

Exogenous lipoid pneumonia following lactulose intake in an infant: Clinical case

Abstract

Exogenous lipoid pneumonia ELP is a rare condition in common pediatric practice. Its clinical presentation is non-specific. Delay in diagnosis and treatment exposes infants to chronic respiratory failure. We report the observation of a 2-month-old infant with respiratory signs that drag on. The imaging showed several negative density pulmonary opacities. Foam cells were isolated on sputum cytological study. Taking lactulose for constipation suggested Exogenous lipoid pneumonia. The aim of herein observation is to draw attention to mineral oil administration drugs risks in infants especially if they are at risk.

Keywords: Exogenous lipoid pneumonia - Lactulose - Infant.

Introduction

Exogenous lipoid pneumonia (ELP) is a rare condition in routine pediatric practice. Its clinical presentation is non-specific. Delayed diagnosis and treatment expose the patient to respiratory complications that can lead to chronic respiratory failure.

Our objective comments draw attention to the risks of administering mineral oil-based medicines to young children, especially if they are at risk.

Case presentation:

G.M., a 2-month-old infant, was admitted to hospital with dyspnea, malaise, cough and tachycardia, which had been evolving for 20 days in a context of apyrexial. This symptomatology did not seem to respond to the treatments administered during several consultations. The patient was born of a well-monitored pregnancy carried to term. There were no incidents during the neonatal period. She has had regurgitation since birth, with untreated breastfeeding and bouts of cyanotic coughing during feedings. Her constipation was treated orally with lactulose. Examination revealed an infant in good general condition, Rose, weighing 5 kg for a height of 58 cm, apyretic, polypnea at 70 cycles/min, with no signs of struggle and snoring. There were no signs of heart failure. Biological workup does not suggest bacterial infection. The abdomen is soft and there is no hepatosplenomegaly. The rest of the examination was normal. No signs of bacterial infection (white blood cell count 8800 c/mm³, CRP 13 mg/l).

A frontal chest X-ray and chest CT scan were performed (figs. 1 and 2). The frontal chest X-ray showed multiple alveolar-type opacities (blurred boundaries with bronchogram), one in the right upper lobe and bilateral para-hilar. Chest CT revealed several bilateral parenchymal para-hilar ground-glass opacities, heterogeneous and containing a few areas of negative density.



Figure 1 Front chest X-ray shows multiple alveolar opacities (blurred boundaries with bronchogram), one in the right upper lobe and bilateral para-hilar.



Figure 2 Chest CT scan reveals several bilateral, heterogeneous, intraparenchymal para-hilar ground-glass opacities, with some areas of negative density.



Figure 3 Chest CT parenchymal opacities with negative density between -154 and -122 HU.

Given the notion of vomiting since birth, we performed PH-metry, which was not in favor of gastroesophageal reflux (acid time (PH<4) less than 2% on a 24-hour recording, gastric tube controlled taking meals into account). The notion of taking kerosene oil to treat constipation prompted us to explore the possibility of exogenous or oily inhalation pneumonitis or lipid pneumonia. To confirm the diagnosis, we reassessed the density of the parenchymal opacities, which were between -154 and -122 HU and therefore lipidic in nature. We were able to demonstrate the presence of lipid foam cells in the sputum with multiple sessions of aspirations. Our patient was treated symptomatically with respiratory physiotherapy and oxygen therapy according to hemoglobin oxygen saturation, and prednisone 2 mg/Kg/d for 2 weeks. Clinical evolution was favorable after 10 days of hospitalization, and radiological clearance was obtained after 3 months.

Discussion

Lipid or lipoid pneumonitis comprises two entities: the exogenous form, also known as exogenous lipoid pneumonitis (ELP) or oily pneumonitis, and the endogenous form, known as endogenous lipoid pneumonitis or cholesterol pneumonitis. The latter is secondary to the release of lipids normally contained in lung tissue destroyed upstream of an obstructive bronchial lesion or by suppuration. More rarely, endogenous lipoid pneumopathy results from fat embolism, alveolar proteinosis or lipid storage disorders[1].

exogenous lipoid pneumonia, a rare condition, is most often the result of repeated micro-inhalations of mineral oil. Lung damage varies according to the type of oil (mineral, vegetable or animal), the quantity and duration of intoxication [1]. The acute form corresponds to significant inhalation, and although it can lead to acute respiratory failure, it is often unrecognized due to the non-specific nature of the symptoms and its rarity. The chronic form

corresponds to repeated, pauci-symptomatic micro-inhalations, leading to fibrosis lesions associated with a granulomatous "paraffinoma" reaction [2].

From a pathophysiological standpoint, mineral oil is a pure, inert hydrocarbon. Its tracheobronchial penetration inhibits the cough reflex and the action of vibratile cilia. At the alveolar level, mineral oil is responsible for an intra-alveolar infiltrate of macrophages associated with an acute inflammatory reaction. The oil is then emulsified and phagocytized, leading to vacuolation of the macrophages, which migrate to the interstitium, forming a multinucleated, vacuolated giant cell granuloma. In the later stages, the disease progresses to fibrosis [3]. In the majority of cases, ELP results from the administration of mineral oil given orally to treat constipation, or from the inhalation of milk [4]. Rabah et al [6], report the occurrence of ELP after rectal administration of mineral oil in a patient with Hirschsprung's disease via an embolism mechanism. De la Rocha et al [7] reported 1 case of ELP following accidental aspiration of mineral oil in a 13-month-old infant who required oxygen therapy for a period of five months. Furthermore, vigorous opposition by infants to the oral administration of mineral oil exposes them to aspiration [6]. Patients with motor delay and/or associated swallowing disorders are particularly at risk of developing ELP [7].

The clinical presentation of ELP is non-specific and depends on the patient's age, the volume of oil aspirated, the presence or absence of risk factors and the chronicity of aspiration. The latent nature of the disease is classic, hence the importance of the anamnesis in authenticating and characterizing exposure to the lipid product and in identifying risk factors. In the series by Hari et al,[8], 41% of cases were discovered incidentally, of which 25% remained clinically latent. Cough, chest pain, fever, weight loss and crepitus are the most frequently observed clinical symptoms. In another study, the same percentage of cases was found to be latent,

asymptomatic in 41% of cases, fever in 39%, weight loss in 34%, cough in 64%, dyspnoea in 50%, chest pain in 21%, haemoptysis in 13% and, on physical examination, 54% of crackling rales [8]. These patients are at risk of non-tuberculous mycobacterial infections, which can be revealing, as lipids increase the growth of these organisms and prevent their phagocytosis by macrophages [9,10 ,19,20].

Standard radiographic signs are nonspecific and variable, ranging from a peri-hilar, bilateral lobar alveolar syndrome affecting mainly the lower and posterior lobes, giving a "ground-glass" appearance, to an interstitial and nodular or even pseudotumoral syndrome. "paraffinoma". The appearance of a bilateral excavated nodule confusing with Wegener's granuloma has been reported, following inhalation of kerosene oil. Chest CT is highly suggestive of the lipidic nature of opacities found on standard radiography, as it measures negative density (between -150 and -30 Hounsfield units) within these lesions [11,12 ,21]. It can also show thickening of the interlobular septa, areas of ground glass or the association of small reticulated opacities within areas of ground glass known as "crazy-paving". CT can also be used to determine the exact number of lesions, their location and size, and to monitor their evolution. Some authors also suggest comparing this density with that of subcutaneous adipose tissue, whose density is comparable [13]. Lee et al [17] analyzed high-resolution CT findings in 25 patients, according to the presence or absence of predisposing factors, and the route and duration of inhalation of a mineral oil "squalene" derived from shark liver. Results were classified into three patterns: diffuse ground-glass opacity, consolidation, and interstitial abnormalities. The ground-glass appearance was mostly found in patients with predisposing factors and after nasal instillation in large quantities. The appearance of consolidation was found mainly in patients with no predisposing factors and taking squalene for several months; the patients with Interstitial syndrome all show "squalene" uptake beyond 1 year. A similar study was carried out by Baron et al [15] on 15 cases of ELP, the aim of which was to distinguish between acute and chronic forms of PLE on the basis of radiological presentation.

He concluded that the features favoring acute forms were the presence of a pleural reaction and enhancement on standard imaging and chest CT ($p < 0.05$). The presence of masses was synonymous with chronic forms ($p < 0.05$). Thoracic MRI can confirm the diagnosis on iso signal to fat of lesions on T1-weighted sequences. Fat suppression sequences show a decrease in lesion signal, enabling differentiation from hemorrhagic infiltrates. In practice, analysis of radiological images (standard radiography and thoracic CT or even MRI) in the light of anamnestic data (exposure to oil with or without risk factors), and of the evolution (reversibility or not) makes it possible to link clinical symptomatology to ELP and to outline a prognosis. Cytochemical analysis of bronchoalveolar lavage fluid can, if necessary, confirm ELP. Bronchoscopy with examination of the bronchoalveolar lavage fluid enables the diagnosis of ELP to be made, as lipid globules are found on the surface of the fluid and lipid-laden macrophages are present, with measurement of the load index [15]. Bronchoscopy has the added advantage of exploring the airways to rule out other causes of dragging pneumonia that could be confused with ELP. On the other hand, invasive investigations such as needle biopsy or thoracotomy are not justified [16] if the clinical history and thoracic CT scan are suggestive enough, unless it is a granuloma or paraffinoma with signs of activity.

Treatment of ELP in children is generally symptomatic, with resolution of symptoms and radiographic abnormalities within months of cessation of exposure [22,23,24]. In cases of diffuse lung damage, high-dose prednisone oral [17] and therapeutic lung lavage may be considered [18]. It is clear that the prevention of another episode of pneumonia should involve not self-medicating, to consulting a specialist in the event of worrying or lingering symptoms, and eliminating lactulose as a 1st treatment for constipation[24].

Conclusion

ELP should be considered in the presence of exposure to lactulose, especially if there are risk factors and radio-clinical discordance. The administration of lactulose for the treatment of chronic constipation should be approached with caution, and preferably avoided in infants resistant to its administration, and in children with or without mental retardation and swallowing disorders.

Consent

As per international standards, parental written consent has been collected and preserved by the author(s).

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