An unusual case of Wegener's granulomatosis with tongue involvement

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ÖZET

Dil tutulumu yapmış nadir bir Wegener granülomatöz


Anahtar Kelimeler: Wegener granülomatosis, dil tutulumu, alveoler opasite.

SUMMARY

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**Wegener’s granulomatosis** is a type of vasculitis characterized by necrotizing granulomatosis respiratory tract lesions and necrotising glomerulonephritis. Nasal, lung and renal biopsies and positive antineutrophil cytoplasmic antibody (c-ANCA) analysis is helpful in diagnosis. Early diagnosis and treatment is critical in prognosis. A 42 years-old male had dyspnea, coughing, hemoptysis, fatigue, loss of appetite, night sweating and arthralgia. Violaceous palpable, purpuric lesions were detected on the sublingual region of the mouth. On chest X-ray, there was nonhomogeneous infiltration in the parenchyma of both lungs. There was alveolar density involving upper lobes of both lungs detected in thoracal computerized tomography as well as patchy densities on the right lung upper-middle lobes. A nasal mucosa biopsy showed strongly destructive vasculitis. c-ANCA test was positive. We report an unusual case with Wegener’s granulomatosis, characterized by a rare presentation of tongue involvement and atypical lung radiology with alveolar opacity.

**Key Words:** Wegener’s granulomatosis, tongue involvement, alveolar opacity.

Wegener’s granulomatosis (WG) is a type of vasculitis characterized by necrotizing granulomatosis respiratory tract lesions and necrotising glomerulonephritis. Nasal, lung and renal biopsies and positive antineutrophil cytoplasmic antibody (c-ANCA) analysis is helpful in diagnosis. Early diagnosis and treatment is critical in prognosis. A 42 years-old male had dyspnea, coughing, hemoptysis, fatigue, loss of appetite, night sweating and arthralgia. Violaceous palpable, purpuric lesions were detected on the sublingual region of the mouth. On chest X-ray, there was nonhomogeneous infiltration in the parenchyma of both lungs. There was alveolar density involving upper lobes of both lungs detected in thoracal computerized tomography as well as patchy densities on the right lung upper-middle lobes. A nasal mucosa biopsy showed strongly destructive vasculitis. Transbronchial and renal biopsies were negative for vasculitis. c-ANCA test was positive.

A diagnosis of WG was established and treatment with methylprednisolone and cyclophosphamide was initiated which resulted in marked improvement of pulmonary symptoms and complete resolution of the sublingual lesions as well as clearances of the pathological findings of chest X-ray graph and CT was achieved with this therapy (Figure 4,5).

**DISCUSSION**

A case report is presented for tongue involvement which proceed to systemic involvement.

In WG the usual targets are extra-oral (1). Unusual oral lesions, including palatal and lingual ulceration, aphthae, non-healing extraction sockets, a friable-granular gingivitis associated with alveolar resorption and tooth mobility have been infrequently described, salivary gland involvement have also been described in the literature (1-4). Frances et al. described oral ulceration in 20% of patients with WG (5). A 48-years-old WG patient with tongue infarction and extensive systemic involvement causing death has also been reported in the literature (6). Rapid resolution of the tongue lesions with immunosuppressive treatment was highly suggestive of tongue involvement of WG in our case.

In WG, the most common pulmonary radiological findings are multiple nodules or masses. In most cases, nodules can scatter in bilateral or in any part of lung. Cavitations can be seen in half of cases. Consolidation and translucent bronchoscopies were normal. A nasal mucosa biopsy showed strongly destructive vasculitis.
opacities are the second most common radiological findings. Widespread alveolar infiltrates, usually confluent due to diffuse pulmonary hemorrhage have been detected in 5 out of 14 (35.7%) patients (7). Besides, areas of consolidation have been obtained in 2 out of 10 (20%) patients and consolidation and ground glass attenuation in 26% patients with WG in various studies (8,9).

We close by suggesting that a diagnosis of WG should be considered in patients with palpable purpuric lesions of the tongue in association with pulmonary symptoms.
REFERENCES


