Int J Pharm Bio Sci 2012 Oct; 3(4): (B) 311 - 315

Review Article **Pathology**

International Journal of Pharma and Bio Sciences **ISSN 0975-6299**

KIMURA'S DISEASE: A DIAGNOSTIC CHALLENGE.

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ABSTRACT

Kimura's disease (KD) is a relatively uncommon chronic inflammatory condition that appears to represent an unusual allergic or autoimmune response. It is extremely uncommon in Indian subcontinent. This case is being presented to make clinicians and ENT surgeons aware of Kimura's disease as a differential diagnosis of swelling in head and neck region, because these are frequently misdiagnosed as angiolymphoid hyperplasia with eosinophilia, Hodgkin's lymphoma, castleman's disease, angioma, lymphangioma or haemangioma.

KEY-WORDS: **Kimura's disease, Angiolymphoid hyperplasia with eosinophilia, Cheek.**

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INTRODUCTION

Kimura's disease (KD) is a relatively uncommon chronic inflammatory condition that appears to represent an unusual allergic or autoimmune response.[1] It was first described in 1937 by Kimm and Szeto in China; who reported seven cases of benign lymph node enlargement with eosinophilic infiltrate, which they termed as eosinophilic hyperplastic lymphogranuloma. Kimura et al. in 1948 from Japan reported a similar finding and described it as an "unusual granulation and hyperplastic changes of lymphatic tissue", and this condition has since become widely known as Kimura's disease.[2] Although the disease can become apparent at any age, most cases occur in the 2nd & 3rd decades of life, with 80 -87% of the affected patients being males, with male-female ratio 3:1.

KD has preponderance in the far eastern countries. It is uncommon in Indian subcontinent.[3] Recurrence is common, occurring in up to 25% of cases treated with surgical excision alone. Clinically as well as histo-pathologically, it can cause considerable diagnostic dilemma.

CASE HISTORY

A 15 year old Indian boy presented with a left sided, painless swelling at the pre-auricular region for nearly one and half year, and over the past 3 months, he noticed a progressive increase in the size of the swelling. There was no discharge, skin changes nor excessive salivation noted, and facial nerve function was intact. The patient's medical history was otherwise unremarkable. The significant clinical findings was left sided cheek swelling measuring about 6x5cm, causing facial asymmetry, but no palpable cervical lymphadenopathy. (Fig-1) Hematological examination showed raised peripheral eosinophil counts (4400 cells/cumm), and elevated serum immunoglobulin E (IgE) levels. Renal function tests were within normal limits. And all other routine invetigations were within normal limits. Fine needle aspiration cytology was performed and was suggestive of cystic lympho-epithelial lesion. Ultrasonography revealed hemangioma.

***PATHOLOGY*:** Mass was excised (Fig-2). It was 5x4cms with unclear borders. A characteristic appearance was the adherence of the skin to the parotid mass, with no clear plane of dissection between the lesion and the parotid tissue.

***MACROSCOPY*:** Received single globular mass measuring 5x4cms with a stalk like structure.

Cut surface solid, firm and grey-white appearance. (Fig-3)

***MICROSCOPY*:** Microscopic examination revealed hyperplastic lymphoid follicles (Fig-4) involving skin and salivary gland tissue with marked infiltration of eosinophils, and proliferation of capillaries. Occasional eosinophilic abscess were also noted. (Fig-5)

So a diagnosis of Kimura's disease was made on histopathology and confirmed with increase in peripheral eosinophilic count and serum IgE levels.

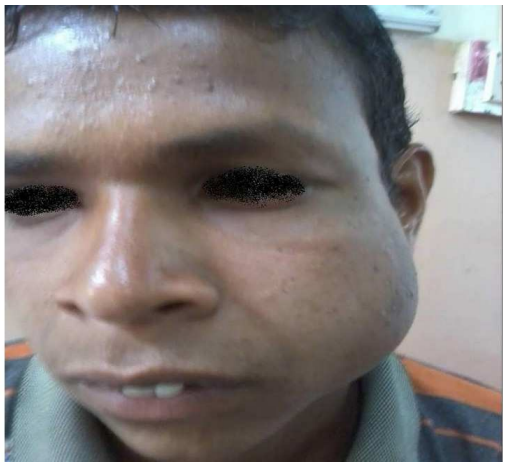

Figure1.
Clinical Photograph showing swelling over left cheek/mandibular region

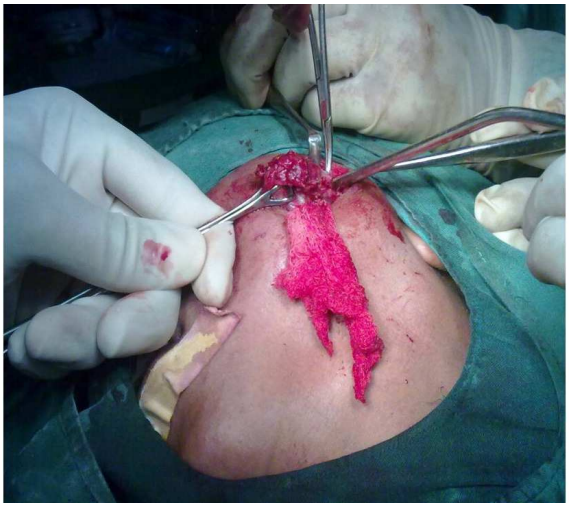

Figure 2.
Intra-operative photograph

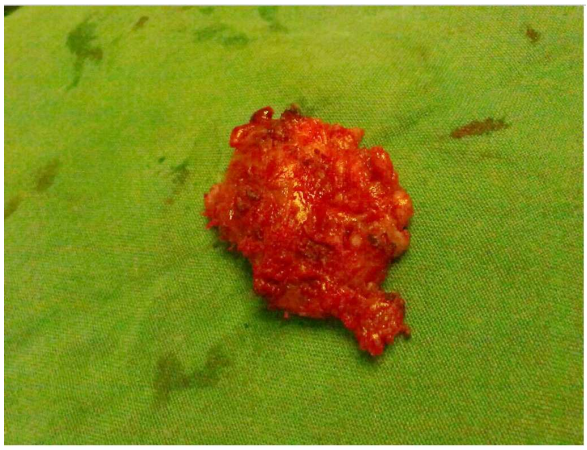

Figure 3
Mass grossly measuring 5x4cms.

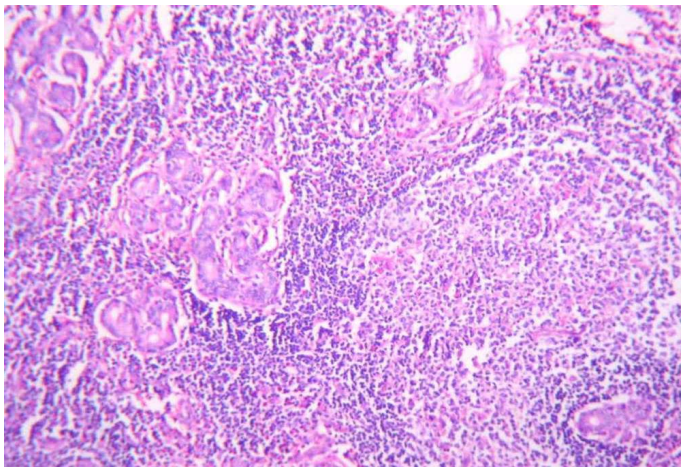

Figure 4.
Photomicrograph showing hyperplastic lymphoid follicles and proliferation of capillaries. Low power (10X)

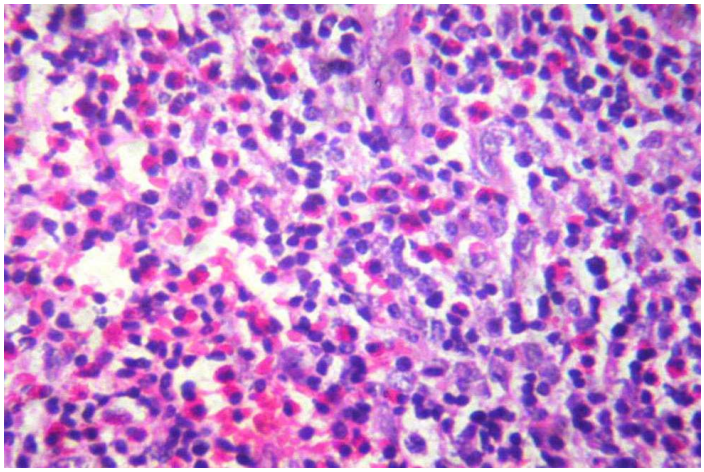

Figure 5.
Photomicrograph showing eosinophilic abscess. High power (40X)

DISCUSSION

Kimura's disease (KD) often involves young oriental males and commonly presents with a solitary or multiple, painless, slow growing subcutaneous nodules.[3] It usually involves the head and neck region, particularly the parotid gland and submandibular areas, and rarely the scalp and orbital region.[1] In approximately 12% of patients, there is a related renal disease, usually presenting as nephrotic syndrome.[3] KD is a benign but disfiguring disease with no evidence of malignant transformation. Under microscopic examination, the lesions of KD are characterized by lymphoid hyperplasia, marked infiltration of eosinophilic granulocytes, and proliferation of capillaries. This is usually accompanied by peripheral blood eosinophilia and elevated serum IgE levels as in our case.[3] Follow up is important because there are reports of recurrence and cases of Hodgkin's lymphoma in patients with KD. Our patient is under follow up and keeping fine. Imaging is often unhelpful. Ultrasound may demonstrate masses that are solid, round or oval, or hypoechoic but they do not really show the nature of the lesion. CT and MRI findings may help to delineate the extent of the disease in more than one

plane.[2] At the moment, surgery is still the treatment of choice as it can be both diagnostic and therapeutic.[4] Differential diagnosis should include angiolymphoid hyperplasia with eosinophilia, lymphoma especially Hodgkin's lymphoma, castleman's disease, salivary gland tumours, lymphangioma or haemangioma, reactive lymphadenopathy, nodal metastasis, hamartoma and Mikulicz's disease. Immunohistochemistry (IHC) can be used to differentiate KD from malignant neoplasms. In our case, IHC was not done because histopathological diagnosis was conclusive and the patient also had increase in peripheral eosinophilic count and serum IgE levels, which were suggestive of KD.

CONCLUSION

This case is being presented because clinically as well as histo-pathologically, it can cause considerable diagnostic dilemma; Also to make surgeons aware of Kimura's disease, as a differential diagnosis of swelling in head & neck region, because in the management, surgical treatment should not be aggressive.

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