



Papillary Cystadenoma Lymphomatosum in a Patient with Psoriasiform Sarcoidosis

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Abstract

A 52 year old male presented with a swelling in the left parotid region and erythematous plaques with silvery scale on the arms. He had a 5 pack-year smoking history. The patient had sarcoidosis for eight, coronary atherosclerosis for five and COPD for three years. Family history did not reveal any disease. Physical examination revealed erythematous plaques with silvery scale on the arms. Electrocardiogram and chest X-ray revealed normal findings. Blood count and serum biochemistry were within normal limits. Thorax CT revealed randomly distributed micronodules in both lungs. PET/CT showed a hypermetabolic lesion in the posteromedial region of the left parotid gland. Pathologic examination of the cutaneous lesions revealed psoriasiform sarcoidosis. Histopathology of the transbronchial lung biopsy revealed non classified granulomatous inflammation while pathology of the needle aspiration biopsy of the parotid gland was compatible with papillary cystadenoma lymphomatosum. Histopathologic examination of the skin true cut biopsy sample demonstrated psoriasiform sarcoidosis.

We present a case of psoriasiform sarcoidosis with coexistent papillary cystadenoma lymphomatosum that has not been reported in the literature previously. The aim of this case report is to investigate the association between the coexistence of the aforementioned parotid tumor and the psoriasiform sarcoidosis.

Keywords: Sarcoidosis; Pulmonary function tests; Prognosis; PFT; DLCO/VA

Introduction

Warthin's tumor, also known as papillary cystadenoma lymphomatosum, is a benign cystic tumor of the salivary glands containing abundant lymphocytes and germinal centers. It is named for pathologist Aldred Scott Warthin, who described two cases in 1929. Benign tumors account for approximately 60% to 80% of parotid neoplasms and among these; Warthin's tumor is the second most common benign neoplasm accounting for approximately 15% of all parotid epithelial tumors [1-4]. Patients with Warthin's tumor have a higher incidence of autoimmune diseases, including insulin-dependent diabetes mellitus, Hashimoto's thyroiditis, and autoimmune hyper- and hypothyroidism [5]. Warthin's tumor may also be associated with IgG4-related disease [6].

We present a case of Whartin's tumor in a sarcoidosis patient with psoriasiform cutaneous lesions. Although there are earlier case reports showing the association of this tumor with autoimmune diseases, its coexistence with sarcoidosis has not been reported in literature previously. Our aim is to investigate the common pathogenetic pathway or mechanism between the papillary cystadenoma lymphomatosum and psoriasiform sarcoidosis.

Case Presentation

A 52 year old male patient was admitted for swelling in the left parotid region and erythematous plaques with silvery scale on the arms (Figure 1). The patient had a 25 pack-year smoking history. Personal history revealed sarcoidosis for eight, coronary artery disease for five and COPD for three years. Family history was excellent. Swelling in left parotid region and erythematous plaques with silvery scale on the arms were detected on physical examination. Blood pressure was 120/75 mmHg. Complete blood count, serum biochemistry and urine analysis were within normal limits. ECG showed a sinus rhythm of 84/min. Chest X-ray was normal (Figure 2). Thorax CT revealed random micronodules in both lungs (Figure 3). PET/CT demonstrated a hypermetabolic lesion

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Figure 1: Erythematous plaques with silvery scale on the arms of psoriasiform sarcoidosis.



Figure 2: Chest X-ray showing normal lung parenchyma.



Figure 3: Thorax CT revealing diffuse and random nodules in both lungs.

with a 7.6 SUV value in the posteromedial region of the left parotid gland. Histopathologic examination of the cutaneous lesions revealed psoriasiform sarcoidosis. Transbronchial lung biopsy revealed non-classified granulomatous inflammation while pathology of the needle aspiration biopsy of the parotid gland was compatible with papillary cystadenoma lymphomatosum that revealed granulomatous inflammation containing lymphocytes, mast cells, oncocytes, amorphous cell groups within an epithelial parenchyma and lymphoid stroma. The final diagnosis was Whartin's tumor and psoriasiform sarcoidosis.

Discussion

Papillary cystadenoma lymphomatosum is the most common tumor of the parotid gland with a double layer of epithelial cells on a dense lymphoid stroma. The tumor arises from incorporation of lymphoid tissue in the parotid gland or by induction of cystic and oncocytic changes in an inflammatory infiltrate. It usually occurs in male smoker's age over forty years. Patients have a higher incidence of autoimmune diseases such as type I diabetes, Hashimoto's thyroiditis and autoimmune thyroiditis [5]. It may also be associated

with IgG4-related disease [6]. Coexistence of papillary cystadenoma lymphomatosum with sarcoidosis has not been reported previously. We present this not only for its coexistence with psoriasiform sarcoidosis but also for the diagnosis of this tumor as an incidental finding in an asymptomatic patient that was evaluated for the investigation cutaneous sarcoidosis lesions by PET/CT.

Papillary cystadenoma lymphomatosum did not lead to any symptoms in our patient because it was small as it did not compress any of the adjacent structures. The diagnosis of the tumor was coincidental during the investigation of extrapulmonary organ involvement of sarcoidosis. PET/CT was performed to identify other organ disease in a sarcoidosis patient that lead to the identification of this tumor. Presence of granulomatous inflammation in the parotid gland strongly suggests that sarcoidosis involvement of the parotid may have lead to the development of the papillary cystadenoma lymphomatosum. Autoimmune mechanisms of sarcoidosis may have played a role in the evolutionary path way of papillary cystadenoma lymphomatosum in this patient.

Since there are previous reports revealing a high incidence of autoimmune diseases in papillary cystadenoma lymphomatosum patients [5,6], the aforementioned tumor may be associated with sarcoidosis because sarcoidosis itself is also an autoimmune disease. Previous personal history of unstable sarcoidosis and the histopathologic features revealing granulomatous inflammation in the parotid gland justifies the association of two diseases but the exact autoimmune pathogenetic mechanism that leads to papillary cystadenoma lymphomatosum is unclear. Smoking is another risk factor for the development of this tumor. The risk may be closely related to the amount and to the duration of smoking. Smokers carry an eight times more risk to develop this tumor compared to nonsmokers [7-9]. The smoking history in our patient was minimal and the risk therefore appears to be negligible if there is any. On the other hand, the coexistence of sarcoidosis and papillary cystadenoma lymphomatosum may have occurred coincidentally and the two disorders may have arisen independently of each other.

Conclusion

This article reveals the coexistence of sarcoidosis involvement and papillary cystadenoma lymphomatosum as the first patient in the literature. This association may just be coincidental or the involvement of parotid gland by sarcoidosis may have caused the development of papillary cystadenoma lymphomatosum as suggested by the previous literature revealing the relevance of autoimmune diseases with this parotid tumor. In our case, there are two factors showing that sarcoidosis may have lead to the development of this tumor, one of which is the presence of an unstable sarcoidosis history and the other is the existence of granulomatous inflammation of the parotid in this patient. A common mechanism for the development for these two disorders is unclear and may only be explained by mechanisms that play a role in the development of sarcoidosis on an autoimmune basis.

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