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CASE REPORT

Pulmonary hyalinizing granuloma involving the diaphragm and the pericardium

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ABSTRACT

Pulmonary hyalinizing granuloma (PHG) is a rare disorder and is a pertinent differential for lung diseases with multiple pulmonary nodules. Natural history of this disease is not known. Although the usual course is benign, a close follow-up of these cases is necessary. We herein report a case of PHG involving the diaphragm and the heart, detected on autopsy in a 57-year-old male who suffered a road traffic accident.

Key words: Diaphragmatic nodules, pericardium, pulmonary hyalinizing granuloma

Introduction

Pulmonary hyalinizing granuloma (PHG) is a rare disease with distinct fibrosing lesions of the lung characterized by central whorled deposits of lamellar collagen. It has been reported that PHG is accompanied by extra-pulmonary fibrous lesions at various sites including the kidney, tonsils and thyroid glands.[1] PHG presents as pulmonary nodules with non specific symptoms of cough, hemoptysis, chest pain and shortness of breath. When asymptomatic, it is usually detected on routine chest radiograph. An immune response to the antigenic stimuli by infection or autoimmune process has been postulated in the pathogenesis but the precise etiology remains obscure [2]. The lesion can be situated in the lung parenchyma or sub pleura. Due to their behavior, a biopsy is required to establish the primary diagnosis of PHG [3]. To the best of our knowledge, this is the first encounter with a case of pulmonary hyalinizing granuloma with involvement of the pleural surface of the diaphragm and heart without involvement of the lung parenchyma.

Case Report

A medico legal autopsy was performed at our institute on a 57-year-old male who died following a road traffic accident. History obtained from his relatives and medical records revealed him to be a chronic smoker for past 30 years and a known hypertensive since 3 years, and on single drug antihypertensive therapy. There was no history suggestive of any infection or autoimmune disease. Autopsy revealed multiple, bilateral, well-circumscribed, rubbery white nodules on the diaphragm, largest measuring 3 × 4 cm and the smallest 1 × 1 cm. There was no pulmonary involvement [Figure 1a] on gross appearance.

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In addition to the diaphragmatic nodules, careful examination of the pericardium also showed presence of two such similar nodules [Figure 1b]. Histopathological examination of these individual nodules showed bundles of lamellar hyalinized collagen arranged in parallel and whorl configuration [Figure 2a], admixed perivascular lymphoplasmacytic infiltration was noted [Figure 2b]. There was no evidence of granulomas or areas of necrosis in the multiple sections that were studied.

Masons' trichrome stain and van Gieson's stain were done, which confirmed the presence of collagen. The nodular section was also subjected to special stains like acid fast and Congo red to rule out mycobacterial involvement and amyloidosis respectively. A final diagnosis of PHG was made that invariably involved the diaphragm and the pericardium.

Discussion

PHG is a rare benign condition first described in 1977, which usually manifests as multiple bilateral pulmonary nodules of lamellar hyaline collagen deposits.[4] It usually affects people of age 19-77 years with a mean age of 44 years at the time of presentation and has no gender predilection [4]. Size of the tumor varies from several millimeters to 15 cm in greatest dimensions and 73% of such patients have multiple lesions [5]. Majority of the patients are asymptomatic, which correlated with the present case.

The etiology of PHG is unknown, but it has been associated with immunologic or infectious diseases such as rheumatoid arthritis, sclerosing mediastinitis, retroperitoneal fibrosis, uveitis, occulopapillitis, tuberculosis, histoplasmosis and aspergillosis [3]. Neoplastic diseases have rarely been reported which include abdominal lymphoma, multiple myeloma, Paget's disease of breast and astrocytoma [6]. The present case did not have any previous history of autoimmune or infective diseases or precisely, no medical records were available for the same. The lesion can be situated in the lung parenchyma or subpleura [3]. PHG is sometimes accompanied by extra-pulmonary fibrous lesions at

CASE REPORT

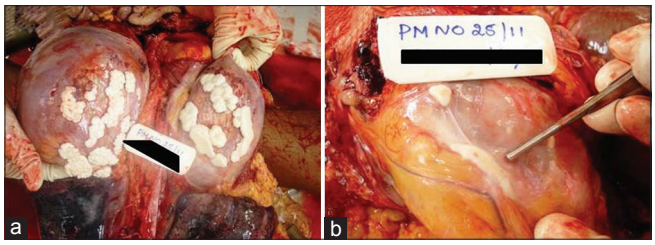

Figure 1 (a) Bilateral irregular multiple white nodules on the diaphragm with normal lung below (b) Irregular white nodules on the pericardial surface of the heart

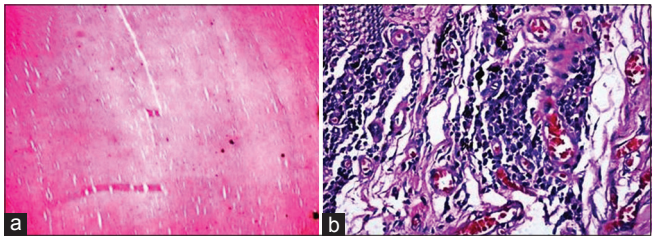

Figure 2 (a) Homogenous hyalinized lamellae of collagen arranged in parallel formation (H and E, ×100) (b) Perivascular lymphoplasmacytic aggregates around hyalinized collagen bands (H and E, ×200)

other sites, and coexistence of PHG along with laryngeal and subcutaneous nodules have been reported [1]. The present case did not show extra pulmonary fibrous lesions at other sites.

The disease follows a relatively benign course with the nodules showing increasing in size over a period of years. There are two reported cases of PHG complicated by lymphoma. Therefore, a follow-up is utmost essential in such cases [2]. Prognosis of PHG is excellent with no significant impact on longevity [4]. There have been reports of patients who responded well to corticosteroid therapy [7]. Surgical resection is the treatment of choice [8].

Conclusion

Diagnosis of PHG requires a histological examination, and therefore, a biopsy is essential. In previously reported cases, most lesions occurred in the lung with extra-pulmonary manifestations. This is a rare case of PHG involving the diaphragm and the heart without involvement of the lung, discovered incidentally at autopsy.

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Authors' Contributions

HRV, HC, GVM participated in the clinical diagnosis, sequence alignment, and drafting of the manuscript and made useful contributions to the review of the literature. HRV and SS participated in writing the discussion section and helped in the revision of the manuscript. All authors read and approved the final manuscript.

Consent

The authors certify that a written informed consent was obtained from the patient's immediate relative/family members for publication of this case report and any accompanying images. A copy of the written consent is available for review by the editor-in-chief of this journal.

Competing Interests

The authors declare that they have no competing interests.

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