Recurrent Kimura Disease in an Asian Male: A Rare Case Report and Review of Literature

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Abstract: Kimura's disease is a rare benign disorder of unknown cause. It presents as multiple swelling in the head and neck region and primarily affects young males of Asian region between the age group of twenty to fourty. Patients can be managed conservatively in asymptomatic cases. Symptomatic lesions can be treated with intralesional corticosteroids, oral cyclosporine, oral pentoxyphylline, all trans retinoic acid and intravenous immunoglobulin. Photodynamic therapy and radiotherapy have also been used in recurrent lesions with success. Though many therapies are available, conservative surgical excision remains the treatment of choice. We present a case of young Asian male who presented with recurrence of Kimura Disease and was treated with conservative surgical excision. The purpose of the article is to focus on the presentations and discussion of the various treatment options

Keywords: Kimura disease, Recurrent, Asian

I. Introduction

Kimura's disease is a rare disorder characterised by involvement of subcutaneous tissue and lymphadenopathy of head and neck region. It is a chronic inflammatory disorder associated with raised eosinophils and serum immunoglobulin E^1 . It is a benign condition endemic in Asians² with a male to female ratio of 3.5-7:1 .most commonly between 20 to 40 years of age.

II. Material and methods

29 year male resident of Nagpur,India presented with recurrent swelling of right cheek for three years . He had underwent excision of a lesion outside 3 years back. Histopathology was suggestive of Kimura's disease. Patient was a known case of Nephrotic syndrome for which he was taking treatment from Nephrologist. Patient started developing swelling of cheek after 5 months of surgery. Swelling gradually increased in size to involve two thirds of the right cheek and and another swelling in right post auricular region. Swelling intermittently increased and decreased in size. Examination revealed a soft tissue swelling of right cheek with ill defined margins. Swelling occurred in the region of the prior surgical scar through which previous excision of lesion was done. Skin over the swelling was adhered to the previous scar. Mucosa below the swelling was free. (Figure 1-4) There was a firm right postauricular lymph node. Magnetic resonance tomography of the lesion revealed lesion in subcutaneous plane abutting right Masseter.

Patient was diagnosed as recurrent Kimura's disease and planned for Surgery after anesthetic fitness. Exploration of the right cheek swelling through the previous incision was done. Intraoperatively fibro fatty tissue encasing masseter was removed. Post auricular lymph node was also send for excisional biopsy. Post operative period was uneventful and patient was discharged on 7th post operative day after drain removal. Histopathology of the facial swelling was suggestive of Kimura's disease. Post auricular lymph node showed reactive hyperplasia. There had no new swelling in early follow up in 6 months.

III. Discussion

H T Kimm and C Szeto were the 1st to describe this disease in 1937. Kimura is credited for the histological description in1948³. Though the exact cause is unknown, type 1 hypersensitivity following infections or allergy may predispose. There is a predominance of T helper 2 cels in these patients⁴. Infections due to candida or other parasites and bites from arthopodes can lead to Kimura's disease due to persistant antigenic stimulation¹. The disease presents as subcutaneous nodules in the region of head and neck which may be associated with lymphadenopathy in 30-40% of cases⁵. They may be associated with enlargement of parotid and submandibular glands.

Renal involvement may be seen in upto 60% of patients which may be in the form of any type of Glomerulonephritis ¹. Involvement of inguinal region and extremities has also been reported ⁶.

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Serum immunoglobulin E levels may be raised. Magnetic resonance imaging will reveal the extent of the lesion with associated vascular involvement. Definitive diagnosis requires biopsy. It's differential diagnosis includes Acute lymphocytic leukemia, Hodgkins disease, Cylindroma, Eosinophilic granuloma, Angioimmunoblastic lymphedenopathy angiolymphoid hyperplasia and Dermatofibrosarcoma protuberens.

Symptomatic lesions can be managed conservatively. Medical management in the form of steroids, cyclophosphamide and antiplatelet drugs may be used for localised lesions, those with renal involvement and in recurrent cases. Surgical resection can be considered as first line treatment though it is associated with relapses³. It can be used for young patients and those with localised recurrence. Radiotherapy can be used for nonresectable, recurrent lesions where medical therapy had been found to be ineffective.

Though patient may present with recurrences, malignant transformation has not been reported till date.

IV. Conclusion

Kimura's disease is very rare and limited cases have been reported. It's presentation is like a lymphoid tumor. The aim of this paper is to repot this rare disease in this part of subcontinent and to familiarise the treating Surgeons and Physicians the nature of it's presentation for proper diagnosis and management.



Photograph 1-Preoperative photograph- Frontal view



Photograph 2-Preoperative photograph- intraoral view



Photograph 3-Preoperative photograph right lateral view



Photograph 4-Preoperative photograph -Right oblique view

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