Case Report

Acute exogenous lipoid pneumonia due to foreign body aspiration in a child- an unusual and sinister presentation: A case report

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ABSTRACT

Background: Acute exogenous lipoid pneumonia is an extremely rare pathology in children and occurs because of aspiration of oil or lipid-containing material. The presentation and radiological findings vary widely and the exact mechanism of injury to the lungs is not clear.

Case Presentation: An 18-month-old girl presented with gradually increasing swelling and subcutaneous emphysema over the chest wall, neck, and face. The investigation revealed pneumomediastinum, pneumorachis, and soft tissue density in the left bronchus. The child underwent rigid bronchoscopy and retrieval of an organic foreign body.

Conclusion: Acute exogenous lipoid pneumonia can occur in children because of aspiration of oil-secreting organic foreign body. The presentation can be atypical, clinical suspicion and early diagnosis with timely intervention are crucial for the appropriate management.

Keywords: Exogenous lipoid pneumonia, Foreign body aspiration, Oil aspiration, Almond aspiration.

INTRODUCTION

Lipoid pneumonia (LP) is a rare pathology and occurs due to the presence of lipids in the alveoli. The lipids can be from endogenous or exogenous sources. It can present as either an acute or a chronic form.[1] Acute exogenous LP (ELP) usually occurs due to inadvertent aspiration of a large amount of lipid/oil and is extremely rare in children without predisposing factors. Foreign body aspiration (FBA) is not unusual in children and the reported literature suggests that 20% of children with FBA receive medical treatment for another pathology. [2, 3] Here, in the index case, the child aspirated an oil-secreting organic foreign body and presented later with a complication of an acute ELP.

CASE REPORT

An 18-month-old female child was presented in the emergency with complaints of swelling over the neck and face for two days. On examination, the child had subcutaneous emphysema which involved the chest wall, neck, and face.

The child was not in respiratory distress and maintained oxygen saturation up to 97% on room air. There was no history of trauma over the chest, neck, or face. There was no upfront history of FBA however, on asking leading questions, the mother came up with a history that "six days back the child was playing with her brother when he was eating almonds". During the evaluation, an X-ray chest (Fig. 1A) was done, which did not show any specific changes but slightly overinflated left upper lobe, a computed tomography (CT) scan (Fig.1B, 2A) of the neck and chest was done which was suggestive of soft tissue shadow in left main bronchus with the possibility of the foreign body along with pneumomediastinum, pneumorachis, and subcutaneous emphysema.
The child was taken for rigid bronchoscopy under general anesthesia. The left main bronchus was filled up with whitish oily secretions. After proper suctioning, an almond as the foreign body was seen (Fig. 2B) and removed. During check bronchoscopy the left bronchus (distal to foreign body location) was full of similar whitish oily secretion, bronchoalveolar lavage was given with normal saline and all secretions were cleared off. The child was extubated and shifted to the intensive care unit for observation. The child was discharged the next day and followed one week later, she was completely asymptomatic and the x-ray chest was also normal. During follow-up, the consent was taken for publication.

**DISCUSSION**

Foreign body aspiration (FBA) is a life-threatening condition in children. Conventionally, after FBA, breathing difficulty and coughing is the commonest presentation.[4] In the index case, the child remained asymptomatic for the initial 4 days and then presented with the unusual complication i.e. subcutaneous emphysema of the neck and face. Initially, penetrating trauma or some intervention at the pharyngeal region were our differentials, however, no history or clinical examination suggested these diagnoses.

The clinical presentation of FBA can vary according to foreign body material, the position of FB in the tracheobronchial system, FB dimensions, and the time duration to aspiration. Inert objects cause less reaction with the surrounding tissue and can remain in the tracheobronchial tree for days to weeks without causing much damage.[2] In the pediatric population, organic FB is the most common aspirated object. These organic FB objects induce more severe mucous inflammation due to their reaction with the surrounding tissue.[2] In the index case, the almond remained in the left bronchus for 6 days, and with time the reaction with surrounding tissue caused the almond to exude oil into the tracheobronchial tree. We suggest that this lipid would have collected and trickled in the lower bronchus leading to the current clinical presentation. The exact pathogenesis of subcutaneous emphysema and pneumomediastinum is not clear. It has been postulated that the aspirated lipid disperses into the tracheobronchial system, and extends to the alveoli. In the alveoli, the macrophages phagocytose this lipid. This stimulation of macrophages, in turn, causes a cytokine storm and activates severe inflammatory reactions that may lead to alveolar damage and rupture of the perivascular alveoli. Following the rupture of alveoli, air enters into the mediastinum via vascular sheath and subsequently deeper along the facial planes into the neck by following the path of least resistance.[5]

There is no reported case of paediatric acute ELP due to almond aspiration in English literature although cases because of aspiration of oil, used for a cultural or medicinal reason have been mentioned.[1,6] The factors that lead to different clinical presentations and radiological findings are types of oil, amount, and duration of exposure.[6] There are certain risk factors and underlying pathology in children for lipid aspiration: intellectually disabled, neuromuscular disorder, cleft palate, and oral administration of oil in a supine position.[1,6] In the index case there was no predisposing condition or risk factor for ELP or FBA.

The radiological investigation of choice to diagnose LP is a CT scan. Usually, CT scan reveals areas of consolidation, ground-glass opacities, and interstitial changes. Acute ELP changes can be noticeable on CT within an hour of aspiration and pulmonary opacities can appear in a day.[1,7] In the index case along with lung changes and pneumomediastinum, CT showed an endobronchial soft tissue shadow which clinched the diagnosis along with the history received on asking leading questions.

The management includes detection and discontinuation of the causative agent, appropriate supportive treatment, and management of complications.[4,6] In the case of FBA, removal should be done as early as possible. Most of the time spontaneous resolution of pneumonia occurs after removal of FB or discontinuation of the exposure. In non-responder cases, management includes frequent bronchoalveolar lavages that can help to improve signs and symptoms.[1] Some cases can develop complications.
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in the form of infection, fibrosis, bronchiectasis, and rarely critical cases can worsen respiratory failure.[1,3]

To conclude, acute exogenous lipoid pneumonia can occur in children because of aspiration of oil-secreting organic foreign body. The presentation can be atypical, clinical suspicion and early diagnosis with timely intervention are crucial for the appropriate management.

Conflicts of Interest: Nil

Source of Support: Nil

REFERENCES

1. Lu G, Xie Y, Huang L, et al. Study of Acute Exogenous Lipoid Pneumonia. Indian J Pediatr. 2016; 83:787-791.
2. Haddadi S, Marzban S, Nemati S, Ranjar Kiakelayeh S, Parvizi A, Heidarzadeh A. Tracheobronchial Foreign Bodies in Children; A 7 Year Retrospective Study. Iran J Otorhinolaryngol. 2015; 27:377-385.
3. Foltran F, Ballali S, Passali FM, Kern E, Morra B, Passali GC, Berchialla P, Lauriello M, Gregori D. Foreign bodies in the airways: a meta-analysis of published papers. Int J Pediatr Otorhinolaryngol. 2012; 14:S12-9.
4. Orji FT, Akpeh JO. Tracheobronchial foreign body aspiration in children: how reliable are clinical and radiological signs in the diagnosis? Clin Otolaryngol. 2010; 35:479-85.
5. Yi MS, Kim KI, Jeong YJ, Park HK, Lee MK. CT findings in hydrocarbon pneumonitis after diesel fuel siphonage. AJR Am J Roentgenol. 2009; 193:1118–21.
6. Marangu D, Gray D, Vanker A, Zampoli M. Exogenous lipoid pneumonia in children: A systematic review. Paediatr Respir Rev. 2020; 33:45-51.
7. Betancourt SL, Martinez-Jimenez S, Rossi SE, Truong MT, Carrillo J, Erasmus JJ. Lipoid pneumonia: spectrum of clinical and radiologic manifestations. AJR Am J Roentgenol 2010; 194:103-109.