bronchoscopy in children with OSA

LTB finding	All children	Children aged <3 years	Children aged ≥3 years
Number of patients	65	34	31
No LTB findings (<i>n</i>)	35	8	27
1 LTB finding (<i>n</i> (%))	23	20	3
– Laryngomalacia	18 (60)	17	1
– Tracheomalacia	3 (10)	2	1
– Supraglottic obstruction due to retrognathia	1 (3.25)	0	1
– Compression of anterior tracheal wall	1 (3.25)	1	0
>1 LTB finding (<i>n</i> (%))	7	6	1
– Laryngomalacia & tracheomalacia	3 (10)	3	0
– Laryngomalacia & SGS	1 (3.25)	1	0
– Laryngomalacia, compression of anterior tracheal wall & bronchomalacia	1 (3.25)	1	0
– SGS & tracheomalacia	2 (7)	1	1

OSA = obstructive sleep apnoea; LTB = laryngo-tracheo-bronchoscopy; SGS = subglottic stenosis

Table 2. Co-morbidities in children with OSA

Co-morbidities	All children	Children aged <3 years	Children aged ≥3 years
Number of patients	20	10	10
≥1 LTB finding (<i>n</i> (%))	12/20 (60)	9/10 (90)	3/10 (30)
- Down syndrome	5	4	1
- Stickler syndrome	1	0	1
- HIE, dystonia, epilepsy	1	1	0
- Ex-prematurity, mitral regurgitation, intraventricular haemorrhage	1	1	0
- Pulmonary stenosis, GERD	1	1	0
- Absent corpus callosum	1	1	0
- Epilepsy	1	1	0
- Epilepsy, periventricular leukomalacia, GERD	1	0	1

OSA = obstructive sleep apnoea; LTB = laryngo-tracheo-bronchoscopy; HIE = hypoxic-ischaemic encephalopathy; GERD = gastroesophageal reflux disease

Fourth, comparison of the mean pre-operative oxygen desaturation index and oxygen saturation in children with and without an airway abnormality revealed a significantly higher mean oxygen desaturation index and significantly lower mean oxygen saturation values (Table 3).

Fifth, comparisons of pre- and post-operative polygraphy or pulse oximetry test were made. Seventeen out of all 65 children (26 per cent of all children) had both pre- and post-operative findings for polygraphy or the pulse oximetry test. When the apnoea-hypopnoea index or oxygen desaturation index were compared, six children (35 per cent) showed resolution of their OSA (apnoea-hypopnoea index of ≤1, oxygen desaturation index of <5), five (29 per cent) had improvement, two (12 per cent) showed deterioration, and four (24 per cent) demonstrated no change and normal oxygen desaturation index values both pre- and post-operatively (Table 4).

Discussion

In this cohort of 65 children referred with symptoms suggestive of OSA, nearly half had an airway abnormality. These were more commonly found in children aged under three years. When both co-morbidity and age were taken into account, children aged less than three years were much more likely to have airway abnormalities identified on laryngo-tracheo-bronchoscopy compared to those aged three years or more. The airway abnormality with the highest frequency was laryngomalacia, and 19 children underwent aryepiglottoplasty or supraglottoplasty to address their OSA. Pre-operative sleep

studies showed more severe OSA in children with an airway abnormality.

The role of laryngo-tracheo-bronchoscopy has been minimally studied in terms of guiding surgical management in children with OSA. Rastatter *et al.*⁴ examined the prevalence of synchronous airway lesions in 110 children younger than three years, with no co-morbidities or with a known pre-existing synchronous airway lesion, who were undergoing adenotonsillectomy for sleep-disordered breathing, using a complete airway evaluation that included rigid laryngo-tracheo-bronchoscopy. Sixty-seven per cent of the children were found to have at least one synchronous airway lesion. The Respiratory Distress Index determined by a polygraphy in 35 of the 110 children was not predictive of the presence of a synchronous airway lesion. The authors concluded that the degree of impact that these lesions have on the severity of the children's sleep-disordered breathing is unclear.

Michelson *et al.*⁵ conducted a retrospective study on 39 children younger than three years with OSA, who underwent adenotonsillectomy, or powered-intracapsular tonsillectomy with adenoidectomy, and had both pre- and post-operative polygraphy findings. Thirty-eight per cent of the total population had an airway evaluation, and synchronous airway lesions were identified in 60 per cent of that cohort. The study was limited in detecting subglottic and tracheal synchronous airway lesions, as only less than half of the patients' airway evaluations included a laryngo-tracheo-bronchoscopy. The study concluded that adenotonsillectomy is effective for OSA in children aged less than three years, and the presence of a

Table 3. Mean pre-operative ODI, AHI and SpO₂ in children with and without airway abnormalities

Parameter	<i>n</i>	Mean Pre-op ODI or AHI (SD)	Mean Pre-op SpO ₂ (SD)
Children with pre-op pulse oximetry test findings	44		
- With airway abnormality	16	ODI = 11.5 (14.22)	79.2 (12.7)
- Without airway abnormality	28	ODI = 3.1 (4.3)	86.9 (7.1)
- Mean difference (95% CI)		8.5 (0.7, 16.2)*	-7.6 (-13.6, -14.8)*
Children with pre-op polygraphy findings	16		
- With airway abnormality	11	AHI = 17.1 (14.9)	76.4 (14.8)
- Without airway abnormality	5	AHI = 14.1 (17.6)	86.8 (4.8)
- Mean difference (95% CI)		2.9 (-15.3, 21.2)	-10.4 (-25.2, 4.4)*

*Statistically significant at 0.05 level. ODI = oxygen desaturation index; AHI = apnoea-hypopnoea index; SpO₂ = oxygen saturation; SD = standard deviation; CI = confidence intervals; pre-op = pre-operative

Table 4. Outcome after surgery for individual children with both pre- and post-operative sleep study findings

Patient number	LTB finding	Surgical intervention	Pre-op polygraphy (AHI/SpO ₂)	Post-op polygraphy (AHI/SpO ₂)	Pre-op pulse oximetry test (ODI/SpO ₂)	Post-op pulse oximetry test (ODI/SpO ₂)	Co-morbidity	Outcome
1	SGS grade I & tracheomalacia	Intracapsular tonsillectomy & adenoidectomy	23.4/82	2.7/87	–	–	Down syndrome	Improvement
2	Laryngomalacia	Supraglottoplasty	–	–	6.99/83	5.45/86	–	Improvement
3	Laryngomalacia	Supraglottoplasty	–	–	6.45/81	3.90/85	–	Resolution
4	Laryngomalacia	Supraglottoplasty	–	–	8.15/78	3.39/75	Down syndrome	Resolution
5	Laryngomalacia	Supraglottoplasty	–	–	13.68/78	20/76	Pulmonary stenosis, GERD	Deterioration
6	Compression of tracheal anterior wall	Intracapsular tonsillectomy	13.3/76	4.2/91	–	–	–	Improvement
7	Laryngomalacia	Coblation adenotonsillectomy & supraglottoplasty	–	–	59.13/36	0.77/88	–	Resolution
8	Laryngomalacia & SGS grade I	Aryepiglottoplasty	30/47	36/73	–	–	Down syndrome	Deterioration
9	Laryngomalacia	Supraglottoplasty	–	–	20.53/77	5.94/78	–	Improvement
10	None	Intracapsular tonsillectomy & adenoidectomy	–	–	7.35/81	0.36/91	ASD	Resolution
11	None	Revision adenoidectomy & lingual tonsillectomy	–	–	1.19/88	4.57/90	Down syndrome	No change
12	None	Revision adenoidectomy	44/88	4.5/89	–	–	–	Improvement
13	None	Intracapsular tonsillectomy & adenoidectomy	–	–	20.08/64	3.08/81	–	Resolution
14	None	Intracapsular tonsillectomy & adenoidectomy	–	–	7.51/76	1.36/92	–	Resolution
15	None	Coblation adenotonsillectomy	–	–	0.56/93	2.71/78	Chromosome 13.1 deletion	No change
16	None	Coblation adenotonsillectomy	–	–	2.62/92	3.08/83	Down syndrome, ex-prematurity, VSD, tricuspid regurgitation	No change
17	None	Coblation tonsillectomy	–	–	1.17/94	0.13/92	–	No change

LTB = laryngo-tracheo-bronchoscopy; pre-op = pre-operative; AHI = apnoea-hypopnoea index; SpO₂ = oxygen saturation; post-op = post-operative; ODI = oxygen desaturation index; SGS = subglottic stenosis; GERD = gastroesophageal reflux disease; ASD = autism spectrum disorder; VSD = ventricular septal defect

synchronous airway lesion does not necessarily predict treatment failure.

Mandell and Yellon⁶ studied 35 children younger than 18 months who underwent adenoidectomy and airway evaluation (30 underwent rigid tracheo-bronchoscopy). Children with craniofacial dysmorphism and congenital syndromes were excluded. The authors reported that 59 per cent of children younger than 18 months had synchronous airway lesions, of which more than 50 per cent were laryngomalacia and laryngeal oedema.

In all the above studies, there was a high percentage of children (59 per cent or more) younger than three years with an airway abnormality other than adenotonsillar hypertrophy. This is comparable with the results of this study, which looked for airway abnormalities in children with OSA, occurring separately or in addition to adenotonsillar hypertrophy. If children with co-morbidities are excluded from our study, then 50 per cent of the cohort presented with an airway abnormality. If the meta-analysis by Friedman *et al.*⁷ is taken into consideration, which reported approximately 34 per cent of children with persistent OSA after adenotonsillectomy, laryngo-tracheo-bronchoscopy will help guide further surgical management in many children aged less than three years worldwide. A change or update of the surgical pathway for children with OSA aged less than three years, which currently prioritises adenotonsillectomy, needs to be considered, wherein laryngo-tracheo-bronchoscopy is included as part of the surgical management.

The role of laryngo-tracheo-bronchoscopy in children aged three years or more has not been studied before. In our study, only 13 per cent of the children had an airway abnormality. A co-morbidity was present in 75 per cent of the children with an airway abnormality.

Camacho *et al.* reported that supraglottoplasty can improve the apnoea-hypopnoea index and lowest oxygen saturation when performed for children with OSA, either with congenital laryngomalacia or exclusively presenting during sleep, with the majority of children not being cured.⁸ This is supportive of the rationale that airway abnormalities such as laryngomalacia are reflected in the respiratory parameters of sleep studies, which are affected when the airway abnormalities are surgically addressed as above.

- In this study, 77 per cent of children aged less than three years with obstructive sleep apnoea (OSA) had an airway abnormality identified on laryngo-tracheo-bronchoscopy
- These abnormalities occurred separately or in addition to adenotonsillar hypertrophy
- Polygraphy and pulse oximetry test analysis suggested that airway abnormalities contributed to OSA severity; this correlation was previously unclear
- Prevalence of laryngomalacia on laryngo-tracheo-bronchoscopy was 76.5 per cent, higher than in previous reports
- Change or update of the surgical pathway, wherein laryngo-tracheo-bronchoscopy is part of surgical management in children aged less than three years with OSA, may be required

Supraglottoplasty is effective for laryngomalacia-induced OSA,^{9–13} and Zafereo *et al.* have confirmed post-surgical improvement on polysomnography.¹⁴ Table 4 shows that OSA improvement (in 50 per cent of cases) and resolution after surgical intervention (in 33 per cent of cases) was attributed to supraglottoplasty or aryepiglottoplasty, which was validated by mean post-operative oxygen desaturation index, apnoea-hypopnoea index and oxygen saturation values.

The role of polygraphy and the pulse oximetry test has not been thoroughly studied regarding the correlation of their values with airway abnormalities causing airway obstruction. All the pre-operative polygraphy and pulse oximetry tests performed in this retrospective study (Table 3) demonstrated a higher mean oxygen desaturation index and apnoea-hypopnoea index, and a lower mean oxygen saturation value, for children with an airway abnormality contributing to the severity of OSA.

Further multi-centre and multi-surgeon studies, with findings validated on polygraphy pre- and post-operatively after adenotonsillectomy, will be required to: confirm the benefit of performing laryngo-tracheo-bronchoscopy in children aged less than three years; determine whether laryngo-tracheo-bronchoscopy has a role in children aged three years or more; and gain statistically significant results regarding the contribution of an airway abnormality to OSA.

There are two advantages of performing laryngo-tracheo-bronchoscopy. Firstly, laryngo-tracheo-bronchoscopy can guide surgical management, under the same general anaesthetic setting, depending on findings. This effectively means that tonsillectomy, which is a higher risk procedure in children aged less than three years, can be avoided or deferred to a later stage. It also means that other causes of OSA are addressed, such as laryngomalacia, especially in children with no signs of adenotonsillar hypertrophy; in addition, supraglottoplasty, which is effective, can be considered. In our series, children scheduled for laryngo-tracheo-bronchoscopy with or without aryepiglottoplasty or supraglottoplasty had signs of laryngomalacia in the clinic; therefore, laryngo-tracheo-bronchoscopy was helpful to confirm the diagnosis of laryngomalacia and of synchronous airway lesions at other airway levels. Secondly, laryngo-tracheo-bronchoscopy can guide management if further surgical treatment or CPAP is required, depending on findings, when it is performed concurrently with adenotonsillectomy and when children present with persistent OSA post-operatively.

There are three disadvantages of performing laryngo-tracheo-bronchoscopy. Firstly, surgical time increases by at least 10 minutes. Secondly, there is the requirement of a tertiary referral hospital setting to have an appropriately experienced surgeon, anaesthetist and operating theatre staff. Lastly, parents or carers might not be fully aware of the surgical intervention to be performed, as laryngo-tracheo-bronchoscopy will guide the surgical intervention intra-operatively, especially in children with no signs of adenotonsillar hypertrophy.

Conclusion

Seventy-seven per cent of children aged less than three years of age had an airway abnormality identified on laryngo-tracheo-bronchoscopy in this case series, that occurred separately or in addition to adenotonsillar hypertrophy. Consideration of a change or update of the surgical pathway, where laryngo-tracheo-bronchoscopy is included as part of surgical management in this cohort of children with OSA, is required. A much smaller percentage, of 13 per cent, of children aged three years or more with OSA had an airway abnormality, with 75 per cent of these children having a co-morbidity; this is the first time that this aspect has been studied. The pre-operative polygraphy and pulse oximetry test analysis suggested that an airway abnormality contributed to the severity of OSA. Further prospective studies of children, validated using polygraphy pre- and post-operatively after adenotonsillectomy, will be

required to: confirm the benefit of performing laryngo-tracheo-bronchoscopy in children aged less than three years, determine if there is a role of laryngo-tracheo-bronchoscopy in children aged three years or more, and examine the correlation of airway abnormalities with respiratory parameters.

Competing interests. None declared

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