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Original Article

The outcome of managing sleep apnea in children with cerebral palsy

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Abstract

Background
Obstructive sleep apnea (OSA) is not an uncommon finding in the pediatric age group patients with cerebral palsy (CP).

Objective
We aimed to evaluate the frequency of occurrence of OSA in children with CP and the most appropriate surgical procedures used in these patients.

Study design
A prospective study was carried out on 67 children with OSA, 36 of whom suffer from CP. The data included the presenting complaint, any coexisting illnesses, preoperative and postoperative polysomnography results, the primary surgical procedure performed, age at the time of surgery, number of postoperative hospitalization days in the ICU, and presence of postoperative respiratory disorders.

Results
In all, 40 (59.7%) children underwent adenotonsillectomy for initial treatment of OSA. Our results reported that the clinical manifestations were more significant in the CP-affected children. We documented a significant decrease in the mean value of the apnea hypoxia index in all included children postoperatively. However, there was no significant difference in apnea hypoxia index postoperatively among children in both groups. Nine (17.4%) children had an adenoidectomy alone and 18 (26.86%) children had tonsillectomy alone as their initial procedure. Neither uvulopalatopharyngoplasty nor tracheostomy was performed as an initial procedure. The mean follow-up was set for 24 months. Eighty-six percent of these children have not required any further surgery. Of the six children who have undergone further surgery, one required a revision adenoidectomy, and two underwent a tonsillectomy and uvulectomy 2 months after the initial adenoidectomy. Four CP children later required a tracheotomy for severe hypoxia.

Conclusion
OSA is a more frequent occurrence in CP children as compared with their healthy counterparts. They ultimately had significant improvement in their clinical presentations postoperatively, thus improving their quality of life.

Keywords: Adenoidectomy, cerebral palsy, obstructive sleep apnea

Introduction

The term cerebral palsy (CP) describes a group of nonprogressive motor, sensory, and neurological disorders with postural disabilities and limitation of movement resulting from lesions of the developing brain. The term is descriptive rather than diagnostic with a group of heterogeneous etiologies, mainly hypoxia, infection, and congenital deformities. CP children suffer several associated disabilities: poor neuromuscular tone, increased oropharyngeal secretions and drooling gastroesophageal reflux, and seizure disorders, most of which have been associated with apneic attacks [1].

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Sleep disorders are more prevalent in children with CP compared with their healthy counterparts. Sleep plays a prominent role in the growth and development of children. Eleven types of sleep disorders have been recognized, commonly difficult morning awakening, difficulties in initiating and maintaining nighttime sleep (night waking), insomnia, nightmares, and sleep anxiety. Obstructive sleep apnea (OSA) is one form of sleep disorder that occurs when the throat muscles relax during sleep, blocking airflow in the nose and mouth. The overall incidence of OSA is 0.7–3.0% of all children [2]. However, the proportion of children with CP who have OSA is assumed to be likely higher than those without CP [3].

The American Thoracic Society defined OSA as 'a disorder of breathing during sleep characterized by prolonged partial upper airway obstruction and/or intermittent complete obstruction (obstructive apnea) that disrupts normal ventilation and normal sleep patterns' [4,5]. Snoring, difficulty breathing during sleep, and parental observation of apnea are the three most predictive symptoms of OSA. It is strongly linked to decreased quality of life, negative health outcomes, and neurocognitive consequences [6]. Untreated severe cases of OSA in children can lead to cardiorespiratory failure and eventually death [7]. Identification of CP children with OSA is important because their treatment improves the life quality in this age group and prevents other comorbid complications.

The gold standard for the diagnosis of OSA is an overnight polysomnogram (sleep study). The American Academy of Pediatrics recommends polysomnogram as the diagnostic test of choice in evaluating children with suspected OSA, and that it can be performed in children of all ages in an appropriate pediatric sleep laboratory [8]. This guides in determining a treatment plan, considering other comorbidities, and degrees of daytime impairment. It can guide therapy by identifying the severity of sleep apnea present. It also guides the anesthesia and safety decisions taken around the potential surgery, such as the postsurgical need for inpatient hospitalization or intensive care unit admission and care.

The polysomnogram typically used in pediatric age groups include electroencephalography, electromyogram (chin, legs), electrooculogram, measures of respiratory movements of abdominal and thoracic excursion, pulse oximetry, end-tidal carbon dioxide measurement, measurement of nasal airflow (nasal air pressure sensor and/or oronasal thermistor), snore volume, body position sensor, and video and audio recording [9].

Typically, a polysomnogram includes measurements of sleep stages, respiratory events [apnea/hypopnea index (AHI)], length of time with oxygen values less than 92% and elevated end-tidal carbon dioxide values more than 50 mmHg, heart rate, and the presence or absence of periodic limb movements. An AHI more than or equal to 1 event/h is considered positive for OSA [10].

Screening children with CP for OSA is important to avoid the health consequences of untreated cases. Treatment options include positional therapy and/or medications or surgical procedures, mainly adenotonsillectomy and other upper-airway surgical interventions. However, in severe cases, tracheostomy may be required. Untreated sleep apnea may cause difficulty in recovering from anesthesia. This presents a unique challenge to both the otorhinolaryngologist and anesthetist, as these patients are prone to surgical procedures requiring anesthesia. The anesthetist must be familiar with the potential anesthetic complications and immediate postoperative problems associated with OSA for a smooth and safe anesthetic course.

Preoperative assessment of these patients requires meticulous evaluation, including history taking and physical examination. History of seizures, respiratory abnormalities, tongue thrusting, and inadequate head movement all pose difficulty to airway management. Nasogastric or gastrostomy tubes may be inserted in position and needs the care of during the surgical procedure. Deformities increase the difficulties of intravenous access and monitoring during anesthesia. Medications taken may cause oversedation. Antispastic medications used during preoperative management may cause respiratory muscle paralysis, lethargy, bradycardia, hypotension, delayed arousal from anesthesia, or seizures [11].

Due to altered thermoregulatory responses in these patients, hypothermia is a common and serious concern, which may cause delayed awakening from anesthesia and prolonged hospitalization.

Excessive salivation and drooling pose a problem in children with CP. Postoperative aspiration is not uncommon and requires frequent suctioning, positioning, and airway management. Again, these candidates are subject to a raised risk of convulsions, and preoperative anticonvulsants should be started early to attain therapeutic plasma levels [12].

Frequently, hypertrophy of the adenotonsillar tissues is the primary cause of OSA in children. Thus, in surgical interventions, the current first line of treatment for pediatric OSA is adenotonsillectomy. While it is effective in the treatment of OSA in the majority of otherwise healthy children, there are no large studies using data from polysomnogram to assess the efficacy of this surgery in children with CP suffering OSA, although several studies report symptomatic improvement based on parental observation [13,14].

Sleep endoscopy is a technique that can enable the surgeon to determine the level of obstruction in a sleeping child with OSA, thus allowing site-specific surgical therapy for persistent and complex pediatric OSA [15]. The procedure is carried out in the operating room under general anesthesia, in conjunction with direct laryngoscopy [16].

Thus, preoperative plans should be clearly formulated beforehand.

Objectives of the study

The primary objective of this study is to determine the risk of developing OSA in children with CP.

The secondary objective is to evaluate the surgical outcome of the offered surgical management on improving the OSA and the perioperative complications in such cases.

Patients and methods

Ethical consent

Parents of the children invited to the study were counseled to a written consent for inclusion of their child. Approval of the Ethics Research Committee was obtained before initiating the study.

Study design

The current prospective study was conducted at Ahmed Maher Teaching Hospital, a member hospital of the General Organization for Teaching Hospitals and Institutes, located in Cairo, Egypt. The study highlights the collaborative efforts of the Pediatric, Anesthesia and Otolaryngology Departments in managing CP and otherwise healthy children with OSA.

All children suffering from OSA, whether healthy or suffering from CP, and requiring intervention for OSA, over 3 years, between July 2018 and July 2021, were recruited and included in the study after parental consent. The bulk of patients with CP and neurodisabled with OSA were referred from the National Institute of Neurological Disorders and Stroke, a member of General Organization for Teaching Hospitals and Institutes organization, to the outpatient pediatric follow-up clinic for the disabled and development at Ahmed Maher Teaching Hospital for the evaluation of sleep disturbances. Other cases without CP were recruited from the pediatric and otolaryngology clinics of the same hospital. All cases with craniofacial disorders or major congenital malformations of the face and neck were excluded from the study due to complex difficulties in management. A total of 67 children were included in the study. The age group ranged from 2 to 12 years. The children were distributed among two main groups based on the presence or absence of CP. Thirty-one children had OSA without associated developmental disabilities (group A), while 36 patients had OSA along with a variable degree of CP (group B).

All patients were assessed by the pediatrician for a comprehensive medical history, clinical examination, and developmental assessment.

The medical history focused on the primary complaint, frequency of apneic attacks, sleep disturbances, age at the first complaint, coexisting illnesses, or disabilities. A full clinical physical examination was performed to all cases, describing all disabilities in CP cases, followed by a developmental assessment.

The severity of CP was determined using the Gross Motor Function Classification System (GMFCS), which assesses patients' gross motor function and categorizes them into five different levels [17]. The pediatrician assessed the level of CP for each affected patient using GMFCS.

The diagnosis of OSA in children was based on a presenting history of snoring and/or apnea and enlarged tonsils on physical examination. However, those who had a suggestive history of OSA but did not have enlarged tonsils underwent a lateral neck radiograph to assess nasopharyngeal obstruction by adenoidal hyperplasia as advised by the otolaryngologist. In CP children, nasopharyngoscopy was not used to assess adenoid hyperplasia due to inability to cooperate during this procedure unless fully sedated.

All patients were then referred to El-Matareya Teaching Hospital for polysomnography in their pediatric sleep laboratory and further management.

To confirm OSA diagnosis, sleep studies and polysomnography using the device Neuron-Spectrum AM/PSM – 00170188 – Neurosoft was performed on children in both groups. Both the pediatrician and the otolaryngologist attended the interpretation of the polysomnography results to decide on further actions.

Preoperative investigations included full blood counts, electrolyte studies, renal profile, blood grouping and cross-matching for blood and blood products, chest radiograph, and ECG.

A structured plan was set preoperatively by the pediatrician and the anesthetist for assessment of each individual child based on history and clinical evaluation. All CP patients in group B received an antacid or anti-reflux medication aluminum hydroxide 3–10 ml oral 4–6 h before the operation due to their liability to gastroesophageal reflux. A sedative Midazolm 0.1–0.2 mg/kg was given shortly preoperatively while monitoring the general clinical state of the patients (vital signs, epileptic and apneic attacks). An anticholinergic atropine 0.4–0.6 mg 30–40 min before anesthesia was given to reduce excessive secretions encountered in these patients. Intravenous access is secured, especially in CP children; some required expert assistance and ultrasound guidance. The application of EMLA cream (lidocaine 2.5% and prilocaine 2.5%) decreased the distress from the pain associated with venipuncture. Standard noninvasive monitoring was instituted for all children in the study. The warming of intravenous fluids plus warm blankets and humidified warm gases were used to conserve body temperature.

Frequent suctioning of the oropharynx is necessary throughout the operative procedure, as pooled secretions in the oropharynx might impair mask ventilation and the glottis view during fiber-optic endoscopy.

Padding of pressure points in CP patients was carried out to avoid pressure ulcers as recommended by the managing pediatrician.

The otolaryngology surgeon decided the type of operative procedure required for each individual case based on their clinical examination, radiography, polysomnography, and sleep endoscopy results.

With the child lying in the supine position, inhalation anesthesia (sevoflurane 2–4 MAC) is delivered through a face mask, delivering oxygen to maintain spontaneous breathing. After establishing a rhythmic respiration pattern, a flexible fiber-optic laryngoscope was passed through the side port of the anesthesia mask, passing posteriorly toward the oropharynx. A digital video camera is used with an endoscope for visualization and documentation. Adenoids, palate and uvula, the base of the tongue, lingual tonsils and pharyngeal tonsils if present, epiglottis, and posterior pharyngeal wall were examined.

Following fiber-optic endoscopy, endotracheal intubation using appropriately sized tubes (based on weight) was done.

The general anesthetic agent propofol 1.5–2.5 mg/kg intravenous is desirable, especially in patients with reactive airway in appropriate doses according to the age and body weight of each patient.

Tonsillectomy using electrocautery-assisted excision was performed on 17 patients. Adenoidectomy using an adenoid curette was performed on 11 patients. Adenotonsillectomy was performed on 36 patients. Uvulopalatopharyngoplasty (UPPP) was performed on three patients.

Twenty-nine patients needed postoperative monitoring in the ICU for upper-airway obstruction, eight patients in group A versus 21 patients in the CP group B.

Postoperative polysomnogram studies were evaluated by the pediatrician for all included children, including AHI, percent of sleep with oxygen saturation below 90%, and percent sleep time with end-tidal partial carbon dioxide more than 50.

A preoperative AHI more than 5 was considered mild and more than 10 considered moderate to severe.

Follow-up for all children in both study groups was recommended in the pediatric follow-up clinic or inpatient for 24 months postoperatively. Five patients failed to continue the follow-up period and withdrew from the study. Long-term outcome was reported by questioning parents about snoring and the nocturnal respiratory difficulty experienced by their children postoperatively. In addition, subsequent surgical procedures undertaken for the treatment of persistent or recurrent OSA were reported.

Statistical analysis

All clinical data were collected, analyzed, and subjected to statistical evaluation. Statistical significance was set at a two-sided *P* value less than 0.05. For each variable, differences in proportions were calculated using χ^2 analyses. To compare preoperative and postoperative AHI, Pearson correlation was used.

Results

A total of 72 children were enrolled in the study over a 3-year period; five cases failed to continue the assigned follow-up schedule and were excluded. The remaining 67 children were distributed among the two groups, group A (31 children) and group B (36 children). Between the two groups, there were no statistically significant differences in sex or age (Table 1). Age ranged from 2 to 12 years (mean age 8 ± 2.3 years). The study group had a female to male ratio of 29–38. Children with CP had significantly lower height, weight, and BMI than the comparison group ($P = 0.005$), as shown in Table 1. According to previous clinical evaluations, there was no significant difference in the prevalence of OSA between the two groups ($P > 0.4$).

Children with CP classified as GMFCS levels II, IV, or V had a higher mean prevalence of OSA than children classified as GMFCS levels I or III (Fig. 1) as determined by patients' medical records, explained by fewer numbers of patients suffering levels I and III.

The most common clinical presentation was snoring and respiratory distress, commonly during sleep. Snoring occurred in 62 (92.5%) patients in both groups, 29 (93.5%) patients in group A and 33 (91.7%) patients with CP in group B. Respiratory distress occurred in six (19.35%) patients in group A versus 24 (66.66%) CP patients. Parents reported sleep apnea in two (6.45%) patients in group A versus 27 (75%) patients with CP. A combined snoring and respiratory difficulty occurred in overall 79.1% of patients, 67.74% in group A patients versus 88.88% in group B patients with CP (Table 2). Documented adenoidal and/or tonsillar hyperplasia were documented in all patients on clinical examination or lateral neck radiography.

There was a significant decrease in the mean value of the AHI in both groups postoperatively, 2.7 compared with 21.3 preoperatively ($P < 0.001$). An AHI less than 5 was observed in 26/28 cases postoperatively compared with an AHI more than 10 in 12 (38.7%) of cases and more than 5 in 19/31 (61%) of cases in group A preoperatively. Children suffering CP had an AHI more than 10 in 75% of cases. Full recovery from OSA occurred in all children with a preoperative AHI more than 5 in group A and in 78% of the CP children group. Children with preoperative AHI more than 10 showed complete resolution in 83% of cases in group A compared with 81% in group B, with no statistically significant difference (Fig. 2).

Table 1: Morphologic characteristics of the studied groups

Characters	OSA without CP: group A (*n*=31)	OSA with CP: group B (*n*=36)	Total (*n*=67)
Male	14	24	38
Female	17	12	29
Age (years) (mean±SD)	9.7 (3)	7.4 (4)	9.8 (6)
Height (cm) (mean±SD)	136.2 (25)	122.7 (12)	135.7 (22)
Weight (kg) (mean±SD)	41.3 (22)	35.4 (19)	38.2 (23)
BMI, (kg/m^2) (mean±SD)	21.1 (5)	17.2 (3)	19.7 (6)

CP, cerebral palsy; OSA, obstructive sleep apnea.

Table 2: Percentage of patients suffering clinical manifestations

Presenting complaint	Group A (n=31) [n (%)]	Group B (n=36) [n (%)]	Total (n=67) [n (%)]
Snoring	29 (93.54)	33 (91.7)	62 (92.5)
Respiratory difficulty	6 (19.35)	24 (66.66)	30 (44.77)
Apnea	2 (6.45)	27 (75)	29 (43.28)
Combined snoring and respiratory difficulty	21 (67.74)	32 (88.88)	53 (79.1)

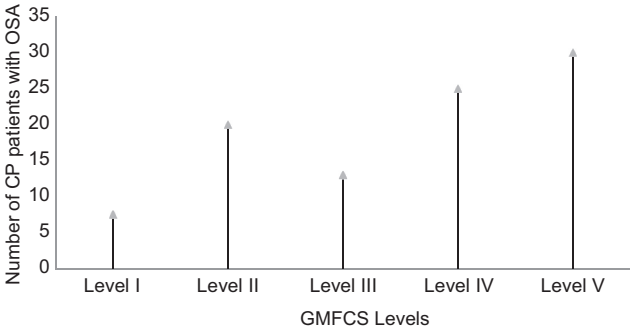

Figure 1: Gross Motor Function Classification System (GMFCS) level versus number of CP patients with OSA. CP, cerebral palsy; OSA, obstructive sleep apnea.

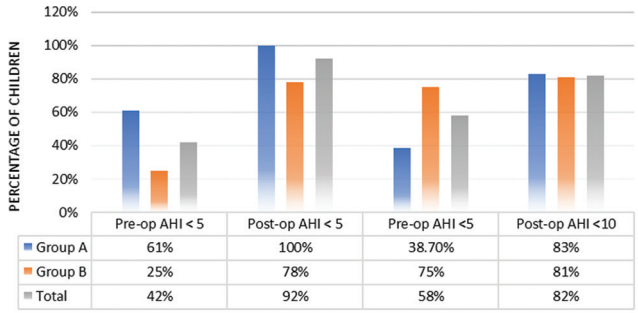

Figure 2: Preoperative and postoperative AHI comparing surgical procedures among both groups. AHI, apnea/hypopnea index.

Patients were allocated to appropriate surgical procedure according to their presenting symptoms, physical examination, polysomnography, and sleep endoscopy. The recommended surgical intervention for patients is shown in Table 3 and Fig. 3.

Postoperative follow-up of all patients showed that 32 (47.76%) patients required postoperative ICU care for an average of 5 days (range, 3–9 days). Three patients from group A, two for mild bleeding that was resolved spontaneously and was discharged from the ICU on the third postoperative day, and the third patient due to delayed recovery from anesthesia and stabilized and discharged from the ICU on the second postoperative day. The three cases were observed in the clinical ward for 24 h and then discharged home.

Most patients (28) in group A were discharged home on the second postoperative day and required no further intervention.

Ten patients in group B were discharged to the hospital ward for further evaluation and observation. The rest of the CP patients were kept under observation in the ICU for stabilization.

Follow-up of all patients was continued for 2 years postoperatively. There were no reported mortalities among any of our treated patients perioperatively and during the clinical follow-up period. All patients were compliant with the follow-up schedule except five patients who failed to show up.

Discussion

Children with CP are at an increased risk of developing OSA. Identifying such children enables clinicians to concentrate their efforts on this population, as improved sleep is associated with improved life quality in patients with CP [7]. Increased oropharyngeal secretions, gastroesophageal reflux disease, and seizure disorders, all of which are common in children with CP, have been shown to contribute to the development of OSA. Given this, even slightly enlarged adenoids and/or tonsils will impair breathing patterns, particularly during sleep, when the pharyngeal muscles are at their most relaxed. This study was designed to (a) determine whether CP children are at a higher risk of OSA compared with the broader population of children without developmental disabilities and (b) to study the effect of surgical correction for OSA in otherwise healthy children versus those with CP.

The bulk of CP children recruited for this study was transferred from the Neurodevelopment and Stroke Institute and presented to a multispecialty pediatric hospital for clinical evaluation of OSA and further management in this group.

All our child population presented with OSA of variable degrees, snoring, and respiratory distress of variable severity. We reported that the higher the level of GMFCS, the more severe is the presenting symptoms, except in level III patients because the patient population in this group was less in number.

We performed sleep studies (polysomnography) on all children in the studied groups, who were suspected of having OSA but with equivocal clinical examination.

The main presenting complaint in our study group was snoring, which occurred in 92.5% of all patients. This complaint was higher in our CP patient population (93.5 vs. 91.7%) than in otherwise healthy patients without CP. This difference was not statistically significant. On the other hand, respiratory distress was statistically significantly higher in the CP group (66.66 vs. 19.35%) than in the healthy group.

This could be explained in part by abnormalities of the upper-airway tone, similar to those observed in those with

Table 3: Surgical procedures recommended for patients

Surgical action and follow-up	OSA without CP ($n=31$) [n (%)]	OSA with CP ($n=36$) [n (%)]	Total ($n=67$) [n (%)]
Adenoidectomy	7 (22.58)	2 (5.55)	9 (13.4)
Tonsillectomy	11 (35.4*)	7 (19.4)	18 (26.86)
Adenotonsillectomy	13 (41.9)	27 (75)	40 (59.7)
Uvulectomy	0	2 (5.55)	2 (2.98)
ICU care	3 (9.67)	29 (80.5)	32 (47.76)

*significant. CP, cerebral palsy; OSA, obstructive sleep apnea.

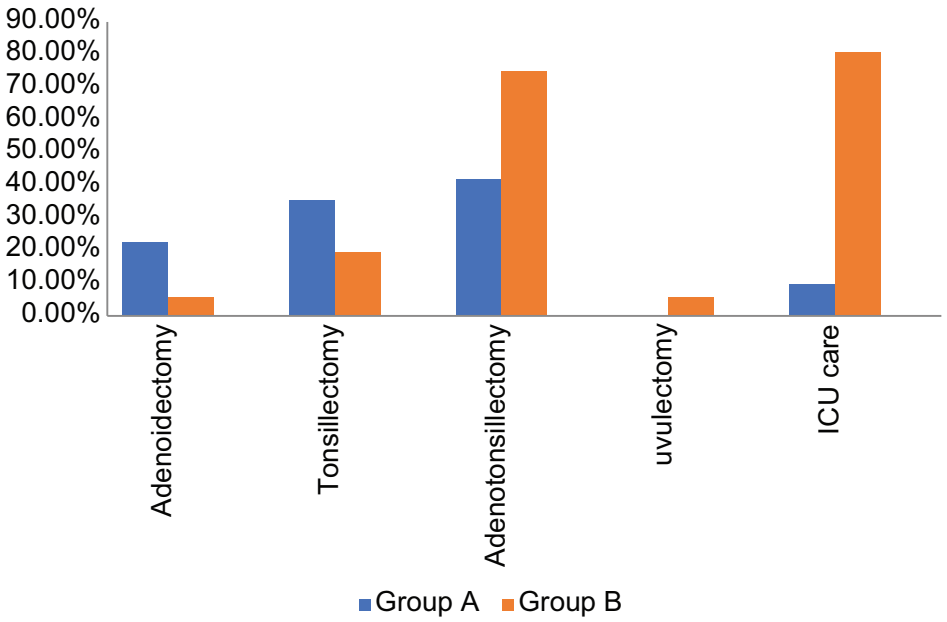

Figure 3: Shows The recommended surgical intervention for Patients were allocated to appropriate surgical procedure

Down syndrome with the low upper-airway tone, of whom 30–60% are diagnosed with OSA [18].

Children with CP suffering OSA may have pulmonary vulnerability, laryngeal dystonia, severe laryngomalacia [19], and concurrent pseudobulbar palsy.

Sleep apnea was documented in all patients through history taking or evidenced by polysomnography. Parents reported it in 6.45% of patients without CP, while parents or health caregivers reported sleep apnea in 75% of patients with CP. This was highly statistically significant ($P < 0.002$). A combined snoring and respiratory difficulty occurred in 79.1% of our study patients, but not statistically significantly higher in CP patients, 88.88 versus 67.74%.

Children with CP and OSA may suffer combined anatomic disturbances such as adenotonsillar hypertrophy and altered physiologic responses causing abnormal muscle control and upper-airway narrowing, compared with normal children. During sleep, these children lose the respiratory drive and compensatory mechanisms of increased airway tone; the airway partially closes, causing obstructive hypopnea and snoring, or complete airway closure, causing obstructive apnea [20].

In addition, underlying and restrictive lung disease, impaired ability to clear respiratory secretions, risk of aspiration and perinatal hypoxic brain injury compromise the central control of breathing during sleep, adding to the severity of OSA in such patients.

The preferred mode of management of OSA in the CP child population is surgical management. Surgical options available include tonsillectomy, adenoidectomy, uvulectomy, UPPP, and tracheotomy.

The most appropriate surgical option was selected based on the clinical, radiological, polysomnography studies, and sleep endoscopy. A total of 59.7% of our study population underwent adenotonsillectomy, 75% in the CP group versus 41.9% in the otherwise healthy group, which did not show statistical significance.

The sleep studies were performed preoperatively and postoperatively to assess the outcome of the surgery on OSA. There were significant reductions in these indices after the first surgery. This was compounded with the reports from the parents on follow-up studies that their childrens' symptoms of snoring, sleep apnea, and respiratory difficulty were markedly improved.

While adenotonsillectomy is the treatment of choice in healthy children suffering from OSA, the efficacy of this surgery in CP patients suffering from OSA has not been adequately assessed. However, several studies report symptomatic improvement based on parental observation [6].

Rates of residual sleep apnea after adenotonsillectomy are around 29% [21]. Children with CP may be at risk due to several factors, including abnormal muscle tone, multilevel airway obstruction, and underlying lung and neurologic disease.

Children with CP have a higher risk for postoperative complications compared with normal children. Postoperatively, children with CP and OSA should be carefully monitored for airway compromise in an ICU. Seventy-seven percent of our children were monitored in the ICU for a mean period of 3 days. Postoperative hypothermia, postoperative pain, and anxiety can all trigger acute muscle spasms, which are often more painful and distressing to the patient than the operation itself.

Postoperative ICU observation for an average of 5 days was reported in 47.76% of patients. Most patients (80.5%) with CP required ICU admission postoperatively and required intervention for upper airway obstruction, compared with those in the healthy group (9.67%) reporting a high statistical significance.

The clinical follow-up to assess for resolution of symptoms is recommended, and a polysomnogram is repeated at 2o months after surgery if symptoms persist.

None of the patients in group A required revision surgery. Five (13.8%) patients in the CP group required a second surgery; namely, three had adenoidectomy and two (5.55%) required uvulectomy followed by tracheotomy for severe respiratory difficulty not responding to other medical measures.

Conclusion

Children with CP suffering from OSA have an impaired life quality. We conclude that OSA is more common in children with CP compared to their otherwise healthy peers who suffer OSA. Surgical management, namely, the combined adenotonsillectomy, is appropriate as an initial surgical procedure of OSA. UPPP may also be useful as a secondary procedure if the primary surgery fails to control symptoms, especially in CP patients. Children with CP have a higher risk for postoperative complications compared with normal children.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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