CASE REPORT

CRYPTOCOCCAL INFECTION OF THE VOCAL FOLDS

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Abstract. Cryptococcosis of the larynx is rare. In this manuscript we present a case of laryngeal cryptococcosis with a clear history of pigeon exposure. Cryptococcal laryngitis may present as hoarseness without other typical symptoms, even in immunocompetent individuals.

Keywords: cryptococcosis, larynx, infection, hoarseness, pigeon, Taiwan

INTRODUCTION

Viral laryngitis is the most common infectious cause of hoarseness (Kinnman, 1975). However, primary mycotic laryngeal infections have been reported; the pathogens include Candida albicans, Histoplasma capsulatum, Coccidioides immitis, Blastomyces dermatitidis, Paracoccidioides brasiliensis and Cryptococcus neoformans (Vrabec, 1993). The disease may occur as a result of dissemination of microorganisms that have already established an infection. Definitive diagnosis is often achieved with culture or histological examination of biopsied tissue in conjunction with special stains such as methenamine silver or mucicarmine if C. neoformans is suspected (McGregor et al, 2003). These microorganisms possess capsules that can be stained with mucicarmine (McGregor et al, 2003). The cell walls also

stain positively with methenamine silver. *C. neoformans* typically presents as a 4~10 μ m encapsulated fungus surrounded by a polysaccharide capsule (McGregor *et al*, 2003). It is globally distributed and commonly found in soil, avian feces (*ex*, pigeons), fruit, wood and vegetables (Medoff and Kobayashi, 1991). Those who work with pigeons are at increased risk for cryptococcal infections (Medoff and Kobayashi, 1991). Here we describe the case of a patient with laryngeal cryptococcal infection with a clear history of pigeon exposure.

CASE REPORT

A 53-year-old man was referred to our department Otolaryngology, Cathay General Hospital because of a 2-month history of persistent hoarseness. He had no history of fever, cough, chest pain, or dyspnea. He had been treated with several courses of antibiotics with limited improvement. The patient was a non-smoker and had no

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travel history. The patient reported he had been exposed to pigeons in his neighborhood for 2 weeks before presentation. The general physical examination was unremarkable. Indirect laryngoscopy revealed a mass on the right posterior vocal cord (Fig 1). His chest radiograph was normal. The patient subsequently underwent direct laryngoscopy with biopsy of the right vocal cord lesion. Histopathology of the mass showed squamous hyperplasia with acute and chronic inflammation along with oval to elliptical microorganisms (Fig 2). The microorganisms were clearly identified using Gomori's methenamine silver (GMS) stain (Fig 3a). A thick capsule that stained positively with mucicarmine was also noted (Fig 3b).

A human immunodeficiency virus test and serum test for syphilis were both negative. A complete blood count and immunoglobulin levels were within normal limits. Sputum, blood, and urine cultures for fungi were negative. A diagnosis of laryngeal cryptococcosis was made.

The patient's postoperative course was unremarkable. He was treated with fluconazole 400 mg once daily for 6 weeks. The patient was symptom free at the end of the treatment course. Fiberoptic endoscopy

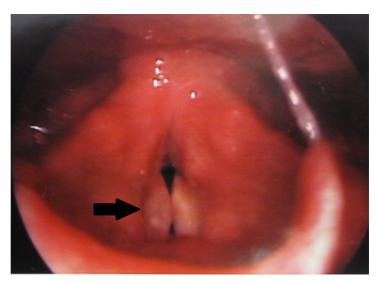


Fig 1–Indirect laryngoscopy revealing a mass on the right posterior vocal cord (arrow).

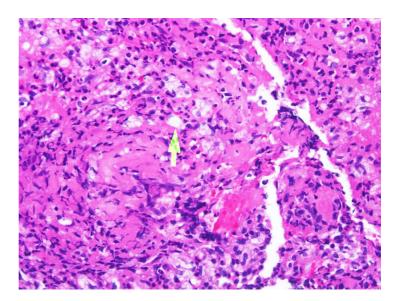


Fig 2–Squamous hyperplasia with acute and chronic inflammation along with oval to elliptical microorganisms (arrow) (H&E, 200x).

at the end of treatment and at a 6-month follow-up visit was normal.

DISCUSSION

Cryptococcosis of the larynx is rare

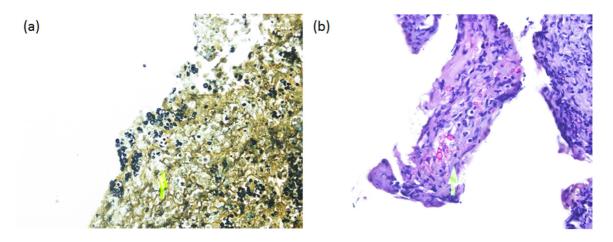


Fig 3–(a) Microorganisms identified using Gomori's methenamine silver (GMS) stain. (b) A thick capsule visualized with mucicarmine stain.

(Nadrous et al, 2004). The first case of laryngeal cryptococcosis was reported by Reese and Colclasure in 1975. Only 15 cases have been reported in the literature (Chongkolwatana et al, 1998; Nadrous et al, 2004; Zeglaoui et al, 2009; Gordon et al, 2010). Of those patients, 13 were immunocompromised: 4 had HIV, 3 had diabetes, and 6 had a history of long-term corticosteroid use for chronic obstructive lung discases or asthma. One patient had been exposed to soil rich in chicken manure (Reese and Colclasure, 1975). As far as we know, this is the first report of laryngeal cryptococcosis with a clear history of pigeon exposure.

Cryptococcal infections occur predominantly in patients with T-cell-mediated immune defects, particularly those with AIDS and transplant-related immunosuppression (Chongkolwatana *et al*, 1998). Corticosteroid therapy and cancer chemotherapy are also predisposing factors for the development of cryptococcal infections (Nadrous *et al*, 2004). The incidence of cryptococcal infections is increased in subjects with hematologic malignancies, sarcoidosis, and diabetes mellitus (Pappas *et al*, 2001).

The treatment of cryptococcal infection depends on the area affected and the immune status of the patient. Fluconazole is effective in treating cryptococcal infections in immunocompetent and immunocompromised patients (Kerschner *et al*, 1995). Treatment of cryptococcal laryngitis in 13 of the cases mentioned previously was accomplishes within 1 to 24 months with antifungal therapy. Three of the patients were treated with surgery only (polypectomy), and 2 of these 3 were disease free by months (Gordon *et al*, 2010). Our patient was treated for 6 weeks and remainded disease free at 6 months follow-up.

No evidence-based guidelines are currently available for treating laryngeal cryptococcosis. We believe fluconazole to be safe and effective for treating cryptococcosis of the head and neck without central nervous system involvement.

Clinicians often encounter patients presenting with hoarseness in daily practice and exposure to pigeons is also common, but cryptococcal infection is seldom considered in the differential diagnosis of hoarseness. Cryptococcal laryngitis can be a cause of hoarseness, even in immunocompetent individuals.

REFERENCES

- Chongkolwatana C, Suwanagool P, Suwanagool S, Thongyai K, Chongvisal S, Metheetrirut C. Primary cryptococcal infection of the larynx in a patient with AIDS: a case report. *J Med Assoc Thai* 1998; 81: 462-7.
- Gordon DH, Stow NW, Yapa HM, Bova R, Marriott D. Laryngeal cryptococcosis: Clinical presentation and treatment of a rare cause of hoarseness. *Otolaryngol Head Neck Surg* 2010; 142: S7-9.
- Kerschner JE, Ridley MB, Greene JN. Laryngeal cryptococcus. Treatment with oral fluconazole. *Arch Otolaryngol Head Neck Surg* 1995; 121: 1193-5.
- Kinnman J. Acute obstructive laryngitis in children. *HNO* 1975; 23: 378-82 (in German).

- McGregor DK, Citron D, Shahab I. Cryptococcal infection of the larynx simulating laryngeal carcinoma. *South Med J* 2003; 96: 74-7.
- Medoff G, Kobayashi GS. Systemic fungal infections: an overview. *Hosp Pract (Off Ed)* 1991; 26: 41-52.
- Nadrous HF, Ryu JH, Lewis JE, Sabri AN. Cryptococcal laryngitis: case report and review of the literature. *Ann Otol Rhinol Laryngol* 2004; 113: 121-3.
- Pappas PG, Perfect JR, Cloud GA, *et al.* Cryptococcosis in human immunodeficiency virus-negative patients in the era of effective azole therapy. *Clin Infect Dis* 2001; 33: 690-9.
- Reese MC, Colclasure JB. Cryptococcosis of the larynx. *Arch Otolaryngol* 1975; 101: 698-701.
- Vrabec DP. Fungal infections of the larynx. Otolaryngol Clin North Am 1993; 26: 1091-114.
- Zeglaoui I, Belcadhi M, Mani R, Sriha B, Bouzouita K. Laryngeal cryptococcosis revealing AIDS: a case report. *Rev Laryngol Otol Rhinol (Bord)* 2009; 130: 307-11.