Case Report

Idiopathic subglottic stenosis in pregnancy: A deceptive laryngoscopic view

Address for correspondence:
Dr. John George Karippacheril, Department of Anaesthesiology Kasturba Medical College, Manipal, Karnataka - 576 104, India.
E-mail: johngeorgedon@gmail.com

Access this article online

Website: www.ijaweb.org

DOI: 10.4103/0019-5049.89894

Quick response code

John George Karippacheril, Umesh Goneppanavar, Manjunath Prabhu, Kiran Bada Revappa

Department of Anaesthesiology, Kasturba Medical College, Manipal University, Manipal, Karnataka, India

ABSTRACT

A 28-year-old lady with term gestation, pre-eclampsia and a vague history of occasional breathing difficulty, on irregular bronchodilator therapy, was scheduled for category 1 lower segment caesarean section in view of foetal distress. A Cormack-Lehane grade 1 direct laryngoscopic view was obtained following rapid sequence induction. However, it was not possible to insert a 7.0 or 6.0 size styleted cuffed tracheal tube in two attempts. Ventilation with a supraglottic device was inadequate. Airway was secured with a 4.0 size microlaryngeal surgery tube with difficulty. Computed tomography scan of the neck following tracheostomy for failed extubation revealed subglottic stenosis (SGS) with asymmetric arytenoid calcification. This report describes the management of a rare case of unrecognised idiopathic SGS in pregnancy.

Key words: Asymmetric, cartilage sclerosis, idiopathic, laryngoscopy, pregnancy, subglottic stenosis

INTRODUCTION

Undiagnosed subglottic stenosis (SGS) is a nightmare for the anaesthesiologist as the conventional preoperative predictors of difficult airway may not raise an alert. Conditions that may increase airway oedema can worsen the SGS, which can manifest as wheezing or breathlessness. Often, this may be the only telltale sign of SGS which can easily be misdiagnosed and treated as bronchial asthma with poor response to bronchodilators.[1-3] When such patients with undiagnosed SGS undergo general anaesthesia (GA), a catastrophic series of events may occur following induction. Multiple attempts at intubation by an unsuspecting team of anaesthesiologists, enamoured by an easy laryngoscopic view, can result in a life-threatening *"cannot ventilate, cannot intubate"* scenario, while forceful attempts may even lead to the damage of the laryngeal cartilages or perforation of the trachea.[4] In this article, we present the management of a case of unrecognised SGS with pre-eclampsia and foetal distress scheduled for emergency lower segment caesarean section (LSCS).

CASE REPORT

A 28-year-old lady with term gestation presented to emergency department with pre-eclampsia and a vague history of occasional breathing difficulty in the past for which she was on irregular bronchodilator therapy; with no history of admission. She was able to provide her history and was comfortably breathing. Her vitals were as follows: Blood pressure, 158/94 mmHg; heart rate, 83/min with respiratory rate 20/min. She was scheduled for category 1 LSCS (immediate) in view of foetal distress. With unknown coagulation status and a normal appearing airway, GA was planned. The anaesthetic induction details are as follows: Preoxygenation, propofol - 2 mg/kg with cricoid pressure *in situ*, suxamethonium - 1.5 mg/kg. Direct laryngoscopy and intubation attempted following 60 seconds of apnoeic oxygenation revealed generalised oropharyngeal and glottic oedema. However, despite a Cormack-Lehane grade 1 view, it was not possible to insert a 7.0 size or subsequently a 6.0 size styleted cuffed tracheal tube in two attempts. A size 3 ProSeal™ laryngeal mask airway (PLMA) was inserted with cricoid pressure maintained,

How to cite this article: Karippacheril JG, Goneppanavar U, Prabhu M, Revappa KB. Idiopathic subglottic stenosis in pregnancy: A deceptive laryngoscopic view. Indian J Anaesth 2011;55:521-3.

while a senior anaesthesiologist was called for help. Considerable upsloping of capnogram, $ETCO_2$ - 55 mmHg and tidal volumes of 70 to 80 ml despite 30 cm H_2O peak inflation pressure were noticed. Auscultation over the chest was unremarkable. Reinsertion of the PLMA did not improve the situation. In view of the compromised foetal status, delivery was done immediately while anaesthesia was maintained with isoflurane in oxygen. After the delivery of a healthy child, the patient regained spontaneous breathing efforts but the tidal volumes were 100 to 130 ml with $ETCO_2$ - 90 mmHg. At this stage, in consultation with the otorhinolaryngologist, it was decided to attempt intubation with a styleted 4.0 size cuffed microlaryngeal surgery tube after deepening with propofol, while tracheostomy was reserved as only an emergent option in view of unknown coagulation status. Intubation succeeded in second attempt, after applying considerable force. Subsequent attempts to inspect the subglottic area with fiberscope were futile as it was impossible to pass it between the tracheal tube and vocal cords. Rest of the anaesthetic management was uneventful. Patient was shifted to intensive care unit with the tracheal tube *in situ*. A provisional diagnosis of airway oedema secondary to pre-eclampsia was made. Patient was nursed in head up position and received dexamethasone and bronchodilators. A trial of extubation was given 48 hours later after cuff leak test showed enough peritubal leak. However, the patient required re-intubation within 15 minutes of extubation, following which she was tracheostomised and evaluated for the underlying pathology. X-ray neck, lateral view, showed no air shadow around the tracheal tube [Figure 1] suggesting possibility of SGS. This was confirmed subsequently by a computed tomography (CT) scan that also showed asymmetric calcification of the left arytenoid cartilage and sclerosis of the cricoid cartilage [Figures 2 and 3]. Further evaluation of the airway under anaesthesia confirmed significant SGS with granulation tissue [Figure 4], the biopsy of which was not suggestive of neoplasm or Wegener's

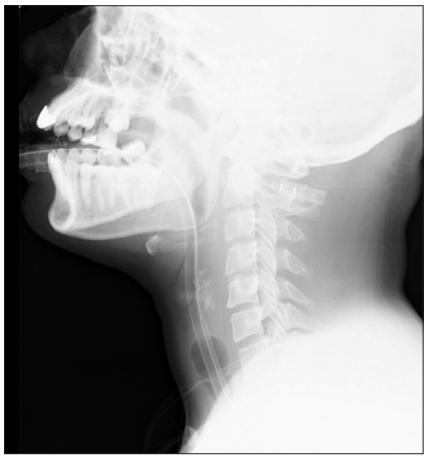

Figure 1: X-ray neck, lateral view, showing absent air column around the endotracheal tube and calcified laryngeal cartilage

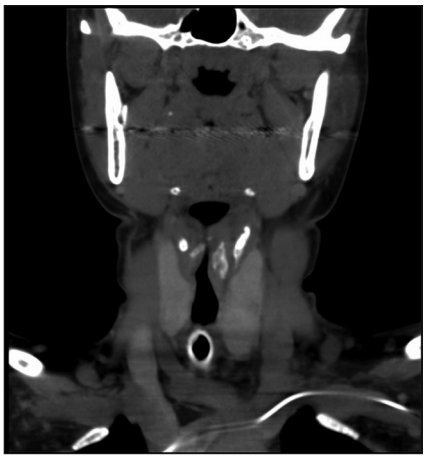

Figure 2: CT scan of the neck. Anteroposterior view, showing severe subglottic narrowing and left arytenoid asymmetric calcification

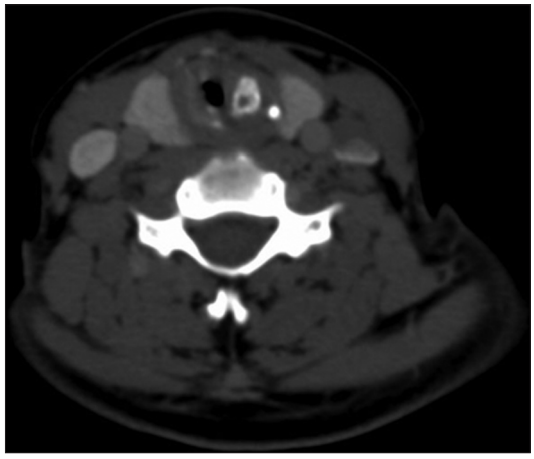

Figure 3: CT scan of the neck. Cross sectional view, show subglottic stenosis with asymmetric arytenoid calcification

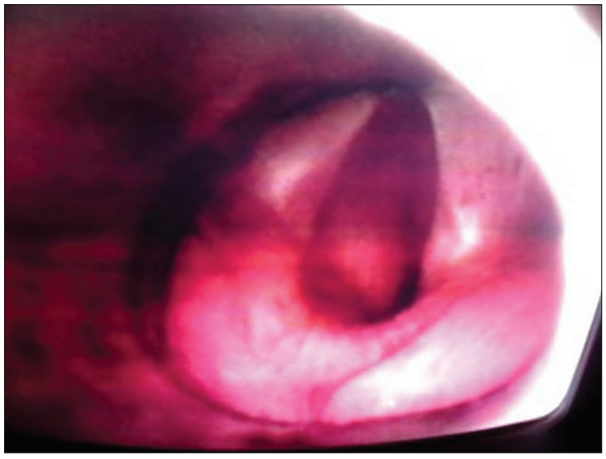

Figure 4: Direct laryngoscopic view showing oedematous vocal cords with severe subglottic narrowing

granulomatosis. Circulating anti-neutrophil cytoplasmic antibody and antinuclear antibody tests were negative. Eight weeks later, she underwent laser excision of the granulation tissue and a silastic keel placement.

DISCUSSION

Trauma, prolonged tracheal intubation, gastroesophageal reflux disease and Wegener's granulomatosis are reported causes for SGS.[1] However, idiopathic SGS commonly occurs in women, which is believed to be related to hormonal factors.[2] Its occurrence during pregnancy is rare and very few cases have been reported in the literature.[1,2,5-7] SGS of pregnancy is probably a misnomer. Conditions that promote airway oedema may result in asymptomatic SGS to manifest with breathlessness.[1] In this case, pregnancy and pre-eclampsia might have contributed to airway oedema, although she remained sub-symptomatic till the last stage of pregnancy. Unfortunately, ability to mask ventilate could not be checked prior to laryngoscopy; thus, it is difficult to ascertain if intubation attempts contributed to worsening of the situation. Despite anti-oedema measures, the patient failed an extubation trial and the presence of granulation tissue noticed during tracheostomy, just 48 hours after initial intubation, indicates the possibility of severe SGS prior to admission itself. The asymmetric calcification or sclerosis of laryngeal cartilages may be a possible predictive marker for underlying neoplastic invasion, especially with calcification of the cricoid cartilage.[8] However, the sensitivity and specificity of this in isolation has been reported to be low.[9] Biopsy of the granulation tissue in our case was negative for malignant cells. Whether sclerosis of the tissue, in proximity to the laryngeal cartilages, contributed to the unyielding nature of the subglottic tissue is not definitely known. Supraglottic airway devices or smaller tracheal tubes have been used successfully to manage SGS.[10] However, in our case, although supraglottic airway was life saving, a need for effective airway resulted in the use of a smaller size tube.

CONCLUSION

Although it is tempting to continue attempts at intubation when a full view of the glottis is visible, initial failure to intubate should lead to suspicion of SGS. In suspicious cases, oxygenation can be maintained with a supraglottic device, while a fiberscope can be passed through this to evaluate the airway below the glottis, which can help formulate a further airway management plan.

REFERENCES

1. Scholz A, Srinivas K, Stacey MR, Clyburn P. Subglottic stenosis in pregnancy. Br J Anaesth 2008;100:385-8.
2. Kuczkowski KM, Benumof JL. Subglottic tracheal stenosis in pregnancy: Anaesthetic implications. Anaesth Intensive Care 2003;31:576-7.
3. Paré PD, Donevan RE, Nelems JM. Clues to unrecognized upper airway obstruction. Can Med Assoc J 1982;127:39-41.
4. Norris BK, Schweinfurth JM. Arytenoid dislocation: An analysis of the contemporary literature. Laryngoscope 2011;121:142-6.
5. Sutcliffe N, Remington SA, Ramsay TM, Mason C. Severe tracheal stenosis and operative delivery. Anaesthesia 1995;50:26-9.
6. Ratner EF, Cohen SE, El Sayed Y, Druzin M. Mask induction with sevoflurane in a parturient with severe tracheal stenosis. Anesthesiology 2001;95:553-5.
7. Salama DJ, Body SC. Management of a term parturient with tracheal stenosis. Br J Anaesth 1994;72:354-7.
8. Nix PA, Salvage D. Neoplastic invasion of laryngeal cartilage: The significance of cartilage sclerosis on computed tomography images. Clin Otolaryngol Allied Sci 2004;29:372-5.
9. Agada FO, Nix PA, Salvage D, Stafford ND. Computerised tomography vs. pathological staging of laryngeal cancer: A 6-year completed audit cycle. Int J Clin Pract 2004;58:714-6.
10. Yavaşcaoğlu B, Tokat O, Basagan EM, Kaya FN, Erisen L, Kutlay O. The use of the laryngeal mask airway in children with subglottic stenosis. J Int Med Res 2001;29:541-5.

Source of Support: Nil, **Conflict of Interest:** None declared