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Case Report

Kimura Disease: A Rare Entity

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Abstract: Kimura disease (KD) is a rare benign chronic inflammatory disorder of unknown etiology and pathogenesis. It is common in Asian population and affects mostly young adults in head and neck region. Differential diagnosis of KD includes Kikuchi disease, Mikulicz's disease and neoplasm like Hodgkin and non-Hodgkin lymphoma. A detailed clinical history along with radiological and pathological correlation is required for the correct diagnosis. Recurrence rate of Kimura disease is very high following any kind of treatment and routine follow up is mandatory. We present a case of KD in a 21-year-old Nepalese male.
Keywords: Case report, Kimura Disease, Pinna.

Introduction

Kimura disease (KD), also known as eosinophilic lymphogranuloma, was first reported by Kimm and Szeto in 1937 in China.[1] But the term" Kimura disease" came into existence after a Japanese doctor named Kimura et al. published a systemic description of the disease.[2] KD is a benign chronic inflammatory disorder of unknown etiology. It is common in Asian population and affects mostly young adults. Infra-auricular and retro-auricular areas are most commonly affected and often accompanied by involvement of major salivary glands and regional lymph nodes. Here, we present a case of KD in right pinna of a 21-year-old Asian male.

Case report

A 21-year-old male presented to ORL-HNS OPD of a tertiary centre with a slowly enlarging mass on right pinna for 9 months. The swelling was painless, slowly growing but associated with mild itchiness. There was no history of trauma and any piercing. Local examination revealed 2x2 cm swelling over the right pinna in cavum concha region obscuring the right auditory meatus (Figure 1).

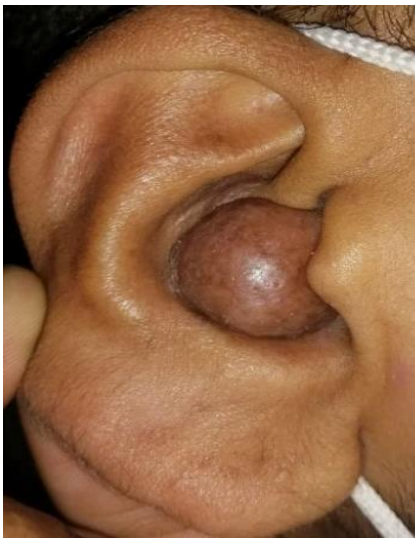

Figure 1: Pre-operative image of right pinna

The swelling was firm, non-tender and non-adherent to the skin and there was no associated cervical lymphadenopathy. His routine full blood count was normal except a slightly higher eosinophils count of 0.6 g/dL. Surgical excision of the mass was done under local anesthesia with a provisional diagnosis of keloid. Histopathological examination of the specimen showed a dense inflammatory infiltrates consisting of large number of eosinophils and lymphocyte forming multiple lymphoid follicles along with neutrophils and plasma cells suggesting a diagnosis of Kimura disease (Figure 2). The patient's recovery was uneventful and there was no recurrence in 24 months' follow-up (Figure 3).

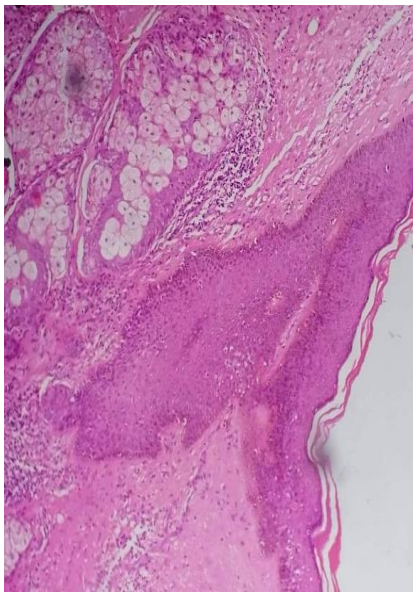

Figure 2: Findings in histopathological examination 10x magnification

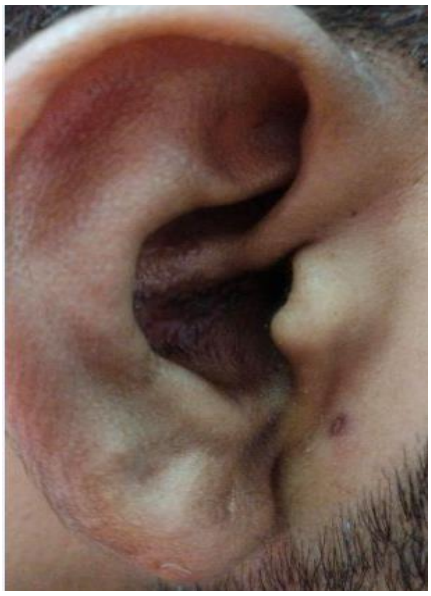

Figure 3: Post-operative image of pinna

Discussion

KD is a rare chronic inflammatory disorder of unclear etiology. Majority of cases are reported in Asia (China, Japan, and Southeast Asia), mainly affecting young Asian males. Disease usually presents as a long-standing and painless subcutaneous mass lesion in the head and neck region (majority in Infra-auricular and retro auricular areas). There may be involvement of salivary glands and regional lymph nodes. Rise in serum IgE and eosinophilia is a common finding. Nephropathy is associated with KD in 12-16% of patients and the reason is not known[3]. Differential diagnosis of KD includes Mikulicz's disease, Kikuchi disease, and neoplasm like Hodgkin and non-Hodgkin lymphoma. A detailed clinical history along with radiological and pathological correlation is necessary for the correct diagnosis, because there are no pathognomonic features of KD.

Histopathology of the subcutaneous lesions, salivary glands and lymph nodes shows a mixed inflammatory cellular infiltrate composed mainly of eosinophils and lymphocytes as in our case. The inflammatory infiltrate contains numerous lymphoid follicles and prominent germination centres. Fine needle aspiration and core-needle biopsy have low diagnostic accuracy, so a histopathological examination of the surgical specimen is a must for diagnosis[4].

There is no standardized treatment protocol for KD till now. Various treatment options are surgical excision, administration of systemic steroids and radiotherapy. Treatment for KD is usually unsatisfactory as new lesions may occur and recurrence after resection is very common. Treatment with intralesional corticosteroid (Triamcinolone acetonide) had been reported to have a better outcome[5].

Conclusion

KD, though rare, should be considered in cases with subcutaneous nodules in head and neck region especially in young Asians. A high index of suspicion is essential to diagnose this condition and confirmation of diagnosis is done with histopathological examination. Although, there is no definitive protocol for management of KD, options available are surgical excision, administration of systemic steroids and radiotherapy. Recurrence rate of KD is very high following any kind of treatment and routine follow up is mandatory.

Declarations
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Informed Consent: Written informed consent was obtained from the patient to publish this case report and accompanying images.
Author Contributions: Sonika Dhari Shrestha, drafting and compiling of article, getting consent from patient; Srijana Pandeya and Puja Sharma, help with HPE image.

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