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# Hoarseness as the initial manifestation of systemic lupus erythematosus

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**Abstract**
A 51-year-old woman complained of hoarseness of two years duration. The patient's past medical history was significant for autoimmunological hepatitis and arthritis for which she had not received treatment. Laryngoscopy and laryngeal stroboscopy revealed 'bamboo joint-like nodules' on both true vocal folds. These nodules resembled rheumatoid nodules and were suggestive of a collagen disease. Previous reports have documented that the treatment for such conditions related to collagen diseases is surgical resection. However, we initially attempted to treat the laryngeal lesions systemically with prednisolone. The hoarseness and the bamboo-like nodules disappeared six months after the treatment. Furthermore, the liver function test returned to normal and arthritis completely resolved. Based on our patient's response to this treatment, we diagnosed atypical-SLE and a lupus laryngitis. This case suggests that studies of the larynx may be helpful in the early diagnosis of collagan diseases and that such conditions may respond to systemic treatment.

**Key words: Lupus erythematosus, systemic; Hoarseness; Vocal folds**

## Introduction

Rheumatoid diseases have been reported to involve the larynx. This is not a rare occurrence and is manifested most commonly as cricoarytenoid joint arthritis (Lofgren and Montgomery, 1962). However, rheumatoid nodules of the larynx are rare (Webb and Payne, 1972; Friedman and Rice, 1975; Schwartz and Grishman, 1980; Hosako *et al.*, 1993). Previous case reports have described specific lesions on the true vocal folds, which are related to systemic lupus erythematosus (SLE) (Hosako *et al.*, 1993). This case report presents a patient with a bilateral vocal fold lesion, located at the middle of her vocal fold resembling a bamboo joint. Hoarseness associated with these rheumatoid nodules was the only symptom of this systemic disease. Generally, the treatment is the surgical removal of these nodules (Webb and Payne, 1972; Friedman and Rice, 1975; Schwartz and Grishman, 1980; Hosako *et al.*, 1993). However, we offer an alternative successful systemic treatment method.

## Case report

A 51-year-old woman was referred by her family physician to our clinic. She complained of a two-year history of gradually progressive hoarseness. The patient had no history of any disorders or trauma to the larynx, but had a three-month history of autoimmunological hepatitis, based on a histological study by a laparoscopic liver biopsy. The laboratory data are shown (Table I).

The patient's hoarseness was concomitant with a flare of arthritis (in the distal interphalangeal joints and proximal interphalangeal joints). For the treatment of her hepatitis she was taking prednisolone 30 mg/day to 8 mg/day and when admitted to our clinic was taking 8 mg/day.

Direct laryngoscopy without anaesthesia revealed bamboo joint-like lesions on the true vocal folds bilaterally (Figure 1). Stroboscopic study showed a different phase of vocal fold vibration between the half anterior to the bamboo joint and the posterior half. We could not find any vibration in the bamboo joint nodules.

The indicated treatment for these lesions was surgical removal. However, we referred the patient to the rheumatology service since the findings were suggestive of a collagen or rheumatoid disease. Careful examination was accomplished using the criteria of Raynaud's phenomenon and arthritis. The diagnosis was atypical-SLE with an auto-immunological hepatitis. As she had already started prednisolone for the auto-immunological hepatitis, we decided to continue that systemic treatment without surgical removal of the lesions. Six months following the treatments, the hoarseness had disappeared, and direct laryngoscopy revealed resolution of the bamboo-like lesions on the vocal folds (Figure 2). Laryngeal stroboscopy revealed normal vibrations of both entire vocal folds. Raynaud's phenomenon, liver function tests, and arthritis also improved.

TABLE I
LABORATORY DATA

| |
|---|
| ESR 62 mm/hour |
| WBC 3900/mm |
| Total Protein 9.8 g/dl |
| γ-globulin 39.9% |
| GOT 83 U/l, GPT 49 U/l, LDH 516 U/l, ALP 273 U/l |
| TC 138 mg/dl |
| CRP (+ve) |
| IgG 4556 mg/dl |
| LE test −ve |
| ANA × 1280 |
| DNAab (−ve), RNAab (−ve), ENAab (−ve), SSAab (−ve) |

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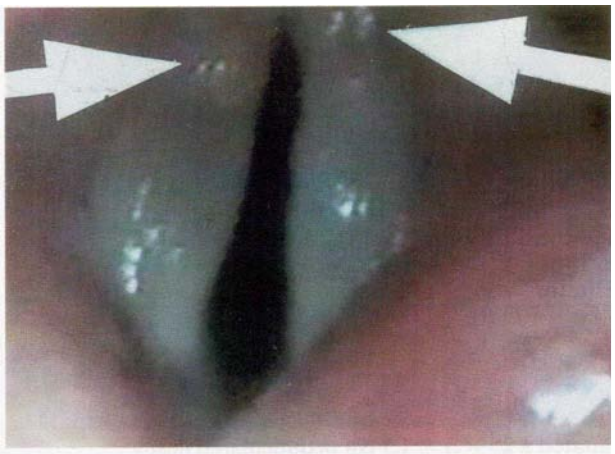

FIG. 1

Laryngoscopic view of the vocal folds prior to treatment. Note the 'bamboo-joint nodules' (arrows) of the vocal folds bilaterally.

## Discussion

Laryngeal involvement in rheumatoid diseases is not rare and is manifested most commonly as cricoarytenoid joint arthritis (26 per cent). Lofgren and Montgomery (1962) and Webb and Payne (1992) have documented vocal fold rheumatoid nodules complicating a progressive case of classical rheumatoid arthritis which occurred 10 years following the diagnosis. Friedman and Rice (1975) have reported three similar cases of rheumatoid nodules involving the larynx. These reports suggest that laryngeal rheumatoid nodules are rare and there has been only one report of vocal fold nodules as the initial manifestation of SLE (Schwartz and Grishman, 1980). Hosako *et al.*, 1993 have reported observing the 'bamboo-joint-like lesion' on the true vocal folds. This particular vocal fold lesion has been localized to the middle of the vocal fold in SLE patients. An excision was performed with improvement of the hoarseness. Histological examination of the lesion revealed a subepithelial location and that it consisted of fibrous tissue and many plasma cells. Immunological analysis revealed IgA and IgG in some of these plasma cells and it was assumed that this lesion was related to SLE. Vocal fold nodules often occur bilaterally at the free edge of the vocal folds. This particular vocal fold lesion involved not only the free edge, but encircled the vocal folds resembling a bamboo-joint.

We first considered vocal fold nodules as the diagnosis, but careful examination with a direct laryngeal scope and stroboscopy revealed the bamboo-joint-like lesions involving both vocal folds. This finding suggested that the laryngeal lesions may be related to a collagen or rheumatoid disease.

The treatment for laryngeal rheumatoid nodules is surgical removal (Webb and Payne, 1972; Friedman and Rice, 1975; Schwartz and Grishman, 1980; Hosako *et al.*, 1993). However, we thought that the nodules were related to a collagen disorder and that systemic steroid treatment would reduce the size of the lesions. After six months of systemic treatment with prednisolone (initially 30 mg to 7 mg/day for the maintenance), the hoarseness resolved. Repeat direct laryngoscopy demonstrated the absence of the bamboo-joint lesion.

This case report presents an early manifestation of atypical SLE. It is the first report of bamboo-joint-like vocal fold nodules successfully treated by systemic medication. Careful laryngeal examination, consisting of direct laryngoscopy and stroboscopy, aids in the diagnosis of vocal fold lesions related to systemic collagen diseases. Such systemic treatment for laryngeal lesions secondary to collagen diseases may reduce the need for surgery. In conclusion, we believe a thorough laryngological examination is helpful in the diagnosis of collagen diseases and that systemic treatment provides a conservative treatment alternative to the resection of such lesions.

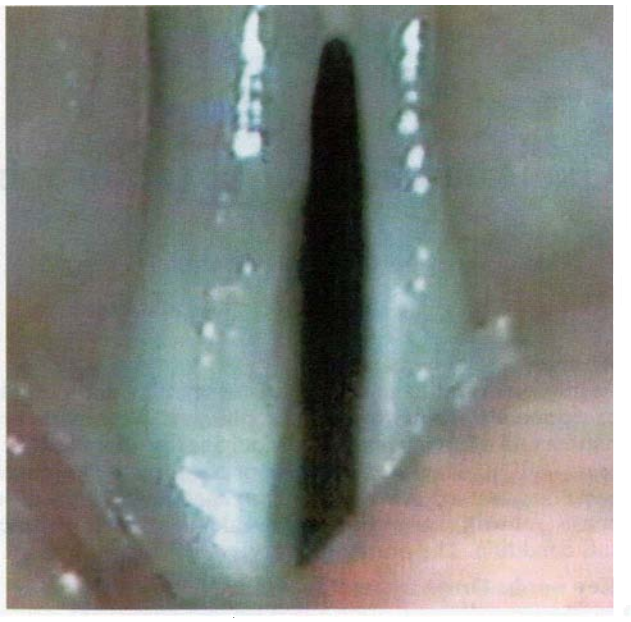

FIG. 2

Laryngoscopic view six months following the systemic treatment, indicating resolution of the 'bamboo-joint nodules'.

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