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Laryngotracheal trauma-induced Macklin effect: A case report

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Abstract

Laryngotracheal trauma is a relatively rare traumatic injury seen particularly in young male adults. Trauma due to strangulation is one of its most frequent circumstances. However rare, pneumomediastinum is a particular complication of severe blunt neck injuries leading to alveolar ruptures. This phenomenon, described as the Macklin effect, requires early diagnosis, and its management varies from conservative to surgical treatment depending on the severity of symptoms. Our aim is to describe the case of a 21-year-old male who presented with blunt neck trauma. Clinical and imaging findings revealed subcutaneous neck emphysema and pneumomediastinum. Treatment was conservative leading to complete resolution of the injuries and the patient was discharged after 2 weeks.

Keywords

Otolaryngology, radiology, respiratory medicine, laryngotracheal trauma, pneumomediastinum

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Introduction

Pneumomediastinum is defined as the abnormal presence of air in the mediastinum, which is the visceral compartment of the chest separating the two pleural cavities. This phenomenon is a rare yet severe condition leading to deadly complications. Many conditions can lead to air leaking in the mediastinum. The etiologies vary from spontaneous ones to iatrogenic situations. The Macklin effect is defined as pneumomediastinum due to air leaking directly from the alveoli to the pulmonary interstitial space due to alveolar rupture, resulting in emphysema tracking along the bronchovascular interstitium. This phenomenon implies the absence of tracheobronchial and esophageal lesions. Instead, this is due to an increase in intrathoracic pressure leading to alveolar rupture. In fact, blunt neck injuries can disrupt airflow pressure inside tracheobronchial tree. This pressure elevation may result in alveoli rupture leading to air dissecting along pulmonary sheath causing pneumomediastinum. This phenomenon is represented in imaging by a linear collection of air, defining the Macklin effect (Figure 1). This serious condition implies diagnostic challenges and requires immediate and prompt management. This essay aims to describe the etiology and physiopathology of the Macklin effect by presenting the case of a severe blunt neck injury, its clinical presentation, and its imaging findings, as well as underlining management strategies.

Case report

A 21-year-old male presented to the emergency department of ears nose and throat after undergoing cervical trauma. There was no medical history besides occasional tobacco smoking. The patient sustained a blunt neck trauma due to strangulation during 1–2 min and was brought to our emergency department a few hours later. On presentation, the patient complained of dysphonia and odynophagia without respiratory distress. On clinical examination, the patient was in a good general status and afebrile. He was hemodynamically and neurologically stable. The patient was also eupneic with adequate oxygen saturation at ambient air. Besides

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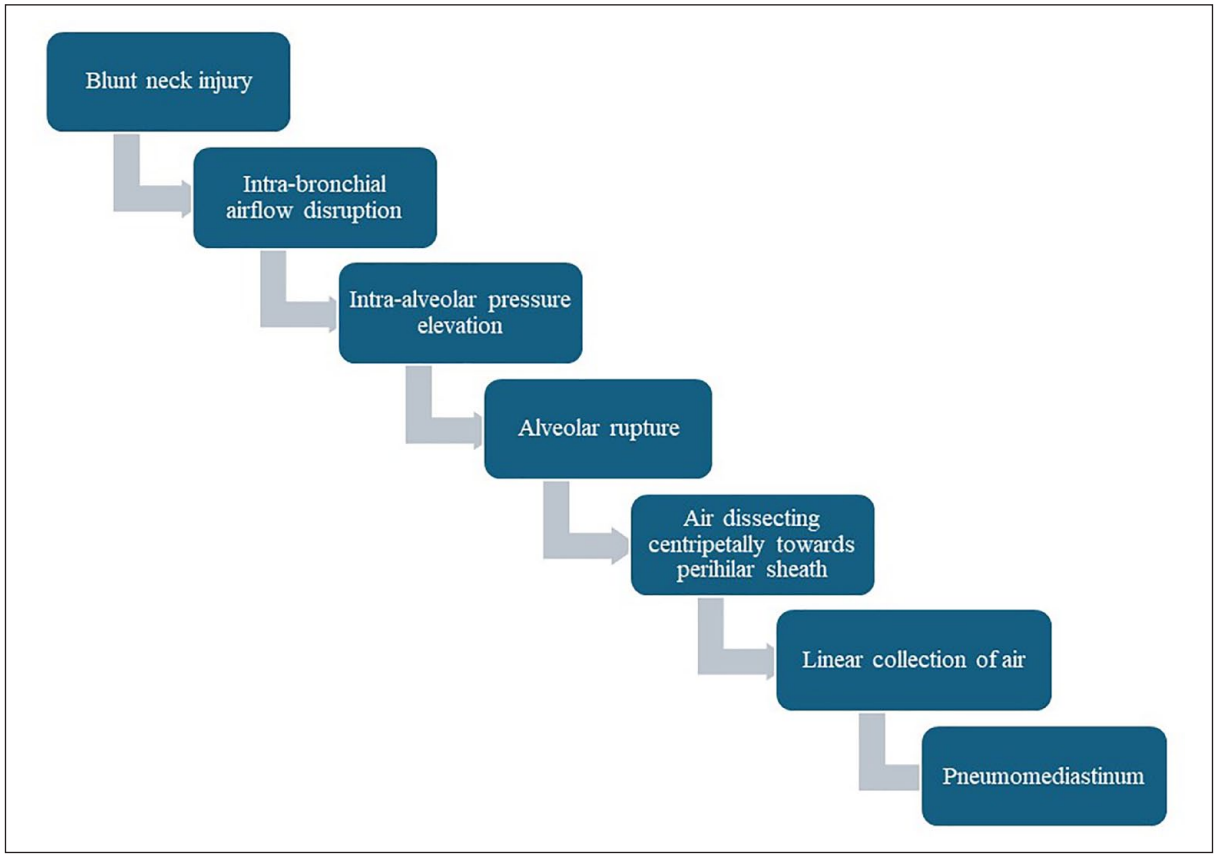

Figure 1. Visual diagram of Macklin effect pathophysiology.

hoarseness of voice and multiple neck ecchymoses, a physical examination revealed extensive subcutaneous cervical emphysema. In palpation, the laryngeal contours were preserved, and the laryngotracheal axis mobility was normal. Auscultation noted decreased breath sounds on the right side of the chest and was normal on the left side. Nasofibroscopy revealed a normal mobile larynx. To complete the lesion assessment, a cervicothoracic computed tomography (CT) scan was performed revealing the integrity of the hyoid bone as well as the thyroid and cricoid cartilages. No cricoarytenoid dislocation was observed. The CT scan revealed significant subcutaneous emphysema (Figure 2). Thoracic sections showed large pneumomediastinum (Figure 3). However, it did not detect any discontinuity in the walls of the tracheobronchial or esophageal structures. This pneumomediastinum is explained by intra-alveolar pressure elevation during strangulation that had led to their rupture, followed by centripetal air dissection from inside alveoli toward perihilar bronchovascular sheath. Due to local pressure conditions, air extended into the neck along the deep cervical fascia, explaining subcutaneous emphysema on CT scan. Upon these findings, the diagnosis of a large-volume pneumomediastinum extending into the cervical soft tissues was made and the patient was admitted to our department for further monitoring and management. An interdisciplinary decision

Figure 2. Computed tomography scan revealing subcutaneous emphysema of the neck.

along with thoracic surgery was made, and conservative treatment was prescribed given the absence of respiratory

Figure 3. Lung parenchyma windowed computed tomography scan revealing pneumomediastinum.

Figure 4. Computed tomography scan confirming the absence of pneumomediastinum.

distress. The treatment involved appropriate analgesia according to the patient's complaint and systemic corticotherapy was initiated along with bronchodilator nebulizations. Close monitoring of vital signs was conducted. There was favorable progression from the early days of management with reduction of cervical emphysema and restoration of normal phonation. On the 14th day of management, a follow-up cervicothoracic CT scan was performed, confirming the resolution of the lesions with complete regression of the pneumomediastinum as well as the subcutaneous emphysema (Figure 4). Given the favorable evolution, the patient was discharged with a follow-up appointment scheduled in 2 weeks, which was unremarkable. Notably, the patient was eupneic, with correct saturation at ambient air. Cervical emphysema has regressed, and nasofibroscopy revealed a normal mobile larynx.

Discussion

The Macklin effect was first described in 1939 after an animal experiment done by Macklin.[1] This phenomenon refers to pneumomediastinum due to air leaking directly from alveoli to the pulmonary interstitial space due to alveolar rupture, resulting in emphysema tracking along the bronchovascular interstitium.[2] Its underlying pathology is a sudden and important increase in pressure gradient between the alveolar and interstitial spaces.[3] The alveoli in question can either be separated or confluent.[1] This results in terminal alveoli breakdown followed by air dissecting along the pulmonary sheaths into the mediastinum.[4] Air extension follows local pressure and depends on multiple anatomical conditions, which, in extreme cases may lead to gas dissecting into the pericardium along blood vessel sheaths[5] resulting in pneumopericardium, or into the neck along the deep cervical fascia like our case. This phenomenon is typically seen after blunt chest injuries, and its prevalence can go up to 39% of all pneumomediastinum following blunt injuries.[1] Pneumomediastinum complicating neck injuries is however not as usual as chest trauma. This is an intriguing complication to discuss as its diagnosis is highly associated with long intensive care stays. While it can be seen in individuals of all ages, literature mainly reports cases in children, and the Macklin effect is rarely seen in the adult population. Higher frequency in children can be due to their smaller and more elastic alveoli, putting them at higher risk of laceration. Moreover, other predisposing factors have been mentioned, such as an underlying chronic lung pathology.[6] This latter condition can range from congenital pulmonary malformation to acquired conditions such as cystic fibrosis.[7] All these factors, along with susceptibility to pulmonary infections, make children more prone to the Macklin effect. Our study describes the case of a young healthy male adult who sustained a blunt neck injury by strangulation which led to an important pneumomediastinum with cervical emphysema. Kuniyoshi et al. reported a similar pediatric presentation in an 8-year-old boy who underwent anterior cervical blunt trauma after falling on a wooden object. Nasofibroscopy revealed swollen arytenoids and mild edema on vocal cord. CT scan detected pneumomediastinum without esophageal or cartilage lesions. Recovery was obtained after conservative management.[8] A more severe presentation has been reported by Steele et al. in a 23-year-old rugby player who

sustained a blunt neck injury after being struck by an elbow. Patient presented with a swollen neck and palpable emphysema extended from zygomatic arch to the sternum. Unlike our case, CT scan revealed important pneumomediastinum extending throughout the right neck to the skull base and down the left arm. Similarly to our patient, no esophageal or tracheal lacerations were detected. Given the abundance of the pneumomediastinum, the patient was intubated and underwent a thorough endoscopic examination by flexible bronchoscopy that ruled out trachea-bronchial injuries.[2] In our case, the treatment course was conservative given the absence of respiratory distress as well as any associated post-traumatic injury. In this case, like any other blunt neck trauma, strangulation led to abrupt pressure increase in the upper as well as the lower airways. Intrathoracic pressure augmentation results in alveolar laceration followed by air accumulation in the mediastinum.[9] Clinical presentation depends on the severity of injuries and is mainly dominated by neck emphysema, shortness of breath, and chest pain. Imaging findings are essential to establishing the diagnosis. Chest X-ray is rarely sufficient in lesion assessment in these cases, however, it can reveal subcutaneous chest and neck emphysema.[2] CT scan is primordial when it comes to exploring cervicothoracic trauma and reveals the pneumomediastinum which can be associated with subcutaneous emphysema, pneumothorax, pneumopericardium, esophageal laceration, or laryngotracheal injury.[10] In addition, CT scan is key in differentiating the Macklin effect from other lesions, especially given the similarity of their clinical presentations and the possibility of their association. Pulmonary contusion, tracheobronchial injury, rib fracture, esophageal rupture, or even cardiovascular contusion can all be confused with a large-volume pneumomediastinum.[11] When managing a patient with chest and neck injury, a thorough clinical and imaging exploration may possibly be completed with bronchoscopy or esophagoscopy to rule out differential diagnosis.[12] Nonetheless, CT scan can be a useful noninvasive alternative allowing proper differentiation between respiratory pneumomediastinum and other causes such as tracheobronchial and esophageal lesions. Murayama and Gibo studied imaging findings of both spontaneous and trauma-induced pneumomediastinum. Macklin effect has been described as a linear air leak along pulmonary interstitium. This particular CT aspect due to air centripetally dissecting toward perihilar bronchovascular sheath is most likely to be seen in non-traumatic pneumomediastinum.[13] In a study conducted by Okada et al.[14] on spontaneous pneumomediastinum, a 64-slice helical CT revealed Macklin effect through air in the perivascular and peribronchial sheaths in all cases. This was defined as a pathognomonic sign of non-spontaneous pneumomediastinum replacing a thorough endoscopic exploration of tracheobronchial and gastro-esophageal walls and ruling out any laceration or injury. Apart from its role for etiological purposes, Macklin effect also has prognostic value, anticipating progression to other, more severe lesions. Belletti et al.[15] conducted a review of literature to assess the clinical role of Macklin. When detected on CT, this entity preceded barotrauma in 94.2% of patients with acute respiratory distress syndrome (ARDS), making it a strong predicting tool when managing these cases. Another study by Palumbo et al.[16] demonstrated Macklin effect's role in anticipating subsequent pneumomediastinum and pneumothorax in COVID-19 ARDS patients, with a specificity of 95.6%. In the same perspective, Paternoster et al.[17] yielded a specificity rate of 99.85%. In another case series study conducted by Paternoster et al.[18] on COVID-19 ARDS patients, Macklin effect was even defined as the most consistent and highly reproducible predictor of barotrauma. Evidence of linear air collection along bronchovascular sheath allowed early detection and accurate management of patients at high risk of pulmonary barotrauma. Maccarrone et al.[19] also validated Macklin effect as a strong radiographic biomarker for pulmonary barotrauma. This correlation was confirmed through a case series study in which 9 out of the 10 patients who had Macklin effect developed pulmonary barotrauma.

When it comes to blunt neck trauma-induced Maklin effect, treatment depends on the associated lesions as well as the severity of respiratory and hemodynamic distress. Interdisciplinary approach is the first step in assessing the management plan for thoracic surgery and otorhinolaryngology. Conservative care allows for control in most uncomplicated cases, as spontaneous resolution is the most frequent evolution of the Macklin effect.[20] Under close monitoring, conservative care involves physical rest and chest pain management which can be obtained by drugs varying from anti-inflammatories to opioids. When presented with respiratory distress, patients should receive adequate respiratory support. In severe injuries with important hypoxemia, mechanical ventilation should be administrated, in other cases, noninvasive ventilation can allow maintaining adequate parameters.[21] Surgical intervention is rarely indicated but becomes necessary in the presence of complications such as a massive tension pneumothorax requiring a drainage thoracotomy. Additionally, in cases of pneumomediastinum infection, surgery may be indicated to control the mediastinitis.[22]

Conclusion

In conclusion, the Macklin effect is attributed to the pneumomediastinum following laryngotracheal injuries, whether they are penetrating, blunt, or iatrogenic. After alveolar rupture by pressure breakdown, the air extends by dissection and spreads along the bronchovascular sheaths into the mediastinum. This rare complication represents a major clinical challenge when dealing with neck injuries as it requires urgent detection and appropriate management. Early diagnosis should be made in case of suggestive signs such as dyspnea and subcutaneous emphysema. Given these symptoms, along with judicious imaging, a diagnosis is made, and the patient should undergo an appropriate therapeutic approach.

An interdisciplinary collaboration among thoracic surgery and otolaryngology is key to ensuring adequate care for each case. Conservative management remains sufficient in most cases, however surgical intervention may be necessary in case of respiratory compromise.

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All authors have contributed to this article.

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Ethics approval

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