Recurring and alternating lung collapses - An unusual presentation of neonatal subglottic stenosis

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Abstract:
Subglottic stenosis is a recognised complication of prolonged ventilation in neonates. Some neonates develop subglottic stenosis even on shorter duration of ventilation. We are reporting a 12 day old neonate admitted for bronchopneumonia. The baby was ventilated for 5 days and extubated. During the post extubation period, respiratory distress persisted without stridor. Recurring and alternating lung collapses were observed on serial x-rays in spite of good pulmonary care. This triggered us further to investigate for airway abnormality which led to the diagnosis of subglottic stenosis and tracheomalacia. An unusual presentation like this post extubation should be evaluated for subglottic stenosis.

Keyword: Subglottic stenosis, ventilation, collapse Recurring and alternating lung collapses

- An unusual presentation of neonatal subglottic stenosis

Introduction
Newborn babies get mechanically ventilated due to various causes like respiratory failure, cardiogenic shock or for neurological reasons. Subglottic stenosis is a known complication of prolonged ventilation. Subglottic stenosis is also an important cause for extubation failure (Parkin et al 1976)¹. Various risk factors and pathophysiological mechanisms are proposed for subglottic stenosis. Risk factors range from size of the tube, nasal versus oral intubation, frequent movement of the tube (Donkle et al 1986)², aggressive versus gentle ventilation (Gaynor et al 1993)³. Follow up of neonates intubated in intensive care detect subglottic stenosis early (Jones et al 1981)⁴. Some studies have shown association with infection and early subglottic stenosis. Bacterial production of a biofilm that coats the endotracheal tube acts as a reservoir for infection, prevents eradication and may play a role in the development of subglottic stenosis (Gibbs et al 2012)⁵. We are reporting an unusual presentation of subglottic stenosis. Recurring and alternating lung collapses post extubation led to the diagnosis of subglottic stenosis and tracheomalacia.
Case Report:

We are reporting a 12 day old male neonate born through vaginal route at term gestation to a 26 year old mother who had regular antenatal visits in the primary health centre where the baby was born. The baby was 2.3 kg at birth and the perinatal period was uneventful. The mother and baby were discharged on the same day of delivery with the baby feeding well at breast. The baby was doing well till 11 days of life when the mother gave Acorus calamus (Vasambu). The baby developed respiratory distress for which he was referred to our hospital on day 12 of life.

At admission the baby had severe respiratory distress (Downes score 6). The baby required oxygen to maintain saturation. Respiratory system examination revealed bilateral crepitations. X ray showed evidence of bronchopneumonia. In view of severe respiratory distress and impending respiratory failure the baby was ventilated. The baby was extubated after 5 days. Blood culture grew Enterococci and appropriate antibiotic instituted. Child had persistent respiratory distress without stridor. Repeated x rays were taken which showed recurring and alternating lung collapses despite good pulmonary care.

Fig.1 Admission X ray

Fig.2 Left lung collapse

Fig.3 Right lung collapse

This led to the suspicion of airway abnormality and we investigated the neonate by direct laryngoscopy and bronchoscopy. Fibre optic bronchoscopy on day 25 of life revealed posterior one third of subglottic region was edematous and stenosed with evidence of tracheomalacia.
The baby was given steroids to decrease subglottic edema and regular chest physiotherapy to treat persistent collapses. Baby recovered completely on day 30 of life. On follow up the baby did not develop any complications.

**Discussion:**

Subglottic region is the narrowest part of the airway in a neonate. This region is highly prone for stenosis. Even a millimetre change can reduce the airway by 60%. The incidence of congenital subglottic stenosis is rare. Acquired stenosis is common in prolonged ventilation. Varying incidence ranging from 1% to 8% of ventilated newborns are reported in studies.

Walner et al (2001) showed decreasing trend of subglottic stenosis from 8% to 1-2%. The decrease in incidence is largely attributed to improved practice of ventilation. Gentle ventilation and non invasive ventilation like CPAP have largely reduced the incidence. Few studies in developed centres have not shown much difference in incidence where the incidence is already low (Choi et al 2000)

We are reporting this case, because this baby has developed subglottic stenosis in shorter duration of ventilation. Preexisting infection might be attributed to its earlier onset in our case. Persistent respiratory distress without stridor is unusual in a case of subglottic stenosis. But recurrent and alternating lung collapses led to the suspicion of airway problems.

Suzumura et al (2000) attributed infection as a risk factor for early subglottic stenosis. If infection occurs within 14 days of intubation, subglottic stenosis can occur early than infection occurring later than 14 days. The endotracheal tube injures the mucosa and causes edema and hyperaemia. Pressure necrosis occurs with initial soft stenosis followed by cicatrical stenosis. Careful monitoring after extubation would help in early identification of airway problem. Direct laryngoscopy and bronchoscopy is the gold standard for diagnosis. Though conservative management is the first line, surgical and laser interventions are needed in some severe cases.

As prevention is better than cure, minimising injury to the airway by non invasive ventilation, proper fixation of tube, aseptic routines, avoiding traumatic intubations will reduce the incidence of subglottic stenosis and improve the outcome. Conclusion

Though rare, unusual presentation like persistent lung collapses after extubation even without stridor should be evaluated for airway abnormalities.

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