Unusual association of diseases/symptoms

Aspergillus epiglottitis in a non-immunocompromised patient

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Summary

The authors present a case that highlights an atypical example of a fungal epiglottitis in an otherwise well adult patient with no medical history of fungal infection or an immunocompromised state. As current medical literature presents this fungus as only manifesting in the immunocompromised, the authors suggest, by this case report, that fungal pathogens be considered as a potential cause of epiglottitis in non-immunocompromised patients.

BACKGROUND

Acute epiglottitis is a condition that needs to be treated in an effective and time efficient manner by senior physicians. This report demonstrates the possible consequences of underestimating the seriousness of this situation and provides an example of a failed initial encounter followed by the proper investigation and treatment.

As current medical literature presents fungal epiglottitis only manifesting in immunocompromised patients, this case represents a patient with no known compromise to her immune system and no preceding illnesses.

CASE PRESENTATION

This case was referred to the on-call ear, nose and throat (ENT) team from her general practitioner with a 4-day history of severe sore throat, inability to eat, fevers associated with flushing, occasional bouts of dyspnoea and a 3-day history of dysphonia. On arrival in Accident and Emergency (A&E), the patient's initial blood pressure and pulse were within normal range, temperature of 37.6°C, with an oxygen saturation of 99% and no signs of respiratory distress. She was taken into a specialised ENT examination room in A&E. During history taking, the patient became very distressed and began to gasp for breath and eventually completely obstructed. Oxygen was immediately placed and she was brought into the resuscitation bay. Anaesthetists were urgently sent for, however, shortly afterwards, the patient was able to breathe again and her saturation came up to 100% on 5 l/min of oxygen. The patient was examined with a flexible nasopharyngeal endoscope with the anaesthetists present. The supraglottic region looked extremely inflamed and oedematous on endoscopic examination. The decision was made to commence the patient on steroids, cefotaxime and metronidazole (on advice of the consultant microbiologist), humidified oxygen and nebulised epinephrine. She was subsequently admitted to intensive care unit (ICU) for observation. Due to the larynx remaining erythematous with minimal change to oedema, the patient was eventually intubated in the operating theatre with the presence of an ENT surgeon and theatre

staff on standby for potential emergency tracheostomy. The further ICU stay after the successful intubation was uneventful.

The patient was extubated 16 h later. She was producing purulent sputum and a sample was sent to microbiology, along with a portion of the endotracheal tube. Heavy growth of *Aspergillus fumigatus* was cultured 3 days later from both the sputum sample and endotracheal tube. Since the patient was improving on cefotaxime and metronidazole, the consultant microbiologist advised us to not introduce any antifungal medications at the time.

The patient gradually continued to improve. Her voice returned to normal around the sixth day of admission and she was gradually building up her diet. Repeat examination by nasopharyngeal endoscope showed the supraglottic region had almost returned to normal. The patient was discharged home on the ninth day from admission.

OUTCOME AND FOLLOW-UP

The patient was discharged on her ninth day from admission as she had greatly improved clinically and examination by nasopharyngeal endoscope showed the supraglottic region had almost returned to normal. Follow-up appointments showed the patient made a full recovery.

DISCUSSION

Epiglottitis is an acute inflammation of the supraglottic region of the oropharynx with inflammation of the epiglottis, vallecula, arytenoids and aryepiglottic folds. Initially starting with inflammation and oedema between the base of the tongue and the epiglottis, it may progress to complete blockage of the trachea leading to complete airway obstruction.¹ First described in the nineteenth century, it was accurately defined by Le Mierre, a French physician, in 1936.

Acute epiglottitis in adults is often 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. It may progress to complete blockage of the trachea.²

Despite its increasing incidence in recent years, epiglottitis remains an uncommon disease with a mean annual incidence of 3.1 per 100 000 in the UK.³ There has been a steady increase in incidence from 0.88 per 100 000 in 1986–1990. Historically reported as a disease in children aged 2–6 years, it can also occur in adults. *Haemophilus influenza* type B accounted for 75–90% of all cases in the prevaccination era.⁴ However, following the introduction of the *H influenza* type B vaccine in 1985 and the subsequent reduction in *H influenza* associated infections including acute epiglottitis, the epidemiology of epiglottitis has shifted to other bacterial, viral and fungal pathogens. *Haemophilus parainfluenza*, *Streptococcus pneumoniae* and group A *Streptococci* are the most common pathogens in acute epiglottitis, but other organisms including *Staphylococcus aureus*, *Mycobacteria*, *Escherichia coli*, *Klebsiella pneumoniae*, *Neisseria meningitidis*, *Moraxella catarrhalis*, *Pseudomonas* species and *Pasturella multocida* are known to cause the disease.⁵ Viral pathogens include Herpes simplex virus and infectious mononucleosis, while fungal pathogens have been reported in immunocompromised patients, namely *Candida* and *Aspergillus*.⁴ Alternatively, epiglottitis may occur following mechanical injury such as the ingestion of caustic material or the inhalation of hot objects, smoke or vapours.⁶ Epiglottitis following illicit drug use has been described as a result of the accidental inhalation of the heated substances.⁷ The causal organism or factor is often less identifiable in adults than in children.

Acute epiglottitis in adults is a rare but potentially lethal disease which usually presents with symptoms of sore throat, dysphagia, odynophagia and hoarseness of voice. Symptoms such as drooling, dyspnoea and stridor are rarely reported in adults and some patients may not report any symptoms or signs of airway obstruction.⁸ Such patients may never seek medical attention and therefore epiglottitis in the adult population may be significantly underdiagnosed given the huge variability in disease presentation and progression.

Diagnosis is essentially clinical and suspicion should be raised if there is any pharyngeal inflammation. Occasionally there may be swelling of the uvula, however, the definitive diagnosis is usually made on direct laryngoscopy, using a nasopharyngeal scope, or on indirect laryngoscopy, where there is typically diffuse swelling of the aryepiglottic structures.⁶ Direct and indirect laryngoscopy should be performed in the appropriate resuscitation area, where there is access to airway management equipment, as both are potentially stimulating procedures which may precipitate sudden loss of a patent airway. Therefore, it is empirical that an anaesthetist and senior ENT specialist are present should intubation or an emergency tracheostomy be required. Lateral neck radiography, performed once the airway has been successfully secured, may show the classical ‘thumb print sign’ due to thickening and oedema of the inflamed epiglottic region.⁸ Ducic *et al* introduced the ‘vallecula sign’ in 1997 to improve the diagnostic accuracy of soft-tissue radiographs.⁹

Once the diagnosis has been made, immediate management involves assessment of the airway, although the role of airway intervention in adults is controversial. There are authors who prefer a conservative management with antibiotics, corticosteroids, humidified oxygen^{10–13} and nebulised epinephrine. Others prefer an aggressive

airway management with early intubation.¹⁴ A mortality of 1–7%^{15–17} among adults has been described, however, 17.6% mortality has been described in patients with acute respiratory obstruction.^{18 19} Prompt initiation of intravenous antibiotics should be started as per the local protocol. Blood cultures should ideally be taken prior to the administration of antibiotics; however, they should not delay airway management. The role of steroids and nebulised epinephrine, although widespread, remains controversial.^{20 21}

A fumigatus is a saprophytic fungus found in nature that has the potential to cause life-threatening infections in humans. *A fumigatus* forms spores, called conidia that become airborne due to its 2–3 µm size and are easily inhaled and typically destroyed by the body’s defences within the lungs. The human body is able to provide a superb environment for this fungus since the internal milieu of humans lies within the optimal range of temperature, pH and nutritional sources for *A fumigatus*’ growth and reproduction, hence studies have shown that this pathogen is the leading opportunistic mould infection with mortality exceeding 50% in high-risk groups.²²

Previous case reports have documented *Aspergillus* infection of the epiglottis in a 42-year-old homosexual male with confirmed HIV positive antibody, who presented with extensive oropharyngeal candidiasis despite treatment with oral itraconazole. He subsequently developed extensive ulceration of the epiglottis with *A fumigatus*.²³ The second case is of a 21-year-old woman with acute lymphocytic leukaemia who also developed epiglottitis with *Aspergillus* grown on culture of throat specimen.²⁴

Learning points

- Being presented with this critical patient that needed immediate intervention, it was unfortunate that blood cultures were not taken as priority was placed on transportation to intensive transport unit, intubation and antibiotic administration. It is fortunate that we were able to isolate the cause of epiglottitis from sputum and endotracheal tube cultures and that blood cultures may not have made an impact on the course of treatment. As previously mentioned, blood cultures should always be taken prior to antibiotic/antifungal therapy so as to help delineate the pathogen and guide therapy.
- A potentially disastrous mistake was made in this case at the initial encounter with the patient in hospital. Any patient with suspected epiglottitis should be interviewed in a well-monitored area with adequate support. Although the patient did not seem to be very ill in the beginning, the case report shows how events quickly escalated. Senior ENT and anaesthetists should be immediately made aware of any patients with the above mentioned symptoms and should be readily available for urgent intervention.
- As current medical literature presents this fungus as only manifesting in the immunocompromised we suggest that fungal pathogens be considered as a potential cause of epiglottitis in the non-immunocompromised patient.

Competing interests None.

Patient consent Obtained.

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