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CASE REPORT

Kimura's Disease: A Case Report

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Abstract Kimura's disease is a rare, benign, slow-growing chronic inflammatory swelling with a predilection for the head and neck region and almost always with peripheral blood eosinophilia and elevated serum IgE levels. Here, we report a 14-year-old male patient with left-sided cheek swelling. It was painless swelling situated anterior to left ear, of 1 year duration, without any other symptoms. Routine investigations were inconclusive. Excision biopsy revealed the diagnosis of Kimura's disease.

Keywords Kimura's disease · Eosinophilia · Eosinophilic lymphogranuloma · Subcutaneous nodules · Local adenopathies

Introduction

Kimura's disease is an allergic inflammatory disorder of unknown cause, endemic in Orientals, but rare in Tropics. It is a benign condition, but may be mistaken for a malignant disease. Renal involvement is its only systemic manifestation. Kimura's disease is an inflammatory disorder that presents as massive painless, subcutaneous swelling in head and neck and the submandibular region in young men with unknown cause. Major salivary glands and lymph nodes may also be involved. Bilateral involvement is rare. Patients almost always have marked peripheral eosinophilia and elevated serum IgE levels. Bilateral involvement is rare. Treatment options consist of surgical excision, corticosteroids and follow-up.

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Case Report

A 14-year-old boy, resident of Bijapur, Karnataka, presented to surgical OPD with complaint of left cheek swelling for 1 year, which was slowly increasing. On physical examination, he was found to be moderately built and moderately nourished. A 6×4 cm, vertically oval, non-tender swelling was found to be present on the left-side cheek, 6 cm in front of left Tragus of the left ear. Non-adherent to skin and mandible, restricted mobility, a 1.5×1 cm left preauricular lymphnode enlargement was present (Fig. 1).

Hematological Examination

Hemoglobin	17 g/dl
Total WBC count	11,600 cells/mm^3
DLC—Neutrophils	66 %
Lymphocytes	30 %
Eosinophils	4 %
Basophils	0 %
ESR	10 mm

Fine needle aspiration cytology revealed a cystic lymphoepithelial lesion.

Excision biopsy was performed under general anesthesia, which revealed features of Kimura's disease: hyperplastic lymphoid follicles with prominent germinal centers. Germinal centers are well vascularized, showing interstitial fibrosis and deposition of proteinaceous material, abundant inflammatory cells compromised of eosinophils with occasional micro abscess, lymphocytes and macrophages. Amidst the cells are many glands, keratin material and dilated and congested blood vessels lined by spindle-shaped endothelial cells (Fig. 2).

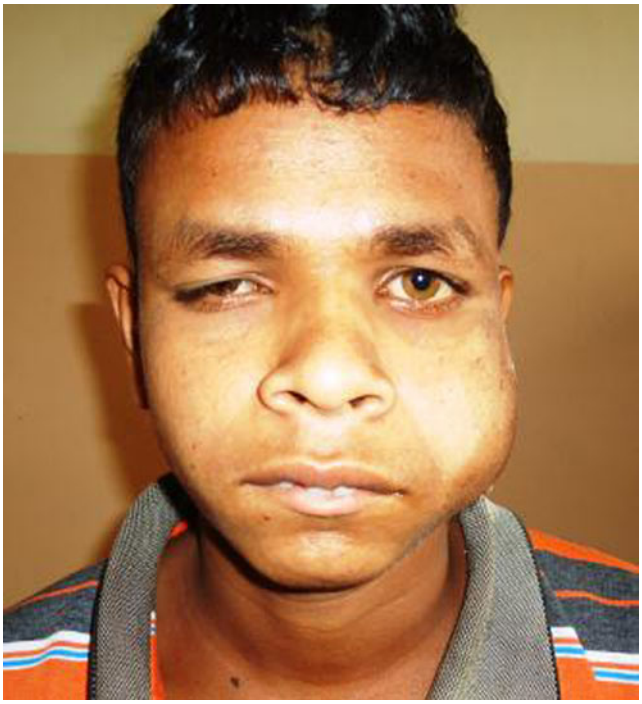

Fig. 1 Pre-operative photograph

The post-operative complication was seroma formation, which was managed by aspiration by a sterile syringe.

Comments

Classical triad of the disease is painless subcutaneous masses in the head and neck region, blood and tissue eosinophilia and markedly elevated IgE. Renal involvement is the only systemic manifestation which may affect up to 60 % of patients as

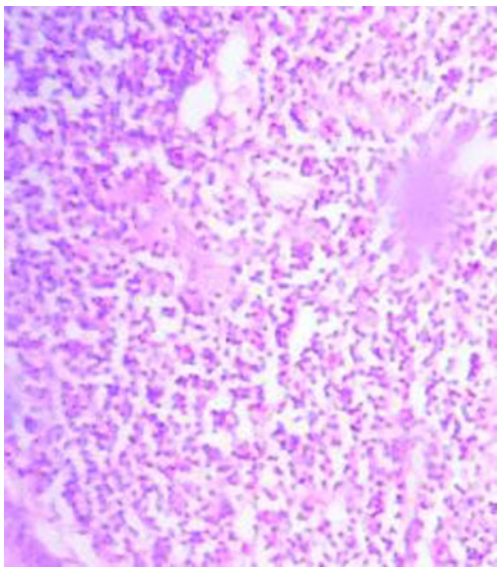

Fig. 2 Biopsy showing lymphoid tissues and germinal centres

membranous glomerulonephritis, minimal change glomerulonephritis, diffuse proliferative glomerulonephritis, mesangial proliferative glomerulonephritis and also nephrotic syndrome [1].

Associated with systemic disease are nephritic syndrome [2], juvenile temporal arteritis and coronary artery spasm [3].

It is interesting to note that calcium channel antagonist or nitroglycerine, which is commonly used for variant angina, shows a little effect in coronary artery spasm associated with Kimura's disease [4].

Glucocorticoids seem to exert strong anti-vasospastic effect on the coronary artery by reducing eosinophilic infiltration and suppressing development of inflammatory mediators [5].

A number of theories about its origin have been proposed, which include interference with immune regulation, atopic reaction to a continuous antigenic stimulus (especially Candida albicans) parasitic infection and neoplasm. The most popular theory is that of Candida acting as a source of persistent antigenemia, although neither hyphae nor spores have been isolated [2].

Kimura's disease is a benign disease which does not require long-term treatment. Surgical resection has been found to be curative although recurrence is common [6]. Cytotoxic agents like cyclosporine, pentoxyfilline and systemic steroids may be indicated in frequent relapses or cases complicated by nephrotic syndrome [7].

Irradiation should be considered not only in patients resistant to steroids, but also in young patients in whom the long-term side effects of steroids may be more deleterious than a limited course of irradiation that may prevent relapse [8].

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