

## Age-Related Outcomes of Sleep Apnea Surgery in Infants and Children

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This study was designed to determine whether age at the time of surgery is an important influencing factor on outcomes following surgical correction of severe refractory obstructive sleep apnea (OSA) in infants and children. Data were collected prospectively on 55 children, all with severe OSA refractory to conservative medical and surgical measures, who underwent combinations of soft-tissue and skeletal procedures aimed at relieving their airway obstruction. The study population was subdivided for analysis into three groups based on age at the time of surgery (>36 months, >12 to <36 months, and ≤12 months). Each child was assessed for clinical outcomes, polysomnography results, and complications. Children in the >36 months group demonstrated a significant improvement in respiratory disturbance index (RDI), apnea index, and lowest overnight oxygen saturation postoperatively. Only RDI improved significantly in the >12 to <36 months group. Although there was a trend toward improvement in the respiratory indices for the children ≤12 months of age, they had a significantly longer intensive care and hospital stay, a greater mean number of extubation attempts, and the highest surgical failure rate (29%). Other complications such as infection, atelectasis, or temporary postoperative nasopharyngeal tube dependence occurred most frequently in the >36 months group. Surgical management of severe refractory OSA in children age ≤12 months is more difficult and less likely to succeed. The reasons for this are discussed and recommendations for management are given.

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The sequelae of untreated upper airway obstruction in children are protean, including failure to thrive, pulmonary disease, neurological impairment and intellectual delay, cardiac failure, and death [1, 2]. Hitherto failure of conservative ther-

apies has often necessitated tracheostomy placement with the attendant risks of tube dislodgment or obstruction leading to death, or late complications and decannulation difficulties relating to chronic infection or granulation tissue formation [3, 4]. Tracheostomy may have a profound negative impact on the child's social and psychological development, and in the long term imposes a financial burden on the parents. A variety of other surgical maneuvers have been employed to treat obstructive sleep apnea (OSA) with mixed results [5-8]. These have included soft-tissue procedures such as choanal dilatation and stenting, tonsillectomy, adenoidectomy, uvulopalatopharyngoplasty (UPP), palatal split, tongue-lip adhesion, and tongue reduction; or skeletal surgeries such as maxillary and monobloc advancements, all aimed at improving airway patency.

The senior authors have developed a treatment philosophy for refractory sleep apnea and for tracheostomy-dependent children with craniofacial deformities that has been reported elsewhere [9]. The approach is based on a multidisciplinary team evaluation to define the problem in terms of the anatomic and functional airway zones affected. From these data a rational, sequential, and case-directed surgical strategy is applied with the aims of improving the respiratory index, avoiding tracheostomy, and preventing the long-term sequelae. This protocol has been applied across all age groups for a variety of underlying conditions. It has been our clinical impression that in the younger children (<12 months) it has been more difficult to achieve our aims. This study was undertaken to determine whether the age at time of surgery is an important influencing factor on outcomes of sleep apnea surgery in children and, if so, the reasons for any observed differences.

## Patients and Method

Fifty-five children were treated surgically between July 1989 and December 1995. There were 35 males and 20 females, and age at time of surgery ranged from 2 to 221 months (mean, 67.8 months). To analyze the impact of age at surgery, the patient population was subdivided into three groups: those older than 36 months, those between 12 and 36 months, and those age 12 months or younger at the time of surgery. Data were collected prospectively for each case and the mean follow-up for the group was 21 months (range, 1–70 months).

All children presented with severe OSA refractory to medical management. Medical failure for each of the three age groups was defined as: (1) persistent upper airway obstruction in spite of supplemental oxygen, CPAP, and tonsillectomy and adenoidectomy if indicated; (2) inability to tolerate CPAP; and (3) ventilator- or tracheostomy-dependent patients with upper airway obstruction who failed conventional weaning and decannulation procedures. Four of the patients already had tracheostomies, which had been placed shortly after birth to relieve airway obstruction. Surgical treatment was aimed at relief of the upper airway obstruction to improve symptoms and respiratory indices, and to avoid either long-term tracheostomy or permit tracheostomy decannulation. Clinical evaluation was carried out in each patient by the craniofacial surgery team and a pediatric pulmonologist. This included a complete history, physical examination, chest and cervical spine radiographs, a lateral cephalogram, computerized axial tomography (CAT) scan of the head and neck, a 12-lead electrocardiogram, cardiac ultrasonography, investigation for gastroesophageal reflux, and, when indicated, flexible endoscopy of the upper airway during spontaneous ventilation. In addition, overnight 12-channel polysomnography (PSG) was recorded before and after surgery, when possible. These sleep data, which included oxygen saturation recordings, allowed calculation of the apnea index (AI; the number of apneic events divided by total sleep time in minutes and multiplied by 60) and the respiratory disturbance index (RDI; the number of apneas *and* hypopneas divided by total sleep time and multiplied by 60).

Based on these data, sleep apnea surgery consisting of various combinations of soft-tissue reduction and skeletal expansion procedures [9, 10] was recommended. The nasal airway was inspected and all sites of obstruction were evaluated. Treatment was directed at septal deviation, turbinate hypertrophy, choanal stenosis, and adenoidal hypertrophy if present. Horizontal, vertical, and transverse nasal dimensions were analyzed to determine if inferior maxillary repositioning and/or transverse palatal expansion by a Le Fort or segmental Le Fort osteotomy was indicated. The soft palate and lateral pharyngeal walls were inspected. If the soft palate was touching or close to the pharynx, or enlarged tonsils were present, UPP and/or tonsillectomy were carried out. The size and position of the tongue were also evaluated. Central tongue reduction (using a carbon dioxide laser) was indicated for macroglossia, which was present in several of the Down syndrome patients and a few of the patients with cerebral palsy. Sagittal advancement of the tongue away from the retropharyngeal wall was accomplished by mandibular advancement and/or genioplasty with a tongue-hyoid suspension. In order to treat the infant and younger child, a variety of modifications to existing surgical techniques were made, and are discussed later. The operative procedures that were used have been broadly grouped into either "skeletal surgery" or "soft-tissue surgery" and further subdivided according to age at time of surgery (Tables 1 and 2).

All 55 patients were included in comparing complication rates among the three age groups. The analyses of changes in PSG results were restricted to patients with both pre- and postoperative PSG data. Of the 32 patients whose age at surgery was >36 months, 28 (88%) had complete data for RDI and low oxygen saturation (LO<sub>2</sub>S), and 29 (91%) had complete data for AI. Of the 16 patients whose age at surgery was >12 to <36 months, 8 (50%) had complete data for the three PSG variables. Of the 7 patients whose age at surgery was <12 months, 3 (43%) had complete data for the three PSG variables. The reason for data variability is that some of the children were intubated, in severe respiratory distress, or had tracheostomies prior to sleep apnea surgery and therefore PSG could not be obtained.

**Table 1. Soft-tissue Surgery**

Type of Surgery	>36 Months	>12- $<$ 36 Months	$\leq$ 12 Months	No. of Surgeries
<b>Naso/Palato/Pharyngeal</b>				
Inferior turbinectomy	13 (41%)	6 (38%)	—	19 (35%)
Septoplasty	2 (6%)	1 (6%)	—	3 (5%)
Nasal dilatation	2 (6%)	1 (6%)	—	3 (5%)
Choanal polypectomy	1 (3%)	—	—	1 (2%)
UPP	16 (50%)	8 (50%)	4 (57%)	28 (51%)
Tonsillectomy	15 (47%)	6 (38%)	—	21 (38%)
Adenoidectomy	15 (47%)	8 (50%)	—	23 (42%)
<b>Tongue</b>				
Tongue-hyoid advancement/suspension	12 (38%)	10 (63%)	6 (86%)	28 (51%)
Tongue reduction	19 (59%)	1 (6%)	1 (14%)	21 (38%)
Floor of mouth/tongue degloving	—	—	2 (28%)	2 (4%)
<b>Miscellaneous</b>				
Suprahyoid myotomies	2 (6%)	1 (6%)	—	3 (5%)
Epiglottoplasty	1 (3%)	—	—	1 (2%)
Wilkes-Brody	3 (9%)	2 (13%)	1 (14%)	6 (11%)
Intraoperative laryngo/bronchoscopy	3 (9%)	—	—	3 (5%)

UPP = uvulopalatopharyngoplasty.

**Table 2. Skeletal Surgery**

Type of Surgery	>36 Months	>12- $<$ 36 Months	$\leq$ 12 Months	No. of Surgeries
<b>Mandibular</b>				
Mandibular osteotomy/advancement	13 (41%)	5 (31%)	1 (14%)	19 (35%)
Distraction osteogenesis	6 (19%)	6 (38%)	1 (14%)	13 (24%)
Genioplasty	5 (16%)	—	1 (14%)	6 (11%)
Hemimandibular reconstruction	2 (6%)	1 (6%)	—	3 (5%)
TMJ release/reconstruction	2 (6%)	2 (13%)	1 (14%)	5 (9%)
Bilateral condylectomies and coronoidectomies	—	—	1 (14%)	1 (2%)
<b>Maxillary</b>				
Le Fort I	3 (9%)	—	—	3 (5%)
Le Fort II	1 (3%)	—	—	1 (2%)
Le Fort III	4 (13%)	1 (6%)	—	5 (9%)

TMJ = temporomandibular joint.

Statistical analysis of the pre- to postsurgical changes in RDI, AI, and LO<sub>2</sub>S was by Wilcoxon signed rank tests. Comparisons between groups were made using Kruskal-Wallis rank sum tests. Analysis of variance was used to compare mean values for other variables and (generalized) Fisher's exact tests were used to compare proportions.

## Results

All children had severe OSA (as determined by clinical and PSG evaluation) that was refractory to traditional medical management. The demographic data for all patients are presented in Table 3. The diagnoses were grouped into five broad diagnostic categories, the distribution of which varied between age-at-surgery groups (Ta-

**Table 3. Patient Population (N = 55; males, N = 35; females, N = 20)**

Patient Data	Mean (SD)	Range
Age at time of surgery (months)	67.8 (62.1)	0.2-221
Length of hospital stay (days)	20.8 (15.0)	2-69
ICU stay (days)	15.0 (13.6)	0-69
Mean number of extubation attempts	1.11 (0.57)	0-3

SD = standard deviation; ICU = intensive care unit.

ble 4). Half the children in the  $>$ 36 months group had either cerebral palsy or Down syndrome, while a mix of diagnoses made up the younger groups, consisting mostly of syndromic craniofacial disorders or fixed skeletal abnormalities. Three of the 7 children aged  $\leq$ 12 months at time of surgery were diagnosed with Pierre Robin syndrome. Table 5 documents the incidence of

**Table 4. Diagnoses Among Age Groups**

Diagnosis	>36 Months	>12-<36 Months	≤12 Months	No. of Diagnoses
Mixed diagnoses	6 (19%)	10 (62%)	3 (43%)	19 (34%)
Cerebral palsy	13 (40%)	3 (19%)	1 (14%)	17 (31%)
Down syndrome	7 (22%)	—	—	7 (13%)
Hemifacial microsomia	4 (13%)	2 (13%)	—	6 (11%)
Pierre Robin anomalad	2 (6%)	1 (6%)	3 (43%)	6 (11%)
Total	32 (58%)	16 (29%)	7 (13%)	55

**Table 5. Related Conditions Among Age Groups**

Clinical Findings and Conditions	>36 Months	>12-<36 Months	≤12 Months	No. of Patients
Gastroesophageal reflux	7 (22%)	6 (37%)	4 (57%)	17 (31%)
Retromicrognathia	9 (28%)	4 (25%)	4 (57%)	17 (31%)
Cleft lip and/or palate	5 (16%)	5 (31%)	2 (29%)	12 (22%)
Midface retrusion/hypoplasia	9 (28%)	2 (13%)	—	11 (20%)
Laryngotracheomalacia	4 (13%)	1 (6%)	2 (29%)	7 (13%)
Tonsillar hypertrophy	5 (16%)	—	—	5 (9%)
Adenoid hypertrophy	3 (9%)	—	—	3 (6%)
Reactive airway disease	3 (9%)	2 (13%)	—	5 (9%)
Cardiac anomalies	—	3 (19%)	2 (29%)	5 (9%)
Polydactyly/limb anomalies	—	1 (6%)	2 (29%)	3 (6%)
Other <sup>a</sup>	3 (9%)	—	1 (14%)	4 (7%)

<sup>a</sup>Diabetes, juvenile rheumatoid arthritis, bronchopulmonary dysplasia, or posterior fossa tumor.

**Table 6. Polysomnographic Results for All Patients**

Parameters	N <sup>a</sup>	Presurgery Median (Range)	Postsurgery Median (Range)	Change <sup>b</sup> Median (Range)
RDI	39	7.6 (0-157.8)	1.9 (0-26.3)	-4.8 (-154.8-5.9)
Apnea index	40	3.0 (0-138.1)	0.6 (0-38.8)	-2.3 (-135.1-11.6)
Lowest O <sub>2</sub> saturation	39	0.76 (0.41-0.97)	0.88 (0.46-0.98)	0.13 (-0.27-0.47)

<sup>a</sup>Only results from patients with both pre- and postsurgery values are included.

<sup>b</sup>Wilcoxon signed rank test used for differences significant to all three parameters ( $p < 0.0001$ ).

RDI = respiratory disturbance index; O<sub>2</sub> = oxygen.

relevant clinical findings and associated conditions. Of particular interest is the relatively high incidence (31%) of gastroesophageal reflux in all age groups and the 22% incidence of associated cleft lip and/or palatal defects.

The PSG results for those patients with complete data demonstrated a significant improvement for all three parameters ( $p < 0.0001$ ). The median change in the RDI, AI, and LO<sub>2</sub>S were -4.8 events per hour, -2.25 events per hour, and +13% respectively (Table 6). Analyzing the same data within each of the three age categories (Table 7), we still found statistically significant improvements ( $p < 0.0001$ ) in all three PSG parameters for patients who had surgery at >36 months of age. In the >12-<36 months group only the changes in RDI were significantly improved, and in the ≤12 months group although

there was an improved trend in the sleep study results this did not reach statistical significance. This may, in part, reflect the small sample in the ≤12 months group.

Comparisons among the three age categories for length of hospital stay, length of intensive care unit (ICU) stay, and number of extubation attempts are summarized in Table 8. Children in the ≤12 months group had longer ICU (mean, 38.3 days) and hospital stays (mean, 41.4 days) compared with the other two age groups ( $p < 0.0001$ ), and also had the largest mean number of postoperative extubation attempts (mean, 1.29).

Surgical failures were defined either by the children subsequently requiring tracheostomy following sleep apnea surgery or by death. In the comparisons between failures vs. nonfailures, the 4 patients with preexisting tracheostomies were

