Case Report

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Anaesthetic management in a rare case of bird face deformity, hypertrophied adenoids and temporomandibular joint ankylosis

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ABSTRACT

Various methods have been described for nasal intubation in children with temporomandibular joint ankylosis with limited mouth opening. As younger children are uncooperative, they require intubation under anaesthesia. When associated with hypertrophied adenoid, there is high risk of airway obstruction and bleeding into the unprotected airway resulting in laryngospasm and/or bronchospasm. We here describe successful intubation in a paediatric patient in whom the airway was obstructed with hypertrophic adenoids.

Key words: Bronchospasm, fibreoptic laryngoscope, obstructive sleep apnoea, temporomandibular joint

INTRODUCTION

Temporomandibular Joint ankylosis with mandibular hypoplasia presents a serious problem for airway management. It can lead to alarming concerns in paediatric age group because of their small or no mouth opening and associated retrognathia with a relatively large tongue and pseudomacroglossia in a confined space that narrows the pharyngeal passage^[1]. In children with long standing bilateral TMJ ankylosis during the active growth phase, a hypoplastic and retrognathic mandible with severe bird face deformity is noted and hence Obstructive Sleep Apnoea (OSA) is commonly associated. As younger children are uncooperative, they require intubation under anaesthesia. When associated with hypertrophied adenoid, there is a high risk of bleeding into the unprotected airway during the fibreoptic laryngoscopy resulting in laryngospasm/ bronchospasm. These children pose anaesthetic risks due to extreme sensitivity to sedatives and opioid. We here describe successful airway management and intubation in TMJ ankylosis case in which the airway was obstructed with hypertrophic adenoids.

CASE REPORT

A thirteen year old male child came with chief complaints of inability to open mouth and was on liquids and semisolid food only. There was a history of difficulty in breathing and

snoring during sleep for the past six years. Patient came to the hospital as the difficulty in breathing and snoring had increased in the past few weeks. On taking radiograph of nasopharynx enlarged adenoids were found. The patient was examined and on investigations was found to have mandibular hypoplasia with left TMJ ankylosis (Figure 1). Paediatric and cardiac consultation were done to rule out any associated congenital abnormalities.

Airway assessment revealed nil mouth opening, Mallampatti Grading and TMJ movements could not be assessed as the patient had severe TMJ ankylosis. Thyromental distance, hyomental and sternomental distances were more than 6.5, 6 and 12.5 cms respectively. Only right side of nares was patent and left side had Deviated Nasal Septum (DNS). Adenoids were reported to be grossly hypertrophied. Computed Tomogram (CT) scan of head and face revealed bilateral TM Joint ankylosis with hypertrophied Adenoids, prognathism and hypoplasia of right mandibular condyle.

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The patient was nebulized with 5 ml of 4 % lignocaine 20 minutes before the procedure and otrivin (Xylometazoline) nasal drops were instilled to decrease bleeding during instrumentation. As per our planned procedure, the first choice included fibreoptic laryngoscopic nasal intubation with flexometallic tube and as a last resort tracheostomy set was kept ready. The pros and cons of the anaesthetic procedure were explained to the patient's parents and informed consent was taken. Presence of an ENT surgeon was also ensured in the operation theater.

The patient was pre medicated with injection glycopyrrolate 0.1mg, pantoprazole 20 mg and 1 mg of midazolam intravenously. The child was little anxious, so subanaesthetic dose of ketamine was admistered (15mg). Glossopharyngeal nerve, Superior laryngeal nerve and recurrent laryngeal nerve blocks were administered using one ml of 1% lignocaine for each block under aseptic conditions. Thereafter, patient was taken up for fibreoptic nasotracheal intubation. The scope was passed through right nares after applying lignocaine jelly along the floor of the nose and was gently advanced slowly through the nose and slided into pharynx after visualizing adenoids in atraumatic way so as to avoid bleeding which would have otherwise obscured the view and led to failure. After going beyond the adenoids, epiglottis, vocal cords and tracheal rings were identified. 4.5 mm flexometallic tube was railroaded to go beyond vocal cords (Figure 2). Inj propofol 40 mg i.v and vecuronium 2 mg were given after confirming the tube placement. Maintenance of anaesthesia was achieved with oxygen, nitrous oxide and halothane. The patient tolerated surgery well and vitals were maintained and monitored throughout the procedure. At the end of surgery, adequate suctioning and clearing of secretions and extubation was done when the patient was fully awake and was able to maintain the airway. Mouth opening of two fingers was achieved after surgery. Postoperatively, the patient recovered uneventfully (Figure 3).

DISCUSSION

The cause of TMJ ankylosis may be congenital, traumatic, infectious or of unknown etiology. Bird face deformities with convex facial profiles can be found in chronic long standing cases with micrognathic mandible and receding chin. Maxilla, soft tissues enveloping the mandible and suprahyoid muscles are secondarily affected. Predictor of difficult intubation like Mallampatti grading was not possible to assess in this case. Awake tracheal intubation is the safest technique but it is not feasible in children and hence requires general anaesthesia^[2].

Options for anaesthetic management remain very few and that too are difficult and challenging which include but are not limited to fibreoptic laryngoscopic nasal intubation, a blind nasal intubation^[3], binasophayngeal airway^[4], fluoroscopic aided retrograde placement of guide wire for tracheal intubation^[5], semiblind technique of nasal intubation and tracheostomy^[6]. All these techniques have got their own merits and disadvantages. Fibreoptic laryngoscope remains the gold standard technique whenever oral route is impossible. Blind nasal intubation has high failure rates. Moreover, in the present case hypertrophied adenoids were present and chances of bleeding were high so blind nasal intubation technique was not advisable.

Compared to flexible fibreoptic laryngoscope, retrograde tracheal intubation is a traumatic experience in the awake patient, uncomfortable and invasive procedure, but may be used where facilities for fibreoptic intubation are not available or not feasible^[7]. Moreover, because of large sized adenoids, it was also not feasible.

Similarly tracheostomy is associated with severe morbidity, mortality and long term side effects so it was reserved as last option in case of emergency. Anaesthetic agents such as opioids and sedatives worsen Obstructive Sleep Apnea (OSA) by several mechanisms. They tend to reduce pharyngeal muscle tone thereby increasing likelihood of upper airway collapse. They reduce arousal and ventilatory response to hypoxemia and hypercapnea^[8]. Therefore, use of any sedative during pre induction period was avoided and only a very small dose of midazolam (1mg) was given.



Figure 1: Showing the bird's deformity due to temporomandibular joint ankylosis and mandibular hypoplasia



Figure 2: Fibreoptic view showing the epiglottis and the adenoids



Figure 3: Post operative view of the child showing a mouth opening of two fingers

CONCLUSION

When TMJ ankylosis is associated with adenoids, blind nasal and other techniques like retrograde intubation should be avoided as they increase the risk of bleeding. Flexible fibreoptic laryngoscope is the method of choice for coping with this type of difficult tracheal intubation techniques. One must acquire skill to operate fibreoptic bronchoscope and it should essentially be available in difficult airway cart to manage such challenging cases.

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