Airway management of an unusual case of recurrent rhinoscleroma

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Abstract

Rhinoscleroma is a rare entity encountered in anesthesia practice. We discuss the management of a patient after its recurrence, involving the upper respiratory tract i.e. nasopharynx and oropharynx, which compromised the airway. The patient was referred for anesthesia on three different occasions with different presentations owing to the recurrence of symptoms. The presence of an oropharyngeal membrane with a small opening made airway management a challenge. The patient was successfully managed on all three occasions. Imaging facilitated assessment and subsequent airway management.

Key words: Rhinoscleroma, recurrence, difficult airway

Introduction

Rhinoscleroma is a rare entity encountered in anesthesia practice. We discuss the management of a patient after its recurrence, involving the upper respiratory tract i.e. nasopharynx and oropharynx, which compromised the airway. The patient was referred for anesthesia on three different occasions with different presentations owing to the recurrence of symptoms. The presence of an oropharyngeal membrane with a small opening made airway management a challenge. The patient was successfully managed on all three occasions. Imaging facilitated assessment and subsequent airway management.

Rhinoscleroma is a chronic, granulomatous infection, caused by Klebsiella rhinosleromatis, affecting the nasopharynx, oro-pharynx, larynx, trachea and bronchi. The disease is known to cause slowly progressive asphyxia. [1,2] It is common in the second and third decade of life, with females more

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frequently affected.^[3] It has three histological stages - catarrhal, granulomatous, and sclerotic/cicatrizing.^[4] It is known to be recurrent but rarely lethal unless it obstructs the airway. Treatment consists of prolonged antibiotic treatment, surgery to remove the obstruction and laser therapy to treat airway compromise and tissue deformity.^[2,5,6] This progressive infection, involving the upper respiratory tract, may pose a challenge in airway management.

Case Report

A 26-year-old male developed bilateral diminished hearing six years ago. He was diagnosed as otitis media and repeatedly treated with antibiotics. However, he had no long-term



Figure 1: Oro-pharyngeal membrane visible at the level of uvula covering the oropharynx with a central opening of approximately 8 mm diameter

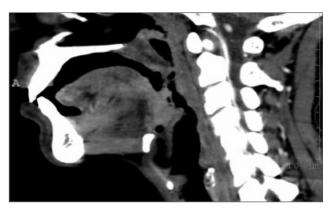


Figure 2: MRI of the airway sagittal section showing the membrane in the oropharynx and no involvement of the airway below

relief and the symptoms progressed. In the last six months, he developed difficulty in breathing and swallowing. Oral examination revealed a membrane at the level of uvula covering the oropharynx with a central opening of approximately 8 mm diameter through which he managed swallowing [Figure 1]. Routine investigations were within normal limits. CT scan and MRI scan showed a concentric narrowing at the level of the oropharynx [Figures 2 and 3] with sparing of the lower respiratory tract. Mallampati grading of airway could not be done. Nasal passages did not seem to be completely patent. Flexible fiber-optic bronchoscopy (with a 6 mm scope) under LA revealed uninvolved vocal cords, but patient had discomfort and was unable to hold his breath during the procedure. Bronchoscopy and biopsy under local anesthesia (LA) confirmed diagnosis as 'rhinoscleroma'. Patient was posted for elective pharyngoplasty to excise the membrane.

Premedication with 20 mg omeprazole and glycopyrrolate 0.2 mg intramuscularly was given. Standard monitoring was initiated which included pulse oximetry and capnometry. Difficult intubation cart was prepared and emergency tracheostomy preparation was done. After preoxygenation, anesthesia was induced with 120 mcg fentanyl, propofol 1mg/kg and sevoflurane in titrated doses. After ensuring adequate mask ventilation, succinylcholine 1.5 mg/kg was given. Although an ideal laryngoscopy could not be performed, moving the tongue gently with the laryngoscope helped visualize the membrane. A 15 Fr bougie was inserted via the hole in the membrane and tracheal rings were well felt. A non-laser shielded 5 mm ID endotracheal tube (ETT) was negotiated, over the bougie, through the opening. Bilateral air entry was confirmed by capnometry and auscultation. Anesthesia was maintained with oxygen, nitrous oxide, sevoflurane and atracurium. Membrane was resected with diode contact laser and the tissue was sent for histopathology evaluation. Patient was extubated and recovery was uneventful.

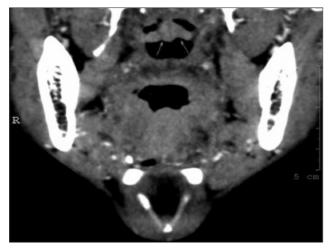


Figure 3: MRI coronal view showing narrowing of the oropharynx due to the membrane (shown by arrow)

Four months later patient had recurrence of the oropharyngeal membrane. Breathing and swallowing difficulties were more severe this time. A repeat pharyngoplasty was planned. The induction was done as in the previous instance but we were unable to negotiate the bougie, and two attempts at blind intubation with 5 mm ID ETT and an attempt with a smaller ETT were unsuccessful. Emergency surgical tracheostomy was done. Intraoperative course was uneventful. Electrical cautery was used. After monitoring the patient in the PACU, he was shifted with the tracheostomy in situ to the ward. Tracheostomy was decannulated on third postoperative day.

One month later, patient presented with stridor. The membrane had recurred with an opening of approximate 5 mm diameter. Emergency tracheostomy was done. Pharyngoplasty was redone with electrical cautery. Post operatively, the tracheostomy was kept and patient discharged on fourth postoperative day with tracheostomy tube *in situ*.

Discussion

Rhinoscleroma leads to fibrosis of the airways with consequent deformity, atresia or complete obstruction of the airway. ^[5] Diagnosis is made on the basis of clinical presentation, histopathology, bacteriological culture and serological investigations. ^[1,3] The extent of oropharyngeal and tracheobronchial involvement can be accurately assessed with CT or MRI scans, although very few studies have described imaging features of rhinoscleroma. ^[7-9] The nasal cavity is the most often affected (95-100%), but the lesions may also involve larynx (15-40%), nasopharynx (18-43%), oral cavity, paranasal sinuses (26%), soft tissues of lips, nose, trachea (12%) and bronchi (2-7%). ^[10] Most patients are symptomatic for many years before they seek medical advice. ^[1]

Sharing the airway with the surgeon is of utmost concern

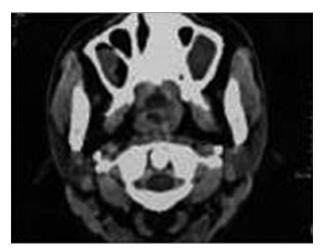


Figure 4: CT scan showing narrowing of the oropharynx due the membrane (shown by arrow)

to an anesthesiologist. In the case reported, a trial at oral intubation guided by a gum elastic bougie was done as the patency of the pharynx distal to the membrane on MRI and CT scan was confirmed [Figures 2-4]. We avoided blind nasal intubation since the nares seemed blocked and the CT image showed some synechiae in the nasal passage. The feel of the tracheal rings by the bougie guided us the insertion of the ETT. A fiberoptic bronchoscope would have helped in visualizing the membrane, as it done in the ENT clinic, but we only had a 6 mm scope, which would have permitted only a larger ETT over it.[11] Trauma to the membrane or bleeding had to be avoided. A supraglottic airway could not be used. Jet ventilation would have made the patient prone to barotrauma as airway pressures cannot be controlled with its use. [12] Retrograde intubation would have been tedious as passing the guide wire through the small opening would have been very difficult. As the pharyngoplasty took only fifteen minutes, ETT was not changed to a bigger size later.

Fatal airway obstruction has been reported after evading tracheostomy in a patient who seemed to be improving following antibiotic treatment. [6] Pharyngoplasty is often a day care procedure performed using laser, and is associated with minimal blood loss, edema, and quick recovery. [4] Routinely, it does not need the patient to have a tracheostomy, as lubricated airways, post-surgery, maintain the airway patency well. In our patient, at the second instance, the opening in the oropharyngeal membrane was smaller. We were however reluctant to do a tracheostomy knowing that the symptoms would be relieved on resection of the membrane. Despite three attempts with smaller sized ETT, the airway could not be secured. An emergency surgical tracheostomy was done followed by pharyngoplasty. The trachea was decannulated on the third postoperative day.

We did not expect recurrence, as the patient had undergone

laser and electrical cautery resection, but there was recurrence. The third time, the patient presented with respiratory distress that warranted an immediate tracheostomy. He had not responded to prolonged antibiotics, anti-inflammatory drugs, steroids or even surgery and finally needed a permanent tracheostomy. [6,8,13] Close observation for prolonged periods of time is essential to recognize reactivation of quiescent lesions. [2] Trauma caused by excision and laser can incite more fibrosis and lead to recurrence of the primary problem with a vengeance. [4] The recurrence rate of rhinoscleroma has been cited to be up to 25% of the treated cases in 10 years. [14]

In patients with an oropharyngeal membrane with a solitary opening, the size of the opening, pathology and involvement of the oropharynx, nasopharynx and the tracheobronchial tree must be evaluated. It is important to consider the site of airway obstruction and ensure availability of equipment to secure the airway. Imaging modalities can help assess the airway at various levels. Early recognition is essential to avoid airway compromise and reduce extensive tissue scaring. Fiberoptic bronchoscope is an important tool in assessing the airway patency but can precipitate respiratory distress and a bougie may be helpful. Tracheostomy should be performed if the opening is small or if the airway is affected at multiple levels.

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