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Case Report

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Isolated chronic dysphonia revealing a systemic lupus erythematosus: 2 case reports

Abstract

Background: Laryngeal involvement in systemic lupus erythematosus may be asymptomatic, and diagnosis may only be made through routine screening or investigation for other symptoms. However, laryngeal involvement can also be potentially serious, with the potential for upper airway obstruction and other complications. Treatment typically involves systemic corticosteroids, which are often effective in managing symptoms. In some cases, more invasive procedures such as tracheotomy or mechanical ventilation may be necessary.

Aim: Emphasize the involvement of the larynx in systemic lupus erythematosus and the crucial role of clinicians in recognizing its symptoms to consider a potential diagnosis.

Case report: We report two cases of isolated dysphonia revealing systemic lupus erythematosus in two young adult females who had no other clinical signs of systemic lupus erythematosus.

Conclusion: Due to the potential seriousness of laryngeal involvement in systemic lupus erythematosus, it is important for clinicians to maintain a high degree of suspicion for this condition in patients with systemic lupus erythematosus, particularly in cases where there are unexplained respiratory or throat symptoms. Early diagnosis and treatment can help prevent potentially life-threatening complications.

Keywords: chronic hoarseness, systemic lupus erythematosus, laryngolupus, diagnosis assessment

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Introduction

Laryngeal involvement in systemic lupus erythematosus is rare, but it can still occur and may present a challenge for diagnosis and treatment.[1] It was described for the first time by Scarpelli DG in 1959.[2]

Laryngeal involvement in systemic lupus erythematosus can present a challenge for diagnosis and treatment due to its clinical polymorphism and unpredictable evolution. The symptoms can vary widely depending on the individual case, but may include hoarseness, difficulty breathing, coughing, and/or pain or discomfort in the throat.

The diagnosis of laryngeal involvement in systemic lupus erythematosus may involve a combination of medical history, physical examination, laboratory testing, imaging studies, and/or laryngeal biopsy.[1]

We report two cases of patients diagnosed with systemic lupus erythematosus revealed by chronic hoarseness. The aim of our study is to emphasize the involvement of the larynx in systemic lupus erythematosus and the crucial role of clinicians in recognizing its symptoms to consider a potential diagnosis.

Case reports

Case 1

Mrs. N.E., a 26-year-old female patient, presented to the external consultation for a sudden onset of an isolated hoarseness 3 weeks before, with neither dyspnea nor dysphagia. She had no medical history. She reported that sometimes she presented joint pain at the end of the day, but no other clinical signs were found on the interrogatory, including the absence of mucosal dryness, chronic fatigue, fever, and oedema.

A physical examination showed a malar rash and left vocal paralysis without mucosal thickening or inflammatory signs on the laryngeal endoscopy. The bone and joint examination showed no pathological signs.

We conducted a cervical CT scan that showed no anomalies.

A blood checkup revealed normocytic normochromic anemia at 9 g/dl, lymphopenia at 700 mm3, and a biological inflammatory syndrome (C-reactive protein at 40 mg/l and erythrocyte sedimentation at 50 mm).

At this point, we decided to perform an immunological assessment. The patient tested positive for antinuclear antibodies (1/320) with a homogeneous fluorescence. Anti-dsDNA was also positive at 200 IU. We completed the biological checkup with a urine protein test that was positive at 3 g/24 h.

A renal biopsy was performed and revealed lupus nephritis class IV. The diagnosis of systemic lupus erythematosus with laryngeal and renal involvement was made. The patient was treated with synthetic antimalarial drugs, steroids (1 mg/kg/day), and monthly cyclophosphamide for renal involvement. The evolution was favorable. Six months later, the patient had no dysphonia.

Case 2

Mrs. A.A., aged 21- years -old, with no notable pathological history, was explored for acute dysphonia presented 2 weeks earlier, associated with photosensitivity with neither dyspnea nor dysphagia. No other clinical signs were found during the interrogation.

Physical examination showed a malar rash, oral ulceration, and right vocal cord paralysis with subglottic mucosal thickening on the laryngeal endoscopy (Figure 1). The bone and joint examination showed no pathological signs.

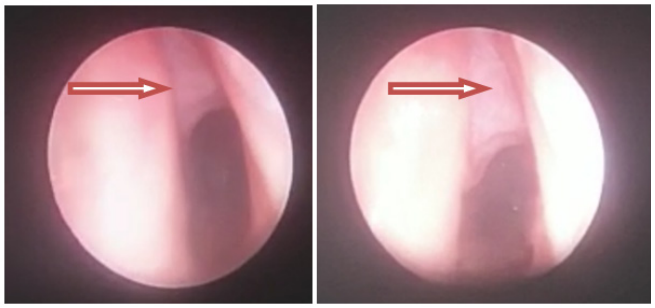

Figure 1 Endoscopic view of the larynx showing subglottic thickening (red arrow).

We conducted a cervical CT scan that showed no anomalies.

A blood checkup revealed normocytic normochromic anemia at 9.4 g/dl, lymphopenia, and a biological inflammatory syndrome (C-reactive protein at 55 mg/l and erythrocyte sedimentation at 65 mm).

At this point, we decided to perform an immunological assessment. The patient tested positive for antinuclear antibodies, which were positive at 1/640 with a homogeneous fluorescence. anti-dsDNA antibodies were positive at 280 IU. We completed the biological checkup with a urine protein test that was positive at 3.4 g/24 hours.

A direct suspension laryngoscopy with a biopsy of the supraglottic thickening and a renal biopsy were performed with a pathological study that revealed, respectively, an inflammatory granuloma and lupus nephritis class IV.

The diagnosis of systemic lupus erythematosus with laryngeal and renal involvement was made. The patient was treated with synthetic antimalarial drugs, steroids (1 mg/kg/day), and monthly cyclophosphamide for renal involvement. The evolution was favorable. Six months later, the patient had no dysphonia.

Discussion

Laryngeal involvement in systemic lupus erythematosus is rare, but it's important to recognize that it can occur and may present with a variety of symptoms, including hoarseness, dysphonia, dyspnea, and/or dysphagia.[3,4]

Laryngeal involvement can be unilateral or bilateral and may recur, adding to the diagnostic challenge. In some cases, it may be the only presenting symptom of systemic lupus erythematosus, highlighting the importance of considering this condition in the differential diagnosis of patients presenting with unexplained laryngeal symptoms.[1]

This laryngeal involvement is usually benign, but it's important to recognize that it can potentially have serious consequences, particularly if it results in obstruction of the upper airways or bilateral vocal cord paralysis,[5] complications that can be life-threatening.[6]

These manifestations typically occur in patients with known and active disease, but they can also rarely complicate inactive or extinct lupus.[7]

Laryngolupus can present with a wide range of symptoms, and the clinical manifestations can be quite polymorphic, including dysphonia, chronic cough, inspiratory dyspnea, laryngeal stridor, or even a sensation of a foreign body in the upper airways.[5,6,8,9]

Laryngoscopic examination may reveal a range of findings, from simple laryngeal edema to more severe inflammation with the formation of pseudo-membranes or laryngeal ulcerations. Vocal cord paralysis is also a possible manifestation of laryngolupus.[3,5,6,8,10]

Additional rare manifestations of laryngolupus, such as supraglottic stenoses, inflammatory pseudotumors of the larynx, epiglottitis, and necrotic vasculitis of the larynx, are all potentially serious complications of laryngolupus.[9]

The range of symptoms and findings underscores the importance of a thorough diagnostic workup in patients presenting with laryngeal symptoms, particularly in those with a history of autoimmune disease or other potential risk factors for laryngolupus.[4]

This may include a combination of imaging studies, laboratory testing, and laryngoscopic examinations, as well as a detailed medical history and physical examination. Early recognition and management of laryngolupus is important to prevent potential complications and ensure optimal outcomes for affected patients.

Tsunoda et al.[11] reported one case of systemic lupus erythematosus that was revealed by chronical dysphonia, while S Bouomrani et al.[12] reported one case of systemic lupus erythematosus revealed by dysphonia and dysphagia, and another case of dysphonia and foreign body sensation and respiratory difficulties was reported by Korbet et al.[7]

In the largest review of the literature made by Teitel AD grouping 97 cases of laryngolupus, laryngeal oedema and vocal cord paralysis were the most common manifestations found, respectively, in 28 and 11% of cases.[9] Vocal cord palsy may be caused by recurrent laryngeal neuropathy, which manifests itself either as vasculitis involving the vasa nervorum, neuritis, or compression by a dilated pulmonary artery.[4]

Systemic corticosteroids are effective in managing laryngolupus, particularly in cases with mild to moderate symptoms.[5,9] However, in severe cases where there is significant airway obstruction or respiratory distress, more invasive interventions such as tracheotomy or mechanical ventilation may be required. [8,12–13]

Conclusion

Laryngeal involvement in systemic lupus erythematosus is rare and often asymptomatic, but it can occasionally present with various symptoms such as dysphonia, cough, inspiratory dyspnea, laryngeal stridor, and the sensation of a foreign body in the upper airways. Therefore, an endoscopic examination is recommended for any patient with systemic lupus erythematosus who presents with laryngeal symptoms. It is important to consider systemic lupus erythematosus as a possible underlying cause in young women who present with such symptoms and have inconclusive investigation results, especially if they have other clinical features of the disease such as joint pain, malar rash, and positive autoantibodies. Early recognition and management of laryngolupus can improve the patient's prognosis and prevent serious complications.

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Author contributions

Mohamed Yafi and Sara Rochd conceived and designed the study. Youssef Lakhdar collected and analyzed the data, Othmane Benhoummad wrote the first draft of the manuscript. Youssef Rochdi

and Abdelaziz Raji reviewed and edited the manuscript. All authors contributed to the interpretation of the data, revised the manuscript critically for important intellectual content, and approved the final version for publication.

Conflicts of interest

The authors declare no conflict of interest.

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Ethical approval

Informed and formal consent was obtained from all individual participants included in the study.

Data availibility

The data used to support the findings of this study are available from the corresponding author upon request.

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