

A Rare Presentation of Kimura's Disease

Sasikumar Pattabi, L. Santhanagopal, G. K. Shreedhar, Dev Krishna Bharathi

Sree balaji medical college and hospital, Chrompet, Chennai, India

Abstract: Kimura's disease is a chronic idiopathic inflammatory disorder presenting as painless swelling in the head and neck region due to unusual granulation with hyperplastic changes. It is predominant in young Asians. It is usually limited to skin, lymph nodes and salivary glands. It rarely affects other parts of the body. We report a rare presentation of this disease in the right gluteal region and right thigh in a 62 year old male.

INTRODUCTION

Kimura's disease is first described in China by Kim and Szeto¹ in 1937 and then in 1948 by Kimura et al² in Japan. It is commonly seen in the young asian males of age group 20 to 40 years. It is commonly seen in the head and neck regions as a painless swelling. It rarely affects other parts of the body⁴. Diagnosis is established by biopsy and histopathological examination.

CASE REPORT

A 62 year old male presented with a swelling in the right gluteal region and the thigh for the past 3 months. He noticed the swelling in the right gluteal region and thigh 3 months back. It was gradually increasing in size. He complained of mild pain over the swelling. He underwent excision of a swelling at the same site in thigh 6 months back. Histopathological report was not available. There was no history of trauma, fever or similar swellings anywhere else in the body. On examination there was a swelling measuring 12x5 cms in right thigh and 8x4 cm in the right gluteal region. The swelling was tender, firm and the edges were smooth. Skin was fixed to the swelling. There were no significant regional lymph nodes. Other side is clinically normal. There was no distal neurovascular deficit.

Baseline investigations revealed hemoglobin: 12g%, WBC: 8600 (neutrophils: 46%, lymphocytes: 40%, eosinophils: 12%, monocytes: 2%). ESR, Urine analysis, sugar, urea and creatinine were within normal limits. X rays showed no bony abnormality. FNAC done elsewhere was inconclusive.

Provisional diagnosis of 'soft tissue sarcoma' was made but trucut biopsy carried out showed no evidence of malignancy. Wide local excision of the mass was done, enclosing the biopsy site and the previous scar with adequate margins. The resected specimen was sent for HPE. The cut section showed white fibrotic area enclosing the brownish area close to posterior margin. The microscopic section showed fibrofatty muscle tissue enclosing areas of intense inflammatory cell collection predominantly showing sheets of eosinophils mixed with mononuclear cells and a few foreign body type of giant cells and histiocytes. Eosinophilic abscesses, lymphoid aggregates, proliferating capillaries and thrombosed blood vessels were also seen. Focal areas of calcification were also seen. These findings are consistent with that of Kimura's disease.

Post operative period was uneventful. Patient was started on high dose steroids. Sutures were removed after 12 days. High dose steroids were weaned off after 6 weeks. Patient was followed up till date for 2 years and there has been no evidence of recurrence.

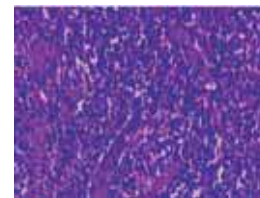
DISCUSSION

Kimura's disease is a chronic inflammatory disorder due to unusual granulation.

Correspondence: Dr. Sasikumar Pattabi, Associate Professor, plot no. 1, bhuvaneshwari nagar first cross street, velachery, Chennai-600042, Tamil Nadu, India
e-mail : psk_66in@yahoo.com



Naked eye picture



Microscopic picture

It was first described by szeto and kim¹ as "eosinophilic hyperplastic granuloma" and kimura² in 1948 as "unusual granulation and hyperplastic changes of lymphatic tissue". It is common in young asian males of age group of 20 to 40 years. It presents as a painless swelling in the the head neck region. But it rarely presents as swelling in the oral cavity, groin, trunk and limbs. It is usually associated with lymphadenopathy. Salivary gland involvement is also seen commonly. There are reported evidences of renal involvement⁶. The aetiology of the disease is unknown, though there are higher incidence of this disease in people with eosinophilia, increased IgE levels and increased antibodies for candida.

Imaging techniques are not of much value in diagnosing this disease though this is helpful in delineating the extent of the disease. FNAC is usually inconclusive. The diagnosis is arrived by either trucut, incisional or excisional biopsy. Differential diagnosis is to be made with Angiolymphoid hyperplasia³ with eosinophilia. In Angiolymphoid hyperplasia with eosinophilia histocytoid blood vessels with vacuolated endothelial cells are seen with intact germinal centers in contrast to kimuras disease where germinal centers are destroyed due to heavy infiltration of eosinophils and there is absence of vacuolated endothelial cells⁵. Also in kimuras disease lymphoid follicles are formed with vascular proliferation fibrosis and micro abscesses. Treatment is by surgery, steroids or radiotherapy. The recurrence is rare after complete resection.

This case is reported because of the occurrences in (1) elderly male (62 yr old man) and (2) in the thigh and gluteal regions.

REFERENCES

1. Kim H. T., Szeto C., Eosinophilic hyperplastic lymphangiomas, Chinese medical journal 23:699-700
2. Kimura T., Yishimura S., Ishikawa E., On the unusual granulation combined with hyperplastic changes of lymphatic tissue. Trans. Soc. Pathol. Japan. 37 : 179-80
3. Briggs PL. Kimura disease is not angiolymphoid hyperplasia with eosinophilia: clinical and pathological correlation with literature review and definition of diagnostic criteria (Portuguese). An. Bras. Dermatol. 2006;81 (2): 167-173.
4. A Hafez, Kimura's disease with atypical musculoskeletal presentation, morthoj, 2010 vol 4 no 2 : 44-6.
5. S R Ranga, Kimura's disease, Indian Journal of Othorhinolaryngology and head and neck surgery vol.56 no 1, January - march 2004 : 43-5.
6. Atar S., Oberman , Recurrent nephrotic syndrome associated with kimars disease; nephron 68 : (2) 259-61.