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CLINICAL RECORD

A new era in supraglottitis? An isolated UK case of supraglottitis secondary to *Neisseria meningitidis*

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Abstract

Objective: We report the first UK case of supraglottitis secondary to *Neisseria meningitidis*.

Method: Case report with review of the current literature on supraglottitis and its aetiology.

Results: An 89-year-old woman was referred with worsening symptoms of dysphagia, hoarseness and neck discomfort. After nasopharyngoscopy and neck X-ray, supraglottitis was diagnosed. Prompt treatment comprised nebulised adrenaline, oxygen therapy and intravenous antibiotics. Microbiology samples grew *N meningitidis*, a notifiable disease in the UK. Public health officials were informed, and full precautions and prophylactic treatment initiated for those at risk. The patient made excellent progress and was discharged several days later.

Discussion and conclusion: Supraglottitis occurs in <4 per 100 000 population. Following a successful UK childhood immunisation programme, most cases occur in adults. Supraglottitis secondary to *N meningitidis* is exceptionally rare, with only seven other reported cases worldwide. Morbidity is exceptionally high; over 60 per cent of patients require airway intervention. To our knowledge, this is the first reported UK case of supraglottitis secondary to *N meningitidis*. This case highlights the important clinical, diagnostic and therapeutic interventions required to prevent complications associated with this potentially fatal condition.

Key words: Epiglottitis; Laryngitis; Public Health; Neisseria Meningitidis; Supraglottitis

Introduction

Supraglottitis, a potentially fatal condition, consists of inflammation of the epiglottis and the surrounding tissues of the larynx. It is now more commonly seen in adults, with children less affected following the introduction of childhood vaccines.

Its aetiology includes infections, with *Haemophilus influenzae* type B infection being especially common.

Early clinical suspicion, with rapid assessment and treatment, is paramount to prevent the life-threatening complications associated with this condition.

We present the first UK case of supraglottitis associated with *Neisseria meningitidis*, including the initial assessment, examination and subsequent management.

Case report

An 89-year-old woman with a past medical history of dementia, metastatic breast carcinoma, hypertension and skin malignancy presented to her general practitioner with a one-day history of left otalgia, for which she was prescribed topical Gentisone® (gentamicin plus hydrocortisone). Her symptoms worsened, and she re-presented to her general practitioner the following day with a rapid onset of dysphagia to both solids and liquids, hoarseness, neck discomfort and 'gurgling' sounds in the back of the throat.

The patient, cared for by family members, was referred to the hospital medical team, where preliminary examinations were arranged, including blood tests, blood cultures and a soft tissue neck X-ray, prior to referral to an ENT specialist.

Initial recorded observations demonstrated a mild pyrexia of 37.6° Celsius with no haemodynamic compromise. Blood tests revealed a neutrophil leucocytosis (white cell count of 21×10^9, neutrophil count of 19×10^9) with a C-reactive protein (CRP) level of 290 mg/l. A classical 'thumb-printing' sign, indicating an inflamed epiglottis, was seen on the soft tissue lateral neck X-ray, with no other relevant signs.

On clinical examination, the patient appeared comfortable, with obvious audible secretions in the back of the throat. A putrid foetor oris was evident during a difficult oral examination, with dry mucous membranes seen.

In light of the patient's questionable stability, careful endoscopic nasopharyngoscopy was performed at the bedside. This revealed significant amounts of thick, putrid secretions in the valleculae and the piriform fossae, with inflamed arytenoids and epiglottis.

A diagnosis of supraglottitis was made and treatment instituted, consisting of nebulised adrenaline (1 in 1000), intravenous dexamethasone (4 mg thrice daily) and oxygen therapy. A rapid review by the anaesthetic team was arranged to assess airway patency (no compromise was found), and to ensure their awareness in case of subsequent airway deterioration. Intravenous antibiotics were changed, from the benzylpenicillin (1.2 g four times daily) and metronidazole (500 mg thrice daily) initially prescribed on admission, to cefuroxime (1.5 g thrice daily).

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The following day, preliminary results from the admission blood culture revealed a Gram-negative diplococcus. The patient's antibiotics[1] were changed again, to cefotaxime (2 g twice daily), to cover for potential *H influenzae* type B and *N meningitidis* infection; the latter was indeed confirmed the following day. The isolate was very sensitive to penicillin; thus, the patient was changed back to intravenous benzylpenicillin, to minimise the risk of developing *Clostridium difficile* infection.

Also on the day after admission, repeated endoscopic examination of the larynx revealed a reduction in secretions, although the supraglottic tissues were still inflamed. On day two post-admission, blood tests revealed a normal white cell count and a reduction in the CRP level.

The patient continued to do well, and was discharged five days post-admission.

As *N meningitidis* is a notifiable infection in the UK, public health officials were alerted and contact tracing (including healthcare workers) instituted, with administration of prophylactic antibiotics (ciprofloxacin 500 mg, single dose, plus rifampicin 600 mg, four doses). Staff who may have been exposed to aerosolised droplets from the patient within 24 hours of commencement of appropriate antibiotic therapy, without appropriate protection, were advised take prophylactic treatment.

At the time of writing, the patient continued to make good progress, with no signs of clinical deterioration.

Further serotyping of the *N meningitidis* strain showed it to be group Y.

Discussion

Acute supraglottitis secondary to *H influenzae* serotype B (Hib) infection was first recognised in the UK by an observant Home Office pathologist, Professor F E Camps,[1] whose meticulous post-mortem examinations of paediatric patients admitted with sore throat, pyrexia and difficulty in swallowing revealed this new clinical entity. His excellent paper,[1] presented to the Royal Society of Medicine, highlighted the clinical characteristics specific to this condition, and recognised that timely intervention could reduce mortality.[2]

Supraglottitis, an inflammation of the epiglottis, arytenoids and surrounding tissues,[3,4] is a recognised condition in adults and children, with an incidence of less than four per 100 000 population.[5] Children are now affected much less than adults, due to the UK immunisation policy offering all babies the combined diphtheria, pertussis and Hib vaccination during the first four months of life.[6] Although bacterial infections have been implicated in this condition,[7] other causes include trauma,[8] viruses,[9] chemicals,[10] recreational drugs[11] and chemotherapy.[12]

Patients often present with a combination of dysphagia, odynophagia, sore throat, hoarseness, and varying symptoms of systemic malaise, especially fever.[3,13] Due to the anatomical location of the inflammation, meticulous assessment and regular monitoring for deterioration is paramount, in order to prevent sudden respiratory catastrophe.[7] Some authors state that there are no reproducible clinical signs indicating potential airway compromise, whilst others suggest that a rapid clinical course (especially within the first 24 hours of the illness) is more predictive of a requirement for airway intervention, which includes tracheostomy and endotracheal intubation.[13–16] Shapiro *et al.*[4] have suggested that a less aggressive infection may occur, without airway signs, when aetiological factors other than Hib are involved.

Our patient had no respiratory compromise and a relatively slow progression of symptoms, and demonstrated *N meningitidis* bacteraemia on blood culture.

Neisseria meningitidis is a Gram-negative diplococcus which is carried asymptomatically in the nasopharynx in 5–20 per cent of the population.[17] It is a rare cause of supraglottitis, with only seven previous cases reported in the literature.[18–24] It is common worldwide, and has five major subgroups (serotypes A, Y, W, B and C), with serotypes B and C being especially prevalent in Europe and North America.[17] However, of the seven reported cases involving supraglottitis, four involved the Y serotype, two the B serotype, and one was undetermined.

All these seven cases involved adults, four women and three men, with ages ranging from 44 to 95 years. Four of the seven cases occurred in North America, one in Singapore[22] and another in Finland.[23] Ours is the first reported case of *N meningitidis* supraglottitis occurring in the UK. In all but two of the previously reported cases, the patient had evidence of respiratory compromise requiring some form of airway management, including tracheostomy (two patients) and endotracheal intubation (three patients). If we include our patient, this means that more than 60 per cent of reported cases have required some form of airway intervention, compared with less than 15 per cent of supraglottitis cases involving Hib infection.[13–16]

- **Supraglottitis is inflammation of the epiglottis, arytenoids and surrounding tissues**
- **An 89-year-old patient with *Neisseria meningitidis* supraglottitis is presented**
- **Rapid onset dysphagia, hoarseness, neck discomfort and gurgling throat sounds should alert the clinician to potential airway compromise**
- **Treatment should be started based on symptoms and early clinical signs, guided by microbiologists with local knowledge**
- **Oropharyngeal examination should only be undertaken by specialists, with full resuscitation equipment available**
- **Neisseria supraglottitis is rare; contact tracing and early treatment are important**

Infection with *N meningitidis* can result in serious morbidity due to systemic effects. However, interestingly, it causes only a localised inflammatory response in supraglottitis,[3] where it affects only the soft tissue of the larynx and pharynx.[4] Only two reported cases had evidence of inflammation spreading beyond the supraglottic region, most notably involving cellulitis of the soft tissues of the neck.[18,24] No cases, including our own, demonstrated any evidence of systemic meningococcal infection, especially meningism or septicaemia. Schwam *et al.* have suggested a possible explanation for such localised inflammation: host virulence factors within the supraglottis area limit interaction between the bacteria and the epithelial cells, thus preventing endothelial cell activation, which is more commonly associated with systemic spread and more serious complications.[24]

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Our patient received intravenous antibiotics, steroids and nebulised adrenaline, and did not exhibit any signs of respiratory compromise despite her delayed admission to hospital. However, supraglottitis is notorious for a rapid decline in patient status and high mortality rates, as first observed by Professor Camps in 1953.[1] Once this condition is suspected, rapid assessment and management are imperative.

We emphasise the importance of blood cultures in all patients suspected of supraglottitis, as their results enable targeted antibiotic therapy and also help prevent a potential public health disaster. Without blood culture results, antibiotic treatment cannot be rationalised to a narrow spectrum, and there is the risk of secondary infection in staff exposed to aerosolised droplets from infectious patients.

Further cases of this rare cause of supraglottitis should be documented, in order to further establish the true epidemiology of this condition.

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