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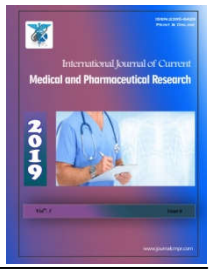

Research Article

PEDIATRIC SLEEP APNEA IN PATIENTS WITH MICROGNATHIA: CASE REPORTS WITH BRIEF REVIEW OF LITERATURE

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ABSTRACT

The diagnosis of pediatric sleep apnea is often missed due to lack of awareness, in association with micrognathia. While the dental specialist tends to focus on the facial especially in developing countries and is observed in patients with craniofacial anomalies, especially deformity and on the resultant malocclusion, attention must also be given to the associated respiratory and feeding problem. Proper and timely diagnosis with prompt intervention if not done tends to hamper the normal growth and development pattern of the child. In this case report two pediatric cases associated with sleep apnea are discussed, one having micrognathia secondary to temporomandibular joint ankylosis and the other with congenital micrognathia. The authors have discussed the management of these casesin different young age group patients with and without distraction osteogenesis along with the brief discussion of literature.

INTRODUCTION

Patients with congenital micrognathia and retrognathia,in addition to malocclusion,display a bird face appearance and jaw hypo mobility leading to dysphagia and pediatric sleep apnea. These patients may have Temporomandibular Joint (TMJ) ankylosis, Treacher Collins or Nager syndromes, craniofacial microsomia, syndromic or non syndromic Robin Sequence.[1]

However not much has been discussed in the pediatric dental literature about pediatric sleep apneaand failure of such children to thrive. OSAS is a complex disorder characterized by intermittent upper airway collapse during sleep[2].Epidemiologic reports have shown prevalence of sleep disordered breathing to be about 2% in children[3,4] and 2.5-6% in adolescents[5].

The authors present two cases of micrognathia due to varying causes, suffering from pediatric sleep apnea and discuss the management of the condition along with the review of literature.

Case Report 1

A four year male patient reported to ourinstitute with aninterincisal mouth opening of six mm (Figure 1) with difficulty in eating andbreathing, along with periods of restlessness associated with breathlessness several times in the night for the past twoyears. The patient had features of micrognathia with deep bite (Figure 2). Computed tomography scan (Figure 3) of the joint area revealed bony fusionof the TMJ and diminished joint space suggestive of TMJ ankylosis. Five episodes of fall in saturation level were noticed hourly on pulse oximetry in the preoperative assessment.

The airway was secured with the tracheostomy and bilateral gap arthroplasty and coronoidectomy performed under general anesthesia. A mouth opening of 40 mm was achieved intraoperatively and 32 mm post operatively (Figure 5).
Post operative monitoring was done with pulse oximetry. The Patient was decannulated after 45 days and was followed up for more than a year.

Case Report 2

An infant(Figure 6) of four months reported for consultation with difficulty in feeding, breathing and limited mouth opening. The patient who was on Ryle's tube feeds since birth was under weight and had episodes of severe obstruction and choking during sleep (8 episodes hourly).

Following examination and investigations includingCT scans (figure 8 and 9),a diagnosis of micrognathia and retrogeniawas made (leading to decreased airway space).Glossopexy was done for the patient but yielded no significant improvement. Later forward positioning of the mandible was planned through distraction osteogenesis.

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The airway was secured though tracheostomy &bilateral mandibular distraction was done under general anesthesia.Due to lack of adequate access through intraoral approach, bilateral orthopedic finger distractors were placed bilaterally via an extra oral approach.Extra-oral monodirectional distractors (Figure 7) were placed at the angle of the mandibleand an oblique mandibular distraction was planned.

Distraction was started actively at the rate of one mm/day on each side after fifth post operative day and continued for 18 days.The patient was kept under continuous monitoring and it was observed that as the mandible was distracted in length, an improvement was noticed in breathing. Patient was decannulated after 45 days after confirminga bilateral gain of 1.8cm was achieved.

The distractors were removed after twomonths under general anesthesia andthe patient was followedupevery 15th day. The child made a noticeable weight gain and the related difficulty in breathing ceased. Further secondary surgery was not required.Patient was followed up for four years without any significant complaints.

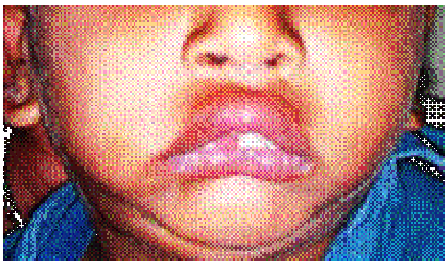

Fig 1 Pre operative photograph

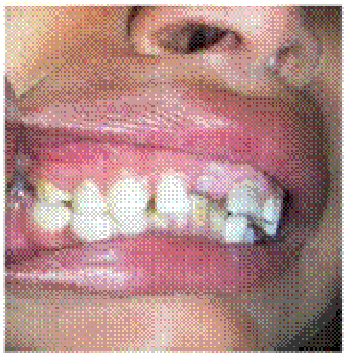

Fig 2 Pre operative mouth opening

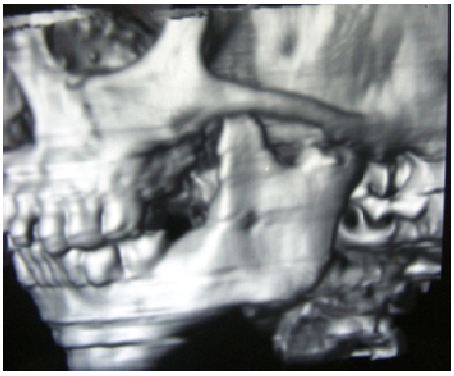

Fig 3 CT Scan

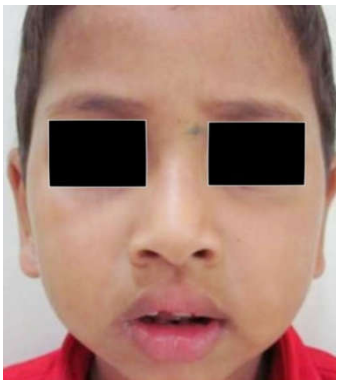

Fig 4 5month post operative photograph

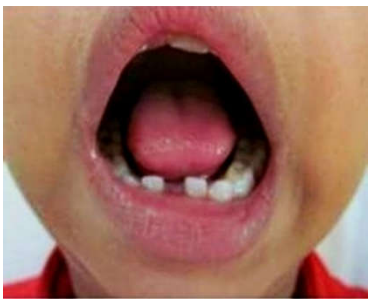

Fig 5 Post operative mouth opening

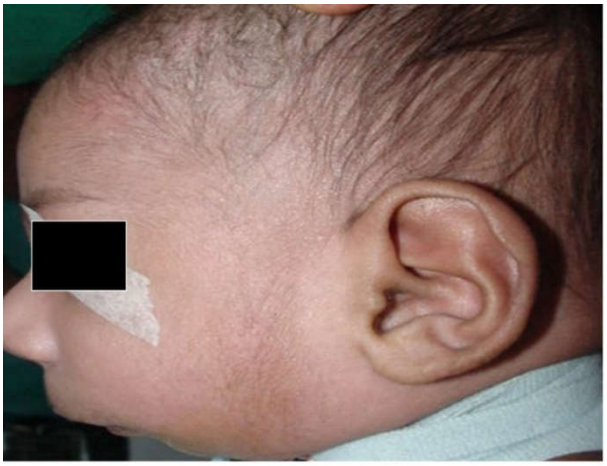

Fig 6 Pre operative profile

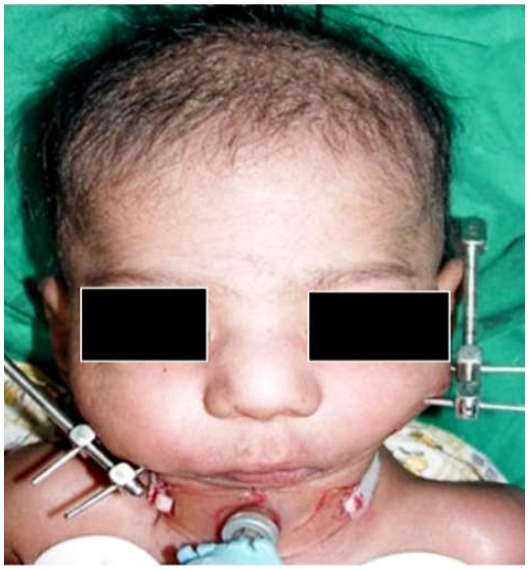

Fig 7 Distractors and Tracheostomy in situ

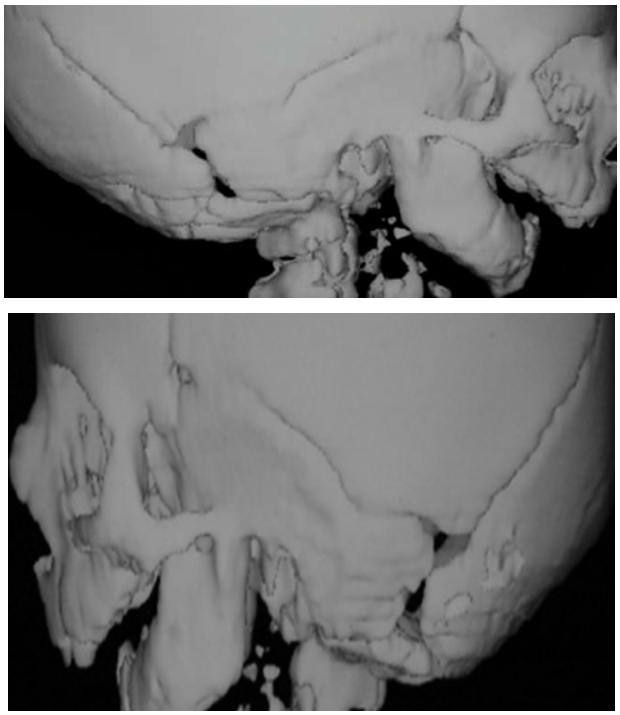

Fig 8- 3 Dimensional Computed Tomography Scan

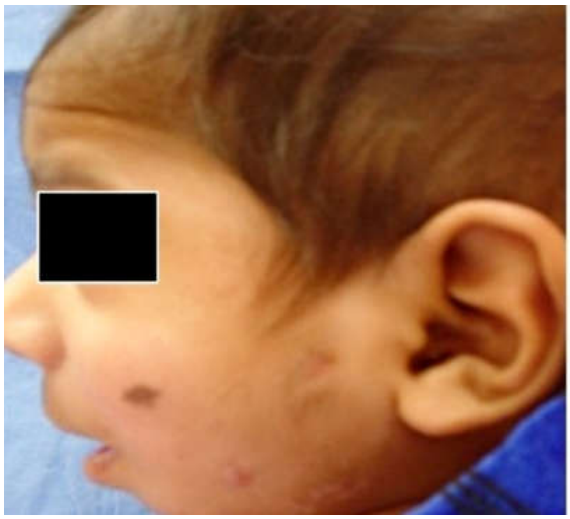

Fig 9 Post operative profile

DISCUSSION

Ever Since the detection of pediatric sleep apnea was reported by Guilleminault at the Stanford Sleep Disorder Clinic, its implications in the children has become the topic of interest in research and clinical investigation[6].

The peak prevalence of pediatric sleep apnea in children occurs in 3-6 years of age. At this age, children have the greatest amount of lymphoid tissue in their upper airways relative to the dimensions of the airways.[7]

Usually snoring and difficulty in breathing are most common complaints of parents of children with pediatric sleep apnea,with reports of these findings in more than 96% of cases.[7,8]

The most common cause of the pediatric sleep apnea is adenotonsillar hypertrophy, almost always amenable to surgery[8,9]. Other reasons for obstruction of the upper airway include obesity, Down's syndrome, various craniofacial anomalies, and neuromuscular diseases[9]. That adenotonsillar hypertrophy is not the sole cause of the syndrome is supported by the fact that affected children do not obstruct when awake, the severity of symptoms is not proportional to the size of the tonsils and adenoids, and sometimes the symptoms persist after removal of these tissues.

OSA in childhood differs from that of adult form. Adults with sleep apnea frequently present with hypersomnia, whereas children often demonstrate short attention spans, emotional liability, and behavioral problems[10]. Certainly, normal children may have a relatively narrow upper airway, but maintain airway patency during sleep because of increasedupper airway neuromotor tone and an increased central ventilatory drive[11, 12]. If children with symptoms of nasal obstruction are found to have both soft tissue and skeletal abnormalities, each may require operative treatment.

Diagnosis of Pediatric Sleep Apnea

Unfortunately, there are very few diagnostic criteria; diagnosis of sleep apnea is based on a spectrum of signs and symptoms and the judgment of the otolaryngologist[13]. Polysomnography (PSG) is considered "gold" standard for diagnosing pediatric sleep apnea. PSG monitoring includes respiration,pulse oximetry, muscle tone electroencephalography , and eye movement[13,14]. But it is problematic not only because of a lack of consensus on the criteria for diagnosing pediatric sleep apnea but also because it is expensive and does not predict morbidity.

Oximetry monitoring alone may identify many elderly patients with mild to moderate sleep apnea , which had been confirmed by the results of previous studies of the young and adults.[13,15]

Vast numbers of surgical and non surgical techniques have been reported in the literature for management of pediatric sleep apnea which tend to improve airway obstruction and associated nutritional difficulties. These includes prone positioning of patient, oral appliances, tongue-lip adhesion or glossopexy, mandibular distraction osteogenesisand tracheostomyhave been used.[16-20] Tonsilloadenoidectomy has been the treatment of choice for pediatric sleep apnea among the surgeons.[8]

Ankylosis of TMJ is a devastating affliction, which usually affects children, the most common cause being childhood trauma and infection.[21-22]While modern antibiotic treatment has reduced the incidence, ankylosing spondylitis, rheumatoid arthritis and psoriasis may also lead to TMJ ankylosis, albeit rarely[22] .The hypothesis has been proposed that in cases caused by trauma, intra-articular hematoma, scarring and excessive bone formation give rise to hypomobility.The afflicted joint gets ankylosed resulting in functional debility and facial deformity due to compromisedgrowth.

The first report discusses a pediatric patient with pediatric sleep apnea secondary to mandibular retrognathia due to TMJ ankylosis, which posed a difficult combination of a respiratory pathology with a functional problem of restricted mouth opening.After a thorough assessment of the primary pathology and pediatric sleep apnea, a simultaneous procedure of gap arthroplasty for correcting both was done but was not followed by distraction osteogenesis, contrary to what had been quoted in the literature.[22-24]

This may be due to the fact that the literature pertained to adult TMJ ankylosis that also required mandibular distraction osteogenesis.

It was observed that in the pediatric patient with bilateral TMJ ankylosis gap arthroplasty led to increase in the mouth opening along with the habitual forward placement of the mandible and the tongue and which would have led to increase in posterior airway space. Sleep apnea in the second case got resolved after distraction osteogenesis as reported in the literature[22-25].As the mandible was distracted in length improvement in the condition correlated with an expanded posterior airway space.

This report is based on a short term assessment and relapse may be kept in mind while evaluating long-term results.

CONCLUSION

In the identification of pediatric sleep apnea the oral surgeon as well as the general dentist can play a major role. A sound and thorough knowledge of such disorders and a multi-disciplinary approach would go a long way in providing a desirable outcome and ensuring better quality of life to the patient. As an oral and maxillofacial surgeon, the emphasis should not only be on the correction of the dentofacial deformities,but it is also important to envisage the underlying disorders like breathing difficulties, sleep apnea and difficulty in feeding. Treatment planning should be done taking into account the age of the patient and the severity of the condition. In infants with congenital micrognathia, distraction osteogenesis would be needed to correct sleep apnea and associated feeding problems as mentioned in the literature. Sleep apnea tends to get corrected after the release of ankylosis as was seen in the case might be due to habitual positioning of mandible under the influence of tongue not necessitating the need of distraction osteogenesis in growing children. However a larger cohort of patients would be needed to be observed to devise a treatment protocol based on specific age group for these patients.

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