*The Journal of Laryngology & Otology*
December 2003, Vol. **117,** pp. 995–997

# Relapsing acute adult epiglottitis following hypophysectomy

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**Abstract**
Adult epiglottitis is an uncommon disease that may become fatal because of sudden airway obstruction. Recurrent adult epiglottitis has been reported rarely in the literature. We present a case of relapsing epiglottitis in a patient with corticosteroid insufficiency secondary to pituitary surgery. It is the first case to highlight this infection occurring in a relapsing manner in a patient with pituitary-adrenocortical insufficiency on long-term steroid replacement therapy.

**Key words: Epiglottitis; Pituitary-Adrenal System; Glucocorticoids**

## Introduction

Acute epiglottitis is an uncommon but dangerous infection of the supraglottic larynx commonly caused by Haemophilus influenzae type b (HIB). Inflammation causes swelling of the supraglottis and predominantly the epiglottis.[1] This can rapidly cause life-threatening airway obstruction necessitating intubation or tracheostomy. After treatment with antibiotics the condition usually resolves promptly without complications or recurrence.

We report a case of recurrent relapsing acute epiglottitis in a patient who had had pituitary surgery and was receiving corticosteroid replacement therapy.

Recurrent acute epiglottitis in adults has rarely been reported in the medical literature. Following a comprehensive Medline literature search only seven reported cases were found. These cases describe epiglottitis recurring over a period of months to years. This case describes a patient with epiglottitis relapsing twice in less than three weeks.

Four of the previously reported cases have highlighted patients who had persistent impaired humoral immunity as demonstrated by decreased immunoglobulin levels.[1]

We have found no previously reported cases of patients with pituitary-adrenocortical insufficiency developing recurrent epiglottitis. This case raises the possibility of such a life-threatening condition affecting such patients and highlights the importance of maintaining adequate corticosteroid replacement therapy during their management.

## Case report

A 53-year-old lady presented to the accident and emergency department with a four-hour history of acute inspiratory stridor. She had complained of a sore throat and dysphagia for the preceding 24 hours.

Eight months previously she had undergone a transsphenoidal decompression of a craniopharyngioma followed by a course of pituitary radiotherapy. Following treatment she was given pituitary replacement therapy of 20 mg hydrocortisone in divided doses and 100 mcg thyroxine daily. She was otherwise fit and well, with no past medical or surgical history.

On admission she was distressed, pyrexial (39.7°C), tachycardic and tachypnoeic. There was audible inspiratory stridor and she was using accessory muscles of respiration. Oxygen saturation was 99 per cent on 10 litres/min of oxygen. Flexible nasendoscopy showed epiglottitis. The patient underwent emergency intubation and was transferred to the intensive care unit.

She was treated with intravenous co-amoxiclav and metronidazole, and her steroid dose was increased to 200 mg hydrocortisone daily in divided doses. Her white cell count was $5.5 \times 10^9$/L and her ESR 81 mm/hr. All other blood results were normal. *Haemophilus influenzae* type b (HIB) was cultured from blood cultures and was sensitive to co-amoxiclav and cefuroxime.

After six days repeat nasendoscopy revealed resolution of the epiglottic swelling and she was successfully extubated. The hydrocortisone was reduced back to her original maintenance dose, but the antibiotics were continued. Two days later she developed stertor without distress. She was apyrexial. Flexible nasendoscopy showed that the epiglottis was once again swollen and obstructing the glottis (Figure 1). After a failed intubation she had a surgical tracheostomy inserted. A repeat white cell count was raised at $20.7 \times 10^9$/L. Her antibiotics were changed to cefuroxime and her steroid dose was increased to 200 mg daily in divided doses.

After a week she was well and able to eat and drink normally. The epiglottis and larynx again looked normal on nasendoscopic examination and the tracheostomy tube was removed. She was allowed home the next day on a gradually decreasing steroid replacement regime. Her cefuroxime was continued orally.

Three days later she presented to the accident and emergency department with acute stridor and marked respiratory distress. She rapidly deteriorated after arrival

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Accepted for publication: 24 September 2003.

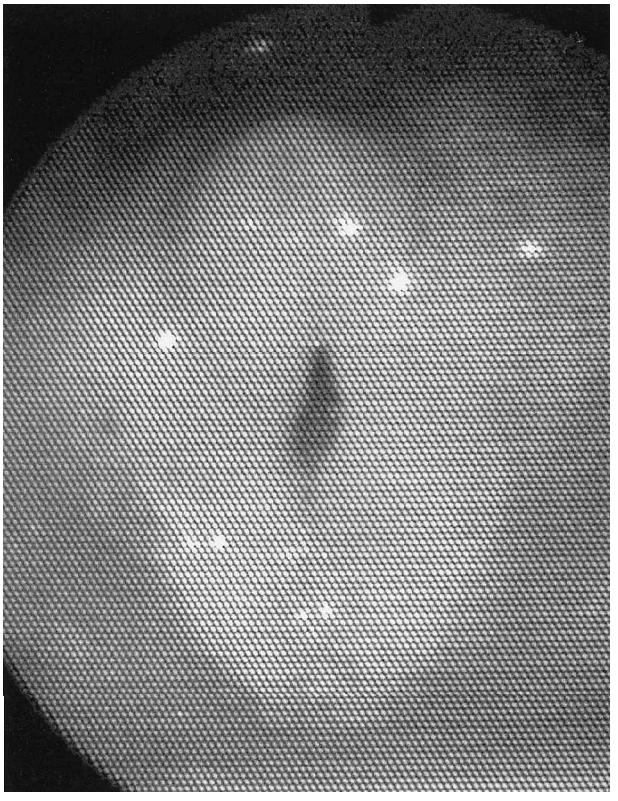

FIG. 1
Endoscopic appearance of the epiglottis.

and a tracheostomy tube was reinserted through the stoma site. Flexible nasendoscopy revealed that the epiglottis was yet again swollen with almost complete airway obstruction. At this stage she was febrile with a white cell count of $13.5 \times 10^9$/L. Her steroid dose was again increased to 200 mg daily and she was restarted on co-amoxiclav. Cefuroxime was stopped. Blood cultures were negative. Four days later the epiglottis appeared normal and after nine days in hospital she was discharged home with a tracheostomy tube *in situ*. Co-amoxiclav was continued for a total of three weeks. She also received a HIB vaccination on the advice of the microbiologist.

- **This is a case report of relapsing epiglottitis**
- **A relative lack of exogenous steroid in a patient with pituitary-adrenal insufficiency and a severe infection appears to have been responsible for the relapses**
- **Relapsing epiglottitis is uncommon but has been previously reported. However this is the first case with rapidly recurrent symptoms**

Her immunoglobulin levels were normal and a human immunodeficiency virus (HIV) test was negative. A magnetic resonance image (MRI) scan of her larynx and neck was normal one week into her second admission.

One month after going home her epiglottis appeared normal and she was well on her original steroid and thyroxine replacement doses. The tracheostomy tube was capped off but left *in situ* for a further month. It was then uneventfully removed. She remains well several months later.

## Discussion

The annual incidence of adult epiglottitis is estimated to be between 8.8–9.7 per million adults per year,[2] with a mean age of 47 years and a male to female ratio of 1.8 to 1.[3] While children usually present with breathing difficulties, adults commonly present with pain and swallowing difficulties. In adults, airway obstruction is reported to occur in 10–40 per cent of cases.[3]

HIB is the commonest cause of infection in children and adults. Other causes include *Streptococcus pneumoniae* and *Haemophilus parainfluenzae*, with isolated reports of *Klebsiella pneumoniae* and *Pasteurella multocida* in blood cultures.[2]

In Great Britain, infection with HIB has considerably declined since the introduction of the conjugated vaccine in 1992.[4] A prospective study of the HIB vaccine in Great Britain to assess its efficacy found that the incidence of epiglottitis had declined by more than 95 per cent in children.[4] A similar study in Sweden showed a substantial decrease in the incidence of epiglottitis in under five year olds from 209 to nine per million after the introduction of large scale vaccination.[5] There was also a tendency towards a decrease in the incidence of adult epiglottitis.[5]

Recurrent epiglottitis has been reported in four patients with impaired humoral immunity secondary to decreased immunoglobulin levels.[1] In one case the patient had had a previous splenectomy and it was hypothesized that this compromised defence against encapsulated bacteria such as HIB. It has been postulated that recurrent epiglottitis could be the first manifestation of a deficit of humoral immunity.[1] All four cases had experienced between two and six episodes of acute epiglottitis over an eight-year period. All of the episodes were successfully treated with intravenous antibiotics.[1]

Of the three other reported cases, one was in a fit and well 19-year-old male who had two episodes of epiglottitis occurring a year apart and resolving rapidly with antibiotics, no cause was found for the recurrence.[6] The second case described a healthy 26-year-old male with epiglottitis recurring after three years. Again the patient responded rapidly to antibiotics, he was found to have an omega-shaped epiglottitis and subsequently underwent elective epiglottectomy.[7] The third case describes a previously healthy 29-year-old male with epiglottitis recurring three times over six months. He responded promptly to antibiotic therapy, although he suffered a relapse after a few days on one occasion.[8] In our case the epiglottitis relapsed twice within 18 days despite an initial response to antibiotic therapy. In a review of 129 cases of acute adult epiglottis in California, the mean hospital stay was just 4.1 days.[3] This is the only reported case in the literature of epiglottitis relapsing over a period of a few weeks rather than recurring over months to years.

There is no evidence that our patient was immunocompromised, her immunoglobulin levels were within the normal range and she had a normal spleen. Before developing epiglottitis she was on adequate anterior pituitary replacement therapy, and while her response to the stress of infection would have been impaired by the absence of pituitary function, she received an increase in exogenous steroid replacement. It is possible that the steroids masked the oedema and that when they were reduced the oedema increased. The precise relationship of her pituitary insufficiency to the relapsing course of her infection is unclear.

The patient eventually responded to an increased steroid dose and prolonged antibiotic treatment. Following two relapses she was left with a tracheostomy for four weeks before its eventual removal.

https://doi.org/10.1258/002221503322683948 Published online by Cambridge University Press

This is the first report in the literature of a case of relapsing epiglottitis, distinct from recurrent epiglottitis. The association of this condition with pituitary-adrenocortical insufficiency is important and should be considered in all patients presenting with epiglottitis.

**Summary**

We report a case of relapsing acute adult epiglottitis in a patient who had had pituitary surgery and was receiving corticosteroid replacement therapy. It is the first case to highlight this form of the disease occurring in a patient with pituitary-adrenocortical insufficiency. It is also the first case of epiglottitis to be reported in the literature as presenting in a relapsing manner over a matter of weeks rather than recurring over months to years.

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S. Latif takes responsibility for the integrity of the content of the paper.
Competing interests: None declared