Spasmodic croup presenting with persistent hiccups: a case report

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ABSTRACT

Background: Croup is a common pediatric respiratory illness characterized by a barking cough, inspiratory stridor, and hoarseness resulting from upper airway narrowing. While most cases are of viral etiology, spasmodic croup can be triggered by non-infectious factors such as allergy, gastroesophageal reflux disease, or psychological stress.

Case Presentation: We describe a previously healthy with no significant past medical history 10-year-old girl, with multiple presentations to the emergency department with persistent hiccups accompanied by inspiratory stridor with the classic "steeple sign". Her clinical course suggested overlapping mechanisms, including reflux-related and stress-related functional triggers, as her symptoms improved with proton-pump inhibition and corticosteroids.

Conclusion: This case illustrates an unusual presentation of recurrent spasmodic croup with persistent hiccups, likely multifactorial in origin. Awareness of atypical features may help in early identification and improve patient outcomes.

Keywords: Spasmodic croup, croup, hiccups, case report, laryngotracheitis.

Type of Article: Case Report **Specialty:** Pediatric Emergency

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Introduction

Croup is a pediatric respiratory disorder and represents an umbrella term encompassing various etiologies [1]. The usual picture includes an acute barking cough, stridor, and hoarseness due to narrowing of the airway [1,2]. Although most cases are infectious, mainly viral, they could be spasmodic croup, which can be due to non-infectious factors. Spasmodic croup is a diagnosis of exclusion and is typically made by exclusion, as it may present with recurrent episodes of croup-like symptoms and admissions. Recurrent croup, defined as more than two episodes, should raise suspicion of non-viral causes [2,3]. In recurrent non-infectious croup, warrants evaluation for allergy, airway hyperreactivity, gastroesophageal reflux disease (GERD), or psychological factors [4]. Recurrence has been reported in approximately 5%-6% of children, and effective management focuses on identifying and treating these underlying contributors [1,5].

Although croup usually presents with its classic symptoms, unusual accompanying features are rarely reported. In the following case report, we describe a child with recurrent croup in whom persistent hiccups drew clinical attention, and we present the subsequent evaluation and management.

Case Presentation

A 10-year-old girl, previously healthy and fully vaccinated, was brought to the emergency department (ED) with a 1-day history of continuous hiccups, mild epigastric discomfort, and heartburn. She had no fever, cough, chest pain, palpitations, syncope, vomiting, trauma, dysphagia, or choking attacks. Family history was unremarkable for asthma, allergic diseases, GERD, or psychiatric disorders.

On arrival, she was alert and well-appearing upon performing the pediatric assessment triangle, with persistent hiccups, with no obvious cyanosis, and stable vital signs on room air. Chest examination showed equal bilateral air entry and intermittent inspiratory stridor during hiccup episodes, and other examinations were unremarkable. Her

initial laboratory investigations and inflammatory markers were all within normal limits, with C-reactive protein being <0.2 mg/l. Her chest radiography showed mild peribronchial thickening.

The neurology team was consulted to evaluate the possibility of a seizure-related cause, and after a detailed assessment, they concluded that the hiccups were partly suppressible with distraction and lacked any associated loss of consciousness or post-ictal features, making psychogenic (functional) etiology more likely. She was discharged after reassurance.

Thirty days after her initial presentation, she re-presented to the ED with continuous hiccups and frequent belching. She described central chest and epigastric cramping pain radiating upward to the throat. On examination, she was alert and oriented, with stable vital signs. Intermittent inspiratory stridor was again noted on auscultation, particularly during hiccup spells.

During this visit, she received 5 ml of an oral antacid (aluminum hydroxide/magnesium hydroxide) and intravenous esomeprazole 20 mg once daily; her symptoms improved, and she tolerated oral intake without difficulty. Chest radiograph (Figure 1) was performed and was unremarkable, showing no new pulmonary, mediastinal, or airway abnormalities. Neck soft-tissue radiography (Figure 2) showed an apparent narrowing of the airway with the classic steeple sign, and dexamethasone 0.6 mg/kg was administered. To further investigate other serious causes of upper airway obstruction, airway endoscopy was performed, confirming airway patency with no evidence of obstruction. Following clinical improvement and confirmation of a stable and patent airway, the patient was discharged home with instructions for outpatient follow-up.

Exactly 31 days after that visit, she presented for the third time due to an episode of hiccups lasting 2 hours with shortness of breath. She was treated with dexamethasone 0.6 mg/kg as a single dose and intravenous esomeprazole 1 mg/kg once daily, which led to the resolution of her symptoms. This time, she reported psychological distress at school due to negative comments from teachers. A psychiatry consultation was obtained, and it confirmed significant emotional distress, with a recommendation for outpatient psychological support.

At the last ED visit, the patient appeared relieved following symptom resolution. Her father shared that the recurrence of symptoms had been frightening and frustrating for the family. He expressed that once the treatment was effective and the likely cause of recurrence was clearly explained, they felt reassured.

Discussion

Croup, also known as laryngotracheobronchitis, is a frequent respiratory condition in children, as they present with stridor, a barking cough, and respiratory distress resulting from the airway narrowing and inflammation [1].

While viral infection remains the most common etiology, multiple non-infectious factors can also contribute to croup, including spasmodic or recurrent forms, in which

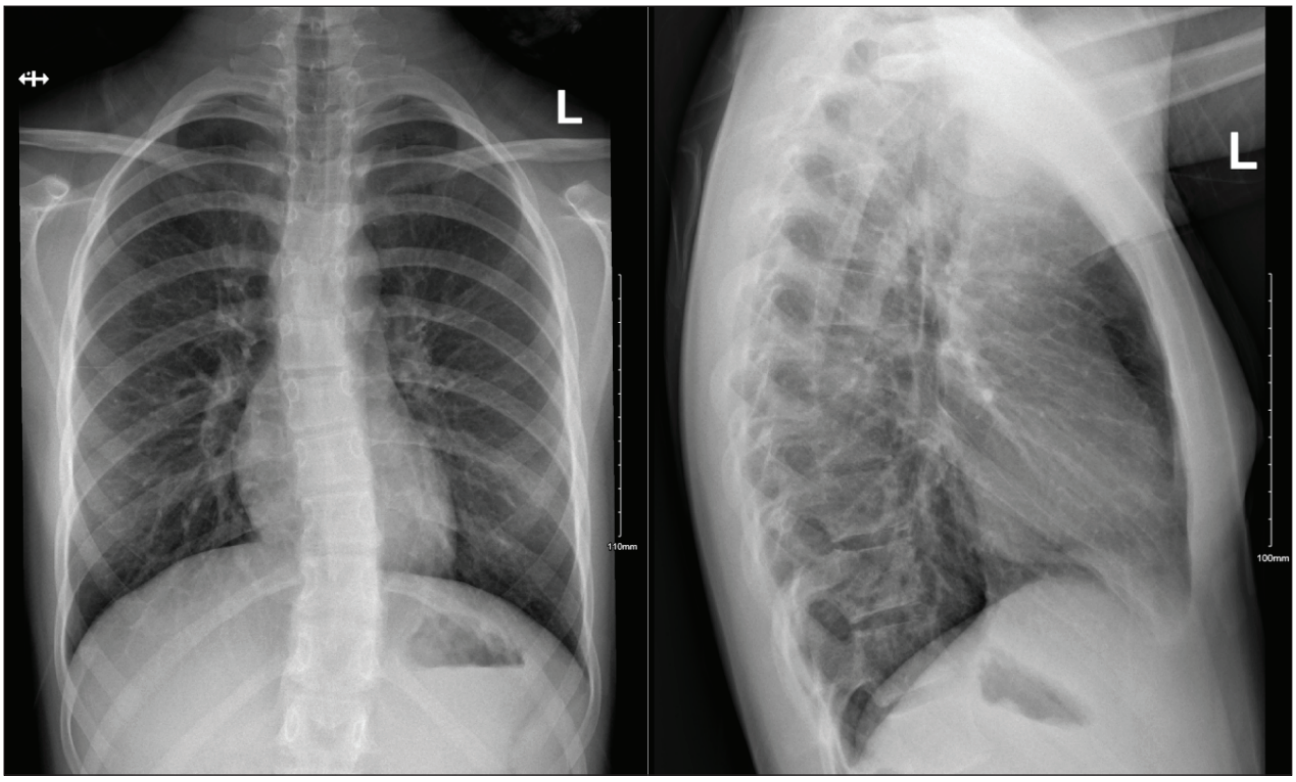

Figure 1. *Chest radiographs from the second visit showing a PA view and a lateral view.*

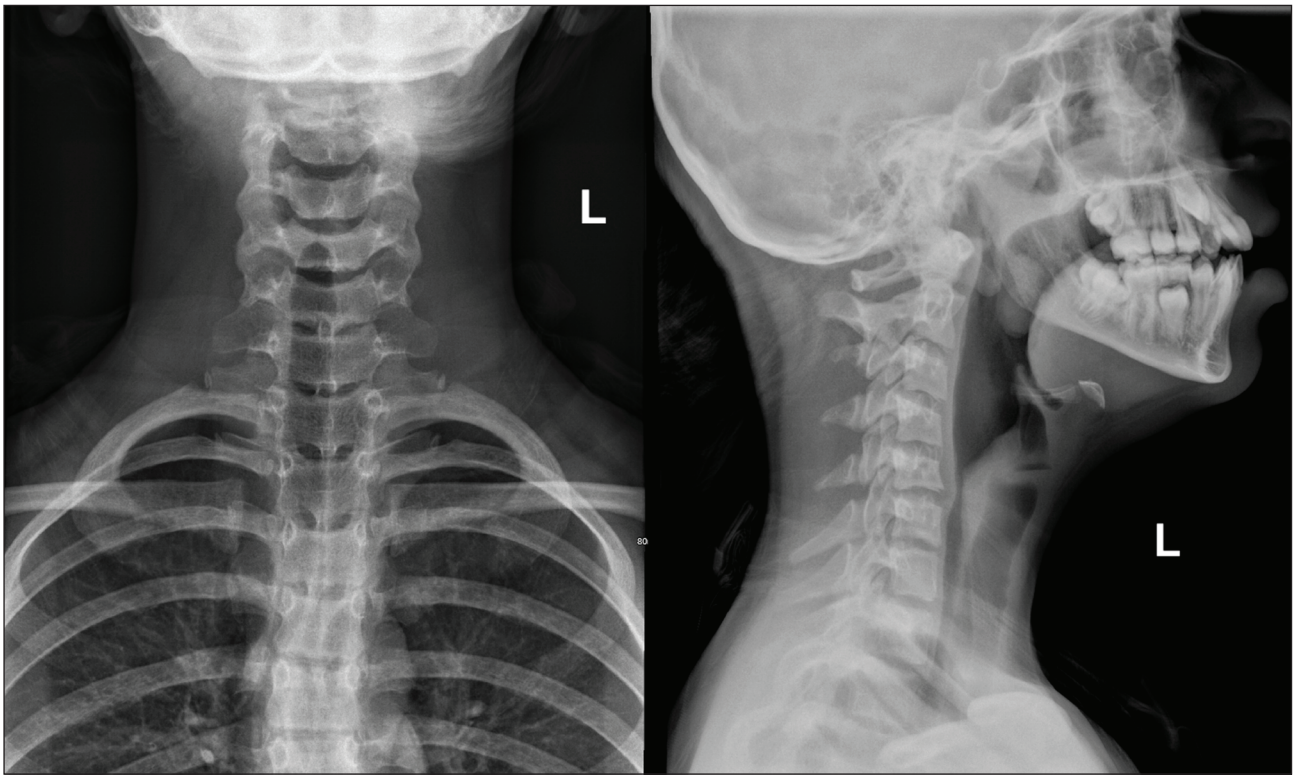

Figure 2. *Neck soft-tissue radiograph from the second visit showing the "steeple sign" on the PA view, indicating subglottic narrowing consistent with croup.*

the classic symptoms may persist without a clear infectious trigger [2,4]. Our case highlights a diagnostic challenge where recurrent croup-like episodes were initially masked by an unusual presenting feature, such as persistent hiccups, drawing attention to possible extra-respiratory etiologies.

Recurrent croup calls for a thorough evaluation to identify underlying causes. Quraishi and Lee [4] emphasized that two or more episodes per year should prompt investigation for conditions such as airway abnormalities, allergic airway disease, or GERD. In our patient, the coexistence of heartburn and epigastric discomfort suggested reflux as a possible precipitating factor, consistent with the established association between GERD and recurrent or atypical croup. A systematic review and meta-analysis by Coughran et al. [2] confirms the significant correlation between croup and GERD, suggesting that acid reflux induces laryngeal inflammation and increases airway reactivity, thereby triggering recurrent stridor. As our patient's symptomatic improvement following the antacid and proton-pump inhibition (PPI) therapy further supports this association.

In our patient, the recurrence of symptoms despite normal airway imaging and endoscopic evaluation, combined with new school-related emotional distress, suggests that psychological factors may have acted as a co-trigger in her presentation. In addition to organic etiologies, psychogenic mechanisms have been described as possible contributors to stridor, which is a functional upper-airway obstruction triggered by stress. Lacy and McManis [6] reviewed 48 cases and reported that most patients exhibited ongoing emotional stress, with more than half diagnosed with conversion disorder, also known as (Functional Neurological Symptom Disorder), which occurs when psychological stress produces involuntary physical symptoms without an identifiable organic cause [6,7].

Acute management of croup involving corticosteroids, as the 2023 review by Aregbesola et al. [1] reaffirmed that glucocorticoids significantly improve symptoms and reduce the need for further medical interventions in children with croup [1]. In our case, dexamethasone administration was associated with an obvious clinical improvement, supporting its role even in atypical presentations.

This report has some limitations. First, as a single case report, the findings cannot be generalized or applied to the pediatric population. Second, GERD was clinically suspected based on symptoms and response to PPI therapy, without objective diagnostic confirmation, which may limit the attribution of reflux as a triggering factor. Further studies are needed to better understand spasmodic croup presenting with persistent hiccups. Larger case series would be beneficial to confirm these observations and guide optimal management strategies in similar presentations.

Conclusion

In conclusion, this case demonstrates that recurrent croup may manifest with unusual symptoms, such as persistent hiccups with intermittent stridor, an association not previously described and likely multifactorial in origin. Recognition of GERD and psychological stress as potential contributors allowed for targeted management and symptom resolution. Physicians should maintain a high index of suspicion for underlying conditions in recurrent croup, ensuring a comprehensive evaluation and multidisciplinary approach.

What is new?

Although croup usually presents with its classic symptoms, unusual accompanying features are rarely reported. In the following case report, the authors describe a child with recurrent croup in whom persistent hiccups drew clinical attention, and they present the subsequent evaluation and management.

Conflicts of interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

Funding

None.

Consent for publication

Informed consent for publication was obtained from the patient's legal guardian.

Ethical approval

Ethical approval is not required at our institution to publish an anonymous case report.

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Summary of the case

1	Patient (gender, age)	Female, 10 years old
2	Final diagnosis	Spasmodic croup
3	Symptoms	Continuous hiccups, epigastric pain, and heartburn
4	Medications	Antacid (aluminum hydroxide/magnesium hydroxide), esomeprazole, and dexamethasone
5	Clinical procedure	Airway endoscopy
6	Specialty	Pediatric emergency