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Comparison of treatment modalities in syndromic children with Obstructive Sleep Apnea—A randomized cohort study

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ABSTRACT

Introduction: Obstructive Sleep Apnea (OSA) is a common medical problem in adults that is becoming increasingly recognized in children. It occurs in the pediatric age group, from newborns to teens. More recently, many specialists have estimated OSA prevalence to be between 5 and 6%. However, in syndromic children, the prevalence of OSA can be from 50 to 100%, having a significant effect on their Quality-of-Life. As they are a challenging population for management, it is essential to evaluate them thoroughly before planning appropriate intervention.

Objective: To compare the efficacy of Adenotonsillectomy (T&A) and Continuous Positive Airway Pressure (CPAP) in syndromic children [Down syndrome (DS) and Mucopolysaccharidoses (MPS)] with Obstructive Sleep Apnea (OSA).

Materials and methods: In a prospective, randomized, cohort comparative study, 124 syndromic children (DS and MPS) aged between 6 and 12 years were recruited from a private MPS support group and the Down Syndrome Society, Chennai. A standard assessment was performed on all children who entered the study including a full overnight Polysomnogram (PSG), Epworth Sleepiness Scale-Children (ESS-C) and Quality-of-Life (QOL) tool OSA-18. The children with positive PSG who consented for the study ($n = 80$) were randomly distributed to two groups, T&A group & CPAP group. The children were followed up with repeat PSG, clinical evaluation, ESS-C and Quality-of-Life (QOL) tool OSA-18 for a period of 1 year.

Observation and results: Follow-up was available for 73 syndromic children. Both the groups, T&A group and CPAP group, showed statistically significant ($p < 0.05$) improvement in Apnea-Hypoapnea Index (AHI), ESS-C, QOL from the intervention. In our study, T&A showed equal outcome compared to CPAP. The contrasting feature between the two groups was that CPAP use gave immediate sustained improvement while T&A gave gradual progressive improvement of symptoms over a period of 1 year.

Conclusion: On average, T&A gives equal outcomes as CPAP and it can be suggested as a first-line treatment in this group of syndromic children.

1. Introduction

Obstructive Sleep Apnea (OSA) is a common medical problem in adults that is becoming increasingly recognized in children. It occurs in the pediatric age group, from newborns to teens. Several studies indicate prevalence rates of approximately 2% [1]. More recently, many specialists have estimated OSA prevalence to be between 5 and 6% [2]. If unrecognized and untreated, OSA can lead to neurobehavioral, growth, and cardiovascular sequelae in childhood [3].

Guilleminault et al. (1976) [4], coined the term "Obstructive Sleep Apnea Syndrome (OSAS)" to emphasize the occurrence of this syndrome in non-obese patients. In the same year, they reported the existence of this syndrome in children. Sullivan et al. (1981) [5] devised the first nasal Continuous Positive Airway Pressure (CPAP) machine and reported its efficacy in the treatment of OSA. In children, an Apnea-Hypoapnea Index (AHI) >1 is defined as OSA [6], with absence of airflow for atleast 2 respiratory cycles, with continuing abdominal and chest movements. AHI grading in children was given by Marcus et al. [7] (1992).

In otherwise healthy children, treatment for short-term outcomes indicates that Adenotonsillectomy (T&A) is the most predictable approach to consider, because irrelevant of the size

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of tonsils or adenoids, it will definitely provide more airway space [8], and is successful in eliminating obstruction in 85–95% [9,10]. On the other hand, CPAP provides continuous pneumatic splinting of the airway and maintains its patency (Rapoport et al., 1996) [11] and is currently recommended as the first-line treatment for OSA children with additional co-morbidity or complex disease (Marcus et al., 2012) [1].

Due to the behavioral problems noted in syndromic children, however, they are a very challenging group to treat with CPAP. They usually require intensive preparation for the first trial. One of the great advantages of nasal CPAP is that it is immediately and demonstrably efficacious in relieving OSA (Lojander et al., 1996) [12]. Another advantage is that it can be offered on a "trial" basis and withdrawn if not tolerated, in contrast to surgical options. This study aims to compare the efficacy of 2 treatment modalities, namely, T&A and CPAP in syndromic pediatric population with OSA and to document if T&A can be advised as a first-line treatment modality for the same.

2. Objective

To compare the efficacy of T&A and CPAP in syndromic children [Down syndrome (DS) and Mucopolysaccharidoses (MPS)] with OSA.

3. Methods

3.1. Participants

The MPS support group started at MediScans, Chennai and the DS Society, Chennai provide health care facilities to the syndromic children by organizing camps and screening for various associated defects/sequelae that occurs due to the disorder. From this pool, along with individual referral cases – study subjects were recruited. One hundred and twenty-four syndromic children aged between 6 and 12 years were referred to our institute for further evaluation. The most common symptoms for which the children were referred were snoring, daytime hyperactivity, urinary incontinence and restless sleep. They were evaluated prospectively by a standardized history, physical examination, audiogram, complete blood investigation, Diagnostic Nasal Endoscopy, Videolaryngoscopy, Dynamic Sleep MRI, Polysomnogram (PSG), X-ray of Nasopharynx, Thyroid function tests, Echocardiography and Electrocardiography (Appendix I). The study was approved by the institute's ethical and scientific committee. The study participants were evaluated by cardiologists/pediatricians/anesthesiologists and considered to be healthy enough to undergo surgery. The total study duration was for 2 years.

3.2. Completion of questionnaire

The parents/caregivers of the enrollee were used as proxy respondents and were provided with the Epworth Sleepiness Scale-Children (ESS-C) [13], OSA-18 [14] questionnaires (Appendices II and III). ESS-C questionnaire was filled on entering the study and on 6 monthly reviews; ESS-C $>$ 10 was suggestive of OSA. OSA-18 questionnaire was filled twice – once during start of the study and once during completion, to assess the effect of intervention on the child's Quality-of-Life (QOL).

3.2.1. Inclusion criteria

1. MPS and DS syndromic children (age 6–12 years) with complaints of snoring and mouth breathing, daytime hyperactivity, urinary incontinence, restless sleep.
2. Obstructive adenoids and tonsils, i.e. grade $>$ 2.
3. Features of OSA on Polysomnogram i.e. AHI $>$ 1.

3.2.2. Exclusion criteria

1. Pediatric patients with previous history of T&A and/or using CPAP.
2. H/o craniofacial reconstruction surgeries/other OSA surgeries.
3. Central apnea.
4. Unfit/unwilling for surgery/medications (Enzyme Replacement Therapy).

3.3. Polysomnogram

The enrollees underwent overnight full PSG. The children with positive PSG who consented for the study were randomly distributed to two groups – T&A group and CPAP group. The PSG machine used in our study was CleveMed Sapphire PSG Type I device, 22 channels unit. The Electrodes were placed by international 10–20 system. Digitized signals were stored on compatible memory storage chips and were analyzed using a computer program (Crystal PSG). Manual scoring based on recent American Academy of Sleep Medicine – AASM guidelines was performed by a trained, certified technician to individually verify the results of the automated scoring system.

3.4. Statistical analysis

3.4.1. Sample size

The study duration was for 2 years during which 124 consecutive syndromic children were recruited and included in the study. Demographic data included age, sex, height, weight, BMI, Neck circumference and associated syndrome. Categorical data is presented as a number (percentage); continuous data is presented as mean ($\pm$SD). The threshold for statistical significance was a p value of $<$0.05. In both the groups, the Student paired t-test was used to compare the mean pre-treatment, 6th and 12th month post-treatment values of AHI, ESS-C and pre- and post-treatment OSA-18 values; the Student unpaired t-test was used to compare the mean post-treatment 12th month AHI, ESS-C and OSA-18 values of both the groups. All of the statistical analyses were performed with a statistical software package (SPSS for Windows, version v20, SPSS Inc., Chicago, Illinois). Initially, treatment was considered successful when the post-treatment AHI $<$ 1. At follow-up, therapy was considered efficacious if 12th month AHI, ESS-C and OSA-18 values showed significant improvement compared to pre-treatment values. Spearman's correlation was used to compute the correlation of OSA-18 scale with AHI.

3.5. Interventions

Coblation T&A was performed for all the subjects in T&A group using Coblator II Arthro Care, Evac 70 Arthro Wand (Arthro Care Corp., Sunnyvale, CA). When compared to conventional Cold dissection, the coblation technique of T&A has been documented to cause less blood loss, pain scale was lower, operative time was less with lesser post-operative bleeding [15]. Since the syndromic children had other co-morbidities, the idea was to use a safe and effective surgical technique so as to provide a faster recovery and uneventful post-operative period. As these children were prone to respiratory depression, they were monitored post-operatively in an Intensive Care Unit (ICU) setup for a minimum of 24 h and then discharged. The post-operative period was uneventful. Post-Tonsillectomy diet instructions and medications were explained to the parents/caregivers.

The children in CPAP group were prescribed ResMed CPAP machines after trial and fitting. The children and parents/caregivers in this group were counseled regarding the CPAP machine and the procedure of CPAP fitting was explained. For the