A case of failed intubation

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A term male infant weighing 2350 g was born to a gravida three mother by an emergency caesarean section for non-progress of labour. The antenatal period was supervised and uneventful. The baby did not cry at birth and was resuscitated with a bag and mask. Since only partial improvement occurred, the baby was intubated and shifted to the neonatal intensive care unit for ventilatory support. On the ventilator, pulse oximetry showed persistently low saturation. Examination revealed respiratory distress and inadequate chest expansion on positive pressure ventilation. Auscultation revealed equal air entry over both the lung fields as well as in the epigastrium. Spontaneous extubation was suspected and reintubation tried. Repeated attempts, however, failed to negotiate even the smallest diameter (2.5 mm) endotracheal tube beyond the vocal cords, though no obvious anomalies of laryngeal anatomy were seen.

Questions
(1) What was the cause of failed intubation?
(2) How was the ventilation carried out?
(3) What other clinical signs you would look for on examination?
Answers

QUESTION 1
The cause of failed intubation was tracheal agenesis, diagnosis of which is suggested by (1) absence of cry at birth; (2) respiratory distress and cyanosis; and (3) failure to negotiate endotracheal tube beyond the vocal cords in the presence of normal looking laryngeal anatomy (see box 1).2

QUESTION 2
The patient had an inadvertent oesophageal intubation and ventilation was carried out through a tracheo-oesophageal fistula.

QUESTION 3
Other clinical signs are:
(A) Failure to palpate the trachea in the suprasternal space.
(B) Distension of the stomach on positive pressure ventilation.
(C) Auscultation of air entry over bilateral lung fields as well as in the epigastrium.

Discussion
Over vigorous attempts resulted in a false passage and surgical emphysema in the neck. Palpation in the suprasternal space failed to locate the trachea. A quick look for other congenital anomalies revealed the presence of imperforate anus, hypospadias, low set ears with malformed left pinna, epicantlic folds, and hypertelorism. Cardiac examination was unremarkable. The baby died from progressive hypoxaemia at 18 hours of life. Autopsy revealed tracheal agenesis with tracheo-oesophageal fistula.

Tracheal agenesis is a rare congenital anomaly occurring with an estimated frequency of two per 100,000 live births.1 Payne first reported it in 1900, and so far about 100 cases have been described.1,4,5 Floyd et al classified tracheal agenesis into three types.6 In type I, only short segment of the distal trachea is present. It arises from the oesophagus and then divides into the main stem bronchi. Type II is characterised by a completely absent trachea but both the main bronchi join in the midline. In type III, the trachea is again completely absent as in type II but both the bronchi rise separately from the oesophagus. The relative frequency of these types, which correspond to the increasing severity of the defect, is 13%, 62%, and 25% respectively.1 A tracheo-oesophageal or broncho-oesophageal fistula is almost always there.1,2 That our patient had type I tracheal agenesis with a tracheo-oesophageal fistula was confirmed on autopsy.

Tracheal agenesis is associated with other congenital anomalies in the majority of cases.1,4,5 These include other anomalies of the respiratory tract, particularly of the larynx and lungs, complex cardiac malformations, genitourinary as well as gastrointestinal tract anomalies. The link between tracheal agenesis and the VATER/VACTERL association remains unresolved.1,4,7,8

Clinical presentation of tracheal agenesis is quite characteristic. At birth, the diagnosis is suggested by (1) absence of cry; (2) respiratory distress and cyanosis; (3) failure to negotiate endotracheal tube beyond the vocal cords in the presence of normal looking laryngeal anatomy; and (4) partially effective ventilation with bag and mask or through an oesophageal tube.1,2 An additional clinical sign, which has been inadequately stressed in the literature, is the failure to palpate the trachea in the suprasternal space, as was observed in our case. This sign may be falsely present in conditions of extreme deviation of the trachea to one side secondary to conditions like pneumothorax, congenital diaphragmatic hernia, pleural effusion, or intrathoracic mass lesions. In none of these conditions is tracheal intubation difficult.

The diagnosis of tracheal agenesis can be quite challenging. The oesophageal intubation is often inadvertent and in such cases ventilation through a broncho-oesophageal fistula brings about clinical improvement. The diagnosis of tracheal agenesis may therefore be overlooked, at least, initially.1,2 In such cases, distension of stomach and auscultation of air entry over both lung fields as well as in the epigastrium should arouse the suspicion of a misplaced endotracheal tube. Presence of associated anomalies should heighten the suspicion of respiratory tract anomalies especially when respiratory distress and difficult intubation are present.11 Although the focus in emergency situations is for resuscitation, a general examination of the patient may be very helpful to establish the diagnosis. In the present case, multiple congenital anomalies were present but were overlooked because of the preoccupation with resuscitation.

Once the diagnosis of tracheal agenesis is suspected laryngoscopy, bronchoscopy, or oesophagoscopy provide visual confirmation of the defect as well as help localise the fistula between the residual airway and oesophagus.1,2 Further information can be obtained by a contrast oesophagogram.1,2 The role of surgical neck exploration is controversial.1 The management of neonates with tracheal agenesis is very difficult and frustrating. Various surgical procedures have provided only temporary solutions. The majority of the patients died within a few hours to a few days.1,2

Final diagnosis
Tracheal agenesis.

Box 1: Tracheal agenesis should be suspected in any newborn who presents with the following features:
- Absence of cry at birth.
- Respiratory distress and cyanosis.
- Failure to negotiate endotracheal tube beyond the vocal cords in the presence of normal looking laryngeal anatomy.
- Partially effective ventilation with bag and mask or through an oesophageal tube.

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