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Idiopathic dendriform pulmonary ossification incidentally found in a patient with gastroesophageal reflux disease: Case report and literature review

Safa Alshaikh, Zahra Alkhunaizy, Hanoof Alabdullattif, Maryam Alkhaja

ABSTRACT

Introduction: Diffuse pulmonary ossification was firstly described by Luschka in 1856. It has two distinct patterns with different clinical, radiological, and histological associations: nodular pulmonary ossification (NPO) and dendriform pulmonary ossification (DPO). Dendriform pulmonary ossification is described by the presence of branching metaplastic osseous spicules usually with bone marrow elements. Dendriform pulmonary ossification is described by the presence of branching metaplastic osseous spicules usually with bone marrow elements. Dendriform pulmonary ossification can occur as idiopathic or in association with chronic lung diseases. Idiopathic DPO is a rare entity and has association with chronic gastric acidity.

Case Report: A 36-year-old gentleman, unknown to have any medical illness, was incidentally found to have bilateral diffuse high-density lung opacities during workup for back pain. The patient was asymptomatic. High resolution computed tomography (HRCT) of the chest showed bilateral ossified tiny branching opacities mainly involving the lower lobes. Histologically, the sections showed lung parenchyma with mature bone formation in the interstitial pulmonary spaces with fatty marrow in some of the bony spicules. Complementary upper gastrointestinal study was conducted confirming the presence of high-volume gastro-esophageal reflux.

Conclusion: Dendriform pulmonary ossification is a rare entity that can cause serious complications such as respiratory failure. Thus, awareness of it as a differential diagnosis of chronic lung disease is necessary. More studies are needed to establish treatment guidelines and long-term prognosis.

Keywords: Dendriform pulmonary ossification, Hematopoietic cells, Nodular pulmonary ossification

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INTRODUCTION

Diffuse pulmonary ossification is an uncommon slowly progressive chronic disease which is characterized by the presence of widespread metaplastic bone within the lung. It has two distinct patterns with characteristic clinical, radiological, and histological association: nodular pulmonary ossification (NPO) and dendriform pulmonary ossification (DPO) [1–4]. Nodular pulmonary ossification is more common and usually associated with underlying cardiac disorders. It is defined by the presence of rounded metaplastic ossifying masses in the alveolar spaces. In contrast, DPO is rarer and is described by the

occurrence of dendriform, also referred to as "branching-like" spicules of metaplastic bone frequently found with bone marrow elements [5–8]. It is commonly seen in association with chronic lung diseases. Nevertheless, idiopathic DPO cases have been reported. Recent studies showed that idiopathic DPO appeared to be associated with gastroesophageal reflux disease (GERD) [5]. Carney et al. stated that less than 10 cases of idiopathic DPO cases had been discussed in the literature [4]. Herein, we report a case of idiopathic DPO in a 36-year-old asymptomatic male who was incidentally found to have bilateral diffuse lung opacities on plain radiograph during his workup for back pain.

CASE REPORT

A 36-year-old gentleman, unknown to have any medical illness, was incidentally found to have bilateral generalized high-density lung opacities during plain radiograph workup for back pain. The patient had no pulmonary related symptoms. His physical examination was unremarkable.

High resolution computed tomography (CT) of the chest showed bilateral ossified tiny branching opacities mainly involving the lower lobes at the sub-pleural, septal, and peri-fissural regions. However, no honeycombing, fibrotic changes or traction were observed.

Then, the patient underwent right thoracoscopic lung biopsy under general anesthesia. Macroscopically, the lung biopsy consisted of two pieces of tan gray tissue, the larger measuring 3.0×1.0×0.4 cm, cut section revealed calcified areas. Histologically, the hematoxylin and eosin (H&E) sections showed lung parenchyma with mature bone formation in the interstitial pulmonary spaces with fatty marrow in some of these bony spicules (Figure 1).

Complementary upper gastrointestinal study was performed confirming the presence of moderate to severe gastro-esophageal reflux disease (GERD). According to the pathological features and CT scan findings, a diagnosis of DPO was established.

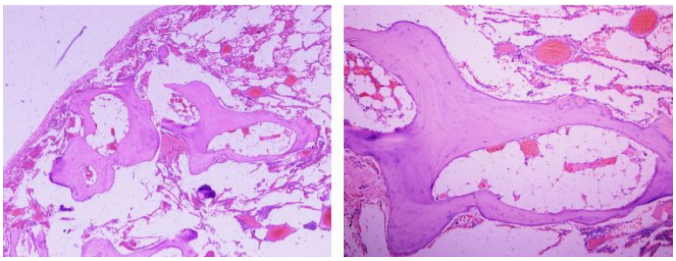

Figure 1: Lung biopsy sections showing irregular branching mature bony spicules intra-alveolar and within the pulmonary interstitium with fatty marrow (Hematoxylin and eosin, original magnification 40× to 100×).

Follow-up

The patient's medical records were reviewed. After surgical intervention and the diagnosis of diffuse pulmonary ossification was made, this patient traveled abroad for seeking treatment and no follow-up at our hospital.

DISCUSSION

Pulmonary metaplastic ossification can be seen as localized or diffuse form, primary or secondary to various diseases. The localized form is mostly found incidentally with dystrophic calcification in many conditions such as abscess, tuberculous scar, or nonspecific areas of lung fibrosis [1, 2, 6].

Diffuse pulmonary ossification is a rare slowly progressive chronic process, firstly described by Luschka in 1856 [9] and is defined by the presence of extensive metaplastic bone within the pulmonary interstitium and alveolar walls [9]. There are two main patterns of diffuse pulmonary ossification with different clinical, radiological, and histological associations: NPO and DPO.

Nodular pulmonary ossification was firstly described by Salivger et al. in association with mitral valve stenosis in 1933 [10]. Since then, it has been documented in patients with pulmonary congestion in the context of cardiac diseases such as left ventricular dysfunction, atrial fibrillation, and post-myocardial infarction [4, 11]. Radiologically, it is recognized as small round nodules. Histological findings include rounded masses of mature bone in the alveolar spaces without any bone marrow elements [6]. The hypothesized pathophysiology behind NPO involves osseous fibroblast metaplasia induced by hemosiderin deposition in chronic venous congestion situations or organization of alveolar exudates [12, 13].

In contrast to NPO, DPO is described histologically by formation of branching—"racemouce" or "reticular"—metaplastic osseous bony spicules with usually bone marrow elements (fatty or hematopoietic cells) in the pulmonary interstitium or alveolar walls [4]. In imaging studies, manifests as tree-like "linear" opacities with predilection to lower lung and is predominantly found in older men in their fifth to sixth decade of life with a male to female ratio 6:1 [5, 8, 14]. Patients with DPO are commonly asymptomatic; therefore, most of the early reported cases in the literature were diagnosed postmortem with occurrence ratio of 1.63 cases DPO/1000 autopsies [15]. Furthermore, DPO can occur as idiopathic or in association with chronic lung diseases. The latter association was reported as idiopathic pulmonary fibrosis, usual interstitial pneumonia, adult respiratory distress syndrome, organizing pneumonia, asbestosis, heavy metals exposure, pneumothorax, and osteogenesis imperfecta [5, 15–17]. Additionally, the coexistence of DPO with pulmonary and extra-pulmonary malignancies has been reported as lung adenocarcinoma, metastatic melanoma [18], mucoepidermoid carcinoma [19], and duodenal adenocarcinoma [20].

Idiopathic DPO is a rare entity in which only osseous branching bony spicules are found within the lung

parenchyma without any other histological findings. It has been theorized that the presence of acidic medium and hypoxia can cause osseous fibroblastic metaplasia [5]. Gruden et al. described that DPO without usual interstitial pneumonia seems to be associated with GERD [5]. Similarly, Fernandez-Bussy et al. identified a case of DPO in a 43-year-old man with history of GERD [15].

Carney et al. stated that less than 10 cases of idiopathic DPO were documented in the English literature [4]. However, we found less than 15 cases of idiopathic DPO in which there were no other clinical, radiological, or histological findings (Table 1).

Definitive antemortem diagnosis of DPO requires lung biopsy by Video Assisted Thoracoscopy (VATS), transbronchial, or by thoracotomy in concurrence with chest computed tomography (CT) appearance [6].

Diffuse pulmonary ossification can produce serious complications such as respiratory failure ending up with lung transplantation. Henceforth, awareness of this entity as a differential diagnosis of chronic lung diseases is necessary because it is, usually, misinterpreted radiologically as pulmonary fibrosis, bronchiectasis, or lymphangitic spread of tumor [4, 19].

No effective management or treatment guidelines of DPO are published. Trails of low calcium diet, calcium-binding drugs, bisphosphates, and warfarin are not shown to be effective [6, 11]. Tomoko et al. suggested possible remission of DPO with corticosteroids [21].

More studies related to pathophysiology, risk factors, and treatment options are required to establish definitive management guidelines and to determine the long-term prognosis.

Table 1: Reported cases of idiopathic DPO

Author	Number of cases	History of GERD
Current case	1	Yes
Enomoto et al. (2021) [1]	3	Yes
Hirai et al. (2019) [2]	1	No
Edahiro et al. (2019) [3]	3	No
Gruden et al. (2017) [5]	1	Yes
Fernández-Bussy et al. (2015) [15]	1	Yes
Mizushina et al. (2012) [14]	1	No
Reddy et al. (2012) [22]	1	No
Bai et al. (2009) [23]	1	No
Jana and Dunton (2001) [24]	1	No

CONCLUSION

Dendriform pulmonary ossification is a rare entity that can lead to serious complications such as respiratory failure. Therefore, clinicians, radiologists, and pathologists should be aware of as a differential diagnosis of chronic lung diseases. More studies are needed to establish treatment guidelines and long-term prognosis.

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Author Contributions

Safa Alshaikh – Conception of the work, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Zahra Alkhunaizy – Acquisition of data, Analysis of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Hanoof Alabdullattif – Analysis of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Maryam Alkhaja – Acquisition of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

The corresponding author is the guarantor of submission.

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Consent Statement

Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.