Case Report

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A very rare pulmonary pathology from symptom to diagnosis: Dendriform pulmonary ossification

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Abstract:

Pulmonary ossification is an uncommon, chronic, and progressive lung disease characterized by the presence of mature bone within the lung parenchyma. Diagnosis during life is rare; most cases are detected incidentally during autopsies. Chronic interstitial inflammation typically occurs in the lungs. This often leads to fibrosis in the parenchyma, though it can sometimes be idiopathic. Dendriform pulmonary ossification (DPO) can be classified as either idiopathic or secondary to an existing lung disease. Even though most patients are diagnosed through autopsy series, we present our case diagnosed using a videothoracoscopic wedge resection. A 49-year-old male patient, who has no chronic diseases and has never smoked, worked in aluminum casting for nine years, approxiately a decade ago. He visited the chest diseases clinic with complaints of increasing chest pain and shortness of breath over the past year. Physical examination revealed rales in the bilateral lung bases, but no clubbing. His oxygen saturation was 98%. Reticulonodular patterns were noted in all zones of both hemithoraces on a chest X-ray. The thorax Computed Tomography (CT) showed milimetric punctate calcific diffuse nodular and septal thickenings accompanied by pleuroparenchymal bands in both lungs. The patient, whose pulmonary function test was limited, was started on a therapeutic dose of 32 mg prednol. Gas exchange was reassessed with Diffusing Capacity of the Lung for Carbon Monoxide (DLCO) after two weeks. A diagnostic biportal videothoracoscopic wedge resection was performed on the patient. Due to the tough nature of the lung tissue, two staples broke during the operation. The patient was discharged on the second postoperative day with a pathology report confirming dendriform pulmonary ossification. The patient's outpatient follow-up is ongoing. DPO is most often diagnosed from surgical samples or during autopsies. It is a rare lung pathology characterized by the presence of mature bone tissue within the lung parenchyma. The disease frequently arises secondary to an underlying lung condition. Chronic inflammation is believed to play a role in its etiology. As seen in our patient's case, professions like aluminum casting can also cause chronic lung inflammation, potentially leading to DPO.

Keywords:

Chronic inflammation, dendriform pulmonary ossification, minimally invasive thoracic surgery

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Introduction

Pulmonary ossification is a rare, chronic, and progressive lung disease,[1] characterized by the presence of mature bone within the lung parenchyma.[1] It was first described by Luschka in 1856.[2] Diagnoses during a patient's lifetime are infrequent, with most cases being incidentally detected during autopsies.[2] For instance, in an autopsy series spanning 26 years with 1,393 cases, only eight instances of pulmonary ossification were identified.[2] This condition is more prevalent in middle-aged and elderly men.[3] Certain metals, extensively used in various industrial settings, have been associated with inflammation and fibrosis in the lung parenchyma.[4]

Pulmonary ossification exists in two forms: nodular and dendriform.[5] The latter, dendriform pulmonary ossification, is less common.[6] It derives its name from its net-like, dendritic, coral tree, or dichotomous appearance in the lungs.[5] Over the years, this pathology has been referred to by various names, inlcuding "branched bony ring formation in the lung" (Daust, 1929), "pulmonary osteopathy" (Ohlinger and Schoch, 1958), and "chronic interstitial ossifying pneumonia" (Arnsperger, 1897).[7] Dendriform pulmonary ossification predominantly affects the alveolar interstitium, displaying dendritic branching that expands the alveolar septa rather than the alveolar spaces. Biopsy specimens often reveal branched bony spicules, many containing fatty marrow, within the alveolar septa.[2] Conversely, nodular pulmonary ossification primarily affects the alveolar spaces and lower lobes. It is also observed in clinical situations associated with passive congestion, such as mitral valve stenosis.[8] Ossification manifests with a mature, lobulated appearance, causing enlargement of the alveoli.[9] Although the histological presentation comprises lobulated bone nodules within the alveolar cavity, these nodules do not contain fat or hematopoietic cells.[2] In clinical practice, distinguishing between nodular and dendriform types is typically deemed non-essential.[10] Sometimes both forms might be present in the same patient.[11] For symptoms like cough and shortness of breath, symptomatic treatment is advised. Follow-up incorporates general clinical findings, pulmonary function tests, and thoracic imaging.[12]

Dendriform pulmonary ossification (DPO) is categorized into idiopathic and secondary types.[12] The etiology of idiopathic DPO remains unclear. In contrast, secondary DPO encompasses conditions like pre-existing idiopathic pulmonary fibrosis, acute respiratory distress syndrome, cryptogenic organizing pneumonia, asbestosis, and heavy metal pneumoconiosis, among other lung diseases.[12]

Computed tomography findings distinctly reveal branched heterotopic bone formation within the lungs, predominantly in the lower lobe. Dendriform pulmonary ossification is identified by a coral-like dendritic pattern on high-resolution computed tomography (HRCT).[13] For nodular pulmonary ossification, HRCT displays lobulated nodules with smooth contours.[13] Although most diagnoses are made through autopsy series, we aim to present a case report of a patient diagnosed via videothoracoscopic wedge resection.

Case Report

A 49-year-old male patient, without any chronic illness and a non-smoker, worked in aluminum casting for nine years, about ten years ago. He approached the chest diseases clinic due to progressively worsening chest pain and exertional shortness of breath over the past year. During the physical examination, bilateral lung base crackles were noted. No clubbing was evident, and his oxygen saturation was 98%. Chest X-ray displayed reticulonodular images across all zones of both hemithoraces [Fig. 1]. The thorax Computed Tomography (CT) revealed milimetric punctate calcific diffuse nodular thickenings, along with pleuroparenchymal bands and septal thickenings in both lungs. The findings suggested the potential for interstitial lung disease in the context of pneumoconiosis with irregular nodularities [Fig. 2].

The patient underwent a fiberoptic bronchoscopy performed by the chest diseases department. The bronchoscopy revealed a normal bronchial system. Bronchoalveolar lavage was performed on the middle lobe. The pathology result was benign cytology with 98% alveolar macrophages and 2% bronchial epithelial cells. No Acid-Fast Bacilli (ARB) was observed in the microbiological examination, and there was no growth of Mycobacterium Tuberculosis. Although Stenotrophomonas Maltophilia was detected, it was considered a contamination, as the patient showed no signs of infection. Based

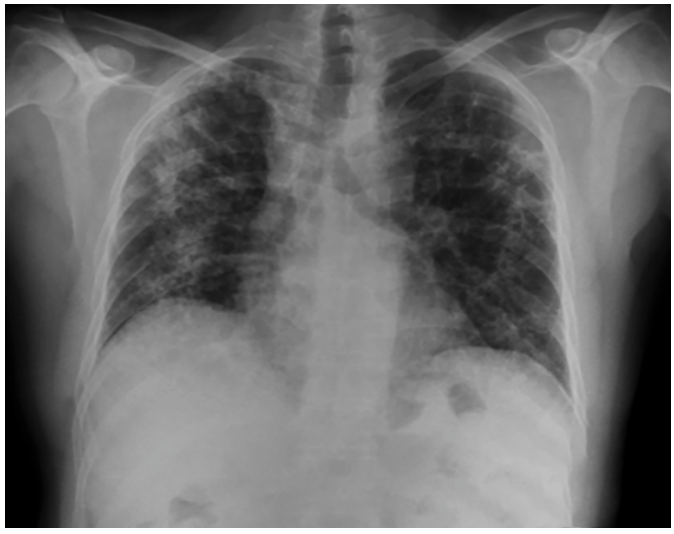

Figure 1: Preoperative chest radiograph

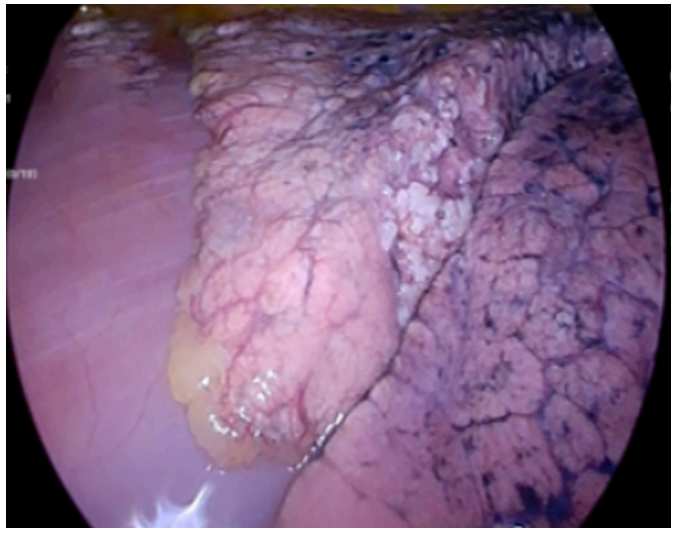

Figure 3: Intraoperative videothoracoscopic view; calcific lesions are observed

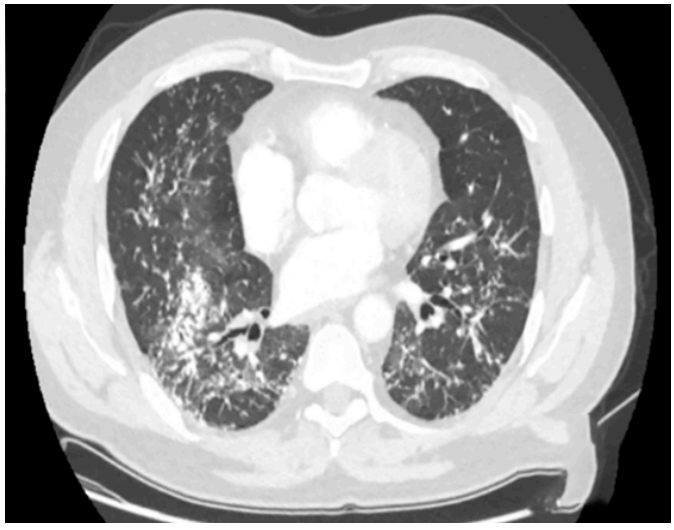

Figure 2: Diffuse punctate calcific appearance on thoracic computed tomography (CT)

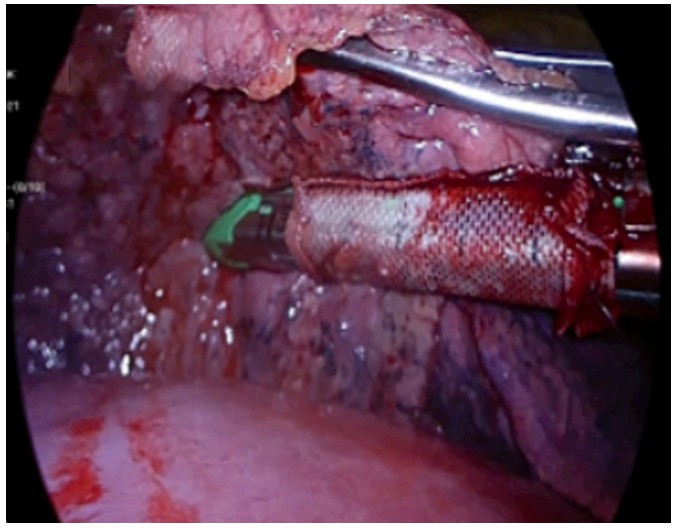

Figure 4: Moment when the first endostapler broke

on a preliminary diagnosis of interstitial lung disease by the chest diseases department, corticosteroid treatment was initiated. The patient, with a limited pulmonary function test, started on a therapeutic trial of 32 mg prednol. A repeat Diffusing Capacity of the Lung for Carbon Monoxide (DLCO) was requested after two weeks, indicating facilitated gas exchange. However, due to a worsening in pulmonary function tests during his follow-up (FEV_1: 2.25lt 61%, FVC: 2.53lt 67%, FEV_1/FVC: 89 114%, DLCO: 8.37 29%), he was referred to our thoracic surgery outpatient clinic for diagnostic sampling. The patient then underwent a right biportal videothoracoscopic wedge resection of the lower lobe. There were many nodules in the lung [Fig. 3]. During the procedure, two staples broke because of the lung's rigid structure before the completion of the wedge resection [Fig. 4]. As the stapler malfunctioned, a stapler of a different brand was employed [Fig. 5]. The wedge resection of the lower lobe was then successfully completed. Protective material was placed on the stapler line to prevent air leakage from the lungs in the postoperative period [Fig. 6]. After the procedure, it was noted that two staples had broken due to the lung's hard structure. In the lung imaging performed after the surgery, the lung was observed to be expanded [Fig. 7]. The patient was discharged on the second postoperative day. The patient has been under the care of the chest diseases polyclinic and has been followed up for six months. Outpatient follow-up continues [Fig. 8].

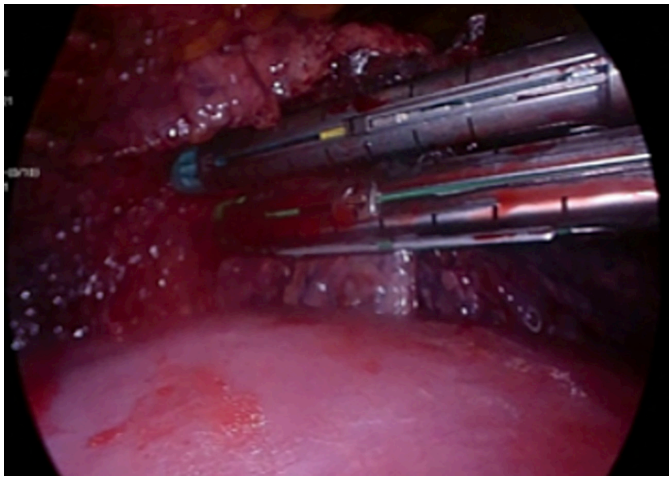

Figure 5: A different brand of endostapler positioned beneath the broken endostapler

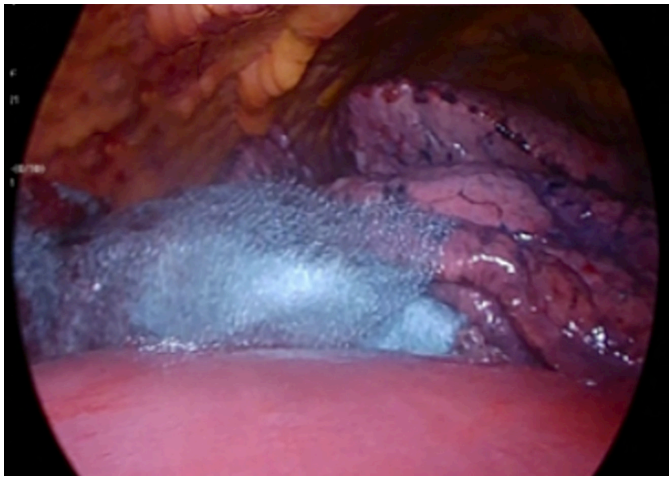

Figure 6: A patch made of polyglycolic acid placed on the stapler line

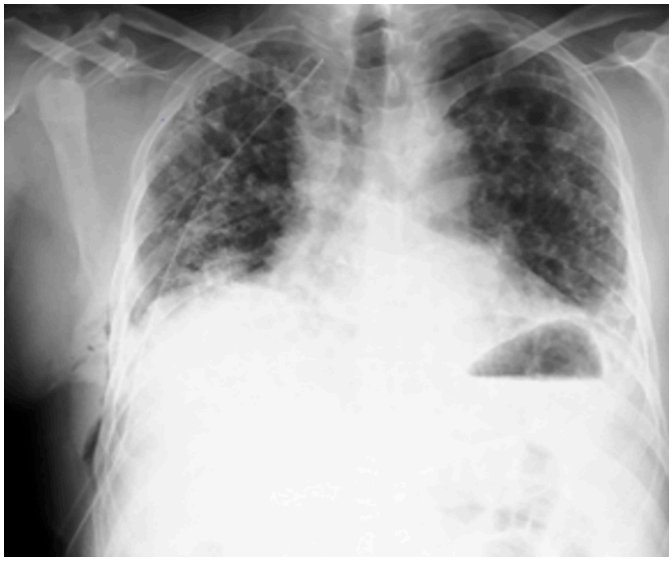

Figure 7: Chest X-ray taken on the immediate postoperative day

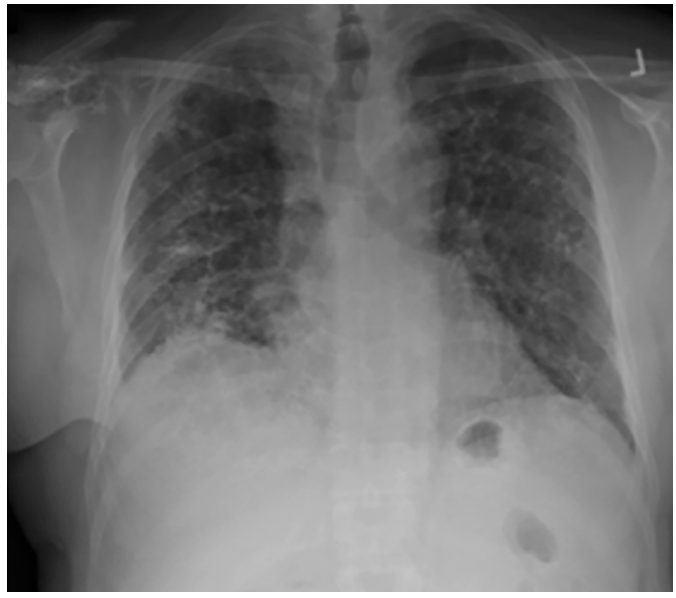

Figure 8: Chest X-ray taken six months after discharge

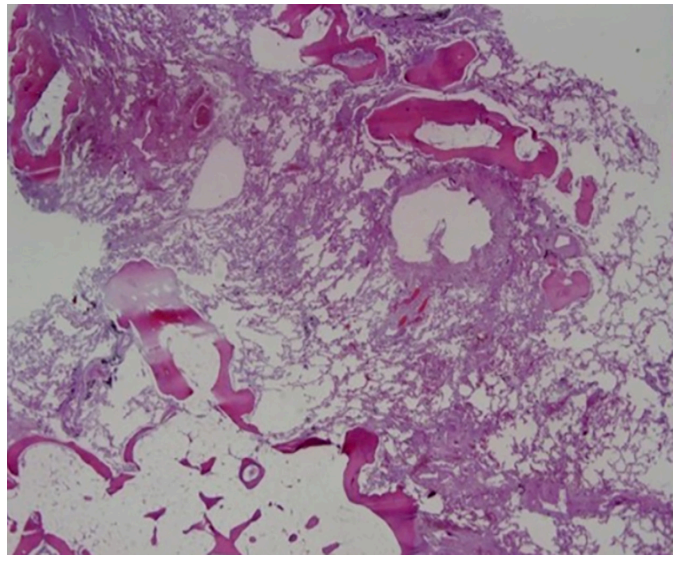

Figure 9: Diffuse dendriform pulmonary ossifications

The final pathology report identified dendriform pulmonary ossification. Macroscopically, the lung wedge resection material measured 9×4.8×2 cm, with an 8.5 cm long staple line. Diffuse, off-white, hard consistency calcification areas, averaging 0.2 cm in diameter, were observed. Microscopic examination did not reveal any specific interstitial lung disease. However, there were findings of diffuse pulmonary ossification, desquamation, reactive type 2 pneumocyte proliferation, alveolar septal thickening, fibroblastic foci in certain areas, fibrosis, and focal bronchiolar metaplasia [Figs. 9,10].

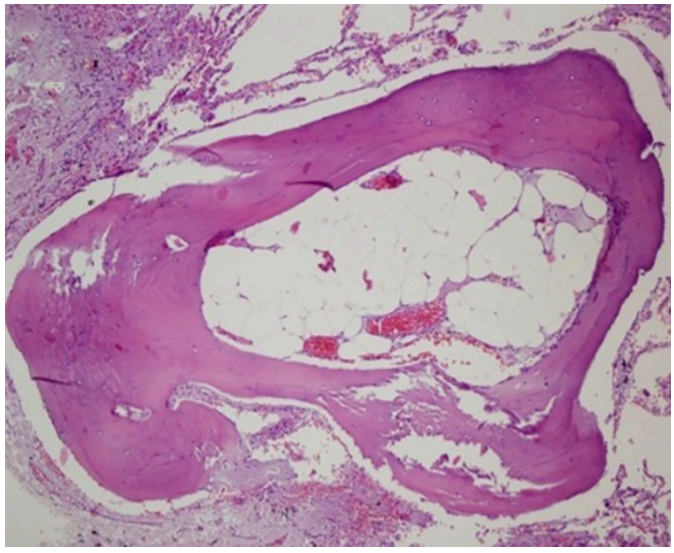

Figure 10: Fat marrow tissue (HEX20, HEX100)

Conclusion

Dendriform pulmonary ossification is a rare pathology characterized by the formation of bone tissue within the lung parenchyma. It is most often diagnosed from surgical specimens or autopsies, with very few cases reported in the literature.

The condition can arise secondary to other underlying lung pathologies. Therefore, a thorough evaluation of the patient's clinical history, radiological, and histopathological findings is crucial. In certain instances, a transbronchial biopsy can secure the diagnosis.

Given the known associations of the disease with idiopathic pulmonary fibrosis, Chronic Obstructive Pulmonary Disease (COPD), organizing pneumonia, and asbestos exposure, the aluminum casting profession's potential role in triggering dendriform pulmonary ossification should not be overlooked, especially in the context of chronic lung inflammation, as observed in our case.

There is no specific treatment for dendriform pulmonary ossification. Patients are managed symptomatically for complaints like cough and shortness of breath. Regular clinical and radiological monitoring is recommended.[14]

We highlight this case to emphasize the rarity of dendriform pulmonary ossification and the significance of its recognition. Future research and extended follow-up outcomes will offer clearer insights into managing and understanding this condition.

Informed Consent

Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

Conflicts of interest

There are no conflicts of interest.

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Authorship Contributions

Concept – M.E.F., B.K., Ö.F.S., Ş.B., E.E., H.V.K.; Design – M.E.F., B.K., Ö.F.S., Ş.B., E.E., H.V.K.; Supervision – M.E.F., B.K., Ö.F.S., Ş.B., E.E., H.V.K.; Funding – M.E.F., B.K., Ö.F.S., Ş.B., E.E., H.V.K.; Materials – M.E.F., B.K., Ö.F.S., Ş.B., E.E., H.V.K.; Data collection &/or processing – M.E.F., B.K., Ö.F.S., Ş.B., E.E., H.V.K.; Analysis and/or interpretation – M.E.F., B.K., Ö.F.S., Ş.B., E.E., H.V.K.; Literature search – M.E.F., B.K., Ö.F.S., Ş.B., E.E., H.V.K.; Writing – M.E.F., B.K., Ö.F.S., Ş.B., E.E., H.V.K.; Critical review – M.E.F., B.K., Ö.F.S., Ş.B., E.E., H.V.K.

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