Management of unexpected subglottic stenosis in a neonate with congenital tracheoesophageal fistula

Sir,

We present a case of 4-day-old full-term female weighing 2 kg with tracheoesophageal fistula (TEF) posted for fistula ligation and repair. On examination, the child was crying, tachypneic with drooling of saliva, and receiving intravenous fluids. Her heart rate (HR) was 165 beats/min and SpO_2 was 85% on room air which improved to 89 with oxygen. Coarse crepitations were auscultated, bilaterally. Her investigations were found to be within normal limits. Chest X-ray showed bilateral opacities and a PaO_2 of 60 mmHg (on oxygen) on arterial blood gas.

After taking high-risk consent and arranging for post-operative ventilation, the patient was taken up for surgery. On attaching monitors, HR was 154/min, blood pressure was 68/40 mmHg, and SpO_2 83% on room air which increased to 95% with FiO_2 - 1.0. Premedication with injection atropine 0.1 mg and injection fentanyl 2 μg was given. After inhalational induction with sevoflurane, mask ventilation was found to be adequate. Hence, injection atracurium 1 mg was given for the best intubating conditions [1]. After ventilating for 3 min, direct laryngoscopy (DL) was performed which showed Cormack–Lehane Grade 1. Intubation with uncuffed 3 mm endotracheal tube (ETT) was attempted; however, it was getting stuck beyond the vocal cords. The same was the case with 2.5 mm internal diameter (ID) ETT; therefore, the ventilation was resumed. Laryngoscopy was done again by more experienced anesthetist. DL findings remained the same. Intubation with stiletted 2.5 mm ID ETT and 2 mm ID ETT was also attempted but was unsuccessful. Ventilation was maintained with bag mask with FiO_2 - 1.0. Provisional diagnosis of subglottic stenosis was made, and otolaryngologists were called for emergency tracheostomy. Appropriate size tracheostomy tubes (TTs) were kept. Meanwhile, I-gel 1.0 was inserted to maintain ventilation. However, ventilation deteriorated and mask ventilation resumed.

After tracheostomy, an uncuffed 3.0 mm ID TT was inserted. Initially, the ventilation was adequate with $EtCO_2$ of 25–35 mmHg and saturation between 95% and 98%. However, within 3–4 min, saturation started falling and ventilation became suboptimal. Position of TT was reconfirmed and it was slightly withdrawn. Suctioning was also attempted, but there was resistance in passing smallest catheter (5 Fr) through TT. Ventilation did not improve. Suspecting that TT was blocked, it was replaced with cuffed 3.0 mmID TT and cuff inflated with 2 ml air. Subsequently, $EtCO_2$ rose to 40–50 mmHg and saturation picked up to 96%. The removed uncuffed 3.0 mm TT was not blocked. Hence, it was assumed that inflated cuff of the TT sealed the fistula and helped in improving ventilation. Only gastrostomy as a life-saving procedure was done.

TEF is a congenital anomaly which requires endotracheal intubation as definitive method to secure the airway since it helps to bypass the fistulous site and ensures effective ventilation. The same would not be possible with a supraglottic device. The diagnosis of subglottis stenosis first needs to be confirmed by mainly radiological or endoscopic means (Fig. 1). There are only surgical treatment modalities present, namely endoscopic dilation, cricotracheal resection, or tracheotomy.

Reena *et al.* [2] described a case of acquired subglottic stenosis; however, there were some risk factors and indicators raising suspicion about the diagnosis in their case of an older child with a history of prolonged intubation. On the contrary, such a condition was not anticipated in our case. Thus, we highlight the difference between the acquired and congenital variety and how one can anticipate either of the two and remain prepared for them.

In retrospective review conducted by Hseu *et al*., multiple airway anomalies were seen to be associated with TEF and tracheomalacia having a highest association. The incidence of subglottic stenosis was found to be 12.9%, whereas it is <2% in pediatric population [3]. Franzen *et al*. reported a case of laryngeal web in a neonate, diagnosed on flexible bronchoscopy by otolaryngologists, and intubation was done by anesthesiologist with a 2.0 mm ETT. . However, the patient desaturated, compelling them to perform emergency tracheostomy [4].

Similarly, after unsuccessful intubation with appropriate pediatric ETT, Kundal *et al*. secured the airway using 6 Fr feeding tube which was connected to 3.5 mm ETT connector and used to ventilate the patient for the entire procedure [5]. This was an innovative way of securing the airway; however, narrow diameter of the catheter would increase work of

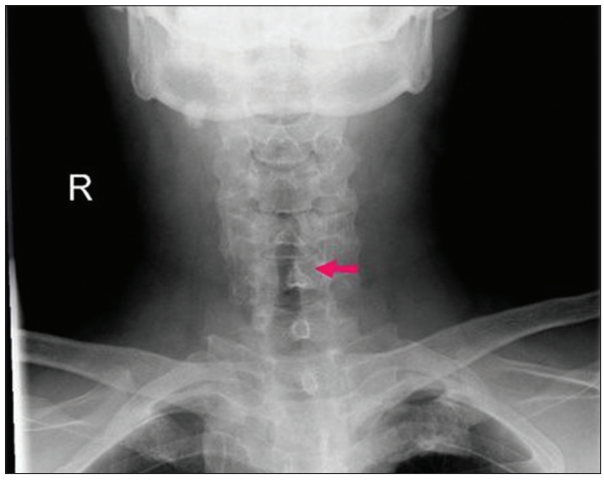

Figure 1: Anteroposterior and lateral X-ray of soft tissue is one of the first screening investigations for subglottic stenosis

breathing, make suctioning difficult, and make it more prone to getting blocked. However, this method may be considered to provide oxygenation on an emergency basis until definitive surgical access is obtained.

Thus, we conclude by saying that neonate with TEF may also have other airway anomalies. While dealing with a case of congenital TEF, appropriate screening tests should be conducted. Furthermore, difficult airway equipment including flexible neonatal bronchoscope should be kept standby. In addition, if unanticipated difficult intubation is encountered, tracheostomy should be the definitive method to secure the airway. Cuffed TT may be preferred to an uncuffed one. Oxygenation in the interim period should be maintained with safest means available and timely multidisciplinary help must be ensured for definitive management.

Megha Kohli, Alphonse Das Aswin, Vishnu Das Panwar, Sonia Das Wadhawan

From [1]Senior Resident, [2]Postgraduate Student, [3]Specialist, [4]Director and Professor, Department of Anaesthesiology and Intensive Care, Maulana Azad Medical College, Bahadur Shah Zafar Marg, New Delhi, India

Correspondence to: Megha Kohli, Department of Anaesthesiology and Intensive Care, Maulana Azad Medical College, Bahadur Shah Zafar Marg, New Delhi - 110 002, India. E-mail: kohli7789@gmail.com

Received - 20 April 2018
Initial Review - 18 May 2018
Published Online - 25 June 2018

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Funding: None; Conflict of Interest: None Stated.

How to cite this article: Kohli M, Aswin AD, Panwar VD, Wadhawan SD. Management of unexpected subglottic stenosis in a neonate with congenital tracheoesophageal fistula. Indian J Case Reports. 2018;4(4):336-337.

Doi: 10.32677/IJCR.2018.v04.i04.028