we may be missing cases that were submitted as claims alleging another adverse event but were, in fact, anaphylaxis. Nevertheless, this review represents the first systematic approach in analyzing the VICP experience with anaphylaxis and will be useful when the VIT is updated.

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1. National Vaccine Injury Compensation Program, Vaccine Injury Table. http://www.hrsa.gov/vaccinecompensation/table.htm. Accessed October 12, 2010.
2. Somsak JG, Johann-Liang R. Charge to the Committee. http://www8.nationalacademies.org/cp/meetingview.aspx?MeetingID=3398&MeetingNo=1. Accessed October 12, 2010.
3. Ruggeberg JU, Gold MS, Bayas JM, et al. Anaphylaxis: case definition and guidelines for data collection, analysis, and presentation of immunization safety data. *Vaccine.* 2007;25:5675–5684.
4. National Vaccine Injury Compensation Program, Statistics Reports. http://www.hrsa.gov/vaccinecompensation/statistics_report.htm. Accessed October 12, 2010.
5. Payne V, Kam PC. Mast cell tryptase: a review of its physiology and clinical significance. *Anaesthesia.* 2004;59:695–703.
6. Pool V, Braun MM, Kelso JM, et al. Prevalence of anti-gelatin IGE antibodies in people with anaphylaxis after measles-mumps rubella vaccine in the United States. *Pediatrics.* 2002;110:e71.
7. Bohlke K, Davie RL, Marcy SM, et al. Risk of anaphylaxis after vaccination of children and adolescents. *Pediatrics.* 2003;112:815–820.

RESOLUTION OF BRONCHOMALACIA PRESENTING AS SEVERE ASTHMA BY ENDOSCOPIC INTERVENTION

Difficult to control asthma should make us think of other concomitant airway diseases, mainly if the treatment for the initial presumed diagnosis is failing. A 42-year-old female smoker was referred to our allergy department because of symptoms of urticaria and facial and eyelid edema in the month before her visit. A detailed allergy history also revealed intermittent cough and dyspnea with no apparent trigger or seasonal changes. The patient had no known allergies or atopic diseases. A physical examination revealed diffuse wheezing in both lungs. The results of a test for dermographism were negative. Results of skin prick tests with a panel of food and aeroallergens were negative. Spirometry showed a severe obstruction of airway flow (forced vital capacity [FVC], 3.73 L [91% predicted]; forced expiratory volume in 1 second [FEV_1], 1.77 L [50% predicted]; and FEV_1/FVC ratio, 47%) with a positive bronchodilator test result (FEV_1, +24.5%) and a fraction of exhaled nitric oxide of 33 ppb. A chest radiograph showed air trapping, and the results of a blood test (immunoglobulins, α_1-antiptripsin, hemogram, and biochemistry) were normal. The patient was diagnosed a having acute idiopathic urticaria and severe persistent nonallergic asthma. We began therapy with salmeterol and fluticasone, 50 to 500 μg. After 1 month the patient experienced partial clinical improvement, although with persisting dyspnea and expiratory wheezing, leading oral steroid treatment to be ruled out. Spirometry did not show improvement. We performed plethysmography and a diffusion test, the results of both of which were normal. Induced sputum analysis showed Curschmann spirals and leukocytes (99% neutrophils, 1% eosinophils).

A chest computed tomogram with contrast showed mild emphysema in the upper lobes of the lungs. Respiratory infections persisted for the following 2 months, and some exacerbation required care from the emergency department. Spirometries showed no improvement. Because of a lack of improvement, a bronchoscopy was performed.[1]

Bronchoscopy revealed a bronchial collapsibility, with significant loss of airway caliber in the left main bronchus, resulting in severe bronchomalacia. Days later, the bronchoscopist placed a Dumon prosthesis in the left main bronchus using an Efer-Dumon rigid bronchoscope (Fig 1).

In subsequent monthly visits, the patient reported good tolerance to the prosthesis and demonstrated clinical improvement. Spirometry also revealed a clear improvement (FVC, 4.07 L; FEV_1, 2.04 L; FEV_1/FVC ratio, 65%), and a bronchodilator test result was negative.

Tracheobronchomalacia is a central airway disease characterized by an increased susceptibility of the tracheal and bronchial lumen to collapse during expiration.[2] Occurring rarely, broncho-

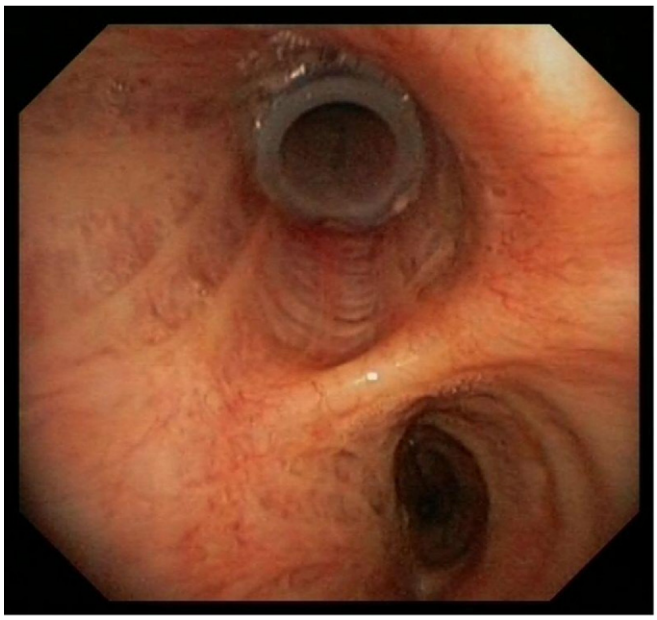

Figure 1. Placement of the prosthesis in the left bronchus.

Disclosures: Dr Sastre reports having served as a consultant to Phadia, Schering-Plough, and GSK, having been paid lecture fees by Novartis, GSK, Stallergenes, and UCB, and having received grant support from Phadia, GSK, and ALK-Abello.

doi:10.1016/j.anai.2011.01.019

malacia is often mistaken for other more common obstructive airway disorders. This disorder is encountered in both pediatric and adult medicine. Its prevalence in adults is unknown because the available data are derived from studies conducted in selected populations and not in the general population,[3] but it can be a more common finding in patients with chronic obstructive pulmonary disease and asthma.[4]

In our case, flow-volume loops did not help to establish the diagnosis because typical findings, such as a rapid reduction in maximum expiratory flow after the initial peak, notching of the expiratory descent, and oscillations in the curve, were shown, likely because the trachea was not affected enough to induce a collapse that can be detected in flow-volume curves.[5] The computed tomogram was not performed in inspiration and expiration; therefore, it was not possible to evaluate the reduction in the cross-sectional area from the bronchi. The bronchoscopy is the "gold standard" technique that allows a definitive diagnosis.[2]

In conclusion, we describe a patient with severe nonallergic asthma whose outcome was poor despite the prescribed treatment. An endoscopic examination revealed severe bronchomalacia. Placing a Dumon prosthesis on the more affected bronchus resulted in clear clinical and spirometric improvement. Bronchial endoscopy should be considered in cases of noncontrolled severe asthma to rule out endobronchial diseases.

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1. Murg SD, Colt HG. Interventional bronchoscopy from bench to bedside: new techniques for central and peripheral airway obstruction. *Clin Chest Med.* 2010;3:101–115.
2. Majid A, Fernández L, Fernández-Bussy S, Herth F, Ernst A. Tracheobronchomalacia. *Arch Bronconeumol.* 2010;46:196–202.
3. Baraldi E, Donegá S, Carraro S, Farina M, Barbato A, Cutrone C. Tracheobronchomalacia in wheezing young children poorly responsive to asthma therapy. *Allergy.* 2010;6:1064–1065.
4. Murgu SD, Colt HG. Tracheobronchomalacia and excessive dynamic airway collapse. *Respirology.* 2006;1:388–406.
5. Loring SH, O Donnell CR, Feller-Kopman DJ, Ernst A. Central airway mechanics and flow limitation in acquired tracheobronchomalacia. *Chest.* 2007;131:1118–1124.

EOSINOPHILIC FASCIITIS MIMICKING ANGIOEDEMA AND TREATMENT RESPONSE TO INFLIXIMAB IN A PEDIATRIC PATIENT

Eosinophilic fasciitis (EF) is a rare scleroderma-like condition that was first described by Shulman in 1974 and characterized by eosinophilic inflammatory fascial infiltrate. The origin of EF remains unclear, and there have been only approximately 200 cases reported to date. Although the onset of symptoms has been linked with severe exercise, use of medication, trauma, and insect bites, no singular inciting event or trigger has been identified. Symptoms characteristically include progressive and symmetric erythema, edema, stiffness, painful extremities, and skin induration. The face, hands, and feet are usually spared. EF is associated with peripheral blood eosinophilia (eosinophil count $>500//\mu L$) in 65% to 80% of patients.[1,2] Hypergammaglobulinemia and elevated acute-phase reactants are also commonly seen.[1,2] Because the mean age of onset is 40 to 50 years,[1,2] EF is extremely uncommon in the pediatric population.

Full-thickness (skin, fascia, and muscle) biopsy is the "gold standard" test to diagnose EF and shows inflammation and thickening of collagen bundles in muscle fascia with infiltration of lymphocytes, plasma cells, and eosinophils.[1,2] Magnetic resonance imaging (MRI) is also a useful tool to diagnose and follow up EF. It detects fascia thickening and shows high signal intensity on T2-weighted sequence of muscle fascia while muscle pattern is normal.[3] Most patients respond to moderate- or high-dose corticosteroids. Methotrexate, azathioprine, cyclophosphamide, and infliximab have been used in corticosteroid-resistant patients.[1,2,4,5]

A 5-year-old, previously healthy boy developed acute, diffuse edema and erythema of the face, abdomen, and extremities, including the hands and feet. The swelling and erythema were painless. There was no precipitating trauma or history of exposure to new foods or medications. His parents reported that he was playing outdoors just before the episode began. On the basis of his symptoms and physical examination results, he was initially diagnosed as having idiopathic angioedema and treated with antihistamines and oral steroids. When the symptoms reoccurred 2 weeks later, further evaluation was conducted. Family history was significant for a first cousin with EF. The patient's white blood cell count was $12{,}600/\mu L$, with 51% eosinophils (absolute eosinophil count of $6426//\mu L$). The eosinophilia and persistence of symptoms led to an inpatient admission. Infections, malignant neoplasm, immunodeficiency, and drug reaction were ruled out. Tests for parasitic infections were unrevealing. Lyme titer results were negative. Bone marrow biopsy result was normal. T-cell and B-cell subsets were normal in number, morphology, and function. C3, C4, C1q, C1 esterase inhibitor, and immunoglobulin levels were normal. To rule out myeloproliferative hypereosinophilic syndrome, genetic testing was performed, and the results were negative for gene deletion associated with *FIP1L1/PDGFRA* fusion. Lower-extremity MRI showed thickened and increased signal in the subcutaneous soft tissue fascia surrounding the muscles. A full-thickness skin biopsy specimen showed fascia and adjacent adipose tissue with perivascular eosinophilic infiltrates and eosinophils in a deep dermal perivascular pattern consistent with EF (Fig 1).

On the basis of the biopsy and MRI results, the patient was diagnosed as having EF. He was initially treated with oral steroids (2 mg/kg) and methotrexate (20 mg) but continued to have progressive subcutaneous induration and developed lower-extremity peau d'orange changes. The therapy was escalated to weekly intravenous methylprednisolone pulses (30 mg/kg), oral steroids (2 mg/kg), subcutaneous methotrexate (25 mg), and infliximab (initially 10 mg/kg and then 15 mg/kg). With this regimen, his edema, erythema, and induration markedly improved and his eosinophil count decreased during several months to 2.5% or $153//\mu L$. During this time,

Disclosures: Authors have nothing to disclose.

doi:10.1016/j.anai.2011.01.021