

# CROUP-INDUCED POSTOBSTRUCTIVE PULMONARY EDEMA

I-Chen Chen,<sup>1</sup> Ko-Hsin Chen,<sup>1</sup> Chien-Ming Tseng,<sup>1</sup> Jong-Hau Hsu,<sup>1,2</sup> Jiunn-Ren Wu,<sup>1,2</sup> and Zen-Kong Dai<sup>1,2,3</sup>

<sup>1</sup>Division of Pediatric Cardiology and Pulmonology, Department of Pediatrics, Kaohsiung Medical University Hospital, <sup>2</sup>Department of Pediatrics, Faculty of Medicine, and <sup>3</sup>Department of Pediatrics, Graduate Institute of Medicine, College of Medicine, Kaohsiung Medical University, Kaohsiung, Taiwan.

Postobstructive pulmonary edema is life-threatening, and results from a sudden episode of upper airway obstruction. Croup is generally thought to be a benign condition, but occasionally it can develop into postobstructive pulmonary edema. We present a case of a 5-year-old boy with recurrent croup, who was diagnosed with postobstructive pulmonary edema. Our experience alerts pediatricians to this easily misdiagnosed disease.

**Key Words:** croup, obstructive sleep apnea syndrome, pulmonary edema, respiratory distress

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Croup, or laryngotracheobronchitis, is generally thought to be a common, short-term and self-limiting illness in children; respiratory distress requires hospitalization only in 1.3–2.6% of cases [1], and less than 1% develop severe croup [2]. Here, we describe a 5-year-old boy with recurrent croup and post-obstructive pulmonary edema (POPE). The rapid resolution of his illness, its differential diagnosis, and effective treatment are also discussed.

## CASE PRESENTATION

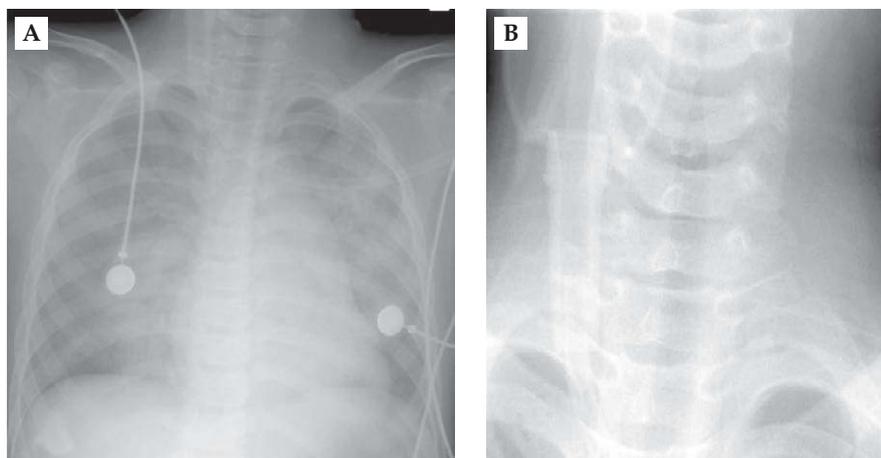
A previously healthy 5-year-old boy developed acute dyspnea in the middle of the night and was sent to our emergency room within a few minutes. He had no history of fever or other illness, but a 2-day history

of cough. In the emergency room, he was noted to be slightly drowsy, with a pulse rate of 120 beats/min, respiratory rate of 40 breaths/min, blood pressure of 116/44 mmHg, temperature of 36.8°C, and pulse oximetry of 44% in room air. Physical examination revealed a child who was slowly responsive to verbal commands. Breath sound was coarse. He had the typical symptoms of croup, including inspiratory stridor, barking cough, subcostal retractions, and a hoarse voice. He was given oxygen via a non-rebreathing mask immediately and treated with nebulized epinephrine, inhaled budesonide and intravenous methylprednisolone (0.6 mg/kg). Intubation was not attempted by the doctor on duty because the boy's pulse oximetry increased to 96% and consciousness recovered within several minutes.

Initial venous blood gas analysis revealed: pH 6.93; partial pressure of arterial carbon dioxide, 83.8 mmHg; partial pressure of arterial oxygen, 23.4 mmHg; HCO<sub>3</sub><sup>-</sup>, 18.8 mmol/L; and base excess, -16.5 mmol/L. Routine blood analysis showed leukocytosis (24.6 × 10<sup>9</sup>/L) with 37.8% neutrophils and mildly elevated C-reactive protein (14 mg/L). Chest radiography showed bilateral alveolar infiltrates in both upper lung fields (Figure 1).



Received: Jan 18, 2010 Accepted: Apr 1, 2010  
Address correspondence and reprint requests to:  
Dr Zen-Kong Dai, Department of Pediatrics,  
Kaohsiung Medical University Hospital, 100  
Shih-Chuan 1<sup>st</sup> Road, Kaohsiung 807, Taiwan.  
E-mail: zenkong@kmu.edu.tw



**Figure 1.** Chest radiographs demonstrating (A) alveolar infiltrates in both upper lung fields and (B) steple sign in a 5-year-old boy with acute dyspnea.

Treatment was continued with nebulized epinephrine (0.3 mL every 4 hours) and inhaled budesonide (0.25 mg every 12 hours), and antibiotic coverage with cefuroxime was initiated after he was admitted to the pediatric intensive care unit. Arterial blood gas analysis at 2 hours after the initial analysis showed: pH 7.26; partial pressure of arterial carbon dioxide, 62 mmHg; partial pressure of arterial oxygen, 58 mmHg;  $\text{HCO}_3^-$ , 27.8 mmol/L; and base excess,  $-0.2$  mmol/L. Repeat chest radiography at 12 hours after the initial examination showed dramatic resolution of alveolar infiltrates (Figure 2). Echocardiography revealed normal cardiac function without cardiac anatomical defects. The possibility of upper airway obstruction or foreign body aspiration was considered, therefore, flexible tracheobronchoscopy was arranged and no foreign body was noted, but adenoid hyperplasia was present.

Further history taking revealed that the patient had an episode of croup with respiratory distress 2 months previously. His parents also mentioned that the child had been snoring for 1 year, but they had never noted apnea. Subsequent polysomnography of the patient was normal. He was discharged in a stable condition.

## DISCUSSION

The hallmark of croup is subglottic edema that results in varying degrees of airway obstruction. POPE, also named negative pressure pulmonary edema, was first described in 1973 [3]. It is a life-threatening condition that results from a sudden, severe episode of



**Figure 2.** Chest radiograph taken 12 hours after initial examination. The radiograph was normal, and there had been rapid resolution of the alveolar infiltrates.

upper airway obstruction (type I), or after surgical relief of chronic upper airway obstruction (type II) [4,5]. The incidence of type I POPE in patients with upper airway obstruction is reported to be 7–12% [6]; the incidence of type II POPE in anesthetic patients is reported to be 0.05–0.1% [7]. The true frequency of type I POPE following croup is not known.

POPE develops when excessive negative intrathoracic pressure is generated by forced inspiration against a closed glottis or obstructed airway [8]. This leads to an increase in venous return to the right heart, which increases pulmonary venous pressure. The increase in pulmonary venous pressure creates a hydrostatic transpulmonary gradient with fluid moving from the pulmonary veins to the interstitial space.

At the same time, low cardiac output might increase afterload. The combination of increased preload and afterload results in a marked increase in hydrostatic pressure in the pulmonary vasculature [9]. The increased hydrostatic pressure changes Starling's force and leads to pulmonary edema. Besides, an acute upper airway obstructive event can lead to severe hypoxia. Severe hypoxia can cause hypoxic pulmonary vasoconstriction, which can lead to hypoxic pulmonary hypertension and right heart failure [10]. Fortunately, the echocardiogram in our patient was normal.

The manifestations of POPE include: (1) signs of upper airway obstruction, such as drooling, hoarseness, stridor, and retractions; and (2) signs of pulmonary edema, such as hypoxemia, frothy sputum (not shown in our patient), and bilateral alveolar infiltrates in upper lung fields on chest radiography. POPE usually occurs within minutes and resolves rapidly within 12–24 hours [5,9,11,12]. Accordingly, our case described above had type I POPE. The common causes of type I POPE include epiglottitis, croup, foreign body aspiration and angioedema [7,9]. In our case, epiglottitis and foreign body aspiration could be excluded by the rapid resolution and flexible tracheobronchoscopy. Because of the history of prior respiratory tract infection and the typical symptoms of croup, our patient was diagnosed with croup-induced type I POPE. However, angioedema still cannot be ruled out, although no skin rash was noted on this patient.

On further consideration of this history of POPE and recurrent croup, we thought that obstructive sleep apnea syndrome (OSAS) might be a factor, given the occurrence of two episodes of respiratory distress at night, a 1-year history of snoring, and adenoid hypertrophy. Our decision to include OSAS in the differential diagnosis was supported by a case report of a previously healthy child with OSAS who developed POPE [13]. In addition, in a study of dogs, recurrent obstructive apnea over an 8-hour period was noted to cause lung edema and deterioration in gas exchange [14]. Fortunately, our patient had normal polysomnography and there was no recurrence of nighttime respiratory distress thereafter. To the best of our knowledge, this is the first report that suggests the need to differentiate OSAS from recurrent croup, which might precipitate POPE, but further studies are warranted.

The treatment of POPE involves early recognition, supportive therapy such as oxygen supplementation by facemask or mechanical ventilation, and treatment

of the underlying disease [5,11]. The use of diuretics in POPE is controversial [11]. As for the treatment of croup, nebulized epinephrine and corticosteroids are the mainstay in relief of airway obstruction [2,15].

In summary, although croup is generally thought to be a benign condition, some cases might develop into POPE. Pediatricians should be alert to this life-threatening and easily misdiagnosed condition.

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# 哮喘導致阻塞後肺水腫

陳怡真<sup>1</sup> 陳可欣<sup>1</sup> 曾建銘<sup>1</sup> 徐仲豪<sup>1,2</sup> 吳俊仁<sup>1,2</sup> 戴任恭<sup>1,2,3</sup>

高雄醫學大學附設醫院<sup>1</sup> 小兒心肺科

高雄醫學大學 醫學院醫學系<sup>2</sup> 小兒科學<sup>3</sup> 醫學研究所

阻塞後的肺水腫通常發生於急性上呼吸道阻塞之後，是一種緊急，且嚴重會影響生命的疾病。而哮喘是一種急性上呼吸道阻塞之疾病，雖多屬良性的病程，但仍有少部份可能會引發阻塞後的肺水腫。在本文中我們提出一個 5 歲男孩，經歷了反覆的哮喘，最後引發了阻塞後肺水腫，透過我們的臨床經驗分享可以提醒兒科醫師在這方面的診斷更加小心謹慎，避免診斷上的疏失。

**關鍵詞：**哮喘，阻塞性睡眠呼吸中止症候群，肺水腫，呼吸窘迫

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通訊作者：戴任恭醫師

高雄醫學大學附設醫院小兒科

高雄市三民區十全一路 100 號