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Case Report

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Desquamative interstitial pneumonia in association with chronic necrotizing aspergillosis, a case report and literature review

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Abstract

Desquamative interstitial pneumonia (DIP) is a form of interstitial lung disease that is directly related to smoking. In addition to smoking, other factors have been implicated in its etiology, including: systemic disorders, dangerous materials in the environment, drugs and infectious agents. By reviewing the literature, we find that there are very rare cases that indicate infections as causing DIP. Here the author report on a 58-year-old male who was addict and complained of a dry cough with dyspnea for one month. TBLB was performed and pathology result was consistent with DIP. He received prednisolone 5 mg twice a day, but his symptoms persisted. Open-lung biopsy was performed and it illustrated *Aspergillus pneumonia* (Chronic necrotizing aspergillosis). He was treated with corticosteroids combined with antifungal agents.

Keywords: Desquamative Interstitial, Chronic Necrotizing Aspergillosis, Lung.

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INTRODUCTION

The idiopathic interstitial pneumonia (IIP) is one of the main types of diffuse parenchymal lung diseases. The new classification divides IIP into three main groups: "Major IIP, rare IIP and unclassifiable IIP. Major IIP is further divided into three groups, namely chronic fibrosing IIPs, smoking-associated IIPs and acute or sub-acute IIPs". Desquamative interstitial pneumonia (DIP) is a great type of smoking-related IIP. It is characterized by the presence of macrophages that fill the alveoli and there is as well as mild interstitial inflammatory reactions. The disease is caused by direct or indirect exposure to cigarette smoke. For the first time, in 1965, Liebow et al. described DIP [1]. It is one of the rarest forms of IIP. Although the link between the DIP and smoking has been established, it is also seen in those who have never smoked or those who have quit it. In addition to smoking, other factors have been implicated in its etiology, including: systemic disorders, dangerous materials in the environment, drugs and infectious agents. Lougheed MD *et al*. claimed that substances such as asbestos, talc, graphite, silica and aluminum can cause the DIP [2]. The association of taking drugs such as macrolides, sirolimus, nitrofurantoin, sulfasalazine and tocainide with DIP has been reported [3]. Margaritopoulos GA *et al*. pointed to the possible role of aflatoxin in causing the disease [4]. There are a few reports about the role of infectious agents in etiology of the disease. For example Iskandar *et al* [5]. has reported a case of DIP in a patient who suffered from hepatitis C. The author believed that immunological reactions to HCV had played a role in causing the DIP. Hasegawa *et al* [6]. also identified a case of DIP in a 72-year-old man with hepatitis C who had no history of smoking. Viral agents, like as CMV has also been introduced as a possible cause of the DIP. In this way an 8-month-old infant who suffered from CMV infection was reported with findings compatible with DIP [7]. Sung *et al* [8]. described an interesting case of DIP following infection with CMV and aspergillus in a kidney transplant patient. Here we are reporting a case of DIP in a 58-year-old male in association with aspergillous infection who had history of active smoking.

CASE REPORT

A 58-year-old male who had 40 pack-years smoking history referred to pulmonary ward with chief complaint of productive cough, dyspnea and once hemoptysis since 30 day ago. The patient had and

swelling of the limbs. The patient was a builder. He denied any history of significant weight loss and night sweats. General physical examination revealed an alert and conscious man with some difficulty in breathing. There was no evidence of cyanosis or clubbing. Peripheral lymphadenopathies were not found. On lung auscultation coarse crackles were heard. Abdominal examination was unremarkable. Laboratory data were unremarkable except for mild leukocytosis ($11.59x10^3$) with neutrophilia (89.2%) and high ESR level (85 mm). A chest CT (Fig. 1) scan revealed a mass consolidation with central necrosis in RUL with alveolar infiltration in the RML and both lower lobes. There was mild pleural effusion in the right side. Sputum direct smear and culture were negative for mycobacterial infection. The patient underwent bronchoscopy and BAL and TBLB were done. Alveolar lavage fluid microbiology was negative. Histopathological examinations showed numerous macrophages in the alveolar spaces with interstitial inflammatory cell infiltration and minimal fibrosis (Fig. 2). The pathologic results were compatible with DIP. Rheumatologic panel tests (ANA, ANCA, Anti ds-DNA, Anti RO, Anti LA) were negative. The patient was advised to quit smoking and received prednisolone 5 mg twice a day. After 40 days the patient came back with chief complaint of abdominal pain, constipation and exacerbation of dyspnea. His abdominal pain was periamblical and didn't radiate to any site. Abdominal sonography was unremarkable. His arterial blood gas analysis showed severe hypoxemia; therefore, he was transferred to the intensive care unit for respiratory support Regarding to his past medical history he underwent a chest CT scan. Chest CT scan showed evidence of irregular thick wall cavitary lesions in the anterior segment of the right upper lobe and a mass lesion in superior segment of RLL. Consolidation with air bronchogram in left lower lobe was seen (Fig. 3). An open lung biopsy was done. Histologic examination on H&E stained slides revealed pulmonary tissue with narrow septate hyphae in the parenchyma, foci of necrosis and foreign body type multinucleated giant cells. There was also evidence of accumulation of numerous macrophages in the alveoli. Septal inflammation and thickening were minimal. Taken together the final diagnosis was chronic necrotizing aspergillosis in the background of DIP (Fig. 4). The patient received Amp ambisome (amphotericin B liposomal) 250 mg/daily for 2 weeks, the patient's condition after 10 days gradually improved. Then the patient was discharged.

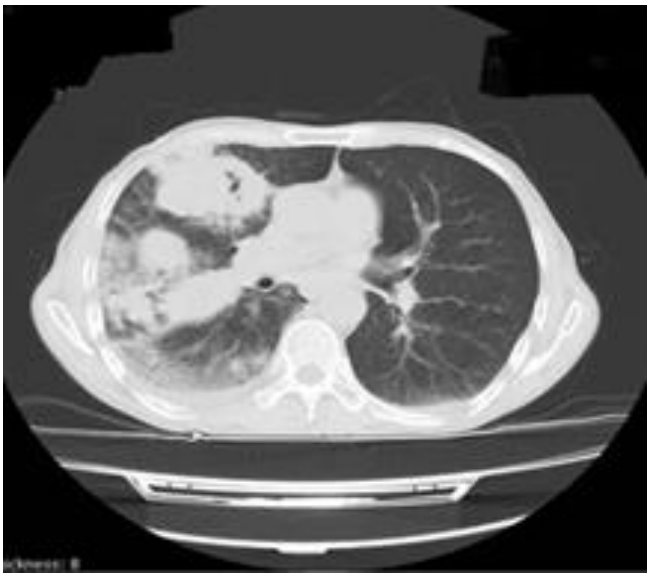

Figure 1: Chest CT scan revealed ground glass and interlobular septal thickening with patchy consolidation in the right upper lobe, right middle lobe, right lower lobe and left lower lobe

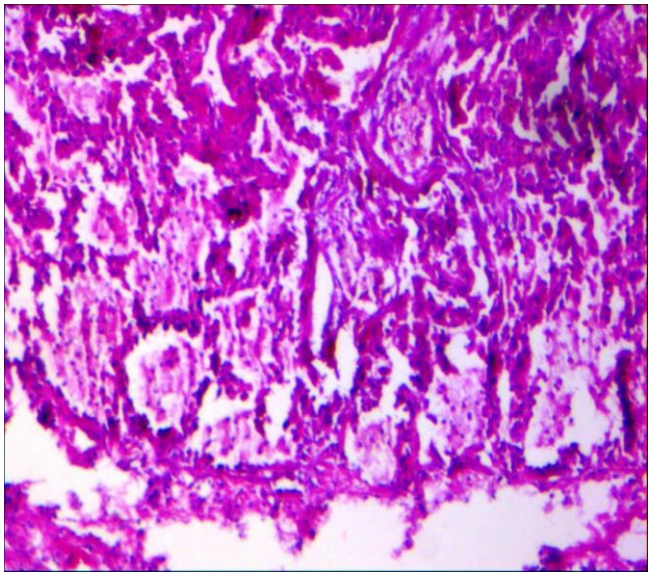

Figure 2: H and E stained slides showed numerous macrophages in alveolar spaces with interstitial inflammatory cell infiltration and minimal fibrosis (X10)

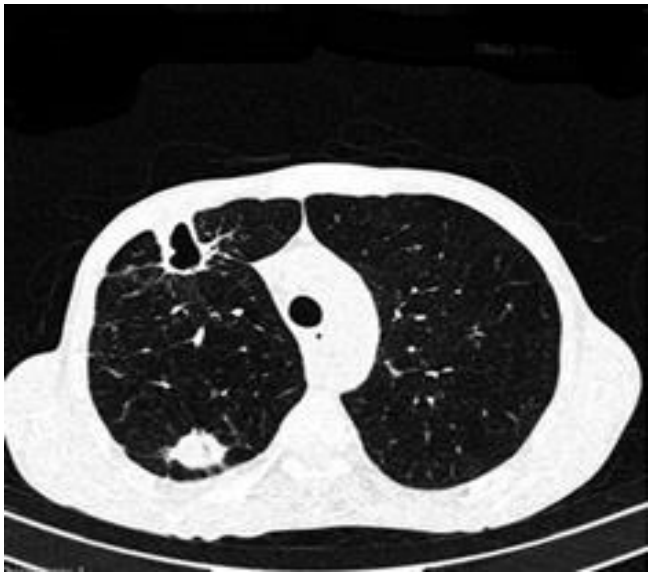

Figure 3: Chest CT scan demonstrated evidence of irregular thick wall cavitary lesions in anterior segment of right upper lobe measures 34*22 mm and right middle lobe measures 50*40 mm due to chronic infection or cavitary pneumonia

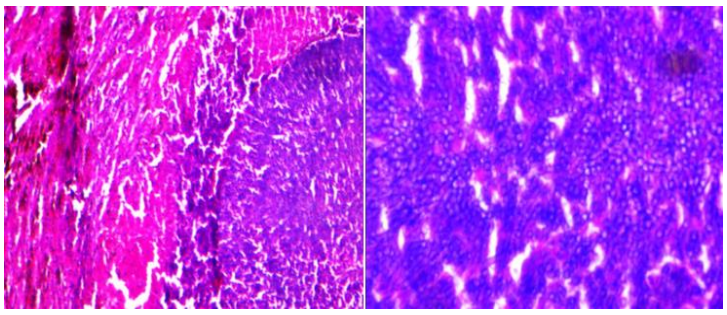

Figure 4: H&E stained slides revealed narrow septate hyphae in the pulmonary parenchyma (X10, left panel, X40 right panel)

DISCUSSION

In fact, IIP is a non homogenous group of diseases characterized by disruption of lung structure through inflammation and fibrosis. Its etiology is unknown. Patients are between 40 and 60 years old at the time of onset of symptoms [1]. Our patient was 58 years old. However, the disease has also been reported in children. In children, defects in

the synthesis of surfactant more than cigarette smoke is implicated. The prevalence of the disease is higher in men than in women [1]. The present patient was a male. The most common symptom is dyspnea, especially with activity and cough. Cough is not necessarily associated with sputum production [9]. It was true about the current patient. Other symptoms such as weight loss, weakness, chest pain and lethargy may also be mentioned [10]. Hemoptysis is rare [1]. On auscultation crackles especially at the base of the lungs are heard. On examination there may be cyanosis and clubbing [1]. Chest X ray findings are non specific. Usually peripheral ground-glass opacities are seen on chest CT scan, however these opacities can be more diffuse [11]. Some degree of emphysematous changes and air ways wall thickening may be present but honeycombing changes are uncommon [11]. The most helpful imaging finding is seen in the HRCT and it is a diffuse and symmetric ground-glass appearance in particularly in the middle and lower lungs zones [12]. Differential diagnoses include RB-ILD, hypersensitivity pneumonia and non-specific interstitial pneumonia (NSIP). Numerous pigment laden macrophages are seen in the BAL fluid. In some cases, the eosinophil population may be prominent. Bronchoscopy is recommended to rule out the mimickers. Owing to the non-specific nature of the imaging and BAL findings surgical biopsy is needed and it is recommended by ATS and ERS [13]. DIP diagnosis is based on the demonstration of pigmented macrophages in the alveoli. The etiology of DIP is not well known. There are a few reports about the role of infectious agents in the etiology of the DIP. An 8-month-old infant who suffered from CMV infection was reported with findings compatible with DIP by Schroten *et al* [7]. Also several other case reports revealed the association of DIP with viral infections such as BK-type polyomavirus [14]. Hepatitis C virus [15]. or influenza [16]. Although our patient has been a smoker and the DIP in him may be due to smoking, the role of the fungus should not be overlooked. As we alluded earlier the microscopic examination related to this patient revealed lung tissue that was invaded by narrow septate hyphae. However there was not any evidence of dissemination of fungal elements in elsewhere in the body. These findings are more indicator of a" chronic necrotizing aspergillosis" (CAN) than an overt invasive aspergillosis. Soubani AO *et al*. introduced CAN as a milder form of invasive aspergillosis which usually occurs in the aged patients who suffered from some degree of immunosuppression or pulmonary underlying diseases such as COPD or old tuberculosis [17]. Whether the etiology of the DIP in our patient is aspergillosis or fungal infection is a secondary phenomen to steroid administration is not clear at this time. We prefer the first theory because the time between taking the steroid and the aggravation of the symptoms was too short. In addition the patient's response to the steroid was not dramatic at first. It suggests that the underlying pulmonary disease has been beyond than just DIP. Since there are DIP-like reactions in adjacent to some pulmonary diseases application of TBLB can be misleading. This happened to our patient and we did not see the fungal elements in the first specimen. Close radiologic assistance and a surgical biopsy help the most [18]. Steroid therapy is the treatment of choice. However, it should not be overlooked to identify the underlying cause and eliminate it. Treatment should be continued until clinical and radiographic signs resolve. In cases that no response was observed, corticosteroids should be discontinued and the underlying disease should be investigated.

CONCLUSION

Astragalo-scapho-calcaneal dislocation is rare. It is a serious medico-Although DIP is directly related to smoking, other factors have been implicated in its etiology, including: systemic disorders, dangerous materials in the environment, drugs and infectious agents. In cases that no response to corticosteroids was observed, it should be discontinued and the underlying disease should be investigated.

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Conflicts of interest

None declared.

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