Long-standing laryngeal rhinoscleroma with rare Mikulicz cells

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Abstract

Rhinoscleroma is a granulomatous infectious disease that typically affects the nasopharynx and upper respiratory tract. Non-endemic (e.g., North American) and laryngeal cases are rare. This case highlights the importance of pathognomonic Mikulicz cells for diagnosis.

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Abstract: Rhinoscleroma is a granulomatous infectious disease that typically affects the nasopharynx and upper respiratory tract. Non-endemic (e.g., North American) and laryngeal cases are rare. This case highlights the importance of pathognomonic Mikulicz cells for diagnosis.

A man in his early-50s presented with worsening stridor, shortness-of-breath, and hoarseness over 2 years. Originally from northwestern South America, he reported with similar symptoms 7 years ago, but work-up did not produce a specific diagnosis. For this presentation, a large obstructing mass on the laryngeal surface of the epiglottis was identified on emergency laryngoscopy that prevented visualization of the vocal cords and lower respiratory tract (Figure 1). Tracheostomy was performed. Biopsies showed squamous

mucosa with submucosal fibrosis and a dense infiltrate, composed mostly of plasma cells and plasmacytoid lymphocytes, with rare/scattered macrophages containing intracellular gram-negative rods (Mikulicz cells; Figure 1) –suggesting a diagnosis of rhinoscleroma[1,2].

Confirmatory cultures showed *Klebsiella ozaenae* . Subsequent ciprofloxacin led to improved stridor, near-complete regression of the supraglottic mass, with improved vocal cord mobility and patent subglottis; corking trials were carried out with the tracheostomy tube, and the patient was safely decannulated. At 8-month follow-up, the laryngeal exam remains stable with no evidence of recurrence/regrowth.

Authorship list: RP, HG, RC, JD, BP-O, and DX analyzed the pathology. MS collected and analyzed the clinical data. BM analyzed the microbiology. All authors drafted, reviewed, and approved the manuscript.

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Figure 1. Laryngeal mass in a gentleman in his early 50s. Top left. Direct laryngoscopic image of supraglottic larynx showing an irregular ulcerative mass. Top right. Axial CT images taken after the patient's tracheostomy was completed. Emphysematous changes in the soft tissue are the result of the surgical procedure. Nasogastric tube can be seen in the esophagus. There is a mass visible in the supraglottic larynx (arrows) that is causing narrowing of the airway. Bottom. On microscopy, H&E (left) and Gram (right) stains show rare Mikulicz cells containing intracellular bacteria (gram negative rods on the Gram stain).