infants at risk for VAE. Patients without VAE had twice the blood loss as those with VAE.

In summary, 66% of infants undergoing craniectomy in the supine position had echocardiographic evidence of embolism compatible with VAE. Only one child became symptomatic, and she responded promptly to iv atropine. No source for the air was identified in any child, and all children left the operating room in good condition. One child with tricuspid atresia had an interseptal defect, and no child had evidence of left-sided air. ECHO appears to be an extremely sensitive, but relatively nonspecific, monitor for VAE. Some of this lack of specificity was minimized by communication between echocardiographer and anesthesiologist. In spite of its shortcomings, ECHO offers valuable information about paradoxical embolism in infants with known or suspected interseptal defects.

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Ligneous Tracheobronchitis: An Unusual Cause of Airway Obstruction

MICHAEL F. BABCOCK, M.D.,* ROBERT F. BEDFORD, M.D.,† FREDERIC A. BERRY, M.D.‡

We recently were involved in the care of a child diagnosed with ligneous conjunctivitis, a rare illness of unknown etiology. The disease consists of growth of conjunctival membranes having a fibrous or woody consistency, and has a variable course often requiring repeated surgical stripping of the membranes. Coexisting respiratory problems, especially frequent pneumonia in children, have often been noted. Our patient suffered from progressive airway obstruction associated with recurrent tracheobronchial growth of ligneous membranes. She required multiple anesthetics for stripping of both the conjunctival and tracheobronchial membranes. Management was frequently complicated by laryngospasm, bronchospasm, and tracheobronchial obstruction. Obstruction of distal airways may have occurred late in the course of her illness.

* Resident in Anesthesiology.
† Professor of Anesthesiology and Neurological Surgery.
‡ Professor of Anesthesiology and Pediatrics.

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Address reprint requests to Dr. Berry: Department of Anesthesiology, University of Virginia Medical Center, Charlottesville, Virginia 22908.

Key words: Airway: ligneous tracheobronchitis; obstruction.

Case Report

The patient was a female infant with a history of normal gestation, term delivery at 3.2 kg, and uneventful perinatal period. Hydrocephalus developed at the age of 1 month, necessitating ventriculo-peritoneal (VP) shunt without associated anesthetic complication. At 7 months, thick membranes covered the palpebral conjunctiva of all four eyelids, and a diagnosis of ligneous conjunctivitis was made. Pathological analysis of the membranes demonstrated typical findings of ligneous conjunctivitis, including acute and chronic inflammation, edema, and mild pseudo-epitheliomatous hyperplasia.[3] Various topical ophthalmic solutions, including antibiotics, cromolyn sodium, chymotrypsin, and hyaluronidase, as well as cryotherapy and electrocautery, were employed to arrest the growth of the conjunctival membranes. None was satisfactory.

The first sign of respiratory involvement occurred after a VP shunt revision, at the age of 28 months, when the patient developed postoperative stridor thought to be associated with endotracheal intubation. This resolved overnight after a single postoperative treatment with nebulized epinephrine. At age 29 months, the child developed acute respiratory distress; physical exam revealed inspiratory stridor. Anesthesia for bronchoscopy was induced *via* a mask in the sitting position with halothane in oxygen, and was complicated by coughing, laryngospasm, and bronchospasm. Ventilation improved with the application of positive airway pressure and by increasing anesthetic depth. Rigid bronchoscopy with a 3.5-mm Storz telescope revealed a subglottic circumferential tracheobronchial cast, which had decreased the tracheal diameter to approximately 2 mm. This was excised and found similar in consistency to the conjunctival membranes. Postoperative examination revealed stridor, wheezing, and retractions, but the child improved over several days with administration of iv steroids and nebulized bronchodilators. This acute response to bronchodilator therapy led to its use in the management of the child's chronic respiratory illness. Pathological analysis of the tracheal membranes revealed acute and chronic inflammatory changes, with fibrinous exudation similar to that seen in the conjunctival membranes.

Periodic episodes of acute respiratory distress continued over the following 10 months, and were associated with regrowth of conjunctival and tracheal membranes. Physical examination usually revealed some combination of inspiratory stridor, wheezing, and retractions. Frequent stripping of conjunctival membranes, alone or in combination with tracheobronchial membranes, was necessary; the patient had a total of 19 anesthetics. Induction of anesthesia was marked by laryngospasm and bronchospasm. This apparent reactive airway disease typically improved upon increasing anesthetic depth. Occasionally, this required iv succinylcholine administration. As the tracheobronchial disease progressed, stridor, chest retraction, and air trapping became more prominent during anesthesia. This airway obstruction necessitated bronchoscopic removal of the ligneous membranes.

Despite temporary improvement following bronchoscopic stripping of the tracheobronchial membranes, and despite continued treatment with bronchodilators, antibiotics, and aggressive pulmonary toilet, the patient's respiratory status gradually declined. At age 39 months, she became ventilator-dependent, requiring high airway pressures and, eventually, tracheostomy. The clinical picture became similar to respiratory distress syndrome; respiratory compliance and oxygenation deteriorated in spite of maximal ventilatory therapy. Distal airway disease was suspected. Hypoxia resulted in brain injury and death at age 40 months.

Discussion

Ligneous conjunctivitis is a rare disease with variable course.[1,2] Its name derives from the peculiar woody consistency of the conjunctival membranes. Pathogenesis of the disease remains obscure, although an immune hypersensitivity mechanism has been proposed.[3] Cases with familial occurrence have suggested a genetically transmitted component.[2] Analysis of the conjunctival membranes reveals replacement of conjunctival stroma with granulation tissue and a characteristic thick hyaline plaque composed of acid mucopolysaccharide. Deposits of this plaque are commonly found surrounding newly formed blood vessels.[3]

Onset typically occurs during childhood, and involvement of other mucous membranes has been noted. There is also an association with hydrocephalus,[2] as in our patient. Although remission occurs, an indolent course with occasional exacerbation is typical. Previous reports of ligneous conjunctivitis are confined mainly to the ophthalmologic literature. Firat reported nine pediatric cases of ligneous conjunctivitis, of which eight had respiratory disease, including one fatal case.[1] The cause of death in that patient, a 2-yr-old child, was reported as bronchopneumonia. Whether the patient had tracheobronchial growth of ligneous membranes was not reported.

Treatment aimed at slowing regrowth of conjunctival membranes has shown some success with topical enzymatic agents, such as hyaluronidase.[4] The number of patients with severe airway involvement has been small, and therapeutic measures beyond bronchoscopic airway stripping and dilatation have not been reported. Cooper *et al.* published a report of ligneous conjunctivitis with subglottic stenosis in an infant who required tracheostomy.[5] This patient had repeated bronchoscopic procedures for airway dilatation and removal of granulomatous tissue. The disease eventually went into remission. Whether their patient developed ligneous tracheobronchitis is not clear; however, their description closely matches the pattern of illness seen in our patient.

The present case demonstrates that tracheobronchitis associated with growth of ligneous membranes in the airway can occur in patients having ligneous conjunctivitis. Anesthetic management was complicated by reactive airway disease, as well as by tracheobronchial obstruction. Stormy anesthetic inductions were marked by coughing, laryngospasm, and bronchospasm. Improvement with deepening of anesthesia and with bronchodilator therapy suggested the presence of reactive airway disease. Distal airway involvement likely occurred as a late complication. This cannot be confirmed, since post-mortem examination was not obtained.

On occasion, conjunctival membrane stripping alone was required. For these procedures, we elected to give general anesthesia *via* mask, since endotracheal intuba-

tion might result in dislodgement of tracheal membranes leading to airway obstruction. Furthermore, an endotracheal tube could aggravate existing edema and inflammation. Anesthesia *via* mask should be considered for these patients when general anesthesia is necessary. Regardless of the choice of airway management, the availability of a skilled bronchoscopist for diagnosis and therapy of airway obstruction is advised.

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Intraoperative Re-expansion Pulmonary Edema

DAWN P. DESIDERIO, M.D.,* MICHAEL MEISTER, M.D.,† ROBERT F. BEDFORD, M.D.‡

Re-expansion pulmonary edema has been reported following evacuation of both chronic pneumothorax[1,2] and pleural effusion.[3] It is an uncommon, but potentially life-threatening, condition which has not been described in the perioperative period. We describe a case of intraoperative pulmonary edema that occurred following rapid re-expansion of a lung that had been chronically collapsed due to a malignant pleural effusion.

Case Report

A 60-yr-old, 88.5-kg, 177-cm man presented with a 6-month history of dyspnea due to a large right pleural effusion caused by a previously diagnosed pleural mesothelioma. The patient's past history was significant for hypertension, a 40 pack-year history of smoking, and a 12-yr exposure to silica. The patient was taking no medications. Preoperative pulmonary function studies revealed restrictive disease with a vital capacity of 1640 ml (45% of predicted), and obstructive disease with a 1-s forced expiratory volume of 1310 ml. Ventilation-perfusion studies revealed a matched defect in ventilation and perfusion involving the entire right lung field both in the anterior and posterior views. Echocardiography showed normal left and right heart chambers with no evidence of pericardial effusion. Preoperative arterial blood gas analysis demonstrated a respiratory alkalosis with a *p*Ha of 7.50, Pa_{CO_2} = 31 mmHg, and a Pa_{O_2} = 79 mmHg. The patient received digitalis preoperatively and iv aminophyline.

After breathing 100% oxygen, anesthesia was induced with thiopental, 250 mg iv, and fentanyl, 100 μg iv. Succinylcholine, 100 mg iv, was given to facilitate endotracheal intubation, and anesthesia was maintained with 1% isoflurane, 50% N_2O in O_2, and incremental doses of fentanyl, 50 μg IV. The patient was placed in the left lateral decubitus position and, shortly after surgery began, 3,200 ml of pleural effusion was rapidly removed through a right thoracotomy incision. The patient underwent a right sub-total pleurectomy with re-expansion of the right lung, implantation of I^{125} needles, and a partial pericardiectomy with marlex mesh reconstruction.

The patient's intraoperative course was stable initially, with the Pa_{O_2} ranging from 100–181 mmHg during controlled ventilation with a FI_{O_2} of 0.5. The central venous pressure ranged between 10–12 cm H_2O, and the urine output was 90 ml per hour. Three hours after the evacuation of the pleural effusion, and while the I^{125} implants were being placed, straw-colored, slightly hemorrhagic fluid was noted to be exiting from the endotracheal tube. Despite frequent endotracheal suctioning, the anesthetic circuit had to be changed four times over the next 1–2 h due to the volume of fluid coming from the endotracheal tube. Chemical analysis of the fluid revealed a protein value of 4.0 g/dl and an albumin of 2.6 g/dl, values consistent with pulmonary edema fluid. Pa_{O_2} was 229 mmHg, while being ventilated with 100% oxygen during this time.

Fiberoptic bronchoscopy was conducted while the patient was in the left lateral decubitus position. There was no sign of airway obstruction,

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* Assistant Clinical Anesthesiologist, Memorial Hospital; Assistant Clinical Professor of Anesthesiology, Cornell University Medical College.

† Resident in Anesthesiology, Cornell University Medical College.

‡ Chairman, Department of Anesthesiology and CCM, Memorial Hospital; Professor of Anesthesiology, Cornell University Medical College.

Address reprint requests to Dr. Desiderio: Department of Anesthesiology and Critical Care Medicine, Memorial Sloan-Kettering Cancer Center, 1275 York Avenue, New York, New York 10021.

Key words: Anesthesia: thoracic. Complications: pulmonary edema. Lung: edema.