Bilateral Lacrimal Gland Disease: From Docile to Deadly

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Abstract

Purpose: Bilateral lacrimal gland (LG) disease represents a rare presentation with prognoses ranging from innocuous to life-threatening. To our knowledge, the literature contains only isolated case reports1 and brief single-diagnosis series2 describing this entity, the largest of which includes 14 cases describing IgG4-related disease.3 Here, we present 80 such patients with pathology ranging from inflammatory, infectious, neoplastic, and other etiologies. Clinical presentation, radiography, histopathology, & treatment outcomes were assessed. Methods: We reviewed the clinical information available from 80 patients who presented with bilateral lacrimal gland disease. Each case was evaluated for patient age, sex, race, signs and symptoms on presentation, duration of disease, radiographic characteristics, serology, biopsy results, treatment, and outcome. Results: Included are 18 males and 62 females with mean age of 46 years (range 8-82). Racial composition is diverse, distributed among Black (n=39, 49%), White (n=31, 39%), and Hispanic (n=10, 13%) patients. Categories of disease include inflammatory (n=44, 55%), lymphoproliferative (n=19, 24%), prolapsed LG (n=11, 14%), dacryops (n=5, 6%), and extramedullary hematopoiesis (n=1, 1%). Specific diagnoses include idiopathic orbital inflammation (pseudotumor, n=22, 32%), sarcoidosis (n=14, 17%), lymphoma (11, 14%), reactive lymphoid hyperplasia (n=8, 10%), Rosai-Dorfman disease (n=2, 2%), Erdheim-Chester disease (2, 2%), Sjogren’s Disease (3, 4%) and less common entities. Clinical features and outcomes were evaluated. In all cases, LG enlargement was clinically and radiologically evident, with the disease process either confined to the LG, or showing adjacent extension. Conclusions: Our findings indicate that bilateral LG disease is most commonly a manifestation of inflammatory and lymphoproliferative diseases. We believe this to be the largest such case series to date and suggest that evaluation and management of bilateral LG disease should focus on the most prevalent diagnoses: idiopathic orbital inflammation, sarcoidosis, lymphoma, reactive lymphoid hyperplasia, and prolapsed lacrimal gland. References: 1. Shinder R, Mirani N, Wu HV, Langer PD. Extramedullary hematopoiesis in the lacrimal gland. Ophthal Plast Reconstr Surg 2008;24:48-50. 2. Derr C, Shah A. Bilateral dacryoadenitis. J Emerg Trauma Shock 2012;5:92-4. 3. Go H, Kim JE, Kim YA, et al. Ocular adnexal IgG4-related disease: comparative analysis with mucosa-associated lymphoid tissue lymphoma and other chronic inflammatory conditions. Histopathology 2012;60:296-