

## An Unusual Airway Complication with Sarcoidosis

MICHAEL H. WILLS, M.D.,\* MARK M. HARRIS, M.D.†

Sarcoidosis is a multisystem disease perhaps best known for its pulmonary involvement. Anesthetic considerations include both parenchymal and airway disease. We report a case of an often overlooked manifestation of this disorder, laryngeal sarcoid, which resulted in an unexpected and difficult airway management problem, and, also, in appreciable postoperative morbidity 36 h postoperatively.

## REPORT OF A CASE

A 37-yr-old woman with chronic granulomatous dacryocystitis presented for lacrimal gland incision and drainage, and placement of a lacrimal duct tube. Significant past medical history included hiatal hernia with reflux, insulin dependent diabetes mellitus, and sarcoidosis. She had received no steroids since 1983, and was a nonsmoker. She jogged regularly and had no respiratory symptoms.

Physical examination revealed a mildly obese, 76 kg female with a quiet voice, breathing comfortably. She was edentulous with adequate airway to external examination. The lungs were clear to auscultation and percussion, and cardiac examination was normal. A chest roentgenogram obtained 1 month prior to the operation revealed significant perihilar adenopathy and a reticular nodular parenchymal infiltrate that was not changed from previous films.

While she was breathing oxygen, anesthesia was induced with thiopental, 300 mg, and succinylcholine, 100 mg iv. After waiting 30 s, laryngoscopy was performed. However, the cords "appeared closed." One minute later, after a further dose of thiopental, a 7 mm endotracheal tube was inserted through the cords with a twisting motion. The endotracheal tube met resistance until the balloon passed the region of the cords. Anesthesia was maintained with isoflurane and N<sub>2</sub>O/O<sub>2</sub>. The trachea was extubated at the end of the procedure with no immediate respiratory difficulty. She was without respiratory problems at the time of discharge to the floor from the recovery room 2 h later.

Thirty-six hours after the operation, the patient had increasing respiratory difficulty. She complained of increasing air hunger and had a barking, honking cough, a barely audible voice, and marked inspiratory and expiratory stridor on physical examination. The lung fields were clear to auscultation, and moderate supraclavicular retractions were visible. Emergency evaluation with fiberoptic laryngoscopy revealed edema of the laryngeal structures and reduced movement of the cords. This was considered to be compatible with granulomatous involvement, and she was treated with dexamethasone and racemic epinephrine with marked resolution of symptoms over the next 48 h.

Further review of the old records revealed an evaluation for hoarse-

ness in March, 1983, which included flexible fiberoptic bronchoscopy. Findings included slight vocal cord edema without nodules or paralysis. Several white raised nodular lesions were noted in the proximal trachea. Biopsies of the lesions showed noncaseating granulomata. Brushings and cultures were negative. Previous biopsies of skin, conjunctival follicle, and scalene node in 1983 had also demonstrated noncaseating granulomata. Thus, the clinical picture was considered to be compatible with laryngeal sarcoidosis.

## DISCUSSION

Sarcoidosis is a chronic granulomatous disease with protean manifestations involving almost any organ system, including the lungs, nasopharynx, tongue, oral cavity, larynx, trachea, and esophagus. The disease occurs most commonly in blacks and females between the ages of 20 and 40 yr. Kirshner and Hollinger<sup>1</sup> report a case of generalized sarcoidosis including involvement of the larynx in a 14-yr-old boy, although middle-aged adults are the usual patients. While pulmonary involvement is well-known to the anesthesiologist, few appreciate that laryngeal involvement may occur in 1-5% of affected patients.<sup>2-4</sup>

Laryngeal sarcoid was first described by Poe<sup>5</sup> in 1940. The seriousness of the disease was emphasized in a review by Devine<sup>6</sup> in 1965 that included eight patients who developed airway obstruction, five of whom required tracheostomies. He also mentions that a few patients were felt to have died from laryngeal sarcoid. The otolaryngology literature notes the potentially life-threatening nature of progressive sarcoid of the larynx.<sup>2-7</sup>

Typically, laryngeal sarcoid is associated with systemic disease, although it may be an isolated finding.<sup>7</sup> Symptoms and signs of laryngeal involvement include hoarseness, dysphagia, dyspnea, stridor, weak voice, sensation of a lump in the throat, and throat pain.<sup>3</sup> The diagnosis of laryngeal sarcoid involves visualization of the laryngeal structures by direct or indirect laryngoscopy. Several clinical appearances may be evident with both generalized or local involvement. Various structures, including the epiglottis, aryepiglottic folds, ventricles, and arytenoids, may have a nodular or smooth character. Exophytic masses have been described.<sup>1,6</sup> There is a predilection for the supraglottic areas, with only rare involvement of the vocal cords. Supraglottic obstruction from granulomas has been reported by Neel and McDonald<sup>9</sup> in two of 13 patients. Ulceration is rare, with the pathologic process involving mainly the submucosa.

Treatment of laryngeal sarcoid depends on the degree

\* Resident in Anesthesiology.

† Assistant Professor of Anesthesiology and Neurosurgery.

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Address reprint requests to Dr. Wills: Department of Anesthesiology, University of Virginia Medical Center, Box 238, Charlottesville, Virginia 22908.

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of obstruction and symptomatology, but most authorities seem to agree to early intervention. The disease has a tendency to spontaneous regression, which complicates the evaluation of the efficacy of treatment. Systemic steroids are used frequently; however, the response may be disappointing. Other treatments have included local injections of depot steroids in the lesion<sup>8</sup> with the risk of worsening the obstruction. Chloroquine-like drugs have been tried,<sup>9</sup> as well as radiation therapy.<sup>10</sup> Surgical approaches have included tracheostomy and the surgical removal of the obstructing lesion.

As this case demonstrates, the anesthesiologist must remain aware of the possibilities of laryngeal involvement in sarcoidosis. We conclude that instrumentation during endotracheal intubation increases the risk of obstructive respiratory arrest. Suspicion of the disorder requires a preoperative evaluation, including laryngoscopy and possible roentgenographic studies of the neck, such as tomography. A flow volume loop, or FEV<sub>1</sub>, may also be useful preoperatively for assessment of airway obstruction. A small endotracheal tube<sup>11</sup> and prophylactic dexamethasone may minimize airway problems. Careful postoperative observation for up to 36 h may be needed to avoid

life-threatening postoperative airway obstruction and respiratory arrest.

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## Submucosal Epiglottic Emphysema Complicating Bronchial Rupture

JOHN J. SACCO, M.D.,\* DOUGLAS W. HALLIDAY, M.D.†

Rupture of the tracheobronchial tree is a relatively rare event when compared to other intrathoracic injuries resulting from blunt trauma. The true incidence of this lesion is difficult to establish, because many patients die from this injury prior to reaching the hospital, or diagnostic failure occurs antecedent to an early hospital death.<sup>1</sup> Curiously, children seem particularly liable to suffer from this injury.<sup>2</sup>

The acute clinical manifestations of this disorder are varied, and include: mediastinal, suprasternal and sub-

cutaneous emphysema, Hamman's sign (percardial crackling sounds synchronous with the cardiac cycle in the presence of mediastinal air), hemoptysis, pneumothorax, tension pneumothorax, atelectasis, cyanosis, respiratory distress, shock, pain on swallowing, hoarseness, airway obstruction and suffocation from disruption, concomitant esophageal and cervical spine injuries, and, very rarely, hemorrhage from major pulmonary vessels.<sup>3</sup>

We describe a pediatric patient who sustained a bronchial tree disruption and presented with some of the above signs and symptoms. Additionally, we believe her presentation is unique, in that submucosal epiglottic emphysema compounded her injuries.

#### CASE PRESENTATION

A previously healthy 7-yr-old 24 kg, female patient was admitted to our Emergency Unit approximately 40 min after falling across the handlebar of her bicycle. She sustained a contusion on the anterior neck at the level of the cervical trachea. This was further complicated

\* Attending Anesthesiologist, St. Joseph's Hospital.

† Assistant Clinical Professor, SUNY Health Science Center.

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Address reprint requests to Dr. Sacco: St. Joseph's Hospital Health Center, 301 Prospect Avenue, Syracuse, New York 13203.

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