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Case Report

An unusual radiologic manifestation of hypersensitivity pneumonia

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ARTICLE INFO

Article history:
Received 29 March 2017
Received in revised form
19 April 2017
Accepted 29 April 2017
Available online 16 June 2017

Keywords:
Computed tomography
Hypersensitivity pneumonia
Organizing pneumonia pattern
Children
Paediatrics

ABSTRACT

Hypersensitivity pneumonia is clinically suspected and can be characterized on computed tomography by its pattern of diffuse lung disease, in children, as in adults. However, identifying the diagnosis is not always as simple. We report an organizing pneumonia pattern of hypersensitivity pneumonia that can be seen in adult patients, but has not been reported in the pediatric population.

Introduction

Hypersensitivity pneumonia is one cause of chronic lung disease seen on computed tomography (CT) and can be seen in adults, secondary to external, and often, occupational allergens [1]. It can be seen in children secondary to environmental allergens, but often, the allergen is not found. The diagnosis of hypersensitivity pneumonia in children can be challenging [1]. The pattern-based approach to chronic interstitial lung disease in children is similar to that in adults where, at first, the typical radiologic pattern is identified with different possible descriptions: unusual interstitial pneumonia, nonspecific interstitial pneumonia, hypersensitivity pneumonia, and organizing pneumonia [2,3]. Second, if the cause (some common and others rare) of this pattern is not identified, a diagnosis of an idiopathic condition is suggested [2,4]. The typical radiologic hypersensitivity pneumonia pattern includes the presence of centrolobular ground glass opacities, seen diffusely in both lung fields. In the chronic phase, fibrotic changes might be seen. In adults, an organizing pneumonia pattern of focal consolidation can be seen with hypersensitivity pneumonitis [2,5]. This pattern has

Competing Interests: The authors have declared that no competing interests exist.

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http://dx.doi.org/10.1016/j.radcr.2017.04.014

not been reported in children. We present a case of an unusual radiologic presentation of hypersensitivity pneumonia presenting with an organizing pneumonia pattern of diffuse lung disease on CT in a 7-year-old child.

Case report

A 7-year-old, otherwise healthy girl, presented with a 6-month history of chronic cough. She had been treated with antibiotics (oral antibiotics, 15 days) for 2 episodes of fever without complete resolution of symptoms. Her initial chest X-ray demonstrated a focal left lower lobe retrocardiac alveolar infiltrate with air bronchograms, consistent with pneumonia (Fig. 1A). Two-weeks later, X-ray showed the appearance of alveolar infiltrates in the right upper and lower lobes and in the left upper lobe with partial regression of the left retrocardiac alveolar infiltrate seen earlier (Fig. 1B). CT (Siemens SOMATOM Definition Edge, 80 kVp, tube current 216 mA, slice thickness 0.75 mm, and CTDIvol 1.5 mGy) confirmed an organizing pneumonia pattern of focal opacities in both lung fields, and one with mixed attenuation (central ground glass and peripheral alveolar consolidation), compatible with the Atoll sign in the left lung fields (Fig. 2). A bronchoscopy showed no anatomic anomalies, and the bronchoalveolar lavage demonstrated a normal cellular repartition with an absence of cellular anomalies (Gold and Arhens score, negative periodic acid-schiff stain, normal CD4:CD8 ratio, and a normal CD1 stain). Bacteriology, virology (adenovirus and cytomegalovirus), and a quantiferon test showed negative results. Owing to the persistence of a chronic cough and increased respiratory effort, an oral short-acting steroid treatment (prednisone 2 mg/kg/d) was initiated. Clinical improvement of respiratory symptoms and in lung function test (functional vital capacity and total lung capacity) was demonstrated post-treatment. After 2 months of corticosteroid weaning, the patient presented again with shortness of breath. CT performed postweaning, demonstrated the appearance of a new focal perihilar apical segment left lower lobe alveolar consolidation with air bronchograms. The diagnosis of organizing pneumonia was retained based on the clinical and CT findings.

Follow-up CT 13 months after presentation, and 6 months after initiation of steroid treatment demonstrated a new

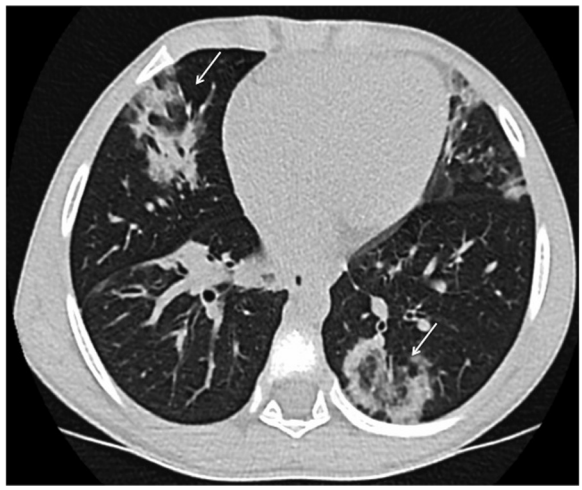

Fig. 2 – CT at treatment initiation demonstrating patchy focal alveolar consolidations in both lung fields with a central ground glass opacity surround by a peripheral alveolar consolidation (the Atoll sign) in the apical segment of the left lower lobe.

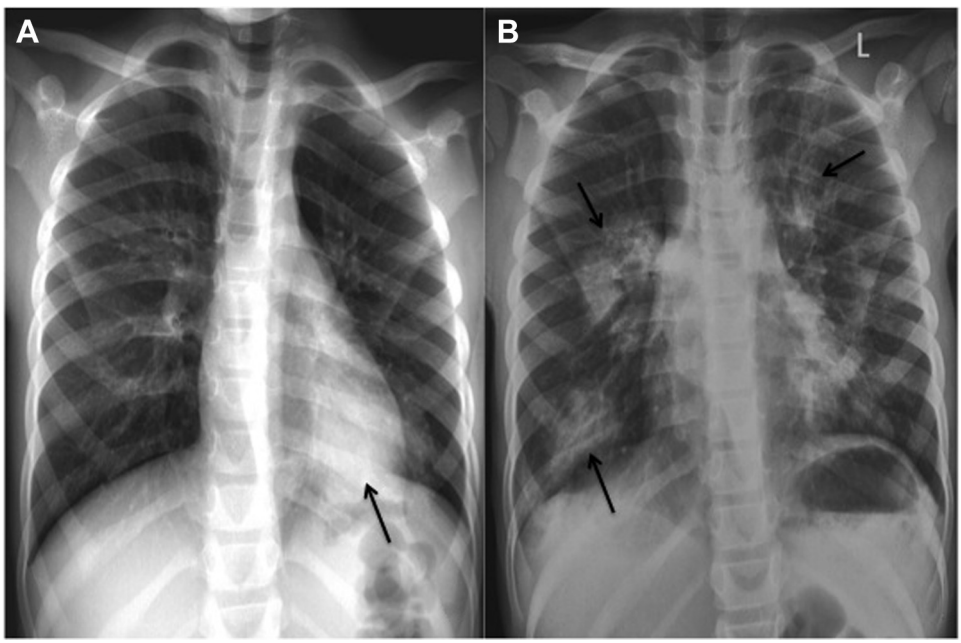

Fig. 1 – (A) Chest X-ray at presentation demonstrating left lower lobe retro-cardiac consolidation (arrow). (B) Chest X-ray 1 month later demonstrating multiple focal lung consolidations in the right and left lobes (arrow), disappearance of the retro-cardiac consolidation.

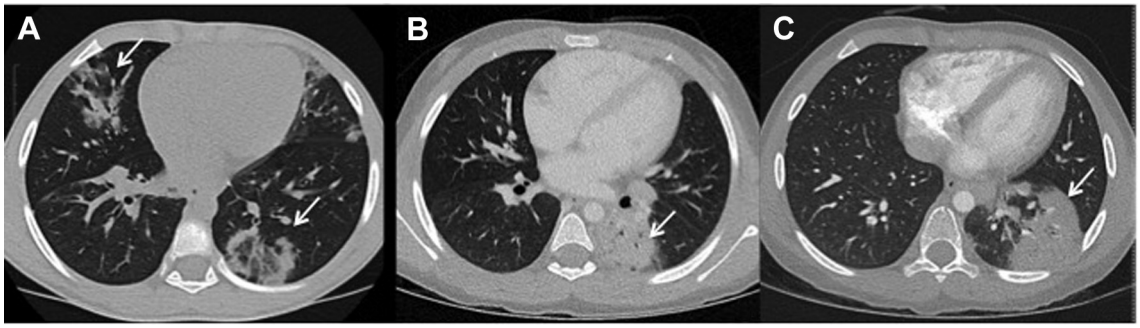

Fig. 3 – (A) Focal mixed attenuation lung consolidations with the Atoll sign in the left lower lobe and patchy focal consolidations in the right lobe, on initial CT at presentation (arrows); (B) new focal alveolar consolidation with air bronchograms in the left lower lung (arrow), postcorticosteroid weaning (8 months after presentation); and (C) new focal alveolar consolidation with air bronchograms in the left lower lobe at follow-up (6 months after initiation of corticosteroid treatment and 13 months after presentation) (arrow).

peripheral posterolateral left lower lobe alveolar consolidation with air bronchograms (Fig. 3). Given the clinical history and the persistence of the pulmonary abnormalities, a biopsy was deemed necessary. A transbronchial lung biopsy was not feasible due to the localization of the consolidation, and an open surgical biopsy was performed. Histologic immune cytology stains demonstrated the presence of interstitial fibrosis, obliterative bronchiolitis, and poorly circumscribed epithelioid granulomas consistent with the diagnosis of hypersensitivity pneumonitis. However, blood work demonstrated the absence of positive precipitin antibodies (indoor pets and fungi) and an absence of cellular abnormality in favor for autoimmune disease. Further questioning showed negative results with respect to environmental triggers such as bird droppings, actinomycetes, fungal, or air conditioner exposition. A treatment with daily corticosteroids was continued in addition to monthly intravenous corticosteroids. At follow-up, progressive diminution of cough was noted.

Discussion

Hypersensitivity pneumonia usually presents with diffuse, bilateral centrolobular ground glass opacities, with or without signs of fibrosis. We present a case of atypical CT findings in a child with hypersensitivity pneumonia. The typical hypersensitivity pneumonia pattern has 3 different presentations: in the acute setting, it presents with nonspecific diffuse centrilobular ground glass opacities [2,5]. In the subacute phase it presents with nonspecific patchy ground glass opacities, centrilobular nodules, mosaic perfusion, and the head-cheese sign (lucent lung regions of air trapping, patchy ground glass opacities, and normal lung) [2]. In the chronic phase, findings include irregular reticulations, traction bronchiectasis, honeycombing (mid-lower lung distribution or a diffuse distribution) [2]. The organizing pneumonia pattern of focal consolidation in the acute or chronic phase can been seen in adults [2,5]. However, this has not been reported in children.

Our patient presented with an organizing pneumonia pattern of diffuse interstitial lung disease on CT. This pattern is most commonly demonstrated by patchy bilateral peribronchovascular opacities. Occasionally, it can present with patchy bilateral ground glass opacities or, more rarely, with interstitial perilobar thickening (crazy paving pattern) [6]. Ring-shaped peripheral opacities with central ground glass attenuation often referred to as the "Atoll sign" or the "reverse halo sign," may be seen (as seen in our patient), and if present is highly suggestive of the organizing pneumonia pattern [2,6].

Both the hypersensitivity pneumonia pattern and the organizing pneumonia pattern have a number of etiologies that need to be investigated. The organizing pneumonia pattern seen in this case can be secondary to infections, such as pneumococcal pneumopathy, iatrogenic, drug- or radiation-induced lung disease (often unilateral) [4]. This pattern may also be seen with connective tissue disorders, in particular dermatomyositis, rheumatic arthritis, and systemic lupus erythematosus [3,4]. In the oncologic context, this pattern may be seen with leukemia, Ewing sarcoma and Hodgkin disease. Occasionally, the diagnosis is proposed on lung biopsy performed for aspergillosis in immunosuppressed patients [4].

In children, the diagnosis of hypersensitivity pneumonia is challenging, and known CT characteristics include diffuse centrolobular ground glass opacities, mosaic attenuation, and eventually, signs of fibrosis. An organizing pneumonia pattern of hypersensitivity pneumonia is described in the adult literature [2,5]. This has not been described in the pediatric population. This case illustrates a pediatric patient with organizing pneumonia pattern of hypersensitivity pneumonia.

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