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IP Journal of Otorhinolaryngology and Allied Science

Journal homepage: https://www.joas.co.in/

Case Report

Isolated fungal epiglottitis: A rare presentation

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ARTICLE INFO

Article history:
Received 06-06-2022
Accepted 05-08-2022
Available online 07-10-2022

Keywords:
Candidiasis
Fungal epiglottitis
Odynophagia

ABSTRACT

Introduction: Primary fungal epiglotitis is the isolated fungal infection of the epiglottis, without affecting the other body parts such as the larynx, lungs, pharynx, and oral cavity. It is an extremely rare clinical entity. Very few cases could be found in world literature. Pain throat and/or odynophagia are common clinical presentations. It can be secondary to inhaled steroid therapy which is usually mild.
Case Presentation: In this article, we introduced a rare case of fungal epiglotitis in a 66 years old immunocompetent male presented with odynophagia without any other symptom or sign. In endoscopic laryngoscopy, using a 70^0 scope, a white lesion on swollen epiglottis was seen and rest of larynx was normal. No cervical lymphadenopathy was seen. Laryngeal area was tender on deep palpation. Patient was put on antibiotics and anti-inflammatory agents. There was no response even after 24 hers of starting treatment. Patient was put on empirically oral antifungal treatment and he improved in 6-8 hrs.
Conclusion: Diagnosis of fungal epiglotitis was clinical, based on patient's history, signs and sypmtoms. Signs included the inflammatory changes of the epiglotis and candida whitish discoloration of it. There was rapid and obvious improvement in condition of the patient, once started on oral antifungal treatment.

1. Introduction

Epiglottis is an acute inflammation of the supraglottic region of the oropharynx with inflammation of the epiglottis, vallecula, arytenoids and aryepiglottic folds. This condition was first described in the nineteenth century, however it was accurately defined by Le Mierre, a French physician, in 1936. Acute epiglottitis in adults is also referred to as supraglottitis as the inflammation is generally not confined to the epiglottis but can also affect the supraglottic structures such as the pharynx, uvula, base of tongue, aryepiglottic folds and false vocal cords. Respiratory fungal infections are usually found in immunocompromised individuals who have received either broad-spectrum anti-microbial therapy or long-term steroid therapy or have predisposing factors, such as underlying chronic diseases, granulocytopenia, diabetes mellitus, and mechanical, chemical, or thermal injury to the mucosal barrier. These infections are often present as a part of bronchopulmonary fungal infections, and their isolated and primary involvement of epiglottis is highly uncommon. Vrabec DP[1] has also observed that isolated laryngeal candidiasis is infrequently recognised and poorly documented It has been described in patients. We report a case of isolated epiglotic candidiasis in an immunocompetent individual, who presented mainly with odynophagia without any other systemic sign or symptom, to highlight the importance of high clinical suspicion of this condition and provide a review of the relevant literatures.

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https://doi.org/10.18231/j.ijoas.2022.022

2. Case Report

A 66 years old male presented with odynophagia without any other systemic symptom or sign for 3-4 durations. It was mild to start with and progressive in nature. There was no other complaint. As his complaints were disproportionate to his clinical presentation and he insisted on having severe odynophagia, an endoscopic laryngoscopy, using a 70^0 scope was done. A white lesion on swollen epiglottis was seen and rest of larynx was normal. No cervical lymphadenopathy was seen. Patient was hypertensive and diabetic for last 10 years, and was on regular treatment. Laryngeal area was tender on deep palpation. His routine blood investigations revealed acute infection (elevated white blood cell count with predominance of neutrophils), ESR was 90 and FBS level was 137mg%. Chest X-ray was normal. Al the viral markers were negative. Patient was hospitalised and put on I/V fluids, I/V antibiotics and anti-inflammatory agents, keeping in mind that white lesion could be candidiasis. There was no response even after 24 hrs. of starting treatment. Patient was put on empirically oral antifungal treatment and preparations for D/L brushing or biopsy were started. But the patient improved considerably in next 12 hrs. Hence plan of any other invasive procedure was dropped. His blood sugar level was further controlled as per treatment by medical specialist. He started taking semisolid on third day of admission and all the treatment was continued for next seven days. His subsequent laryngoscopies revealed improvement as well. He was discharged on 9^{th} day with antifungal for next seven days. On follow up, he is asymptomatic for last 6 months.

3. Discussion

Haberman RS et all[2] probably reported first case of candida epiglottitis in a healthy, non-debilitated patient. They have mentioned that literature search did not produce a previously reported case. Even now, we could not found any other case of isolated involvement of epiglottis by candida in apparently healthy person, making this case a rare presentation.

Sharma N et all[3] have presented a case of Candida Epiglottitis in an adolescent patient with human immunodeficiency virus infection and observed that atypical organisms can cause epiglottitis in immunocompromised adolescents and they can have mild symptoms. Fungal pathogens must be considered in such cases. Even in their immunocompromised patient, they observed that symptoms significantly resolved with appropriate treatment within 2 days. Similar was the scenario in our case where the patient improved within 12 hours of starting oral anti-fungal treatment.

Beil EP et all[4]reported a case of a case of acute Candida epiglottitis in an otherwise healthy 4-year-old child and observed that it is very rare to find this condition in immnocompetent patients. They observed that the disease is commonly found in the patients who are immunocompromised due to various problems including human immunodeficiency virus disease and lymphoma and leukemia etc.

Walsh TJ et all[5]observed that Candida seldom had been reported to be a cause of epiglottitis. They presented a series of three patients with this condition but all patients were granulocytopenic. They suggested that fungal epiglottitis should be considered, especially in immunocompromised patients with symptoms of refractory pharyngitis. As per their study, Candida epiglottitis occurred either as a localized infection, as a source of Candida bronchopneumonia, or as a manifestation of disseminated infection. But our case is unique as it was isolated involvement of epiglottis.

Ng HL et all[6]in their retrospective study of 106 patients have considered direct visualisation of the inflamed epiglottis the gold standard of diagnosis. Of these patients only seventy-one patients had a lateral neck radiograph performed and 55 showed the classical thumb sign. Ninety-nine of their patients did not require any airway support during their illness. As our patient never had difficulty in respiration and laryngoscopy revealed only epiglottitis, we reserved the option of lateral neck radiograph to minimise the x-ray exposure. They further stressed that as active airway intervention is not without risk, so they recommended a conservative approach with endotracheal intubation in selected cases if needed.

Ibrahim J I et all[7] has proposed that biopsy and swap cultures were not needed for diagnosis in fungal laryngitis cases due to the resolution of the infection before the expected time of biopsy results. They have suggested that with high clinical suspicion and laryngoscopy details, there is no need for invasive procedure which have potential complications. Their observation was supported by the fact that Candida responds extremely well to treatment, which makes the option of a biopsy unappealing.

Wong KKet all[8]in their retrospective review of literature studied has observed anatomic involvement of larynx. Their, 18 patients (33%) showed Candida involvement in all three anatomic locations: the hypopharynx, the supraglottis, and the glottis. In 15 patients (28%), the Candida was isolated to the glottis. The remaining patients showed subglottic and glottic involvement. This makes our case a unique one as there was only involvement of epiglottitis.

Liu YC et all[9]has observed in their study that candida, aspergillosis, cryptococcal, histoplasmosis, and Blastomycosis are the common fungi causing laryngitis.

In their study of seven patients of fungal laryngitis, Swain SK et all[10] found that the common clinical presentations of fungal laryngitis are variable and could be hoarseness, dysphagia, dysphonia, stridor, odynophagia,

and respiratory distress. They further recorded that even in immunocompromised patients with noninvasive lesions, first-line treatment is the oral antifungal agent and the treatment duration ranges from 1 week to 1 month depending on the extent of the clinical improvement.

Ravikumar A et all[11] suggested in their article that certain conditions affect the mucosal barriers and predisposes the person to fungal infection. They cited prior radiotherapy, gastroesophageal reflux disorders, inhaled corticosteroids, smoking, and trauma as such conditions. They also observed that usual mode of presentation in an immunocompromised patient is with the change of voice, dysphagia, odynophagia, dyspnoea.

In their retrospective chart review analysis of 93 patients during 1999-2016, Ibrahim J I et all[7] found that steroid inhalers were the main risk factor that is closely related to the occurrence of fungal infection of larynx. They further opined that other risk factors include the use of oral steroids, nasal steroids spray, broad spectrum antibiotics, diabetes mellitus, smoking, radiotherapy or chemotherapy exposure, acid reflux disease with symptoms like heart burn. They mentioned that trauma to larynx during intubation can be one of the risk factor. They proposed that having one or two vocal cord palsy that affect the normal touching between the vocal cords, may predisposes the possible growth of candida over vocal cords.

Valente P et all[12] reviewed various articles available on Pubmed on laryngeal candidiasis and observed that females are more commonly affected. Their observation was similar to Ibrahim J I et all[7] who also found that fungal infection of larynx affects females more than males. Valente P et all[12] further observed that diagnostic approach to fungal laryngitis remains controversial, since some authors recommend prompt lesion biopsy and others rely on empirical antifungal treatment that showed effective results regarding symptoms and lesions resolution. But they concluded that the conservative diagnostic approach with antifungal treatment provide effective outcomes and must be tried first.

4. Conclusion

Diagnosis of fungal epiglottitis in this case was clinical, based on patient's history, signs and symptoms. Signs included the inflammatory changes of the epiglottis and candida whitish discoloration of it. There was rapid and obvious improvement in condition of the patient, once started on oral antifungal treatment. As the patient's general condition was very good without any sign of toxicity or compromised respiration, the take home lesson is that no complaint of the patient to be ignored. Even in literature, it is found that few cases may present as unresolving throat pain. Laryngoscopic examination should be done more often to diagnose such conditions which have potential to turn serious. Candidiasis respond very well to oral antifungals and must be given trials for being less toxic and free availability.

5. Source of Funding

None.

6. Conflict of Interest

None.

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Cite this article: Singh H, Sharma D, Mahajan M, Saini A, Guleria TC, Ranot B, Kaur R. Isolated fungal epiglottitis: A rare presentation. *IP J Otorhinolaryngol Allied Sci* 2022;5(3):87-90.