CASE REPORT

Laryngeal leishmaniasis in a patient taking inhaled corticosteroids

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SUMMARY

We present a case of a man in his late 60s, who had spent 3–4 months of the year in rural Spain, presenting with intermittent hoarseness of voice. He had a background of asthma and bronchiectasis, and was taking inhaled corticosteroids. His dysphonia was initially managed as bronchiectasis with little improvement. Bronchoscopy revealed a cystic lesion on his left vocal fold, and tissue biopsy revealed *Leishmania* amastigotes. This confirmed a diagnosis of laryngeal leishmaniasis. We propose that this is likely secondary to his inhaled corticosteroid therapy. The infection was treated with a 30-day course of miltefosine, and at most recent follow-up the patient was deemed free from leishmanial infection.

BACKGROUND

Laryngeal leishmaniasis is a very rare diagnosis in immunocompetent individuals, and often presents with hoarseness of voice. We present the case of a man with a background of bronchiectasis, who had spent several months of the year in Spain. This man presented with voice hoarseness, and a perceived worsening of his bronchiectasis.

CASE PRESENTATION

A retired policeman in his late 60s noted some intermittent dysphonia, starting in 2011. He had a medical history of bronchiectasis diagnosed in 1994. This worsened at around the same time as the onset of the hoarseness, and in May 2013, *Pseudomonas aeruginosa* was grown in one of his sputum cultures. The patient's voice was intermittently husky, and of decreased pitch and volume. Further, he demonstrated worsening hoarseness with continued usage, improving on vocal rest. He had neither dysphagia nor otalgia.

With regard to further medical history, he had experienced lifelong asthma, and had contracted whooping cough at the age of 5 years. Otherwise he was normally fit and well. He was taking a combination of long-acting β_2 agonist and inhaled corticosteroid (seretide), a leukotriene receptor antagonist (montelukast), a long-acting muscarinic antagonist (tiotropium) and a proton pump inhibitor (omeprazole). He had no history of hypertension nor of diabetes, and was an ex-cigarette smoker with a 15 pack-year smoking history.

Regarding travel history, he had previously visited the Gambia, Borneo and a few other African countries. None of these countries are known to have leishmaniasis reservoirs. Further to this, he had regularly travelled to Spain, where he

had bought a home near Granada, 9 years prior. This was situated in an olive grove and in an agricultural area, with many dogs living nearby. Two friends in the village had developed leishmaniasis, one in 2010 and the other in 2013. The patient, at the time, had lived in Spain for 3–4 months each year.

On examination, the patient had no skin lesions and no hepatosplenomegaly. He had no palpable lymphadenopathy. Flexible nasendoscopy showed speckled candida effectively involving the whole pharynx. The right vocal fold was normal, and on the left there was a smooth swelling on the cranial surface of the true vocal fold (figure 1). This had features more consistent with a cyst than a polyp. There was no overt candidiasis affecting the larynx. Normal movement of both vocal folds was seen.

INVESTIGATIONS

By September 2013, the patient underwent a CT of the thorax, which demonstrated evidence of bronchiectasis in lower lobes and the right upper lobe, as well as some tracheal thickening. Bronchoscopy, performed in January 2014, was abnormal, showing a white plaque on the trachea and vocal folds, and the patient was referred to ear, nose and throat (ENT) department. In March 2014, examination of the larynx under anaesthesia revealed a raised ulcerative process on the superior aspect of the left vocal fold. Tissue analysis of a biopsy sample revealed *Leishmania* amastigotes. This confirmed the diagnosis of laryngeal leishmaniasis. This infection was likely to have been acquired in Spain, as the result of a single bite from an infected sand fly. This infection could have started months or years ago. The laryngeal lesion may have resulted from

Figure 1 A view of vocal folds prior to miltefosine treatment, demonstrating a smooth nodule on the left fold.

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Figure 2 The vocal folds after miltefosine treatment, showing some residual scarring on the left fold.

corticosteroid inhaler use creating a local site of decreased immunity.

TREATMENT

The patient was prescribed miltefosine 100 mg once daily for 30 days, prochlorperazine 5 mg as required and prednisolone 10 mg and 5 mg on alternate days. He discontinued his seretide. With regard to his pharyngeal candida, he was advised to use hypertonic saline (Sterimar) and nystatin.

OUTCOME AND FOLLOW-UP

In October 2014, the patient was reviewed by ENT department and found to have neither residual nor recurrent disease at the laryngeal level (figure 2). He was reviewed by the respiratory team, due to exacerbation of his bronchiectasis and asthma. He was advised to restart his seretide inhaler using a spacer. He remains under respiratory medicine follow-up.

DISCUSSION

We have found 20 case reports of laryngeal leishmaniasis in the English literature. The most frequently described presenting symptom is hoarseness of voice, which can lead to a wide range of differential diagnoses.^{1–6} Many case reports feature immunocompromised patients, with comorbidities including HIV infection^{4 7} and previous renal transplantation.⁵ In immunocompetent patients, some of the reported cases had a previous history of cutaneous leishmaniasis.^{2 8} Two cases identified the

use of corticosteroids as creating reduced immunity.^{1 6} We propose that in our patient, treatment with inhaled corticosteroids led to local immunosuppression in his larynx. This, we believe, resulted in the localised manifestation of the laryngeal leishmaniasis, in a patient who might have otherwise had a transient subclinical infection.

Learning points

- ▶ Treatment with inhaled corticosteroids can create local areas of decreased immunity.
- ▶ Laryngeal leishmaniasis is an atypical presentation of a disease that most commonly presents with cutaneous ulceration.
- ▶ When treating a patient with a common disease that is not responding to conventional therapies, it is important to explore and consider alternative diagnoses.

Contributors The case was identified and managed by JM and DP. The manuscript was drafted by RMR, and the version submitted approved by all the authors.

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