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Case Report

Isolated Laryngeal Plasmacytosis: An Unusual Cause of Laryngeal Dyspnea Case Report and Literature Review

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Abstract

Introduction: Laryngeal plasmacytosis (LP) is a rare benign lesion of mature polyclonal plasma cells, which should be differentiated from extramedullary plasmacytoma.

We describe a case of isolated laryngeal plasmacytosis leading to upper respiratory tract obstruction in which an endoscopic supraglottic laryngectomy was performed.

The patient had near complete clinical response after surgery with inhaled budesonide

Keywords: Laryngeal plasmacytosis; Laryngeal dyspnea; Partial laryngectomy

Introduction

Laryngeal plasmacytosis is a rare benign lesion, corresponding to an infiltrate of the laryngeal mucosa by mature polyclonal plasma cells, as opposed to clonal plasma cell proliferation [1,2]. It may be confused with extramedullary plasmacytoma because of its high cell density, homogeneity and predominance of plasma cells. We report a case of supraglottic endoscopic laryngectomy in a 65-year-old patient with isolated supraglottic laryngeal plasma cell disease (PL).

Case Presentation

A 65-year-old black patient with no previous pathological history presented with intermittent dyspnea since 2008, treated as asthma by several pulmonologists.

The CT scan showed homogeneous thickening of the epiglottis and the rest of the supraglottic region.

The patient was referred to the ENT emergency department for severe laryngeal dyspnoea.

Nasofibroscopy revealed a homogeneous thickening of the epiglottis, completely obstructing the laryngeal margin (Figure 1).

well-defined, regular contours obstructing 90% of the laryngeal lumen (Figure 2).

The patient underwent a tracheotomy under local anesthetic.

PET-SCANNER showed hypermetabolism of this thickening (Figure 3).

Laryngeal MRI showed intermediate T1 and T2 laryngeal thickening with cystic areas in hypersignal T2 (Figure 4).

three direct laryngoscopies with biopsies were performed.

Microscopically, there was a dense infiltration of mature plasma cells beneath the squamous epithelium, separated by thick collagen bundles. A pronounced lichenoid reaction was seen in the overlying squamous epithelium (Figure 5).

IHC studies showed that the plasma cells were positive for CD138; and negative for CD20, and CD3. immunofluorescence with antilight-

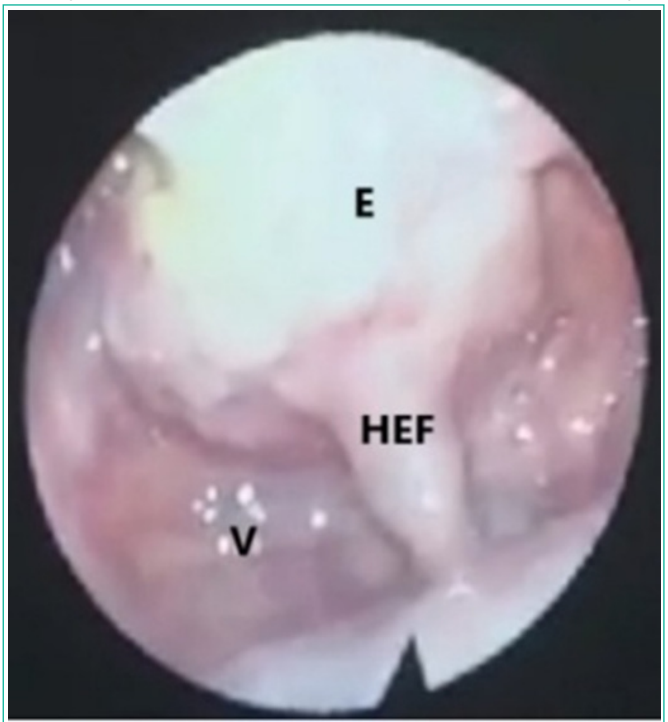

Figure 1: Indirect laryngoscopy showing an epiglottis thickening completely obstructing the laryngeal margella. epiglottis(e),Vallecula()V, hyo epiglottic fold (HEF).

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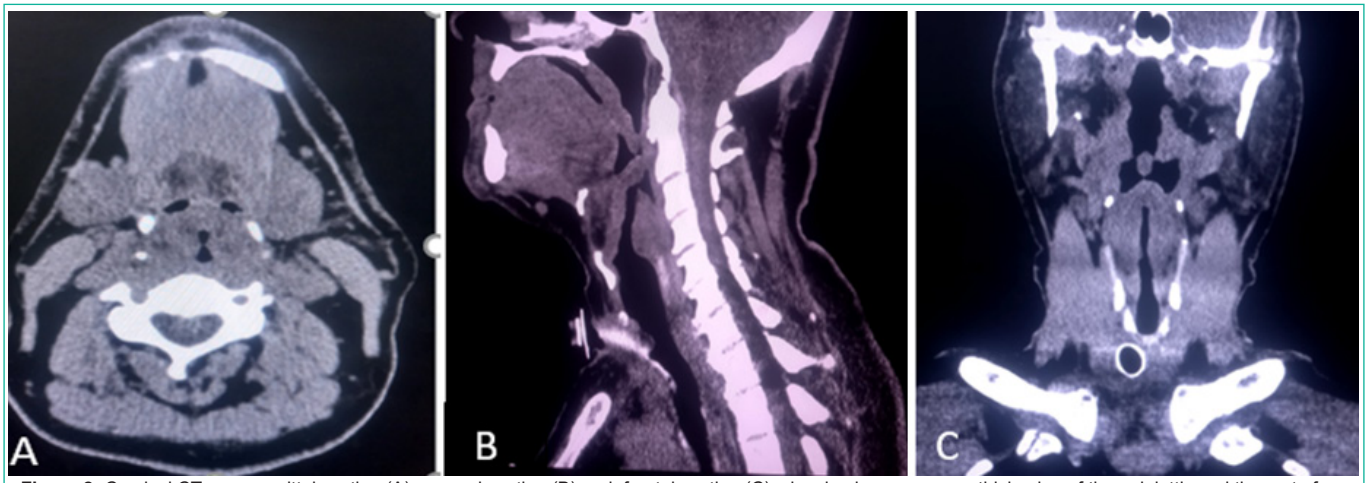

Figure 2: Cervical CT scan, sagittal section (A) coronal section (B) and frontal section (C), showing homogeneous thickening of the epiglottis and the rest of the supraglottic region, with well-defined, regular contours.

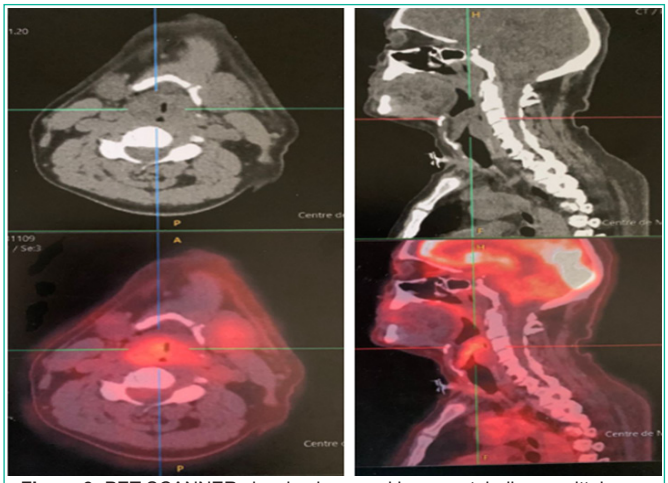

Figure 3: PET-SCANNER showing laryngeal hypermetabolism sagittal section (A) and coronal section (B).

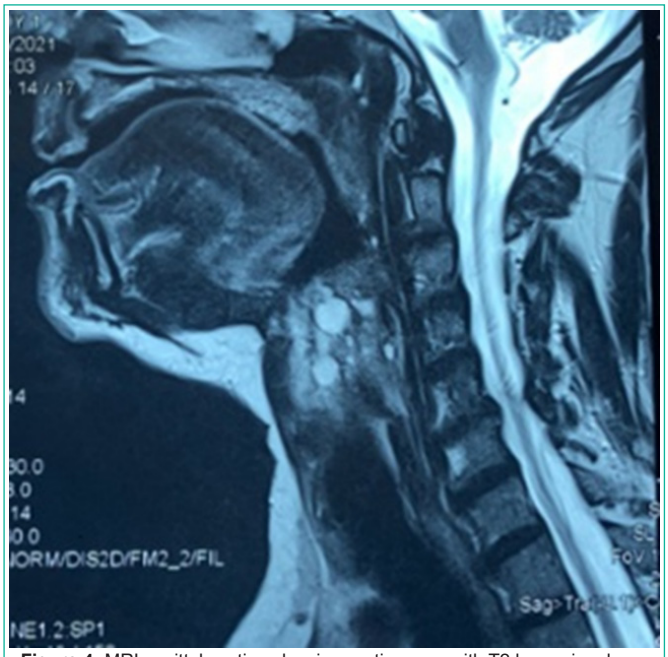

Figure 4: MRI sagittal section showing cystic areas with T2 hypersignal.

chain and antibodies confirmed the polyclonal nature of the plasma cell population (Figure 6).

The patient underwent an endoscopic supraglottic laryngectomy followed by a prescription for budesonide-based inhaled corticosteroids.

A follow-up nasofibroscopy was performed 3 months later, showing good progress with a permeable laryngeal airway (Figure 7). The patient was decanted and monitored every 6 months.

No pathogens were found, either on special staining (Ziehl-Nilson, Periodic Acid-Shiff and Grocott), microscopy or culture. No amy loid deposits were found on Congo red staining.

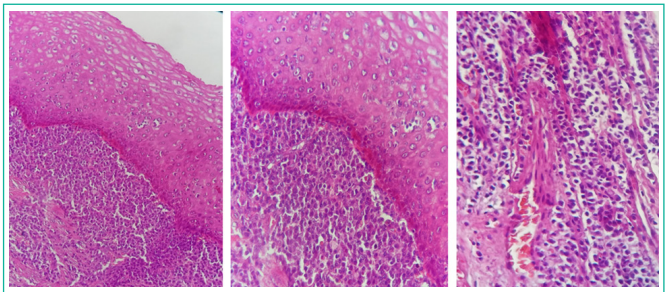

Figure 5: Laryngeal biopsy, showing mononuclear cellular infiltrate composed of plasma cells under a regular Malpighian mucosa.

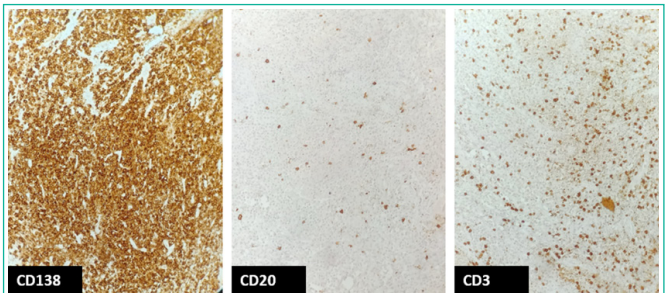

Figure 6: Selected immunohistochemical stains demonstrated in cells. Positive staining for CD138 and negative staining for CD20, CD3.

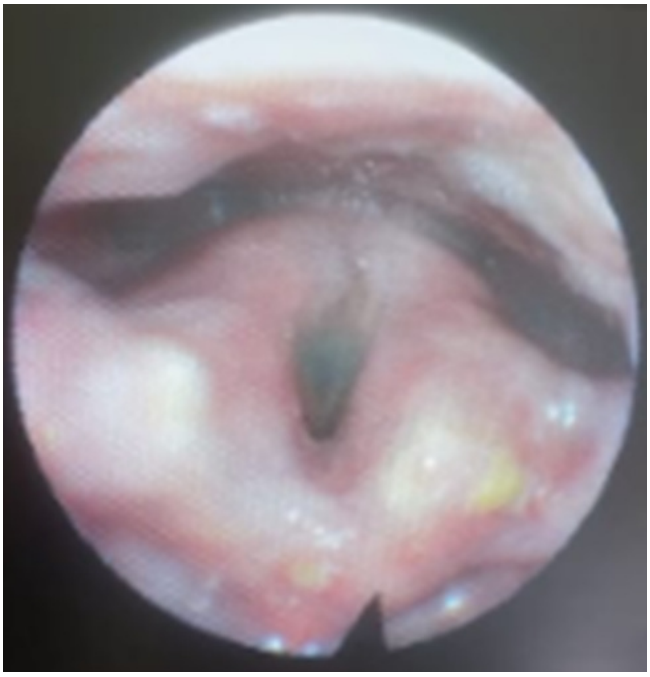

Figure 7: Indirect laryngoscopy 3 months after surgery showing good improvement in the laryngeal tract.

Discussion

To the best of our knowledge, only 11 cases of upper aerodigestive tract mucosal plasmacytosis have been reported in the international literature: seven males, four females, aged 40—67 years. Ferreiro et al. reported a series of nine patients, including seven with multiple locations (larynx, pharynx, lips, tongue and nose) and two with isolated laryngeal location [3]. Lee et al. reported a case of isolated LP in a patient with non-Hodgkin lymphoma. The most recent report was of oral and supraglottic mucosal plasma cytosis [4]. In most (8/11) of these cases, several regions were involved, with associated symptomatology. Only three patients, other than the present case, had isolated laryngeal (notably, supraglottic) involvement. Reported clinical signs (hoarseness, dyspnea, dysphasia, oral pain) vary with location [5]. Flexible laryngoscopy typically finds a verrucous or "cobblestone" mucosal aspect [3]. One patient, with non-Hodgkin lymphoma, showed vocal fold paralysis, but this was more strictly related to the lymphoma. The contribution of CT and MRI remains a subject of debate. Biopsy was systematic in the literature reports. Definitive histologic diagnosis found homogeneous inflammatory infiltrate, mainly composed of polyclonal plasma cells and lymphocytes [6,7]. Although there are no pathognomic criteria for diagnosing upper aerodigestive tract mucosal plasmacytosis, Ferreiro et al. described a characteristic set of clinical and histological criteria: mucosal hyperplasia with a "cobblestone" aspect, psoriasiform epithelial hyperplasia, dyskeratosis, and distinct plasmacytosis [3]. This description matches the present clinical and histological observations very well. Lee et al., however, do not consider these lesions as being characteristic [4]. There are no specific clinical signs other than endoscopic aspect. A pre dominance of regular non-tumoral polyclonal plasma cells in the infiltrate rules out myeloma and extramedullary plasma cytoma. In difficult cases, differential diagnosis may concern lymphoma. If the clinical aspect is suggestive of infection, absence of Mikulicz's spumes cells and negative culture rule out Klebsiella infection or rhinoscleroma. Other infectious etiologies are ruled out on Warthin-Starry and Gram staining, protein electrophoresis, syphilis serology and bacterial and/or mucosal culture. Congo red staining should be performed to rule out amyloidosis [3,8].

The etiology of laryngeal plasmacytosis remains largely unknown and has been rarely reported except in conjunction with oral plasma cell gingivitis [9]. Associating with immune-mediated disease, long-term denture wear, and smoking has been suggested [10.11]. It is notable, however, that the patient in this case has no known history of these suggested risk factors. The clinical presentation of chronic partial upper airway obstruction and histologic diagnosis of extensive subepithelial plasma cell infiltrate and proliferation are consistent with the patient's presentation in this case.

There is a lack of consensus on the management of this condition. Topical, intra-lesional, and systemic corti costeroids are used most often, but the results are inconsistent, and many patients experience a chronic waxing and waning of their disease [5,10]. Other treatment options, including immunosuppressive agents, antibiotics, irradiation, ablative therapy, and surgical excision, have been described with varying success [5,10]. Nebulized cortico steroids for laryngeal MMP involvement have not been described but were chosen as a treatment in this case given their well-established safety profile and efficacy for improving general airway inflammation [2]. Corticosteroids act to reduce inflammation through the translation of glucocorticoid receptor (GR) dependent genes and direct interactions with nuclear factor-kappa B to inhibit the production of cytokines and other pro-inflammatory molecules [2]. Intranasal corticosteroids have been shown to downregulate inflammatory cell recruitment and infiltration, which provided further rationale for the use of nebulized budesonide in this case of laryngeal plasmacytosis.

Conclusion

Laryngeal plasmacytosis is a rare benign pathology of unknown etiology, in which the clinical aspect is completely non-specific.

Diagnosis proceeds by elimination, and should be considered in case of polyclonal plasma cell infiltration of the submucosa [5].

Treatment is purely symptomatic; no effective medical treatment has as yet been reported [3,12].

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