Am J Transl Res 2026;18(7):5829-5832
www.ajtr.org /ISSN:1943-8141/AJTR0172974

Case Report

Rapidly progressive air leak syndrome after uneventful extubation in a pediatric patient: a case report

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Received March 30, 2026; Accepted May 18, 2026; Epub July 15, 2026; Published July 30, 2026

Abstract: Background: Air leak syndrome following uneventful extubation is rare in pediatric patients undergoing nonthoracic surgery; however, it can lead to rapid airway compromise and respiratory failure. Case presentation: A previously healthy 8-year-old girl developed rapidly progressive generalized subcutaneous emphysema and dyspnea shortly after extubation following surgery for a right radial fracture. Intraoperative intubation was uneventful, the peak airway pressure remained within normal limits, and extubation proceeded with no coughing or bucking. After transfer to the ward, the oxygen saturation declined to 40%. Manual ventilation through a supraglottic airway device was associated with high resistance and air leakage. On arrival at the emergency department, severe cervicofacial swelling from subcutaneous emphysema precluded videolaryngoscopic intubation. Endotracheal intubation was eventually achieved with a Cormack-Lehane grade IV view. Imaging revealed extensive subcutaneous emphysema, pneumomediastinum, and bilateral pneumothorax. Needle decompression and bilateral chest tube insertion were conducted immediately, followed by mechanical ventilation in the intensive care unit. Bronchoscopy revealed no definite tracheobronchial injuries. The patient recovered without complications. Conclusion: Rapidly progressive air leak syndrome should be considered in children who develop neck swelling or unexplained respiratory distress shortly after extubation. Repeated forceful ventilation should be avoided when ventilation is ineffective and airway control should not be delayed.

Keywords: Subcutaneous emphysema, pneumomediastinum, pneumothorax, air leak syndrome, extubation, pediatric

Introduction

Extubation after routine pediatric surgery is generally considered a low-risk procedure. However, when air leak syndrome occurs in this setting, clinical deterioration can be rapid and severe [1, 2]. Subcutaneous emphysema, pneumomediastinum, and pneumothorax are well-recognized complications of thoracic surgery or airway trauma. However, their occurrence after uneventful extubation in children undergoing orthopedic surgery is rare and may not be immediately suspected [1-3]. Here, we describe a child whose post-extubation neck swelling rapidly progressed to respiratory failure and difficult airway management.

Case presentation

An 8-year-old previously healthy girl underwent surgery for a right radial fracture at another hospital, with an operative time of approximately 1 hour. The attending anesthesiologist reported that endotracheal intubation was achieved on the first attempt without difficulty. Anesthesia was induced with propofol, rocuronium and maintained with sevoflurane. The endotracheal tube cuff pressure was not measured, but the peak airway pressure remained within normal limits throughout the procedure. No intraoperative anesthetic or surgical complications were reported. Extubation was conducted without coughing or bucking. She was transferred to the ward approximately 20 minutes after arrival at the post-anesthesia care unit.

Thereafter, neck swelling and progressive dyspnea were noted. The swelling rapidly extended to the chest and abdomen, and oxygen saturation declined to 40%. Endotracheal intubation failed due to severe airway edema. A size 3 I-gel

https://doi.org/10.62347/CYNQ4622

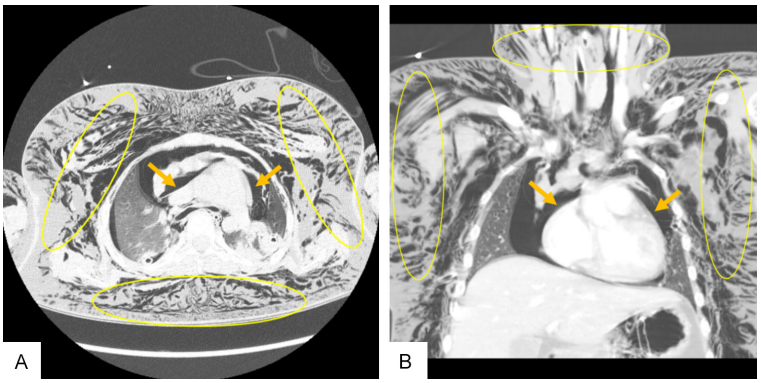

Figure 1. Chest computed tomography images showing extensive subcutaneous emphysema, pneumomediastinum, and craniocaudal air dissemination. A. Axial view. B. Coronal views. White ovals mark areas of subcutaneous emphysema; yellow arrows indicate pneumomediastinum.

was inserted; however, manual ventilation was ineffective, with oxygen saturation ranging between 40% and 60%. The patient was then transferred to the emergency department. Vital signs were not documented during transport.

Upon arrival, severe cervicofacial swelling caused by subcutaneous emphysema was observed. Ventilation via a supraglottic airway device revealed marked leakage and high resistance, and oxygen saturation could not be reliably measured despite monitoring. After device removal, video laryngoscopy failed because edema of the lips, tongue, pharynx, and larynx prevented adequate blade insertion. Arterial blood gas analysis demonstrated the following: pH, 7.112; arterial carbon dioxide tension ($PaCO_2$), 78.9 mmHg; arterial oxygen tension (PaO_2), 23.0 mmHg; and lactate, 5.5 mmol/L.

An anesthesiologist was immediately called, and tracheal intubation was attained using a pediatric laryngoscope with a 6.0-mm cuffed endotracheal tube secured at 20 cm. The laryngoscopic view corresponded to Cormack-Lehane grade IV. After intubation, the oxygen saturation improved to 87%. Extensive subcutaneous emphysema with palpable crepitus was noted throughout the body.

Tension pneumothorax was suspected, and immediate decompression using an 18-gauge needle released a large air volume. Bilateral pneumothorax was subsequently confirmed, and 20-French chest tubes were inserted bilaterally and connected to wall suction at -20 cmH_2O. Chest computed tomography revealed extensive whole-body subcutaneous emphysema, bilateral pneumothorax, and pneumomediastinum (**Figure 1**). Bronchoscopy demonstrated no definite tracheal or bronchial injury. The patient was admitted to the intensive care unit for mechanical ventilation.

By the following morning, subcutaneous emphysema significantly resolved. The patient was extubated on day 7 of hospitalization. Both chest tubes were removed 3 days later. The patient was discharged without complications on hospital day 15.

Discussion

The defining feature of this case was the pace of deterioration. Within approximately 20 minutes of ward transfer, the localized cervical swelling had progressed to generalized subcutaneous emphysema, and marked swelling of the lips, tongue, pharynx, and larynx rendered laryngoscopy nearly impossible, ultimately yielding a Cormack-Lehane grade IV view. Previous reports and reviews have linked air leak syndromes with thoracic procedures, airway trauma, vigorous coughing, high airway pressure, barotrauma, and underlying pulmonary disease [1-4]. In this patient, no such factors were identified before the sudden onset of neck swelling or respiratory distress. An apparently uncomplicated anesthetic does not exclude this possibility.

The mechanism could not be confirmed; however, the findings were consistent with the Macklin effect: increased intraalveolar pressure can cause alveolar rupture, allowing air to track along the bronchovascular sheaths toward the mediastinum before dissecting through the cervical fascial planes into the subcutaneous tissue [5, 6]. In the present case, pneumomediastinum, bilateral pneumothorax, extensive subcutaneous emphysema, and no definite tracheobronchial injury on bronchoscopy supported an alveolar or distal airway

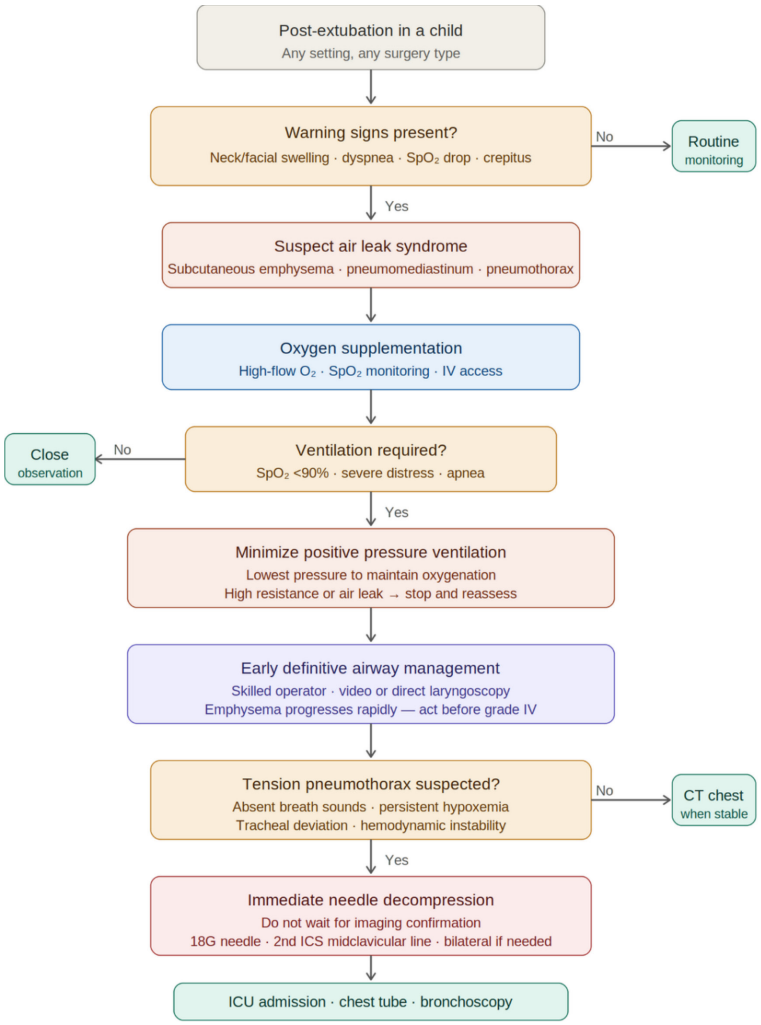

Figure 2. Suggested emergency management algorithm for pediatric patients with post-extubation cervicofacial emphysema with respiratory compromise. Early airway control should be considered when ventilation is ineffective, and decompression should not be delayed when tension pneumothorax is suspected.

source, although microscopic injuries could not be excluded.

This rapid course may be partly attributed to pediatric anatomy and physiology. The pediatric airway is inherently narrow, and the combination of compliant thoracic structures with limited oxygen reserves means that there is little physiological margin during acute respiratory compromise [7, 8]. The anatomical continuity between the cervical fascial planes and mediastinum further facilitates air migration [6]. Therefore, neck swelling after extubation in pediatric patients warrants prompt reassessment rather than watchful waiting.

The role of positive-pressure ventilation in driving the deterioration deserves attention. Intraoperative airway pressures were reportedly unremarkable; however, the conditions during recovery and subsequent rescue attempts were less certain. When a supraglottic airway device was utilized, ventilation was achieved with high resistance and significant leakage, indicating partial upper airway obstruction. Applying further pressure under these circumstances likely worsens mediastinal and subcutaneous air dissections [4, 9]. When supraglottic or bag-mask ventilation is ineffective, continued forceful ventilation may be harmful, and prompt establishment of a definitive airway should be prioritized.

The same urgency applies to decompression. Cervicofacial emphysema progressively distorts airway anatomy, and the window for successful intubation narrows quickly [7]. When tension pneumothorax is suspected in patients with refractory hypoxemia, decompression should precede imaging [10]. Based on the management sequence in this case, we propose an algorithm for pediatric patients presenting with post-extubation cervicofacial emphysema and respiratory compromise (**Figure 2**).

This study had some limitations. The initial surgery was conducted at another institution, and detailed intraoperative records, cuff pressure data, and vital signs during transfer were not available. Thus, precipitating events could not be identified with certainty. Nevertheless, the clinical sequence, imaging findings, negative bronchoscopy for major airway injury, response to airway control, and pleural decompression supported the diagnosis.

Post-extubation neck swelling or unexplained dyspnea in a child should raise the suspicion

of air leak syndrome until proven otherwise. This finding should not be dismissed, particularly when ventilation is ineffective. In that situation, repeated forceful ventilation and delayed airway control or decompression may worsen the clinical course.

Disclosure of conflict of interest

None.

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