BILATERAL PAROTID SARCOIDOSIS RECOGNISED BY FINE NEEDLE ASPIRATION CYTOLOGY

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ABSTRACT

Sarcoidosis is a systemic granulomatous disease of unknown origin. The most common sign is bilateral lymphadenopathy. Parotid involvement is very rare. In case report we present a case of bilateral parotid sarcoidosis recognised by fine needle aspiration cytology in 33-year-old male. Fine needle aspiration cytology showed aggregates consisting of epithelioid cells, histiocytes, giant cells and lymphocytes. Diagnosis was confirmed by serum angiotensin-converting enzyme level and negative tuberculin skin test. The patient had a good response to the steroid therapy.

Keywords: Parotid Glands; sarcoidosis; fine needle aspiration cytology

INTRODUCTION

Sarcoidosis (sarcoïd, Besnier-Boeck disease or Besnier-Boeck-Schaumann disease) is a systemic disease of unknown origin, characterized by granuloma formations in multiple organs. Most common involvement is seen in lung and lymph nodes (1). In a recently reported sarcoidosis series, the frequency of parotid gland involvement was 6% (2). We report a case of bilateral parotid sarcoidosis and discussed fine-needle aspiration cytology (FNAC) results with clinical findings.
CASE REPORT

A 33-year-old male patient was referred to the hospital, with a 6-month history of a painless, bilateral parotid swelling and xerostomia. Diffuse and non-nodular bilateral parotid enlargement and normal periglandular anatomy, and bilateral cervical lymphadenopathy were found by magnetic resonance imaging (Figure 1).

FNAC applied to the patient in fine needle aspiration unit of pathology department and six smears was prepared. Two of them were air-dried; four of them were fixed in alcohol.

Air-dried smears stained with May-Grünewald & Giemsa, alcohol-fixed smears with Hematoxylene&Eosin and Papanicolau. FNAC shows aggregates consisting of epithelioid cells, histiocytes, giant cells and lymphocytes without caseous necrosis. Necrotic material in the background was not detected. The specimen was reported as granulomatous inflammation compatible with a sarcoidosis (Figures 2A-C). The serum level of angiotensin-converting enzyme (ACE) was 62 ng/ml. Tuberculin skin test (TST) was performed to rule out tuberculosis and was negative. Radiologically, involvement of other organs was not found.

After the prednisone and non-steroidal anti-inflammatory drugs therapy, which lasted for 2 months, significant reduction in the parotid glands and elimination of xerostomia was seen in patient. Level of ACE decreased. There were no complications or recurrence during 7-month follow-up.

DISCUSSION

Sarcoidosis is a systemic disease characterized by granulomatous inflammation. Lung and lymph node involvement is seen more commonly than other organs. Sarcoidosis is slightly more prevalent in women than men and it is more common in adults (1). Parotid gland involvement occurs in 0.5-15 % of patients of sarcoidosis (3). The main clinical signs are painless swelling of parotid glands, cervical lymphadenopathy and xerostomia (4). In our case, all three findings were present.

Biopsy, radiography and serum ACE level are important in the diagnostic process. Histologically and cytopathologically noncaseating granulomas, composed of epithelioid cells, histiocytes and giant cells are characteristic features for sarcoidosis (5, 6). Within the granulomas are epithelioid cells, which may contain stellate inclusions (asteroid bodies) and concentrically laminar calcifications (Schaumann bodies) (1). There are a few case reports in the literature illustrating asteroid bodies and schaumann bodies in cases of
sarcoidosis in cytological material (7-9). Any these type specific signs were absent in our case. Radiologically conventional sialography usually demonstrates normal proximal ducts, a few fragile distal ducts and non-specific parenchymal nodules. Computed tomography, with or without simultaneous sialography, defined and characterised such nodules and demonstrated normal periparotid anatomy (10). Seventy per cent of patients presented elevation serum ACE (11). Elevation of this enzyme in conjunction with a positive chest radiograph has a high diagnostic reliability (1).

The differential diagnosis includes tuberculosis, crohn’s disease, tuberculoid leprosy, cat-scratch disease, fungal infections, and toxoplasmosis. The results of bacterial culture are important to exclude diseases with bacterial etiology. In the tuberculosis granulomas often contain caseous necrosis, but sometimes granulomas without necrosis and the TST (tuberculin skin test) usually is positive. Tuberculoid leprosy is characterized by the presence of non-necrotizing granulomas with acid fast bacilli absent or few in number and the lepromin skin test is positive. In the cat-scratch disease "suppurative" and pus forming granulomas, containing large numbers of neutrophils are usually seen. Fungal infections of parotid gland usually seen in patients with acquired immunodeficiency syndrome and cytopathologic image characterized by necrosis and scattered fungal forms. The salivary glands involvement, in a range of other diseases that makes up epithelioid granulomas, is possible. There are few cases of intra-glandular toxoplasmic lymphadenitis of parotid gland and Crohn's disease of minor salivary gland ducts in the literature (12, 13). In our case, the serum level of ACE was examined and TST and radiological methods were applied for differential diagnosis.

The main treatment of sarcoidosis is corticosteroids and nonsteroidal antiinflammatory drugs although in the literature some patients have been found to be treated surgically (14, 15). Spontaneous regression of this disease has been reported (16). In our case, prednisone and antiinflammatory drugs were used for treatment.

**CONCLUSION**

FNAC can be used as a cost-effective, rapid and less invasive diagnostic method for differential diagnosis of such diseases with granulomatous inflammation in addition to clinical, radiologic and laboratory findings.
REFERENCES

Figure 1: Magnetic resonance imaging shows diffuse and non-nodular bilateral parotid enlargement and normal periglandular anatomy.
Figure 2: FNAC shows aggregates consisting of epithelioid cells, histiocytes, giant cells and lymphocytes (Papanicolaou, x400).