**Tracheolith: a case report of a new disease entity**

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This case reports a stone in the trachea of a patient with tracheostomy. The presence of stone in the airway is uncommon condition. Broncholithiasis is characterized by formation of the stone due to erosion of calcified lymph node in the bronchus. There is no reported case of a stone in the trachea in the literature. The pathophysiology of this condition is still unknown, but we think that it is related to prolong tracheostomy. We recommend performing chest radiography in all cases of tracheostomy obstruction. More investigation and data collection of same cases are needed to have a more clear picture regarding the pathophysiology and the underlying cause.

**Keywords:**
airway, broncholith, stone, tracheolith

Introduction

The presence of stone in the airway is uncommon condition. Broncholithiasis is characterized by formation of the stone due to erosion of calcified lymph node in the bronchus. There is no reported case of a stone in the trachea in the literature. This case reports a stone in the trachea of a patient with tracheostomy. We would like to propose the term tracheolith to describe this condition.

Case report

A 13-year-old girl was diagnosed as a case of oculocerebral syndrome (Lowe’s syndrome). She was admitted to the ICU because of respiratory problem. During her stay of more than 1 month, tracheostomy was performed using silicon (portex) tracheostomy tube with cuff. Later, gastrostomy was performed for feeding purposes. Tracheostomy care was given either at hospital or by qualified nurse at home after discharge. About 2 years and 9 months later (post-tracheostomy), she was brought to emergency department with difficulty in breathing, despite adequate cleaning of tracheostomy tube and suctioning of secretions as per standard guidelines. Patient was taken to operation room and tracheostomy tube was replaced, but surprisingly dyspnea aggravated. Plain radiograph of the neck and chest revealed an opaque shadow in trachea between carina and tip of tracheostomy tube. A bronchoscopy showed a gritty mass, which was removed in piecemeal with difficulty. Slight oozing was noted at the site and was controlled. Postoperatively, airway obstruction was completely alleviated. The removed specimen was sent for analysis, which concluded to be a stone. Since then, she is under periodic observation and no similar complaint has been reported.

Discussion

It is uncommon to have stone formation in the airway. Broncholith is a term used to describe presence of stone in the bronchus due to erosion of calcified lymph node to the airway [1]. The most common underlying causes are tuberculosis and histoplasmosis [1,2]. Hemoptysis is the most common symptom and lithoptysis is almost peculiar for the disease [2,3]. Computed tomographic scan can help in the diagnosis and knowing the site to plan for removal [3,4]. Lobectomy is the traditional method of treatment but there are many reported cases managed using bronchoscopy or moreover laser and cryotherapy [2,5–7]. We want here to introduce a new term, tracheolith. In this case, we reported a presence of stone in the trachea of a patient with tracheostomy for 2 years. It is unusual cause of tracheostomy tube obstruction. Chest radiography showed the finding of radiopaque mass and bronchoscope was used to remove the mass (Figs. 1 and 2). The analysis of the mass confirmed the diagnosis of stone (Table 1). The patient was followed for 1 year after that without recurrence.

It is unclear what the underlying cause is, because the patient does not have history of tuberculosis or histoplasmosis. We suggest two theories to explain this phenomenon: to have calcified granuloma after prolong tracheostomy or formation of stone from stagnant secretion. It is unlikely to have lymph node eroding to trachea because it will be obvious on the bronchoscopy. As the stone is formed in
the trachea, it can be directly removed by the rigid bronchoscope.

Conclusion
Tracheolith is a stone formation in the trachea. As it leads to airway obstruction, it must be removed. The most effective and easy way is to perform rigid bronchoscopy. The pathophysiology of this condition is still unknown, but we think that it is related to prolong tracheostomy. We recommend performing chest radiography in all cases of tracheostomy obstruction.

Table 1 Chemical analysis of the stone

| Elements          | (%) |
|-------------------|-----|
| Calcium oxalate   | 16  |
| Calcium phosphate | 26  |
| Calcium           | 30  |
| Oxalate           | 10  |
| Ammonium          | 0   |
| Phosphate         | 15  |
| Magnesium         | 0   |
| Uric acid         | 0   |
| Cystine           | 0   |

The pathophysiology of this condition is still unknown, but we think that it is related to prolong tracheostomy. We recommend performing chest radiography in all cases of tracheostomy obstruction.

More investigation and data collection of same cases are needed to have a more clear picture regarding the pathophysiology and the underlying cause.

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Conflicts of interest
There are no conflicts of interest.

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