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Research Article

Association of obstructive sleep apnea on surgically treated cleft anomalies

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ABSTRACT

Background: Palatoplasty is frequently employed to correct the many anatomical and physiological effects of cleft disorders in children, including Obstructive Sleep Apnea (OSA). It is as yet unclear, whether Sleep Disordered Breathing (SDB) and OSA are encountered post palatoplasty. This study attempts to analyze the occurrence, if any, of the same.
Methods: The study is a retrospective analysis, involving patients who had undergone palatoplasty. Patients were subjected to overnight Polysomnography (PSG), and data pertaining to Apnea Hypopnea Index (AHI), Desaturation Index (DI) and Periodic Limb Movements (PLMS) were recorded.
Results: Of the 21 patients evaluated for post palatoplasty OSA, eleven patients were found to have mild OSA (52.3%), and four patients had moderate OSA (19.04%). Of the patients with mild OSA, five patients had complaints of postoperative snoring (45.4%).
Conclusions: A sizeable risk of post palatoplasty OSA does exist, and the incidence of postoperative OSA may be higher than previously noted. However, further studies are warranted, in this regard.

Keywords: Cleft disorders, Obstructive sleep apnea, Palatoplasty

INTRODUCTION

Sleep apnea in infants and children was first described in 1975, and this was in reference to the entity called sudden infant death syndrome. It was in 1976, that obstructive sleep apnea (OSA) was described in school children.[1]It has been established that there has been a significant upturn in the recognition and identification of sleep disorders in the paediatric age group. Sleep disordered breathing (SDB) broadly seeks to include OSA and Upper Airway Resistance Syndrome (UARS). OSA can be defined as a disorder of breathing during sleep, characterized by prolonged partial upper airway obstruction, intermittent complete or partial obstruction (obstructive apnea or hypopnea) or both prolonged and intermittent obstruction that disrupts normal ventilation during sleep, normal sleep patterns, or both.[2] It has been agreed upon that an apnea-hypopnea index greater than 1, may be considered abnormal in a child. The International Classification of Sleep Disorders 2nd edition (ICSD 2) defines apnea as a cessation of airflow over two or more respiratory cycles. However, since normal respirations in children may vary from as many as 60 breaths per minute in new-borns to 12 breaths in adolescents, specific time period in seconds may not be applicable. Most sleep centers agree that a reduction in airflow by a minimum of 30 per cent is a prerequisite, with or without an arousal and/or oxygen desaturation of 3-4 per cent.[3]

In addition to important risk factors such as obesity and upper and lower respiratory tract problems, hypertrophy of the adenoids and tonsils have been postulated to be the

most common cause of sleep-disordered breathing in infants.[4] Children with craniofacial anomalies and certain malformations such as micrognathia and maxillary hypoplasia, that have been known to concern anatomic or functional narrowing of the airway are at significant risk for development of obstructive events during the period of sleep.[5] Such children are subjected to various procedures such as palatoplasty to repair anatomical defects. It has been reported that the risk of developing upper airway obstruction after palatoplasty is considered very minimal.[6] That being said, various other recent reports do indicate an increased incidence of sleep-disordered breathing in patients secondary to palatoplasty.[4] Further studies are required in this regard, as knowledge remains limited in this population.

METHODS

Post palatoplasty patients belonging to the age group up to 20 years were included in the analysis. All these patients had undergone palatoplasty before 2 years of age. History and General Physical Examination were duly taken and the patients were subjected to overnight Polysomnography. The study was performed by a polygraph system with a technician in attendance. Electroencephalogram (EEG), Electro-oculogram (EOG), chin Electromyogram (EMG), Electrocardiogram (EKG), and Periodic Limb Movements (PLM) were monitored to score sleep stages using standard methods. Periodic Limb Movements were monitored by left and right anterior tibialis. Nasal and Oral airflow were measured separately using pressure transducers. Arterial oxygen saturation (SpO_2) was monitored using a pulse oximeter. Respiratory sounds were recorded with a snore sensor. Thoracic and abdominal respiratory movements were recorded by respiratory inductive plethysmography.

A Central Apnea was defined as a cessation of oral and nasal airflow with simultaneous cessations of respiratory movements for at least 10 seconds (2 respiratory cycles in children).

An obstructive apnea was defined as a cessation of airflow for at least 10 seconds (2 respiratory cycles in children) in the presence of continuous respiratory movements.

Hypopneas were scored and severity of Obstructive sleep apnea was classified as per guidelines.[7] Number of apneic, hypopneic and desaturation episodes were noted as was also snoring episodes. Arousal statistics were noted. The desaturation index was calculated as the number of desaturation episodes amounting to less than 3 percent of the mean oxygen saturation per hour of sleep.

RESULTS

We evaluated 21 patients diagnosed to have cleft palate and who underwent palatoplasty, for symptoms of post palatoplasty obstructive sleep apnea. Eleven of these patients had mild OSA (52.3%); four had moderate OSA (19.04%). None of the patients were found to have severe OSA (Table 1).

Table 1: Distribution of cases.

OSA symptoms	Number	Percentages
Mild OSA	11/21	52.38%
Moderate OSA	4/21	19.04%
Severe OSA	0	0
No OSA	6/21	28.57%

Of the patients with mild OSA, five patients had complaints of postoperative snoring (45.4%); one of these patients did have complaints of severe snoring. It was also noted that six patients had normal AHI, although five of these patients did report post palatoplasty snoring.

Table 2: Parameters studied.

Parameters studied	Mild OSA Mean ± s.d	Moderate OSA Mean ±s.d
Apnea hypopnea index	9 ± 2.32	19.92 ± 8.95
Desaturation index	2.99 ± 3.62	4.67 ± 5.25
PLMS index	137.91 ±79.47	149.27 ±106.15
PLMS arousals	42.63 ± 68.26	24.50 ±24.39
Minimum spo_2 (%)	91.09 ± 5.10	90.75 ±6.34

The mean AHI for patients diagnosed to have mild OSA was 9±2.32, whereas for moderate OSA, the mean AHI was found to be 19.92±8.95. The mean Desaturation Index in patients with mild OSA was 2.99±3.62, whereas in patients with moderate OSA, the mean Desaturation Index was 4.67±5.25. In those diagnosed to have mild OSA, the mean PLMS index and arousals was 137.91±79.47 and 42.63±68.26 respectively, whereas in those diagnosed with moderate OSA, it was 149.27±106.15 and 24.50±24.39 respectively. The minimum spo2 during sleep was noted for all patients. The mean value of minimum spo_2 in case of patients with mild OSA was 91.09±5.10 %, and for patients with moderate OSA, it was found to be 90.75±6.34% (Table 2).

DISCUSSION

Sleep apnea is a vital cog, in the spectrum of sleep disordered breathing. No definitive population-based study has been successful in evaluating the incidence of OSAS in the pediatric population. It has been reported previously that as much as 8-12 percent of individuals under the age of 18 years reported heavy snoring. Initial studies did estimate the prevalence of OSAS to be between 1 and 3%. Recent studies attempting to describe prevalence have arrived to the conclusion that it could

vary between 5 and 6%, which is sizeable, considering the population affected.[8]

As stated previously, defects such as cleft palate are one of the common causes for obstructive sleep apnea. Robison et al did report an increased prevalence of SDB and/or OSA among the cleft palate population, with an even greater prevalence noted in patients diagnosed with Pierre Robin sequence.[9] Muntz et al also theorized that there was a notably high incidence of SDB and definable OSA among the cleft population.[10]

The gold standard for diagnosis of SDB is by use of laboratory polysomnography (PSG),and this includes electroencephalogram (EEG) leads, electro-oculogram (EOG), electromyogram (EMG), nasal pressure/oral thermistor, electrocardiogram (ECG), pulse oximetry, chest and abdominal excursion belts, plethysmography, limb leads, end tidal or transcutaneous CO2, esophageal manometry as well as audio/video taping.[11] Information thus generated can help in the evaluation and analysis of sleep architecture, breathing events recorded during sleep (including period of apnea, hypopnea, flow limitation, respiratory effort related arousals), desaturation and periodic limb movements. An apnea/hypopnea index (AHI) of greater than 1 event in an hour has been postulated to be abnormal in the pediatric population. SDB is characterized by frequent episodes of arousal, with increased respiratory effort, hypercapnia, apnea accompanied by desaturation or markedly negative esophageal pressure fluctuations (ICD 2).[3] It must be noted that although night to night variation has been documented in the adult population, one night of polysomnography is usually sufficient to make a diagnosis in the pediatric populace.[12] In instances where PSG may be unaffordable, primary care physicians have looked to alternative avenues for diagnosis of SDB. This includes the use of various Questionnaires that have been developed to try to determine those at an increased risk of SDB, however, evaluation of these questionnaires are accompanied by the caveat that they may not reliably differentiate those children with SDB from those without. This may be attributable to unreliable parent reports.[13]

A multidisciplinary team effort is of utmost importance, to manage the many facets that concern orofacial clefting and the treatment stratagem for repair involves palatoplasty, the challenge being not merely the successful closure of the cleft palate, but to prevent postoperative compromise of speech. Cleft patients do have the potential for normal facial skeleton development; however, corrective surgical procedures have been known to impair maxillary growth and may also lead to midface retrusion.[14]

The primary goals of palatoplasty are to restore velopharyngeal functionality and to ensure development of normal speech, concurrently avoiding palatal fistulas and maxillary growth restriction. Performing this procedure at the most optimal time is of paramount importance, and this factor may have bearing on various postoperative complications. An early repair of the defect has been proven to benefit speech development; however, this may result in inhibition of facial growth to a great extent, attributable to the fact that the transverse facial growth is not completed until 5 years of age. It must be noted that while previously surgery on the palate was often delayed until maxillary growth was completed, of late, speech outcomes have taken prominence and as a result of this change in perspective, most experts advocate the repair of cleft palates commencing at about 10 months of age.[15,16] Some surgeons do propose waiting until 2 years of age to operate on large cleft palates. Surgery on isolated soft palate clefts however, may be attempted as early as 3 months of age.[14]

Surgical techniques for repair of the soft palate most often include the Furlow double-opposing Z-plasty and the intravelar veloplasty. Repair of the bony palate is by employment of the Von Langenbeck palatoplasty, the Veau-Wardill-Kilner palatoplasty, or a Bardach two-flap palatoplasty. Vomer flaps are used for the repair of the nasal mucosa, in tandem with hard palate repair.[17,18] The impact of surgical technique on development of postsurgical obstructive sleep apnea is formidable. Madrid et al compared the efficacy of pharyngoplasty and palatoplasty for reducing the incidences of OSA in children with VPI. It was noted that highest indices of OSA were in the pharyngoplasty category.[1]Certain techniques have also been proven to reduce incidence of OSA drastically. Furlow et al attempted a trial of Teflon injections to reduce incidence of postsurgical OSA and found good response to the same.[20]

The outcome of surgical intervention is open to debate and depends on perspective, as to whether postoperative OSA is considered significant enough. Crockett et al. did theorize that OSA is a possible postoperative complication in patients with 22q11.2 deletion syndrome who underwent surgery for Velo Pharyngeal Insufficiency (VPI).[21] Spruijit et al in a systematic review, mentions a 1 percent incidence of postoperative OSA, among patients treated for VPI, associated with 22q11.2 deletion syndrome.[22]

Liao et al. attempted to longitudinally investigate the incidence and severity of obstructive sleep apnea (OSA) following Furlow palatoplasty for velopharyngeal insufficiency (VPI) in children with cleft palate. It was noted that a high incidence of mild OSA (100%) did occur during the early postoperative timeframe, although resolution of the same was noted within 3 months, in all but two of the patients (20%). In one patient (10%), OSA was found to persist as long as 6 months postoperatively. Their study concluded that children with VPI associated with cleft palate; undergoing Furlow palatoplasty, did invite the possibilities of temporary and mild OSA.23

In a three year retrospective study that involved 539 children, Muntz et al theorized that syndromic children had significantly more symptoms of SDB and were more likely to require a PSG. Of these subjects that did require PSG, only six had a normal obstructive apnea–hypopnea index (OAHI). It was hypothesized that though there was a statistically significant improvement in symptoms post intervention, some were not cured. It was concluded that PSG should be done more frequently, and post intervention PSG must be given its due importance.[10]

MacLean et al. has also reported that a number of studies assessing the impact of palatoplasty, pharyngoplasty, and pharyngeal flap did conclude that this surgery may increase the risk for development of OSA.[24] Rose et al states that although the risk of developing upper airway obstruction post palatoplasty may be considered to be minimal, there were recent reports of increased instances of sleep-disordered breathing in patients secondary to palatoplasty; although it is also conceded that knowledge of this entity remains limited.[4]

Our retrospective analysis revealed eleven patients to have had mild OSA (52.3%), and four to have moderate OSA (19.04%). It also revealed that postoperative snoring was a common problem reported by parents. This finding was further augmented by the reporting of PLMS arousals, during overnight PSG, which points to disturbed sleep patterns. The finding of significant desaturations post palatoplasty supplements these findings.

The development of post palatoplasty OSA is important with regard to its implications on growth and development of the child. It has been evident that sleep fragmentation, such as found in paediatric OSA may promote the occurrence of reduced daytime functioning.[25] Behavioural and neurocognitive dysfunction and a reduced academic potency are well-described morbidities of OSA in children.[26] Paediatric OSA has now been linked with cardiovascular complications as well, such as alteration in blood pressure regulation, systemic hypertension, and changes in the left ventricular function.[25] Among the proposed mechanisms for alterations in somatic growth of children, reduced levels of insulin-like growth factor-I, insulin-like growth factor–binding proteins, and growth hormone release have been implicated.[27]

Our study is a retrospective review that hopes to supplement current hypothesis pertaining to post palatoplasty OSA. However, as per the author's best knowledge, related literature has yet to be published in India. That being said, it does highlight the possibility that a sizeable risk of post palatoplasty OSA does exist, and the incidence of postoperative OSA may be higher than previously noted. It is reiterated that clinicians must have a low threshold to perform polysomnography on such patients following surgery, bearing in mind associated neurocognitive morbidities.

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