A case of atypical croup

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CASE PRESENTATION

A previously healthy eight-month-old girl was admitted to the pediatric intensive care unit at Stollery Children's Hospital (Edmonton, Alberta) following a waxing and waning course of respiratory distress. She had initially presented 11 days earlier to the emergency room with stridor and a brassy cough, and laryngotracheobronchitis was diagnosed. Treatment with racemic adrenaline and one dose of oral dexamethasone was effective and she was discharged after overnight observation. Nasopharyngeal aspirate was negative by direct fluorescent antibody and by viral culture for influenza A and B, parainfluenza and respiratory syncytial virus. Over the next three days, the child had increasing stridor and was admitted and again treated with racemic adrenaline and a four-day course of oral dexamethasone. She was subsequently discharged. White lesions were noted on the tongue and were treated with topical nystatin. Four days later, the child presented to the emergency room with recurrence of stridor and fever and new onset of drooling. She had a respiratory rate of 44 breaths/min, heart rate of 170 beats/min, blood pressure of 90/58 mmHg, oxygen saturation of 98% and temperature of 39.6°C. She was in moderate respiratory distress, with marked stridor. Examination of the oropharynx revealed ulcerative lesions on the anterior tongue, hard and soft palate, and posterior pharynx. Laboratory studies revealed a white blood cell count of $18.1 \times 10^9/L$ (74% neutrophils, 23% lymphocytes and 3% monocytes). Other hematological values were normal. A laryngobronchoscopy was performed and revealed that the

Figure 1) View of laryngopharynx of a child with recurrent croup. E Epiglottis; T Tongue

mucosa of the laryngopharynx was studded with discrete white lesions on a background of mucosal inflammation with severe reduction of airway calibre requiring intubation (Figure 1). The subglottis and the tracheobronchial tree were severely inflamed but had no discrete lesions.

What was the diagnosis?

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DIAGNOSIS AND DISCUSSION

The diagnosis was herpetic croup. Swabs taken from the lesions in the laryngopharynx (Figure 1) were positive by direct fluorescent antibody test and by viral culture for herpes simplex virus type 1 (HSV-1). The patient was placed on a 10-day course of intravenous acyclovir. An extubation attempt failed four days following admission. She was successfully extubated two days later after a two-day course of dexamethasone to reduce the subglottic swelling.

Previous case reports have described protracted upper airway signs and symptoms due to HSV infection in immunocompetent children (1-4) with descriptions of epiglottitis (5), laryngitis (2,3) and laryngotracheobronchitis (1,6) due to HSV. Most cases are caused by HSV-1, but HSV-2 has also been implicated (2). In a retrospective series of ulcerative laryngitis (7), only one of 15 patients did not require airway intervention, with the median duration of intubation being four days (range three to 11 days). Five children required a tracheostomy.

Two hypotheses have been proposed to explain the development of herpetic croup. The first hypothesis suggests that

the virus spreads contiguously down the respiratory tract following an initial symptomatic or asymptomatic oral infection, but it is not clear why this would occur in a normal host. Our patient had no clinical or laboratory evidence of an immunodeficiency. In favour of this hypothesis is the fact that respiratory viruses were not detected in our patient's initial nasopharyngeal sample. The second hypothesis suggests that oral corticosteroids used to treat croup (caused by a nonherpes virus) lead to immunosuppression and, therefore, more severe infection with HSV if the child has a concurrent exposure (2). In favour of this hypothesis is the fact that our patient's mouth lesions were not noted until several days after the onset of her stridor.

Herpetic croup should be suspected if a child presents with atypical croup, particularly if stridor is prolonged in the setting of mouth lesions that are compatible with herpetic gingivostomatitis. However, the absence of ulcerative gingivostomatitis does not exclude the presence of lesions lower in the respiratory tract (7). The role of antivirals and corticosteroids in hastening recovery is not clear.

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