CASE STUDY

Idiopathic Ulcerative Laryngitis: An uncommon disease

Laringitis Ulcerativa Idiopática: una entidad poco común

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Received 12 September 2015; accepted 21 September 2015
Available online 31 December 2015

Case report

We present the case of a 39 year-old woman who was working at that time as an Emergency Room nurse of a hospital. She was a mild smoker and had no past medical history of interest. The patient was referred from another center with diagnosis of persistent laryngitis and severe dysphonia for over three months duration despite treatment with oral corticosteroids, antibiotics and proton-pump inhibitor (PPI). The patient reported recent history of tuberculosis infection in some staff members. Chest radiography showed no significant alterations, and Purified Protein Derivative (PPD) for tuberculosis testing was negative.

Examination revealed a severe dysphonia with woody voice. The abbreviated Voice Handicap Index (VHI-10) was 30. During laryngoscopic exploration ulcerated lesions were seen in the middle and anterior third of both vocal folds with ligament exposure in some areas (Fig. 1a). Stroboscopic examination showed absence of the mucosal wave bilaterally. Evaluation with Narrow Band Image (NBI) showed no significant increase in vascularity or specific patterns that could suggest malignancy (Fig. 1b).

With an initial diagnosis of non-specific chronic laryngitis, the differential diagnosis should be made with granulomatous diseases, premalignant or malignant lesions, viral infections or severe pharyngolaryngeal reflux (Table 1).

Laryngeal microsurgery was performed obtaining biopsies of both vocal folds (Fig. 2) and cultures (acid-alcohol fast bacilli, Periodic Acid-Schiff (PAS) and Red Congo staining) without showing the presence of bacilli or granulomas, nor signs of malignancy. Given these results, the patient met all diagnostic criteria for Idiopathic Ulcerative Laryngitis (IUL): Preceding upper respiratory infection with cough, bilateral mid-membranous ulcerative lesions of the vocal folds, prolonged course of ulcer resolution >6 weeks and lack of response to pharmacologic management (no acute resolution with PPIs, corticosteroids, antibiotics, antifungal medications).

Six months after the beginning of the dysphonia, the patient had an spontaneous speech recovery, with normalization of the VIH-10 to 5. Laryngeal examination showed disappearance of the laryngeal ulcers, with recovery of the mucosal wave in the stroboscopic exam and a NBI pattern of normal vascularization in the larynx (Fig. 3a and b).

Discussion

“Laryngitis” is the general term that describes an inflammatory condition of the larynx. There are several known causes, including viruses, bacteria, harmful substances, trauma, and systemic inflammatory processes involving the larynx. Often various noxa are identified simultaneously. The course of laryngitis may be acute or chronic, and the clinical manifestations include hoarseness, cough, sore throat, foreign body sensation, dysphagia, even dyspnea. The symptoms usually improve within a week.

In recent years, a clinical entity that presents as acute laryngitis has been described. In 2000, Spiegel et al. reported a case of a 35 year old with dysphonia and presence
of ulcers in the membranous portion of the vocal cords. The course of the disease persisted for several months. Later on, the same group published a series of 14 patients and this new entity was named Prolonged Ulcerative Laryngitis. Beaver et al. reported another case report also characterized by ulceration in the membranous portion of the vocal folds with a prolonged course, unrelated to systemic diseases or attribution to a particular cause.

More recently, Tzu-Yu Hsiao characterized a series of 39 patients diagnosed with prolonged ulcerative laryngitis between 1999 and 2008, who were treated with an observational approach without applying specific treatments. The age of patients ranged from 26 to 76 years with a median of 49.5 years. The recovery time of the patients ranged from 4 to 20 weeks, with an average of 9.4 weeks.

Simpson and Sulica, published in 2011 a retrospective study with multicenter data from eight institutions in the United States. They called the disease Idiopathic Ulcerative Laryngitis and established diagnostic criteria. The series included 15 patients who had multiple therapeutic attempts made during the course of the disease without clinical improvement. After the resolution of the ulcers, 60% of the patients had persistent vibration abnormalities.

IUL is a rare clinical entity that usually presents with persistent hoarseness accompanied by cough, which is established after an extended upper airway infectious condition. It is characterized by the development of bilateral ulcers in the membranous portion of the vocal folds that remain a long period of time before healing. It is more common in middle age females. Typically the lesions do not respond to any treatment used to this day. Clinical improvement and disappearance of ulcerative lesions are commonly found between two and four months after onset.

It has not been set an etiology for this disease yet and there are no definitive treatments. Further studies are needed to establish a more complete understanding of this disease.

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<thead>
<tr>
<th>Table 1</th>
<th>Differential diagnosis of Ulcerative Idiopathic Laryngitis.</th>
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<tr>
<td>Granulomatous diseases</td>
<td>Diagnostic findings</td>
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<tr>
<td>Tuberculosis</td>
<td>Positive PPD. Biopsy: Isolation of acid-alcohol fast bacilli, granulomas with or without giant cells and caseous necrosis.</td>
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<tr>
<td>Sarcoidosis</td>
<td>Consistent clinical and radiological findings (bilateral hilar lymphadenopathy, pulmonary infiltrates), non-caseating granulomas, Kveim–Siltzbach Test.</td>
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<tr>
<td>Premalignant lesions and tumors</td>
<td>Biopsy: dysplasia/infiltrating carcinoma.</td>
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<td>Pharyngeal–laryngeal reflux</td>
<td>Larynx erythema, vocal cord edema, hypertrophy of the posterior commissure. Biopsy specific chronic inflammation.</td>
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<tr>
<td>Viral infections</td>
<td>Culture, PCR, serology (antibodies HSV-1 and HSV-2).</td>
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needed to establish the causative/risk factors for this condition and to determine the optimal medical treatment.

**Conflict of interest**

The authors declare no conflict of interest.

**References**