Case Report

Management of Compromised Airway due to Unusual Presentation of Cystic Hygroma

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Abstract

The successful management of a unique case of huge cystic hygroma in a fourteen month old child who presented in the emergency room with severe respiratory distress as a result of nearly almost complete upper airway obstruction, is presented.

Introduction

Cystic hygroma is a benign, painless, soft and compressible tumor of lymphatic system. Its frequency is one case per 6000 births in USA.[1] The head and neck region is the most common site of presentation. Forty percent of the lesions appear in the newborn, 50% present by the end of the first year of life, and 75% by the end of the second.[2]

Cystic hygromas have rarely been reported as a cause of acute upper airway obstruction with sudden neck swelling as a result of infection and haemorrhage into the lesion.[3] Anaesthetic concern includes bleeding, difficulty in visualizing the airway, extrinsic and intrinsic pressure on the airway causing distortion and obstruction of the airway. Therefore a strategy needs to be developed in order to anticipate and manage patients with these types of difficult airways. Multi specialty approach can ensure the survival of a child presenting with dyspnoea due to airway obstruction. This includes identifying the potential problems and considering different options and selection of an appropriate plan in the particular scenario. We are reporting an airway crisis in a paediatric patient admitted with severe respiratory distress due to a huge cystic hygroma arising from the neck and extending into the base of tongue, pharyngeal space, mediastinum and diaphragm. In addition to involving the laryngeal inlet, it also causes deviation and compression of the trachea.

Case Report

A fourteen months boy presented in the emergency room with respiratory distress due to a large swelling situated in the anterior and posterior triangle on the right side of the neck. It was associated with fever and stridor. On examination respiratory rate was 52 breaths per minute, heart rate 130 beats per minute and arterial haemoglobin oxygen saturation (SpO_2) was 75-80% with 10-15 liter per minute oxygen via face mask. X-ray chest revealed large swelling extending from neck to mediastinum causing airway narrowing as well as displacing trachea towards the left side. However, lateral X-ray of the neck showed almost complete airway compression at the level of epiglottis (Figure 1). MRI showed a sharply defined lesion in front of the

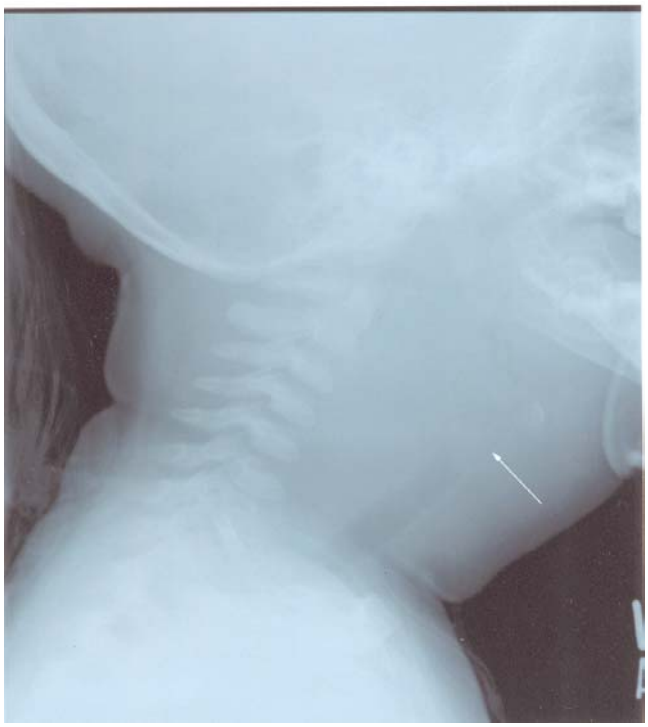

Figure 1. Marked swelling surrounding larynx and trachea, almost occluding the airway at the level of epiglottis

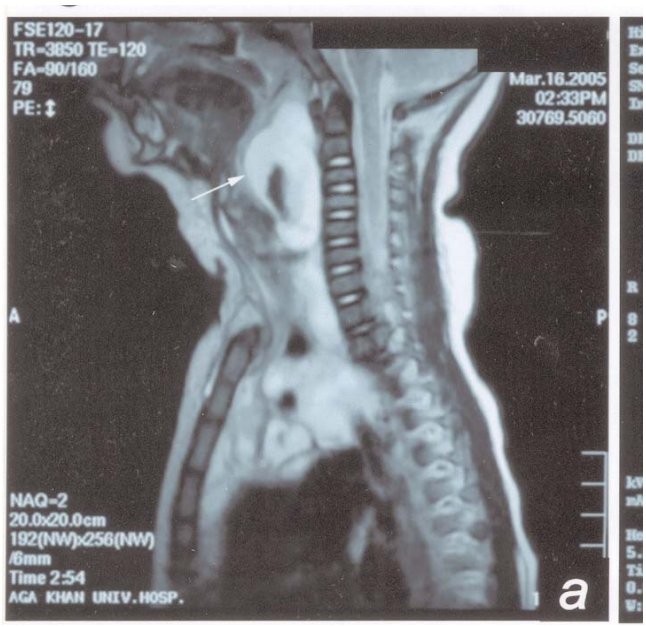

Figure 2a. a: MRI showing sharply defined lesion in front of cervical vertebrae and behind the base of tongue, narrowing the airway with pinching effect at the level of Laryngeal inlet

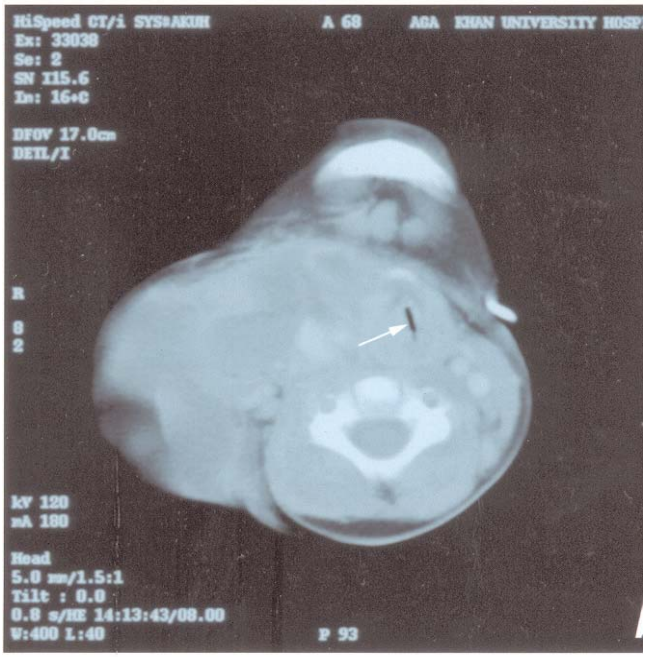

Figure 2b. Slit like glottic opening showing the compromised airway

cervical vertebra and behind the base of the tongue (Figure 2a), making a slit like glottic opening and was compromising the airway (Figure 2b). On the basis of the clinical findings and MRI appearance he was diagnosed as a case of cystic hygroma. Considering the severity of the acute airway obstruction and respiratory distress, emergency tracheostomy was planned and the child was brought to the operating room.

In the operating room electrocardiograph, pulse oximeter, and blood pressure cuff were attached. Patient baseline SpO_2 was 75% and he was hypoxic (SpO_2 75-80%) since his admission in ER which was approximately one hour before coming to the operation theater. Intravenous cannula was already in place in the dorsum of right hand. Inhalation induction was started by using sevoflurane along with 100% Oxygen. After five minutes SpO_2 rose to 90% but the patient was not anaesthetized adequately. To increase the depth of anaesthesia a small dose of thiopentone (10 mg) was given intravenously. This resulted in apnea and airway collapse. However, positive pressure ventilation via face mask was started which kept the SpO_2 to 90%. Laryngoscopy revealed marked swelling in the oropharynx and hypopharynx and laryngeal inlet was not visible. Mask ventilation resumed and laryngeal mask airway (LMA) size 1.0 was inserted. Inflation of LMA cuff displaced it to the left side of the mouth which is possibly due to the anatomical distortion of the oral cavity. Keeping LMA in this position positive pressure ventilation was started and kept inspired concentration of sevoflurane to 2 - 2.5% with 100% Oxygen. Further increase in sevoflurane concentration resulted in airway collapse, causing difficulty in ventilation and dropping of SpO_2 to 70%. In order to prevent this we kept sevoflurane concentration between 2-2.5%. Oxygenation was improved by applying continuous positive airway pressure (CPAP) of 10 cm of H2O and obstruction was relieved but SpO_2 could not be increased more than 90%. Due to inadequate ventilation, end tidal carbon dioxide concentration (ETCO2) remained 75 - 80 mm of Hg. Trachoestomy was started after infiltration of 2% Xylocaine for local anaesthesia. No muscle relaxant was used throughout the procedure. Breathing Circuit was attached to the Tracheotomy Tube (TT) as soon as it was inserted. Ventilation via TT improved oxygenation markedly and SpO_2 rose to 98% while ETCO2 decreased to 40 mm of Hg. Sevoflurane was discontinued after the completion of the procedure. Patient regained consciousness after five minutes and was shifted to the recovery room.

In the recovery room breathing was normal and SpO_2 remained 98%. After meeting the discharge criteria child was shifted to paediatric special care unit.

Discussion

Airway compromise due to cystic hygroma, although rare, but remains an important cause especially in an emergency situation. Airway obstruction can occur in cystic hygroma due to infection and haemorrhage causing rapid increase in the size of swelling.

Different options are available for intubation. Blind

nasal intubation is associated with bleeding that would further compromise the airway. Fiberoptic intubation is an attractive option but expertise is required to handle paediatric airway emergency. While tracheostomy under local anaesthesia is not only distressing for the child but also difficult for the surgeon, because of indistinguishable landmarks.

Inhalation anaesthesia remains the preferred technique for management of a difficult pediatric airway.[3] We also tried this technique in our patient along with keeping the patient spontaneously breathing but the airway obstruction worsened and he became restless.

The exact mechanism of obstruction in these patients under general anesthesia remains unclear. The probable explanation is that during normal breathing intra pleural pressure and intra alveolar pressure is always more negative than intra tracheal pressure and thus airway remains open. When patient tries to breathe out forcefully to overcome resistance, the intra pleural and intra alveolar pressure increases markedly subjecting the trachea to compress transmural pressure especially at the lower trachea level. When positive pressure is applied to airway during forced expiration, the gradient between intra thoracic and intra tracheal is reduced therefore tracheal collapse is less likely.[4] Application of continuous positive airway pressure in our patient with intermittent positive pressure did alleviate tracheal collapse. This has been described by several authors.[5]

The neuromuscular blockage agent may relieve airway collapse during forceful expiration and straining but they may worsen the airway obstruction if the cause of obstruction is mechanical such as mediastinal mask.[6] Therefore we did not use muscle relaxants.

In our patient, the causes of airway obstruction were not only pressurizing effect of primary pathology on trachea but also the loss of muscle tone during general anaesthesia aggravated the pressurizing effect of the mass.

The treatment and prevention of symptoms associated with acute neck swelling like cystic hygroma has not been established. Certain anaesthesia considerations will help in the management of such airway emergencies. Firstly airway obstruction may get worse during general anaesthesia, as airway dynamic (intrapleural - intrathoracic pressures) may markedly change during this period. Secondly, it is important for the patient not to cause straining, since forced expiration worsens the narrowing of airway. Thirdly, positive pressure ventilation with CPAP during expiration will prevent collapse of upper and lower airway. In addition to these, awareness of potential airway complications, coordination and expertise of an anaesthesiologist, paediatric otolaryngologist or paediatric surgeon are needed to manage these complex situations.

References

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