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Laryngeal Web in an Infant with Tracheoesophageal Fistula

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Infants who need surgical repair of a tracheoesophageal fistula frequently have other congenital abnormalities. Some of these abnormalities may be obvious, while others are suspected only when unexpected problems arise during anesthesia and operation. The following case report is an example of the second type.

REPORT OF A CASE

A 2-day-old, 3.3-kg infant girl was admitted to the Montreal Children’s Hospital for repair of a Type I tracheoesophageal fistula. At birth the infant had had an Apgar score of 5, which had improved to 7 and then to 9 after oxygen administration and pharyngeal suctioning. She was in no apparent respiratory distress on admission. Roentgenogram of the chest showed minimal pneumonic streaking of the right upper lobe of the lung. There was no obvious congenital abnormality.

In the operating room, atropine, 0.05 mg, was given intravenously. The upper esophageal pouch was suctioned and the patient was given pure oxygen by mask in preparation for awake intubation. Laryngoscopy using the infant-sized Miller blade was easy. A size 14 Cole tube was readily inserted through the glottis, the infant breathing spontaneously throughout. However, on advancing the tube further, the infant showed severe substernal retraction and no entry of air into the lungs was heard on auscultation. Gentle manual positive-pressure ventilation was attempted, without success. Examination showed no evidence of gastric distention, thus eliminating the possibility of having intubated the fistula. The tube was withdrawn, and pure oxygen by mask was reapplied, with dramatic improvement of oxygenation and airway.

Intubation was reattempted using progressively smaller tubes, but the same problem recurred. It was noted that when the tube was just past the glottis, the airway was satisfactory. But sliding the tube as little as 5 mm past the glottis caused obstruction. That level was well above the level of the fistula, which was just above the carina as outlined radiologically—and thus intubation of the fistula was rather unlikely. Some form of subglottic narrowing was suspected. After repeated attempts, a size 10 Cole tube was successfully passed past the obstruction, secured in place, and the fistula repaired through a right thoracotomy incision. The anesthetic was nitrous oxide—halothane with controlled ventilation. No muscle relaxant was used.

At the end of the operation, the infant was awake and breathing spontaneously. It was then decided that since the airway had been adequate prior to operation, the trachea should be extubated and the infant watched carefully. Steroids were given to try to prevent laryngeal edema that might result from the repeated attempts at intubation.

Following extubation, the infant did very well initially. However, during the next 12 hours she developed retractions, and became tachypneic, and eventually hypercarbic ($P_{aCO_2}$ 66 torr). The trachea was reintubated with a size 10 Cole tube without difficulty. This restored the airway and improved ventilation ($P_{aCO_2}$ 40 torr). Three days later the trachea was again extubated. There was good entry of air, but severe subcostal and suprasternal retractions developed. The infant was given a trial of racemic epinephrine inhalation to reverse any superimposed laryngeal edema. The initial response was good, but she became progressively tired, and a roentgenogram of the chest showed collapse of the left lower lobe. It was decided to perform a tracheostomy and examination of the larynx. After oxygenation and topical spray of the larynx with lidocaine, 1 per cent, an infant-sized anterior commissure laryngoscope was used to open the moderately hyperemic, but otherwise normal, vocal cords. A web-like structure of firm fibrous tissue was seen lying immediately below the anterior commissure. The web appeared almost triangular in shape, with the anterior end under the anterior commissure, and somewhat grooved as a result of prior intubation. A gastrostomy was also performed because of a continuous leak through the chest tube draining the repair site. The patient did very well for two days following the tracheostomy, but on the third day she suffered a spontaneous left tension pneumothorax, resulting in cardiorespiratory arrest. The infant died following unsuccessful...
ful attempts at resuscitation. Postmortem examination of the larynx confirmed the presence of a fibrotic congenital web over the anterior surface of the cricoid cartilage (fig. 1).

**DISCUSSION**

Congenital anomalies of the larynx associated with the tracheoesophageal fistula are rare. However, they are of special interest, both from the developmental point of view and in relation to airway management and possible intubation during anesthesia and resuscitation.  

Embryologically, the larynx is formed between the fourth and tenth weeks of gestation. The upper portion arises as a ventral growth from the pharyngeal floor, formed mainly by the fifth branchial arch, while that part from the vocal folds caudally forms around the stem of the trachea.  

Like many other embryologic lumens, the laryngeal cavity becomes obliterated by epithelial ingrowth during development. At the tenth week of gestation this epithelial union is dissolved, and the lumen is re-established. Incomplete recanalization of the primitive laryngeal airway results in web formation. The incidence of laryngeal webs in general has been estimated at approximately one in 10,000 births. Most webs are thin bands extending across the anterior margins of the vocal cords, i.e., glottic in location (75 per cent of cases). Only 15 per cent are subglottic. Symptoms will depend upon the extent of the web, and are usually present at birth. Frequently the infant has a hoarse or weak cry, or a complete absence of sound with crying effort. Respiratory distress may be the first sign. The definitive diagnosis usually depends on direct laryngoscopy, although a lateral roentgenogram of the neck may be very suggestive. Treatment varies from a lifesaving perforation of an obstructing web to incision or dilatation of partial webs. Tracheostomy may be necessary until the laryngeal airway is adequate.

**REFERENCES**