CASE REPORT

Multipathogenic necrotising supraglottitis in an immunocompetent patient

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SUMMARY

Supraglottitis is a potentially life-threatening condition. It is now uncommon due to the *Haemophilus influenzae* type B vaccination and is more recently caused by *Streptococcus pneumoniae*, *S. pyogenes*, *H. influenzae* non-type B, *H. parainfluenzae*, *Staphylococcus aureus* and *Pasteurella multocida*. Very rarely, it can cause necrotising supraglottitis/epiglottitis, and this has been reported in immunocompromised individuals. We present a unique case of multipathogenic supraglottitis causing laryngeal fibrinoid necrosis in an immunocompetent patient. During his admission, the patient was critically unwell and required surgical intervention and tracheostomy. However, he made a full recovery with no persisting morbidity. We believe that this was owed to the aggressive antimicrobial therapy, timely surgical management of the disease process and the patient's immunocompetency.

BACKGROUND

Supraglottitis in adults is an uncommon infection with the potentially life-threatening sequelae of enlargement of the epiglottis and false cords, which can lead to complete airway obstruction.¹ Until recently, *Haemophilus influenzae* type B was the most common aetiological micro-organism. Its incidence has greatly reduced since the introduction of national vaccination programmes.² Increasingly, supraglottitis is being caused by *Streptococcus pneumoniae*, *S. pyogenes*, *H. influenzae* non-type B, *H. parainfluenzae*, *Staphylococcus aureus* and *Pasteurella multocida*.³ It has been associated with necrotising epiglottitis in the immunocompromised population with only one case reported in a healthy individual.⁴ We present a unique case of necrotising epiglottitis in a healthy adult with mixed viral and bacterial pathogens. In doing so, we discuss the importance of treating patients with multipathogenic supraglottitis (and other multipathogenic infections) aggressively due to the potential for laryngeal fibrinoid necrosis and rapid, clinical deterioration. This case is important as it highlights the value of obtaining surgical tissue for analysis in cases of multipathogenic infections so that antimicrobial therapy can be optimised. We also note the limitations of diagnostic imaging and the importance of maintaining an open mind until surgical intervention.

CASE PRESENTATION

A 38-year-old man presented to our emergency department following a 10-day history of symptoms

suggestive of upper respiratory tract infection, which over the preceding 24 hours had progressed to sore throat, drooling, dysphonia and stridor. He had no medical history or relevant drug, family or social history. The primary survey revealed biphasic stridor and dyspnoea. He was tachypnoeic, tachycardic and normotensive, with normal chest auscultation. Neck examination revealed tender laryngeal structures. Intraorally, the palate and oropharynx were erythematous and oedematous. Fibreoptic nasendoscopy (FNE) revealed airway compromise due to inflammatory epiglottic and supraglottic oedema, along with left-sided laryngopharyngeal swelling. Given these specific findings, the patient was treated as having severe infective supraglottitis. He was intubated via the awake transnasal fibreoptic method, due to concerns regarding the severity of the visualised supraglottic swelling, his clinical condition and the potential for further deterioration. He was subsequently admitted to our intensive care unit.

INVESTIGATIONS

Routine biochemistry investigations revealed a high neutrophilia and C reactive protein (CRP). Throat swabs and blood cultures were taken on admission. The patient's chest X-ray on admission was unremarkable.

TREATMENT, FURTHER INVESTIGATIONS AND CLINICAL COURSE

From admission, the patient was initially treated with regular intravenous ceftriaxone and intravenous metronidazole as per our local antimicrobial guidelines for supraglottitis, prior to any culture results being available. He was also given regular intravenous dexamethasone. After 3 days of level 3 care, he was transferred to the level 0 ENT ward. Antimicrobial treatment was then rationalised to benzylpenicillin and oseltamivir in light of the admission throat swab eventually culturing a heavy growth of group A β -haemolytic *Streptococcus* and influenza A virus. However, the patient then began to show systemic signs of acute illness, including fever and tachycardia. Repeat examination with FNE revealed mucosal slough on the left-sided aspect of the epiglottis, which over the following 24 hours progressed and coalesced to an ulcerated and exudative appearance, coupled with the return of his original presenting symptoms. A subsequent CT scan reported possible left parapharyngeal abscess and laryngeal abscess (figure 1). The patient

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Figure 1 Axial and sagittal views of the patient's CT neck scan, depicting (a) the process at the level of the epiglottis, (b) the process at the level of the hyoid, (c) the process at the level of the vocal cords and (d) the severity and level of airway compromise.

was taken to theatre for emergency microlaryngoscopy and tracheostomy. The microlaryngoscopy showed membranous, necrotic slough over the left hemilarynx, which extended to the right false cord; however, no pus was found (figure 2).

The necrotic area was debrided and sent for further urgent microbiology and histology. Postoperatively, the patient was readmitted to the intensive care unit. Based on the available evidence of macroscopic necrosis at surgery and previously cultured group A β -haemolytic *Streptococcus* (throat swab), antimicrobial therapy was changed to intravenous ceftriaxone, clindamycin and metronidazole on the recommendation of our microbiology colleagues.

The histology was soon reported as 'fibrinoid necrotic tissue with inflammatory cells and overlying reactive squamous

Figure 3 Histology slides of left hemilaryngeal, debrided tissue. (a) Low-power light microscopy. (b) High-power light microscopy.

epithelium, no evidence of malignancy or vasculitis; conclusion=necrosis' (figure 3).

The aforementioned antimicrobial therapy continued until eventual enrichment broth culture from the operative samples grew *Enterococcus faecalis* (sensitivities, teicoplanin and vancomycin). Antimicrobial therapy was further rationalised to intravenous teicoplanin, clindamycin and metronidazole. This rationalisation was justified by *Enterococcus faecalis*' intrinsic resistance to cephalosporins, the sensitivities we established, concomitant streptococcal throat culture, and the relative severity of the infection. In total, he received 2 weeks of intravenous antibiotics and was ultimately discharged with a 10-week course of oral clindamycin only.

OUTCOME AND FOLLOW-UP

Two days after his operation, the patient was discharged back to the ENT ward from the intensive care unit. Since this episode, the patient has thankfully made a complete recovery. He no longer has a tracheostomy, his larynx is intact and he has suffered no lasting morbidity or disability.

DISCUSSION

Fibrinoid necrosis, visible by light microscopy, is typically a result of immune reactions involving deposition of complexes consisting of antibodies and antigens. It usually occurs in the arterial walls of immunologically mediated diseases such as polyarteritis nodosa, when fibrin leaks out of vessels and combines with the immune complexes to form a bright pink, amorphous appearance.⁵ Fibrinoid necrosis of the larynx following supraglottitis has been reported in the literature in relation to the immunocompromised population. The most recent case to develop in an immunocompromised patient was caused by a non-toxicogenic *Corynebacterium diphtheria* infection, and the paediatric patient was found at presentation to be pancytopenic due to acute lymphoblastic leukaemia.⁶ However, only one case has been reported in an immunocompetent patient in whom α -haemolytic *S. pyogenes* was the only pathogen isolated.⁴

Figure 2 Intraoperative views of the supraglottis, obtained via microlaryngoscopy.

The present case reports an immunocompetent patient with a multipathogenic infection, including group A β -haemolytic *Streptococcus*, *E. faecalis* and influenza A virus. *E. faecalis* is a known commensal of the gastrointestinal tract and is associated with nosocomial infections. Due to its resistance, it is usually difficult to treat and can often cause systemic complications of infection.³ This may have contributed to the severity of this infection; however, it remains speculation until the case numbers expand. In this case, timely surgical intervention leads to preservation of the majority of the laryngeal skeleton.

Normally, necrotising supraglottitis is seen in immunocompromised individuals. However, in this patient, full blood count showed an appropriately reactive neutrophilia with elevated CRP (11.5 and 238 on admission, respectively). There was no evidence of neutropenia or leucopaenia throughout admission. Full immunology screen, including immunoglobulin levels, was organised to look for evidence of deficient immunological function. This screen was prompted as the supraglottic biopsies were reported as a non-malignant, non-vasculitic, inflammatory necrosis; the subsequent immunology screen was normal. Finally, regarding investigations, radiological appearances in cases such as this can prove difficult to interpret and may differ from the intraoperative findings (figure 1).

Following investigation, we have no reason to doubt this patient's immunocompetency. Although he ultimately survived the condition, he was certainly critically unwell, requiring intensive care admission. The fact he survived may partly be attributed to immunocompetency, as this condition often leads to lethal complications in immunocompromised patients. Survival was also likely facilitated by good operative source control. Furthermore, the cultured *E. faecalis* may have in fact been a contaminant.

Importantly, necrotising supraglottitis has been observed in the presence of multiple pathogens, and given the presentation above and the literature, it is important to identify the presence or absence of viral involvement in addition to bacterial cultures in both the immunocompetent and immunocompromised patient. Patients presenting with multipathogenic infections are usually immunocompromised. This is important and requires investigation as the natural history of such cases can be of rapid, life-threatening deterioration despite appropriate antimicrobials. Equally, one should consider the potential for multipathogenic infections in certain at-risk groups (the elderly, malignancy, polypharmacy) and target antimicrobial investigation and treatment accordingly.

In the context of upper airway infections, this case highlights the importance of an appropriate management plan in terms of prompt, reactive escalation to the patient's changing clinical state. This included securing the airway early, timely

surgical debridement and seeking regular microbiological advice when confronted with a multipathogenic disease process. This is especially relevant in the era of ever-increasing antimicrobial resistance. Cases where viruses are identified alongside bacterial infections should be managed as above due to the aggressive natural history displayed. Indeed, this approach to multipathogenic infections resulting in necrosis could be applied to conditions other than supraglottitis.

Learning points

- ▶ Rapid, life-threatening deterioration can occur in upper airway infections (eg, supraglottitis) despite the normally adequate response to antimicrobial management.
- ▶ Decreased immune function and, therefore, potential for deterioration should be suspected in numerous circumstances (eg, multipathogenic infections, the elderly, malignancy, polypharmacy).
- ▶ The unique multipathogenicity underlying this patient's supraglottitis is likely to have contributed to the severity of his clinical course.
- ▶ With increasing antimicrobial resistance, aggressive and timely management (hospitalisation/intubation/surgery) of these patients with common upper airway infections should be considered.

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REFERENCES

- 1 McGill TJ, Jones D. *Non-malignant conditions of the paediatric larynx*. Bath: Oxford University Press, 1998:p598.
- 2 Senior BA, Radkowski D, MacArthur C, et al. Changing patterns in pediatric supraglottitis: a multi-institutional review, 1980 to 1992. *Laryngoscope* 1994;104:1314–22.
- 3 Gorbach SL, Bartlett JG, Blacklow NR. *Infectious disease*. 3rd edn. Taunton: Quebecor World, 2004:p460.
- 4 Klcova J, Mathankumara S, Morar P, et al. A rare case of necrotising epiglottitis. *J Surg Case Rep* 2011;2011:5.
- 5 Kumar V, Abbas AK, Aster JC. *Robbins Basic Pathology*. Philadelphia: Elsevier, 2013:p11.
- 6 Lake JA, Ehrhardt MJ, Suchi M, et al. A case of necrotizing epiglottitis due to nontoxicogenic *Corynebacterium diphtheriae*. *Pediatrics* 2015;136:e242–e245.

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