

Kimura's Disease of the Parotid Gland: A Case Report

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Abstract

Kimura's disease is a chronic inflammatory disorder of unknown etiology mostly affecting subcutaneous tissue, lymph nodes and salivary glands and usually limiting to head and neck region. A 36-year-old man presented to the Department of Surgery, Regional Institute of Medical Sciences, Imphal with painless swelling involving the superficial lobe of left parotid, fine-needle aspiration cytology of which showed polymorphic population of lymphoid cells, plasma cells and histiocytes suggestive of lymphoma. Peripheral blood gave a picture of marked eosinophilia. With a provisional diagnosis of pleomorphic adenoma, superficial parotidectomy of the left side was done. Final histopathology picture was of Kimura's disease. Post-operatively, the patient was put on oral cetirizine 10 mg daily, and there was no evidence of recurrence until last follow-up. Kimura's disease should be considered in the differential diagnosis of parotid swelling.

Keywords: Eosinophilic micro abscesses, Immune mediated disease, Kimura's disease, Parotid gland

INTRODUCTION

Kimura's disease is a chronic inflammatory disorder of unknown etiology mostly affecting subcutaneous tissue, lymph nodes and salivary glands and usually limiting to head and neck region. It is believed to be an immune-mediated disease and TH2 cells are suspected to play an important role.¹ Microscopically, there is lymphoid nodules with marked infiltration of eosinophil, formation of eosinophilic microabscesses, vessels with hobnail endothelial cells. The exact prevalence of Kimura's disease is not known, but is more reported in Asian populations. Here we present a case of Kimura's disease involving left parotid gland.

CASE REPORT

A 36-year-old man presented to the Department of Surgery, Regional Institute of Medical Sciences, Imphal with painless swelling over left jaw (Figure 1). Clinical examination revealed nontender, soft to firm swelling involving the superficial lobe of the left parotid along with left cervical lymphadenopathy (Level II and III). Computed tomography scan of proliferative nodules suggested pleomorphic adenoma of the left parotid. Fine-needle aspiration cytology



Figure 1: Swelling of left parotid gland

(FNAC) of the parotid showed polymorphic population of lymphoid cells, plasma cells and histiocytes suggestive of lymphoma (that required histopathological examination for

confirmation). Peripheral blood gave a picture of marked eosinophilia (differential count of 46% and absolute count of 7,800/cumm). With a provisional diagnosis of pleomorphic adenoma, superficial parotidectomy of the left side was done (Figure 2). Final histopathology showed lymphoid follicles with marked eosinophilic infiltrates, eosinophilic abscess and few hyalinized vessels. Overall picture was of Kimura's disease (Figure 3). Post-operatively the patient was put on oral cetirizine 10 mg daily till last followup. There was no evidence of recurrence till last follow-up.

DISCUSSION

Kimura's disease is a rare inflammatory disease of unknown etiopathogenesis, first reported from China in



Figure 2: Superficial parotidectomy of left parotid

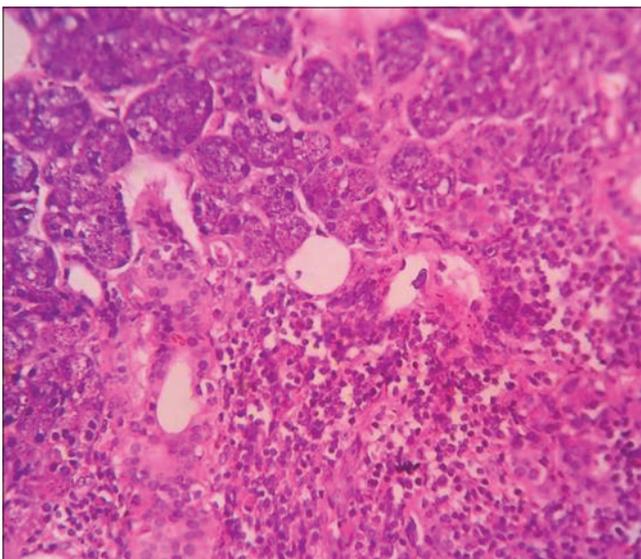


Figure 3: Histopathological examination of superficial parotidectomy specimen reported as Kimura's disease

1937 which was then termed as "eosinophilic hyperplastic lymphogranuloma."² It shows higher predilection of male patients in the third decade of life. More reported in Asian populations, it is, usually, confined to head and neck region. A closely related entity is angiolymphoid hyperplasia with eosinophilia (ALHE). These two diseases are distinguished on the basis of clinical and histopathological features. Lymphadenopathy and eosinophilia are more commonly seen in the case of Kimura's disease as compare to ALHE and in histopathology, Kimura's disease shows sparse vascular component and more of lymphoid proliferation with prominent eosinophilic cell infiltrate.³

Kimura's disease of the parotid is often misdiagnosed with more common lesions of the parotid such as neoplastic lesions, benign lymphoepithelial lesions (Mikulicz's disease), angioimmunoblastic lymphadenopathy, etc. When there is associated cervical lymphadenopathy, clinicians are often prompted with a diagnosis of a malignant lesion. In our case, FNAC of the parotid swelling showed feature of lymphoma that prompted biopsy for confirmation. FNAC of cervical lymphadenopathy was non-specific lymphadenitis. A minimum biopsy of parotid was superficial parotidectomy and hence we proceeded with the procedure.⁴

The optimal treatment of Kimura's disease is not defined. Observation has been advised for asymptomatic lesion. Medical treatment has been described using cetirizine, steroids, cyclosporins, retinoids with variable degree of success.⁵⁻⁷ However, recurrence on cessation is a problem. Intravenous immunoglobulin has been reported to give good remission. Surgery has been performed for primary, isolated lesion. Radiotherapy has been occasionally used. Hareyama *et al* reported the use of radiotherapy at dosages of 26-30 Gy with local control rate of 74%.⁸ However, the use of radiation for the treatment of benign disease would be rationale is not known.

CONCLUSION

Kimura's disease is a rare, chronic inflammatory condition of unknown origin that is more commonly seen in Asia with more male sex preponderance. It often involves subcutaneous tissue, salivary glands and lymph nodes in head and neck region, and there is associated marked eosinophilia. Kimura's disease, even though rarely encountered, must be kept in mind in the differential diagnosis of salivary gland tumors, especially when there is marked eosinophilia. Especially, when associated with cervical lymph node enlargement it is likely to be mistaken for a malignant lesion. A correct diagnosis is important because of its reported responsiveness to nonsurgical treatment and high rate of recurrence.

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