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Kimura's Disease: A Case Report and Literature Review

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Abstract

Kimura's disease, common pathology in the East, responsible of chronic neck swelling is rarely reported in sub-Saharan Africa. We reported a case which was observed in the internal medicine department of Aristide Le Dantec hospital in Dakar. This was a young 15-year-old, with no particular disease history, who had recurrent non-inflammatory swelling next to the left zygomatic bone associated with itching. Biology revealed an inflammatory syndrom, eosinophilia and increased serum IgE. The histological examination of the mass biopsy concluded to Kimura disease. The corticosteroid has reduced the size of the mass within a few weeks of treatment. Kimura's disease is unknown in our regions. His painless character and chronic evolution delay the time of diagnosis. This case proves the reality of this disease, which must find a place in the diagnosis approach of cervical swelling.

Keywords

Cervical Lymph Nodes, Kimura's Disease, Sub-Saharan Africa

1. Introduction

Kimura's disease or eosinophilic hyperplastic lymphogranuloma is a chronic inflammatory disease of unknown etiology. It is manifested by swelling of subcutaneous sitting in the cervical region; they are accompanied by satellite lymphadenopathy associated with hyper eosinophilia and elevated serum IgE [1]. The prognosis is good; no malignant transformation is observed.

It was described for the first time in China in 1937. It is a frequent pathology in the Far East rare in west and

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exceptionally rare in Africa, where it has been reported only one single case in the black population [2].

We report a case of Kimura's disease in a young Senegalese child.

2. Observation

It was a 15 years old child without any special medical history or concept known personal or family atopy. He consulted for a painless soft facial swelling, movable relative to the deep plane of 6 × 5 cm next to the zygomatic bone left (**Photo 1** and **Photo 2**). This swelling evolved for about 4 years. There was no fever and general condition was kept. The remainder of the physical examination was normal including the specialized examination of the ENT; there was no adeno-splenomegaly.

Laboratory analysis showed an inflammatory serum markers with accelerated erythrocyte sedimentation rate of 65 mm in the first hour, C reactive protein 20 mg/l. polyclonal hypergammaglobulinemia to 19.1 g/l, anemia

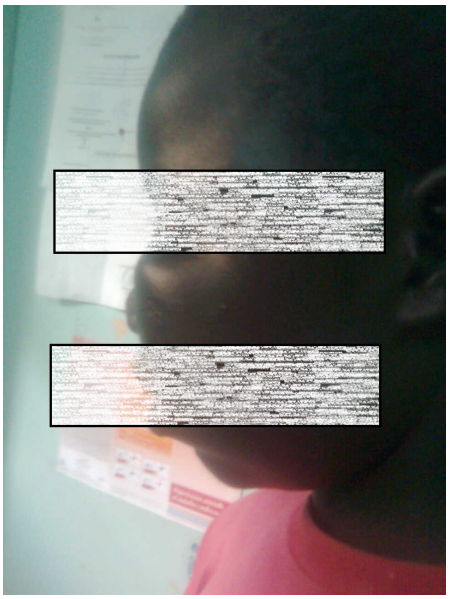

Photo 1. Left cheek swelling (lateral view).

Photo 2. Left cheek swelling (frontal 2/3 view).

with 11.2 g/dl hypochromic microcytic type of inflammatory; the leukocyte count was 6960/mm^3 with 30% eosinophilia (or 2080/mm^3). The renal function tests showed a creatinine 10 mg/l, uremia at 0.43 g/l and 0.42 g proteinuria/24; there was no urinary infection or leucocyturia. Total serum IgE was 642 kUI/l four times more than normal value.

An excisional biopsy was performed. The pathological examination of the excised sample showed a fibrous tissue invaded by a dense follicular lymphoid hyperplasia with clear center separated by dens eosinophilic infiltrates sidling between striated muscle fibers associated with a dense vascular hyperplasia. The combination of vascular and follicular lymphoid hyperplasia with abundant eosinophil raised eosinophilic lymphogranuloma or Kimura disease.

Local recurrence was observed a few months later which had justified corticosteroid based treatment. A mild regression of the lesion was noted after 2 months of treatment. Unfortunately the patient was lost to follow.

3. Discussion

Kimura's disease is a chronic disease of unknown etiology. It is typically manifested by painless subcutaneous nodules in head or neck.

Described for the first time in China by Kimm and Szeto about 7 patients, Kimura and colleagues in 1948 in Japan have made a more detailed description. Since then, about 200 cases have been published to date [3] mostly from the Far East.

Kimura's disease predominantly affects young Asian adult male especially the Japanese and Chinese. A few cases have been published in Western literature [1]. This disease is very rare in the black race. Herrero-Basilio and Co [2] have reported a 23 years young Nigerian case who presented a subcutaneous swelling taking all the left cheek. One case was reported by Dib [4] in North Africa.

The usual clinical presentation is that of a painless swelling in skin sitting in the neck area and head. They are often associated and quasi steadily satellite lymphadenopathy, eosinophilia and elevated serum IgE as was the case in our patient. However it was not of satellite nodes.

Differential diagnosis is difficult to make sometimes [5] mainly with hyperplasia with eosinophilia Angiolymphoid (HALE). Both conditions have long been considered two clinical expressions of the same entity. Current studies have shown that these are two distinct conditions. In tropical region, eosinophilia associated with cutaneous swelling must discuss certain parasites especially onchocerciasis, african human trypanosomiasis and schistosomiasis [6] conventionally, the different lesions observed by their seat, appearance and evolutionary mode.

Angiolymphoid hyperplasia with eosinophilia is a benign vascular proliferation associated with eosinophilic dominant inflammatory infiltrate. It mostly affects middle-aged women between the 3rd and the 4th decade of Caucasian origin. This condition is characterized by nodules or isolated or multiple cutaneous papules or pinkish beige often itchy sometimes painful electively sitting at the head and in the peri-auricular region. Peripheral lymphadenopathy satellite channels and eosinophilia are rarely found in only about 5% of cases [7].

Also given the insidious installation mode of the lesion, its painless and soft character, its chronic course and the absence of constitutional symptoms, benign soft tissue tumor in particular lipoma, cannot be excluded formally to clinic.

Exceptional locations have been described [8]-[11] in particular palate, parotid, orbital and members. Kyung-Jim Song [10] reported the case of Kimura's disease diffuse on arm and Manish [11] reported a femoral localization of the disease.

Renal involvement is not uncommon. Yuen HW and Co [3] found membranous glomerulonephritis in 60% of their series. It is mainly manifested by a significant proteinuria or a full blown nephrotic syndrome and sometimes precedes the disease in some cases [12]. Sze-Kit and Co [13] reported a series of 8 cases of Kimura's disease presented nephrotic syndrome and 6 of them had nephrotic syndrome before establishing Kimura's disease diagnosis. However, the relationship between renal disease and Kimura's disease remains controversial. Chauvel [14] meanwhile found a case of necrotizing extracapillary glomerulonephritis.

The etiology of Kimura's disease remains unclear. Several hypotheses have been raised. The most frequently proposed hypothesis is that of an aberrant delayed hypersensitivity reaction to prolonged autoimmune or infectious (fungus or a parasitic) antigenic stimulation [15]. According to another hypothesis, it would be a Th2 type allergic reaction leading to eosinophilia and elevated IgE. Mast cells play a central role not only through IgE

synthesis but also in promoting the migration of eosinophils into the lesion.

Treatment options range from a surgical excision to radiotherapy and immunosuppressive agents and corticosteroids. Spontaneous remissions may be observed. However, the excision of the mass remains the treatment of choice especially in young [11] although local recurrences are frequent as is the case in our patient. Systemic corticosteroids may be indicated in case of relapse or in patients with renal impairment. Cetirizine has achieved a complete remission in a steroid-dependent patient [16].

Our patient had received in the first time of a local resection and given tumor recurrence after 6 months, a corticosteroid due to 1 mg/kg/day had been undertaken which allowed a moderate regression of the lesion after 2 months of treatment. Local radiotherapy should be reserved for non-operable lesions, refractory recurrent, medical treatment and surgical treatment.

4. Conclusion

This observation proves the reality of Kimura disease in Africa especially among the black Africans. Although it is very rare, nevertheless it shows the universal distribution of this disease. Because of the painless nature and chronic evolution of the lesion which may last several years but also the absence of malignancy, diagnosis is exceptionally mentioned in our context given the predominance of infectious etiology. Yet should we think about it in order to guide the pathologist.

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