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Prikaz slučaja /
Case report

HYPERSENSITIVITY PNEUMONITIS - „ BIRD BREEDER'S (FANCIER'S) LUNG"- *A Clinical case*

HIPERSENZITIVNI PNEUMONITIS - „PLUĆA ODGAJIVAČA PTICA - GOLUBOVA"- *Prikaz slučaja*

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Ključne reči

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Abstract

Hypersensitivity pneumonitis (HP) or extrinsic allergic alveolitis (EAA) is an inflammatory disease caused by repetitive inhalation of antigens. Given that HP development is influenced by many etiological factors and the clinical presentation is diverse, it can be viewed as a syndrome with highly variable and very difficult to assess prevalence. It is relatively infrequent disease, which accounts 2% of cases of interstitial lung diseases. Radiological findings, when present, may include ground glass opacities and mosaic attenuation on computed tomography. Clinical improvement upon removal of antigen helps cement diagnosis and facilitate the treatment of antigen avoidance and corticosteroids. We present an interesting case of hypersensitivity pneumonitis, particularly bird breeder's (fancier's) lung in a 67-year-old man old man exposed to birds which was his hobby (pigeons) whose symptoms occurred much later than the usual timeframe reported in the literature.

INTRODUCTION

Today, HP is most commonly used to depict illness previosly reported as external allergic alveolitis [(1)]. Nowadays, it is illness that happen after presentation to a large variety of environmental settings and antigens (organic dust, it is combined with farming (mouldy grain or hay handling) calling farmers lungs, birds, humidifiers, mouldy wood and a diversity of other settings where moulds abound [(2, 3)]. One population study assessed that the number of newly ill of interstitial lung illness was 30 per 100,000 persons for one year [(2)]. Several diagnostic criteria have been published for HP, but the most used is the one set by Richerson and associates [(4, 5)]. Clinical, there could be discussed acute, subacute and chronic manifestation of disease. Which form will be represented it depends on the nature of the attacking agents, their physical characteristics and concentration, the intensity of individual exposures and individual changeability. The acute answer after inhalation is a nonspecific diffusive pneumonitis with inflammatory cell infiltration of the bronchioles, alveoli and interstitium. In the subacute and chronic phase, loosely shaped, barren, epithelioid cell granulomas can be spilled in the interstitium. Given the image of a single specified HP, its eviction is obligatory. Corticosteroid treatmet is major, in spite of the leave of long-standing utility proof [(4, 5, 6)].

CASE REPORT

A 67-year-old man was sent from his general practicioner (GP) to our clinic under clinical presentation of severe and persistent dry cough, night sweats, and weight loss of 15 kg over the preceding four months. The dry cough and night sweats lasted for three months in spite of curing by his GP. On admission, he denied symptoms of shortness of breath on exertion, weakness, or increase in body temparature. His cardiological findings including echocardiography

was within normal range. He was a smoker, but last months since his difficulties started, he stopped it. Significantly, he had not treated any pulmonary disease. He did not use any drugs or herbal medicines. He was retired military solder. There was nothing special about the physical examination the findings and its vital signs were stable. Examination of the chest demonstrated clear breath sounds without rales or wheezing. The cardiac and abdominal examinations were also without significance. During examination skin and nails, skin rash or nail clubbing was not noticed. Also he had no musculoskeletal abnormalities. We investigated several reasons for chronic cough, such as upper respiratory cough syndrome and gastro-esophageal reflux syndrome, but no specific findings have been reported. Chest X ray was within normal range according to age. Next step was to perform the pulmonary function test which were in the normal range, except for blandly reduced diffusion capacity of carbon monoxide. A chest computed tomography (CT) scan showed mosaic pattern: a combination of secundary lobules with ground-glass opacity due to lung infiltration and secundary lobules some with patchy lucency due to bronchiolitis with air trapping (Figure 1), which is consistent with HP. Laboratory results were within normal range. Sputum was sterile. Urin analysis did not reveal proteinuria or glucosuria. We also wanted to check it out total serum IgE level slightly increased to 125 kU/L (normal range, <100 kU/L). The most commonly tested autoimmune markers such as antinuclear antibody, rheumatoid factor, cytoplasmic anti-neutrophil antibody, perinuclear anti-neutrophil cytoplasmic antibody, anti-Scl 70, anti-SS-A/Ro, and anti-SS-B/La were all in the normal limits.

After all examinations, somehow he mentioned that he has dovecote since his childhood and that pigeons are his passion. He thought that such important fact was irrelevant. We were close to being diagnosed, suspected that the HP was connected to pigeon exposure. He was advised to change his hobby, but he was persistent since it was his passion from childhood. We decided to start with 30 mg oral prednisolone daily. After oral prednisolone treatment, the dry cough was a slightly reduced, but it was not completely resolved. After this treatment, we wanted to know more about the survival of dry cough so we decide to send patient for more precisely diagnosis. He was underwent surgical lung biopsy via video-associated thoracoscopy (VATS). Pathologic findings were showed, which made of multifocal, patchy fibroblastic proliferations and chronic inflammatory cell infiltration around the respiratory bronchiole. Considering the results of all the findings, including the biopsy, we came to the conclusion that it was chronic HP. We recommended again strongly to change occupation, and since the cough did not stop, he quitted his hobby and we narrowly reduced the oral prednisolone slowly until discontinuation. After that the symptoms improved and he felt well.

DISCUSSION

According to US administrative claims-based data, between years 2004-2013. HP was more commonly reported in women and population over 65 years of age. In most cases it is described as chronic HP which is simillar to data from our study [10]. One author and his associates found that HP is tipically subacute in their presentation [11, 12]. Our patient has symptoms like dry cough, night sweats and 15 kg body weight loss. According to research by some authors, the most common symptoms are fever, chills, dyspnea, productive/nonproductive cough, malaise, myalgia, weight loss and rales [13]. Our patient had a normal echocardiography findings. A retrospective study of 73 cases of chronic HP with an echocardiography evaluation was described by some authors. Pulmonary hypertension which consider systolic pulmonary arterial pressure >50mmHg, was noted in about 20% of patients [14]. In our study patient was smoker but he did not suffer from any pulmonary disease. With all symptoms which he had, our patient was not taking any medications. Some authors described a cases of few patients with similar age, with similar symptoms, like our patient, who was initially treated with antibiotics for community-acquired pneumonia [15, 16]. In our study there was no significant findings in physical examinations and chest examination showed clear breath sound without rales or wheezing which is an unsual finding in such patients. Some studies had shown pathological finding in even 88% examined patients [17]. Generally, chest X ray has no significance in such cases but in study done by some author and his associates, it was showed that in two patients, both had patchy opacifications and reticular nodular shadows in middle zones of both pulmonary fields, described by chest X ray [12]. According to some authors, lung function is more severely impaired in the youngest group of patients (<30) with diffusing capacity of the lungs for carbon monoxide (DLCO) <40% in 69.2% of these patients. A restrictive pattern was present in 92.3% of patients in these group [18]. Our patient had normal lung function with slightly reduced diffusion lung capacity of carbon monoxide. A CT scan presented combination of secondary lobules ground-glass opacity and secondary lobules some with patchy lucency. Some author and his co-workers showed some clinical presentations of chest CT, in acute phase: normal CT or some ground-glass opacities with centriolobular micronodules, subacute phase: ground-glass opacities with centriolobular micronodular pattern (<5mm), mosaic pattern and chronic phase: pharenchymal fibrosis including bronchiectasis [13]. Described changes from acute and subacute phase have similar chest CT changes in our case. Our patient had normal laboratory and urin results which was not in accordance with previous results [19]. Our patient had sterile sputum, also. Recent examinations had discovered that patients who were divided into two groups /one group were exposed to bird (BHP), other to fungi (FHP) inhalations/, the FHP group had higher sputum neutrophilia levels and in increase IL-8 levels than in those with BHP [20]. Serum total IgE level was slightly elevated to our patient. Some papers showed laboratory data for twelve patients where was included measurement of IgE level where 11 patients significant increase of IgE [21]. In the research done by some authors, they found that hypersensitivity pneumonitis screening tests were negative, as were tests for rheumatoid factor (RF), antinuclear antibodies, anticyclic citrullinated peptide (anti-CCP), anti-Smith antibodies, antineutrophil cytoplasmic antibodies (ANCA-s), antiribonucleoprotein and antibodies to La (SSB), Ro (SSA), Scl-

70 and anti Jo-1 which is the same data like in our research [19]. After all examinations, our patient mentioned that pigeons are his passion which help us to connect with exposition. The similar data gave two patients from Sri Lanka, exactly they gave information about red-vented-bulbul birds [12]. We treated him with oral prednisolone which is the same treatment like in the one research [12]. The best way to confirm our suspicions was lung biopsy via VATS. Video-assisted thoracoscopic surgery (VATS) was performed to a 68-year-old woman with dry cough in 2010 and specimens showed prominently interstitial pneumonia pattern [22]. The biopsy presented multifocal stunting fibroblastic proliferations and chronic cell infiltration towards the respiratory bronchiole. According to data from Sri Lanka VATS biopsy showed patchy and focal interstitial callosity with lymphocitic infiltrate, minimal fibrosis and fewest noncaseating granulomata in the interstitium [12]. We continue to treat HP with oral prednisolone until discontinuation. Terras and coworkers treated their patients with Azathioprine (AZA). AZA treatment showed improve of forced vital capacity (FVC) at 12 and 24 mounths, a slightly increase in total lung capacity (TLCO) and 6-minute walking test (6MWT) [23].

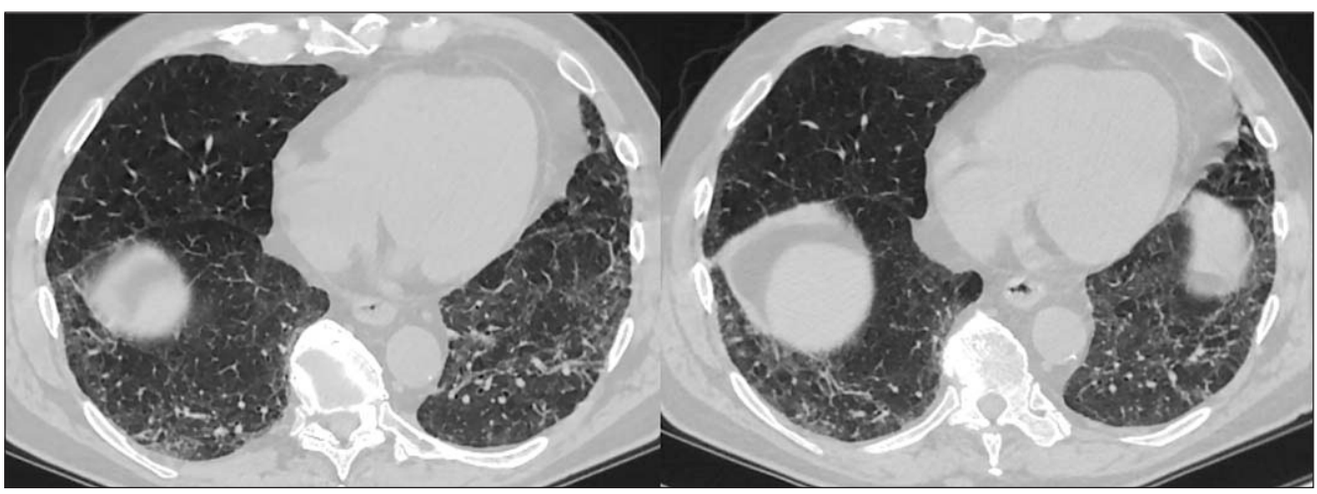

Figure 1. *Lungs CT demonstratelobules some with patchy lucency due to bronchiolitis with air trapping.*

Sažetak

Uvod: Hipersenzitivni pneumonitis (HP) ili spoljašnji alergijski alveolitis je zapaljenska bolest pluća prouzrokovana ponavljajućim udisanjem antigena. Uzimajući u obzir da na nastanak HP utiču različiti faktori, da je klinička prezentacija raznolika, može se posmatrati i kao sindrom sa veoma promenljivom i vrlo teškom procenom prevalence. HP je relativno retka bolest, sačinjava 2% intersticijskih bolesti pluća. Radiografski nalazi, kada su prisutni, prezentuju se kao "ground glass" opacifikacije i prigušenje mozaika na kompjuterizovanoj tomografiji. Kliničko poboljšanje nakon uklanjanja antigena pomaže postavljanju dijagnoze i olakšava lečenje izbegavanjem antigena i kortikosteroida. Prikazujemo slučaj HP, "pluća odgajivača ptica-golubova" kod 67 godina starog muškarca koji je stalnom kontaktu sa pticama, a što je inače i njegov hobi, čiji su se simptomi ispoljili mnogo kasnije nego što je uobičajeno prema podacima iz literature.

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